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# **Place de la recherche qualitative dans l'évaluation des interventions non-médicamenteuses. : Exemple dans le champ de l'autisme**

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**Place de la recherche qualitative dans  
l'évaluation des interventions non-  
médicamenteuses. Exemple dans le champ  
de l'autisme.**

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## RESUME ET MOTS-CLES

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**Place de la recherche qualitative dans l'évaluation des interventions non-médicamenteuses.**

**Exemple dans le champ de l'autisme.**

L'autisme est un trouble neurodéveloppemental caractérisé par des altérations dans la communication, les interactions sociales et les comportements. Bien qu'aucun traitement médicamenteux spécifique ne soit actuellement disponible, un large éventail d'interventions non médicamenteuses a été développé pour répondre aux besoins des personnes autistes. Parmi ces interventions, l'intervention précoce est aujourd'hui un modèle plébiscité, rendu possible par l'amélioration du dépistage et du diagnostic dans la petite enfance.

Bien que la qualité des études dans le domaine ait largement évolué ces dernières années, des lacunes méthodologiques persistent. Celles-ci sont en partie expliquées par la difficulté à évaluer ces interventions non médicamenteuses, souvent complexes. Par ailleurs, la littérature s'est particulièrement concentrée sur l'évaluation de l'efficacité de l'intervention, négligeant d'autres aspects importants tels que la faisabilité, l'acceptabilité et l'implémentation. Dans ce contexte, la recherche qualitative, ainsi que les méthodes associant des approches quantitatives et qualitatives (recherche avec une méthode dite « mixte »), ont un rôle à jouer en permettant une compréhension approfondie des expériences, du contexte des interventions, des facteurs en lien avec l'implémentation....

Cette thèse vise à explorer la place de la recherche qualitative dans le domaine de l'évaluation d'interventions non médicamenteuses complexes, au travers de quatre exemples d'étude autour de l'intervention médiée par les parents dans le champ de l'autisme, et plus spécifiquement le modèle du PACT (Pre-School Autism Communication Therapy).

Après une introduction sur l'évaluation des interventions complexes et sur les méthodes qualitatives, une première étude évaluant la faisabilité de la méthode PACT en visioconférence nous permettra d'illustrer la place de la recherche qualitative pour l'évaluation de la faisabilité. Une deuxième étude, explorant l'implémentation du PACT au niveau mondial via une étude à méthodes mixtes, nous permettra de discuter de l'intérêt de ces méthodes dans le champ des sciences de l'implémentation. Une troisième étude, une métasynthèse sur l'expérience des parents ayant participé à une intervention médiée par les parents, nous permettra d'explorer

le domaine de la synthèse des évidences qualitatives et son intérêt dans le champ de l'évaluation des interventions. Enfin, une quatrième étude (en cours, dont le protocole a été publié) contrôlée randomisée multicentrique visant à évaluer l'efficacité du PACT en visioconférence et l'impact de cette méthode sur la famille, nous permettra d'aborder la complexité de l'évaluation d'une telle intervention et les méthodes utiles dans ce champ.

Ces différentes méthodes seront discutées et mises en perspectives, en utilisant le modèle global du PACT et les différentes études ayant permis, à ce jour, d'évaluer le modèle. Nous ouvrirons sur les perspectives méthodologiques de l'évaluation des interventions non médicamenteuses dans le champ de l'autisme.

**Mots-clés :**

Autisme ; interventions non médicamenteuses ; recherche qualitative ; méthodes mixtes ; évaluation

**Keywords:**

Autism; non-pharmacological intervention; qualitative research; mixed methods; evaluation.

**Laboratoire :**

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## ABSTRACT AND KEY WORDS

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### **The Role of Qualitative Research in Evaluating Non-Pharmacological Interventions: An Example in the Autism Field.**

Autism is a neurodevelopmental disorder characterized by alterations in communication, social interactions, and behaviors. Although no specific medication treatment is currently available, a wide range of non-pharmacological interventions has been developed to address the complex needs of individuals with autism. Among these interventions, early intervention is now a widely favored model, made possible by improved early detection and diagnosis.

However, evaluating these interventions proves to be complex. Despite significant improvements in the quality of studies in the field in recent years, methodological gaps persist. These gaps are partly explained by the challenge of assessing these so-called complex interventions. Additionally, the literature has primarily focused on evaluating the effectiveness of the intervention, neglecting other crucial aspects such as feasibility, acceptability, and implementation. In this context, qualitative research, as well as mixed quantitative-qualitative methods, have a role to play in providing an in-depth understanding of experiences, intervention contexts, and factors related to implementation.

This thesis aims to explore the role of qualitative research in the evaluation of complex interventions through four case studies centered around parent-mediated interventions in the field of autism, specifically focusing on the Pre-School Autism Communication Therapy (PACT) model.

Following an introduction to the evaluation of complex interventions and qualitative methods, the first study assessing the feasibility of the PACT method via video conferencing will illustrate the role of qualitative research in assessing feasibility. A second study, exploring the global implementation of PACT through a mixed-methods study, will discuss the significance of these methods in the field of implementation science. A third study, a meta-synthesis on the experiences of parents participating in parent-mediated interventions will allow us to delve into the realm of synthesizing qualitative evidence and its relevance in the field of intervention evaluation. Finally, a fourth study outlining the protocol for a multicenter randomized controlled study aiming to evaluate the effectiveness of PACT via video conferencing and its

impact on families, will address the complexity of evaluating such an intervention and the relevant methods in this field.

These different methods will be discussed and contextualized using the overarching PACT model and the various studies that have, to date, evaluated the model. We will also explore methodological perspectives for evaluating non-pharmacological interventions in the autism field.

**Keywords:**

Autism; non-pharmacological interventions; qualitative research; mixed methods; evaluation.

## PUBLICATIONS ET PARTICIPATION A DES CONFERENCES

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### 1. LISTE DES PUBLICATIONS EN LIEN AVEC LA THESE

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#### ARTICLES PUBLIÉS :

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**Jurek, L;** Leadbitter, K; Falissard, B; Colin, C; Touzet, S; Geoffray, M. Parental experience of parent-mediated intervention for children with ASD. A systematic review and qualitative evidence synthesis. *Autism*, juillet 2022. doi: 10.1177/13623613221112204

**Jurek L,** Ocelli P, Denis A, Amestoy A, Maffre T, Dauchez T, Oreve MJ, Baghdadli A, Schröder C, Jay A, Zelmar A, Revah-Levy A, Gallifet N, Aldred C, Green J, Garg S, Touzet S, Geoffray MM, the IFPAD study group. *Efficacy of Parent- mediated communication-focused treatment in toddlers with autism (PACT) delivered via videoconferencing: a randomized controlled trial study protocol.* *BMJ Open.* 2021;11:e044669. doi:10.1136/ bmjopen-2020-044669

#### ARTICLES SOUMIS :

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Balanovska, M; Leadbitter, K; **Jurek, L;** Mengarelli, F ; Falissard, B; NGAN, N ; Aldred , C ; Green, J; Geoffray, MM. Parent and professional perspectives on implementing the Paediatric Autism Communication Therapy (PACT): a mixed-methods analysis. En cours de reviewing par le journal *Autism*.

Geoffray MM, Bourgeois-Mollier M ; Maleysson-Baste M ; Gallifet N ; Dochez S ; Bonis G ; Jay A ; **Jurek L** . *Feasibility of a videoconferencing-based parent-mediated intervention: a mixed-method pilot study.* Soumis au journal *Autism* en septembre 2023.

### 2. LISTE DES CONFERENCES EN LIEN AVEC LA THESE

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#### PRESENTATION AFFICHEE SOUS FORME DE POSTER :

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Parental experience of parent-mediated intervention for children with ASD. A systematic review and qualitative evidence synthesis. Jurek, L et al.

- Congrès français de psychiatrie, Montpellier, décembre 2021, sélectionné pour une présentation orale dans le cadre des dix meilleurs posters

- Congrès du GIS Autisme, Lyon, mars 2022
- 1<sup>er</sup> congrès international du GNCRA, Lyon, mai 2022
- Congrès européen de psychiatrie de l'enfant et de l'adolescent, ESCAP, Copenhague 2023

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PRESENTATION ORALE :

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Journée de l'EDISS. Présentation de la méta-synthèse. Prix de la meilleure communication.

Congrès européen de psychiatrie de l'enfant et de l'adolescent, ESCAP, Copenhague 2023, présentation de l'étude pilote et du protocole de l'étude IFPAD dans le cadre du symposium « Working with parents and carers to help autism development: the Paediatric Autism Communication Therapy (PACT) intervention. »

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# PREAMBULE

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## 1. CONTEXTE DU TRAVAIL

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L'autisme, un trouble neurodéveloppemental caractérisé par des altérations dans la communication, les interactions sociales et les comportements, a suscité un intérêt croissant de la part des chercheurs, des cliniciens et des familles ces dernières décennies. Si à ce jour aucun traitement médicamenteux spécifique n'est disponible, un large éventail d'interventions non médicamenteuses s'est développé. Ces interventions, ciblant le développement cognitif, social et émotionnel, peuvent être considérées comme des interventions complexes qui intègrent plusieurs composantes interdépendantes mais agissant de façon synergique pour répondre aux besoins complexes des personnes autistes.

Cependant, l'évaluation de ces interventions n'est pas aisée, et bien qu'une évolution notable soit observée sur le plan de la qualité des études d'évaluation disponibles aujourd'hui, de nombreuses lacunes persistent et aucune méthode ne permet de capter l'ensemble des réponses et/ou difficultés qu'entraînent ces interventions. De plus l'évaluation des interventions, souvent réduite à l'évaluation de l'efficacité de l'intervention, est également un champ vaste, nécessitant en réalité une évaluation graduelle, multimodale, avec des objectifs d'efficacité mais également de faisabilité, d'acceptabilité ou d'implémentation.

Dans ce contexte, la recherche qualitative, et par extension les méthodes mixtes quantitatives-qualitatives, se révèle être un outil essentiel pour appréhender la richesse et la complexité de ces interventions, en permettant une compréhension approfondie des expériences, des perceptions et des résultats des individus impliqués. Cette thèse explore la place cruciale de la recherche qualitative dans l'évaluation des interventions non médicamenteuses, en mettant en lumière la manière dont elle contribue à la compréhension et à l'optimisation de ces approches.

Les réflexions présentées dans ce travail ont pris leur origine lorsque l'étude IFPAD (Impact de la Formation des Parents à Distance), que nous aborderons ultérieurement, a été mise en place au sein du CEDA (Centre d'Évaluation et de Diagnostic de l'Autisme). L'étude IFPAD est une étude contrôlée randomisée visant à évaluer un modèle existant, le PACT (Pre-School Autism

Communication Therapy), délivré par le biais de visioconférences pour permettre aux familles résidant éloignées des centres spécialisés de bénéficier de cette intervention.

Au sein du groupe de travail, de nombreuses questions ont alors émergé : Comment pouvait-on efficacement transformer cette intervention en présentiel en une intervention à distance ? Du point de vue technique, quelles mesures anticipatoires étaient nécessaires pour garantir que toutes les familles puissent bénéficier de cette intervention en visioconférence ? Dans un contexte plus général d'intervention médiée par les parents, comment les parents percevaient-ils cette intervention, et existait-il des ajustements préalables à envisager ? Rencontrait-on des obstacles à sa mise en œuvre dans la vie quotidienne ?

Ces interrogations, bien qu'elles ne fussent pas les objectifs principaux de l'étude IFPAD, ont été jugées essentielles à traiter. Les essais cliniques randomisés (ECR) sont souvent longs et demandent des ressources considérables, ce qui rend impératif une planification minutieuse de leur mise en œuvre et garantit que leurs résultats puissent être appliqués dans la pratique clinique. Par ailleurs, les ECR ne permettent de répondre qu'à une question principale, principalement celle de l'efficacité.

Afin de répondre à ces questions supplémentaires découlant de l'étude IFPAD, nous avons eu recours à la recherche qualitative pour obtenir des informations sur les éléments critiques affectant la faisabilité de l'intervention basée sur la visioconférence et pour décrire les obstacles et les facilitateurs du point de vue des parents. Au moyen de quatre études distinctes, nous tenterons d'apporter des réponses à ces questions tout en réfléchissant au rôle de la recherche qualitative à différentes étapes de l'investigation.

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## PLAN DE LA THESE

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Après une introduction sur l'autisme et les interventions non médicamenteuses disponibles, un premier chapitre visera à explorer le champ de l'évaluation des interventions dites complexes.

Nous définirons dans un second chapitre, la recherche qualitative et ses méthodes.

Le troisième chapitre permettra d'explorer la place de la recherche qualitative dans le champ de l'évaluation des interventions complexes, à l'aide de quatre exemples autour des interventions médiées par les parents et notamment l'intervention PACT.

Enfin, une discussion générale sera proposée ainsi que des perspectives pour l'avenir de la recherche dans le champ des interventions dans l'autisme.

## 2. REMARQUE SUR LA TERMINOLOGIE UTILISEE

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### CONCERNANT L'AUTISME

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Au cours des deux décennies récentes, il y a eu un accroissement des débats autour des terminologies adéquates à employer pour décrire les troubles du spectre autistique (TSA, ci-après désigné sous le terme "autisme"). Ces échanges ont principalement engagé des défenseurs autistes ainsi que des chercheurs universitaires. Le débat s'est concentré sur la pertinence d'un langage orienté vers la personne (tel que "individu touché par l'autisme") versus un langage orienté vers l'identité (c'est-à-dire "personne autiste") (Keating et al., 2023; Kenny et al., 2016).

Le langage orienté vers la personne, fréquemment utilisé dans les contextes cliniques et de recherche, a pour intention de mettre l'accent sur l'individu plutôt que sur sa condition, en établissant une distinction nette. Néanmoins, de nombreux individus argumentent que ce type de langage, en mettant en avant cette distinction, involontairement renforce la stigmatisation. Ce faisant, il continue de véhiculer l'idée que l'autisme est une "déficience" qui doit être dissociée de l'individu (et implicitement suggère que le handicap est intrinsèquement négatif).

En contraste, le langage orienté vers l'identité vise à reconnaître l'autisme comme un élément fondamental de l'identité d'une personne, tout comme son genre ou son origine ethnique. Puisque beaucoup estiment qu'il est inapproprié et contre-productif de dissocier une personne autiste de son autisme, de nombreux militants optent pour un langage orienté vers l'identité (par exemple, "personne autiste").

Une récente étude, réalisée sur des populations anglophones, ont mis en évidence une préférence des termes « autisme » ou « de trouble du spectre de l'autisme » pour évoquer le diagnostic (Keating et al., 2023). Pour parler des individus, l'étude met en évidence une préférence pour le terme « personne autiste », suivi de « personne neurodivergente » ou « autiste ». Les terme « handicap » est préféré aux termes « trouble » ou « maladie ». Enfin, pour évoquer les habilités, les termes « challenges », « difficultés » et « différences » sont préférées aux termes « déficit » ou « déficience ».

La terminologie utilisée dans ce manuscrit, suivra les résultats de ces études.

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## CONCERNANT LA METHODOLOGIE

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De nombreux termes utilisés dans ce manuscrit sont des termes anglophones, n'ayant pas toujours de traduction satisfaisante en français. Afin d'éviter une perte de sens en traduisant abusivement ces notions, nous les utiliserons dans leur forme anglaise, accompagné d'une description en français, entre parenthèse.

### 3. LE TROUBLE DU SPECTRE DE L'AUTISME CHEZ L'ENFANT

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La description des principales caractéristiques de l'autisme (ou trouble du spectre de l'autisme), à savoir un déficit de communication sociale et des comportements sensori-moteurs répétitifs et inhabituels, n'a pas beaucoup changé depuis sa définition initiale par Leo Kanner en 1943 (J. Harris, 2018). Toutefois, l'autisme est désormais considéré comme un spectre avec des manifestations légères à sévères dans les deux domaines évoqués ci-dessus (Lord et al., 2018). Il est intégré, dans les classifications diagnostiques, aux troubles du neurodéveloppement et il n'est pas rare que d'autres troubles du neurodéveloppement (tels que la déficience intellectuelle, les troubles de la coordination motrice, ou des apprentissages) soient co-occurent du diagnostic d'autisme (Lai et al., 2019).

L'autisme est causé par une combinaison de nombreuses variantes génétiques rares et communes, dont certaines peuvent être associées à d'autres troubles neurodéveloppementaux ou psychiatriques, et la plupart des personnes atteintes d'autisme n'ont pas encore de profil génétique connu (Bai et al., 2019). Il n'existe par ailleurs pas encore de biomarqueurs diagnostiques fiables et le diagnostic doit être posé sur la base du comportement (Lord et al., 2022).

Sur le plan médical, le diagnostic de « trouble du spectre de l'autisme » est établi selon les critères du Manuel diagnostique et statistique des troubles mentaux (DSM)-5 de l'American Psychiatric Association, publiés en 2013 (American Psychiatric Association, 2015). Les critères sont décrits dans le **Tableau 1**.

**TABLEAU 1. CRITERES DSM-5 DU TROUBLE DU SPECTRE DE L'AUTISME.**

Déficit persistant dans la communication sociale et l'interaction sociale dans de multiples contextes, se manifestant par

- Un déficit dans la réciprocité socio-émotionnelle
- Un déficit dans les comportements de communication non verbale
- Un déficit dans le développement, le maintien et la compréhension des relations

La caractéristique restreinte et répétitive des comportements et intérêts avec :

- Des mouvements répétitifs ou stéréotypés
- Une intolérance au changement ou une adhésion inflexible à des routines ou modèles ritualisés de comportement verbal et non verbal
- Des intérêts restreints ou fixes, anormaux dans leur intensité ou leur but
- Hyperréactivité ou hyporéactivité aux stimuli sensoriels, ou intérêts inhabituels pour les aspects sensoriels de l'environnement

Les symptômes doivent être présents au début de la période de développement (mais peuvent ne pas se manifester pleinement jusqu'à ce que les exigences sociales dépassent les capacités limitées, ou peuvent être masqués par des stratégies apprises).

Pour évoquer le diagnostic, l'individu doit présenter des déficits de communication sociale (passés ou présents) dans chacun des trois domaines définis ci-dessus et deux des quatre éléments restreints ou répétitifs (passés ou présents).

Les symptômes doivent entraîner une déficience cliniquement significative dans le fonctionnement social, professionnel ou dans d'autres domaines du fonctionnement de l'individu. Le niveau de sévérité est décomposé en trois niveaux : nécessité de soutien, nécessité de soutien important ou nécessité de soutien très important.

Selon une récente méta-analyse d'études de prévalence au niveau mondial, la prévalence de l'autisme serait estimée à 119,25 sur 10000 (soit environ 1,2%) (Zeidan et al., 2022). En France, les dernières estimations de santé publique France, évoquent une prévalence de 22,4 sur 10 000 chez les enfants de 4 ans et moins et de 72,2 sur 10000 chez les enfants de 5 à 9 ans en 2017 (Santé Publique France, 2020). Il s'agit d'un trouble fréquent, avec un impact économique

significatif liés à l'éducation spécialisée, aux soins de santé, et à l'accompagnement continu tout au long de la vie (Rogge & Janssen, 2019).

Bien que le diagnostic soit possible et assez stable dès la deuxième année de vie, le diagnostic précoce de l'autisme est récent. De façon historique, l'autisme était rarement diagnostiqué avant 3 ans, et l'âge moyen au diagnostic variait de 3 à 10 ans en fonction de facteurs tels que la gravité des symptômes, le niveau de langage, le sexe, mais également le statut socio-économique (Fletcher-Watson et al., 2017; Solmi et al., 2022).

De nombreuses études ont mis en évidence des inquiétudes parentales bien plus précoces dans le développement, généralement avant l'âge de 24 mois, avec un repérage des déficits et des retards dans la communication non verbale, l'intentionnalité communicative, l'attention conjointe et l'orientation vers les signaux sociaux (Young et al., 2003). Ces difficultés sont généralement persistantes, hautement prédictives des résultats sociaux et éducatifs futurs et fortement associées à la sévérité des symptômes comportementaux (Mundy et al., 1986; Wetherby et al., 1998). La mise en évidence de ces signes précoces, a permis d'entraîner une réorganisation du repérage et du diagnostic précoce de l'autisme offrant aujourd'hui la possibilité d'intervention très précoces auprès de ces enfants (Charman & Baird, 2002).

#### 4. INTERVENTION PRECOCE

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##### INTERET DE L'INTERVENTION PRECOCE

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La notion d'intervention précoce découle de l'hypothèse selon laquelle, en raison de la plasticité du développement cérébral, l'enrichissement de l'environnement par des interventions peut exercer une influence importante sur le comportement et le neurodéveloppement au fil du temps (Lord et al., 2022). Certaines cibles d'intervention précoce, permettraient grâce à un effet en cascade, de modifier la trajectoire développementale et de permettre à l'enfant d'acquérir des compétences pivots, nécessaires à son développement cognitif et social, et prévenir les difficultés liées à l'autisme avant qu'elles ne se manifestent pleinement (Dawson, 2008; Lord et al., 2022; Zwaigenbaum et al., 2015).

En outre, la recherche a montré que différents aspects du développement émergent et pourraient peut-être être plus facilement modifiés à différents moments. La deuxième année de vie est une période de développement particulièrement critique pour les enfants atteints

de TSA. Il s'agit une période dynamique de croissance du cerveau, au cours de laquelle les augmentations du volume cérébral et la connectivité atypique associée aux TSA apparaissent pour la première fois (Courchesne et al., 2011), mais c'est aussi une période de plasticité neuronale importante qui offre un plus grand potentiel pour modifier le cours du développement (Dawson, 2008).

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## MODELES D'INTERVENTION PRECOCE

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De nombreux modèles d'intervention précoce ont été évalués et sont sous-tendus par différentes orientations théoriques.

De façon historique, les modèles comportementaux, basés sur théories opérantes de l'apprentissage et sur l'analyse appliquée du comportement (ABA en anglais, Applied Behavioral Analysis), ont été les premiers modèles utilisés dans ce champ. Ces interventions, réalisées en centre ou à domicile, sont principalement délivrées par un thérapeute et ont pour but de diminuer les comportements problématiques ou de cibler des apprentissages précis (Leaf et al., 2021). Plusieurs modèles tels que les interventions précoces comportementales intensives (EIBI en anglais, Early Intensive Behavioral Intervention) (Brignell et al., 2018) ou le PECS (Bondy & Frost, 2001) font partie de cet ensemble.

Certains modèles sont basés sur un cadre éducatif, comme le projet TEACCH (Treatment and Education of Autistic and related Communication handicapped CHildren), qui met l'accent sur la structuration des environnements de classe par le biais de repères visuels, de routines de communication et de tâches individuelles (Mesibov & Shea, 2010). Le projet TEACCH vise à accroître l'indépendance des enfants et est conçu pour travailler sur leurs forces existantes plutôt que de se concentrer sur leurs faiblesses.

Au cours des dernières décennies l'intervention précoce est devenue plus naturaliste et plus adaptée au développement avec une plus grande importance accordée à l'enfant en tant que partenaire actif dans la communication (Lord et al., 2022). Ces interventions, basées sur les théories constructivistes de l'apprentissage, mettent l'accent sur la création d'opportunités de communication naturelle, l'amélioration de la réciprocité entre les partenaires de communication, le renforcement de la motivation des enfants pour l'interaction sociale et l'incitation à des comportements sociaux spécifiques, en combinant des techniques comportementales et développementales (Schreibman et al., 2015).

Ces interventions visent faciliter le développement de compétences fondamentales, ou pivot (par exemple, l'attention conjointe et l'imitation), qui ont des effets développementaux en cascade sur différents domaines du développement (Sandbank et al., 2020; Schreibman et al., 2015).

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## L'INTERVENTION MEDIÉE PAR LES PARENTS

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L'implication des parents dans la mise en place de stratégies d'intervention visant à aider leurs enfants atteints d'autisme remonte aux années 1970. Depuis lors, de multiples études ont évalué diverses approches de formation parentale pour résoudre des problèmes comportementaux, améliorer les interactions entre parents et enfants, ainsi que pour faciliter la communication (Oono et al., 2013). Impliquer les parents dans l'intervention permettrait une diffusion de l'intervention dans le quotidien de l'enfant, avec l'idée d'une généralisation des compétences.

La justification de cibler l'interaction sociale précoce parent-enfant (ICP) dans le contexte des interventions médiées par les parents dans l'autisme découle d'une recherche s'étendant sur des décennies sur le développement de l'enfant, mettant en avant l'importance capitale d'interactions sociales positives et dyadiques précoces pour la communication et l'adaptation sociale ultérieure de l'enfant. La recherche a pu, par exemple, mettre en évidence que lorsque les parents interagissent de manière "synchrone" avec l'attention et les intentions de leur enfant, cela peut se traduire par des améliorations en termes de langage et de communication (Siller & Sigman, 2008).

Se basant sur un modèle transactionnel de l'interaction précoce réciproque, entre les particularités développementales inhérentes à l'autisme et l'environnement social précoce, il est suggéré que les perturbations dans ces interactions pourraient potentiellement amplifier les altérations de l'environnement social et l'impact des symptômes au fil du temps (Sameroff, 2009). Dans l'autisme, les signaux de communication de l'enfant peuvent être peu perceptibles, peu fréquents ou mal synchronisés, ce qui augmente les initiatives de l'adulte pour réorienter l'attention de l'enfant. La communication "asynchrone" qui en résulte peut entraîner une réduction des possibilités d'apprentissage du langage. Le risque étant d'entraîner par la suite un style de discours parental de plus en plus directif qui, bien qu'il ne soit pas incompatible avec une sensibilité, peut entraîner une diminution de la communication réciproque. Les

réponses parentales positives, contingentes et sensibles semblent être essentielles au développement de l'attention conjointe, et du langage dans un second temps (Green et al., 2010; Green & Garg, 2018).

Former les parents en tant que "thérapeutes" permet d'instaurer une intervention précocement, avec pour objectif d'améliorer les stratégies d'interaction parent-enfant qui contribuent à renforcer les premières relations sociales de l'enfant. L'intervention médiée par les parents ciblant les interactions précoces, présente l'avantage théorique d'être considéré comme « naturaliste » (c'est-à-dire qu'elle opère au sein du cadre de développement quotidien de l'enfant) et potentiellement plus aisément généralisable que les interventions cliniques directes auprès de l'enfant (Green & Garg, 2018).

Le PACT (Preschool Autism Communication Therapy), que nous développerons dans la deuxième partie, découle de cette théorie (Aldred et al., 2004).

Si les aspects théoriques sont particulièrement importants dans l'élaboration d'intervention actualisée pour les jeunes enfants autistes, leur évaluation par des méthodes structurées et standardisées l'est également.

# PARTIE 1 : L'ÉVALUATION DES INTERVENTIONS NON-MÉDICAMENTEUSES DANS L'AUTISME

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## 1. PLACE DES ÉTUDES CONTRÔLÉE RANDOMISÉE

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L'évaluation de l'efficacité des interventions dans le champ de l'autisme a évolué sur ces dernières décennies. Initialement menée sous forme de séries de cas ou d'étude mono-bras, évaluant notamment les interventions comportementales, les études ont progressivement utilisé des méthodes considérées comme plus robustes, le modèle des études contrôlées randomisées (ECR) (Lord et al., 2022).

Les ECR ont de nombreux avantages méthodologiques dans l'évaluation de l'efficacité des médicaments, interventions non-médicamenteuses ou encore politique de santé, et sont considérées comme le « gold-standard », à condition de respecter certains critères de qualité (Maughan, 2013). Dans le champ de l'évaluation des interventions dans l'autisme, si l'ECR est un modèle aujourd'hui plus utilisé, la qualité des études restent en dessous des standards méthodologiques et de nombreuses études ne respectent pas les principes fondamentaux des ECR. Les études dans ce champ utilisent des échantillons de petite taille, ne garantissant pas la puissance nécessaire pour les études, et le calcul de nombre de sujet nécessaire se base parfois sur des études n'ayant pas la puissance statistique nécessaire (French & Kennedy, 2018; Green & Garg, 2018). L'utilisation d'ECR s'avère parfois difficile, du fait par exemple de la durée des interventions, qui peuvent compromettre le maintien d'un groupe contrôle indemne de l'intervention sur une longue durée ou le maintien de l'aveugle pour les évaluations (T. Smith et al., 2007).

L'ECR implique par ailleurs l'utilisation d'outils d'évaluation fiables pour mesurer l'effet sur un critère principal, cependant, dans le champ de l'autisme les outils d'évaluation de l'efficacité font actuellement débat (McConachie et al., 2015). Les mesures utilisées dans les ECR sont très variées et peuvent rendre difficile la synthèse de la littérature (et notamment la réalisation de méta-analyses) du fait de résultats sur des concepts trop différents pour être synthétisés (Provenzani et al., 2020). Si certains affirment que l'efficacité doit être mesurée sur de la sévérité des symptômes de l'autisme, d'autres soutiennent l'utilisation d'évaluations plus « adaptatives » et plus « globales ». Cette évaluation plus globale, viserait à mieux capter les

bénéfices pour l'individu dans son quotidien, que par l'unique évaluation de la symptomatologie autistique (Green & Garg, 2018).

La plupart des recherches sur les interventions en matière de santé ont jusqu'à présent adopté une perspective d'efficacité ou d'efficience et, pour certaines questions de recherche, ces perspectives continueront d'être les plus appropriées et l'ECR restera un modèle de choix. Cependant la recherche centrée sur l'évaluation de l'efficacité des interventions ne permet pas de répondre à certaines questions tout aussi pertinentes pour les besoins des décideurs. Au-delà de la question de l'efficacité, il est nécessaire de savoir si une intervention peut être applicable, efficiente, transférable et évolutive dans des conditions réelles. Cette évaluation nécessite donc l'utilisation d'une autre approche. Le cadre de l'évaluation des interventions dites « complexe » nous permet d'approfondir cette réflexion.

## 2. L'INTERVENTION NON MEDICAMENTEUSE DANS L'AUTISME, UNE INTERVENTION DITE « COMPLEXE »

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Les interventions non médicamenteuses dans l'autisme, et notamment les interventions développementales médiées par les parents, que nous développons plus spécifiquement dans ce travail, peuvent être considérées comme « complexes » à plusieurs niveaux : il s'agit d'interventions proposant d'associer différents outils thérapeutiques (videofeedback, techniques développementales, comportementales...), qui nécessitent généralement une formation soutenue des professionnels, pour délivrer les « ingrédients » essentiels de l'intervention, mais également des capacités du côté du parent formé (apprentissage, attention, implication). Leurs cibles sont multiples (langage, réciprocité sociale, etc.), et des effets en cascade sur le développement sont souvent attendus. Si ces interventions ne sont pas toujours flexibles dans leur construction, elles s'appuient sur le profil des parents pour permettre d'avancer dans la thérapie, et sont donc modelées pour chaque famille. Les éléments de complexité spécifiques de l'intervention médiée par les parents PACT seront détaillés dans la partie 3, grâce à l'illustration de l'étude IFPAD.

Selon le Medical Research Council (MRC) Guidance (Craig et al., 2008; Skivington et al., 2021), une intervention peut être considérée comme « complexe » en raison de ses caractéristiques telles que :

- Les nombre de composantes entrant en interaction dans l'intervention évaluée et l'intervention contrôle
- L'expertise et les compétences requises par les personnes qui délivrent ou reçoivent l'intervention
- Le nombre de groupes, d'environnements ou de niveaux organisationnels ciblés
- Les résultats ou mesures sont nombreux ou variables
- Le niveau de flexibilité autorisé de l'intervention ou ses composantes

Les interventions médiées par les parents dans l'autisme semblent répondre à cette définition.

### 3. ÉVALUATION DES INTERVENTIONS DITES COMPLEXES

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De manière classique, le processus allant de la conception à la réalisation d'une intervention non médicamenteuse dite complexe suit un cheminement qui comprend plusieurs étapes organisées de manière hiérarchique (T. Smith et al., 2007; Vivanti et al., 2018).

La première étape consiste généralement à développer une théorie justifiant la nécessité d'une approche spécifique. L'évaluation d'une intervention débute ensuite par une phase de validation du concept, visant à obtenir des éléments préliminaires étayant ladite approche.

La faisabilité, l'acceptabilité et la sécurité de l'approche proposée est ensuite évaluée dans des essais pilotes.

Une fois le modèle validé, l'évaluation se poursuit avec des études d'efficacité plus larges, impliquant un échantillon de taille adéquate pour garantir une évaluation rigoureuse de la capacité de l'intervention à atteindre ses objectifs au sein de la population ciblée. Cette étape utilise principalement des études contrôlées randomisées, dans l'idéal multicentriques.

Les interventions ayant prouvé leur efficacité (*efficacy* en anglais) sont ensuite soumises à des évaluations d'efficience (*effectiveness* en anglais), lesquelles mesurent dans quelle mesure les bénéfices escomptés persistent lorsque l'intervention est mise en œuvre dans un environnement clinique général, qui diffère par son organisation avec les services spécialisés ou universitaire, souvent terrains des études d'efficacité.

Enfin, la science de la l'implémentation évalue les méthodes permettant d'assurer une adoption réussie et une mise en œuvre fructueuse d'un programme ciblé au sein des contextes communautaires pertinents.

L'aspect hiérarchique de ces étapes est aujourd'hui critiqué par plusieurs auteurs (Moore et al., 2015; Stahmer et al., 2017; Vivanti et al., 2018), qui proposent un modèle plus itératif, permettant une discussion entre les différentes étapes. Ce modèle vise principalement à éviter de mener « à tout prix » une étude d'efficacité ou d'implémentation, lorsque les étapes préliminaires montrent un besoin de réaffiner l'intervention, de revoir les mécanismes d'actions prétendus, ou de mieux sélectionner le contexte de son évaluation. L'approche basée sur la personne (Person Based Approach) proposent de compléter ce modèle en intégrant à ces différentes étapes, la participation active des usagers et des praticiens qui apportent l'expertise de leur expérience dans les questions d'acceptabilité, de perception de l'intervention ou de mise en œuvre en pratique (Yardley et al., 2015, 2015). Un schéma récapitulatif est proposé en figure 1.

Le MRC par exemple a proposé un ensemble de guidelines pour l'évaluation des interventions « complexes » (Moore et al., 2015; Skivington et al., 2021). Premièrement, il encourage les concepteurs et les évaluateurs d'interventions à avoir une bonne compréhension théorique de la manière dont l'intervention provoque le changement pour faciliter le transfert de l'intervention dans des contextes ou des populations différents en conservant ses composantes essentielles. Par ailleurs, les auteurs insistent sur l'évaluation de l'efficacité en lien avec le contexte spécifique, c'est-à-dire tous les éléments extérieurs à l'intervention mais qui pourraient avoir une incidence sur le résultat. Les interventions fondées sur la recherche sont souvent perçues comme étant incompatibles avec la pratique clinique existante parce qu'elles ne reflètent pas la réalité des services communautaires qui ont leur propre culture et leur propre contexte (Goering et al., 2008; Lord et al., 2022). L'étude de l'implémentation de l'intervention, est ainsi essentielle, afin de s'assurer, au-delà de l'efficacité, que l'intervention puisse être transférée en pratique courante et intégrée dans les systèmes de soin.

L'utilisation mixte de recherches quantitatives et qualitatives est, de fait, plébiscitée pour l'évaluation de ces différentes approches (Craig et al., 2008; Goering et al., 2008; Skivington et al., 2021).

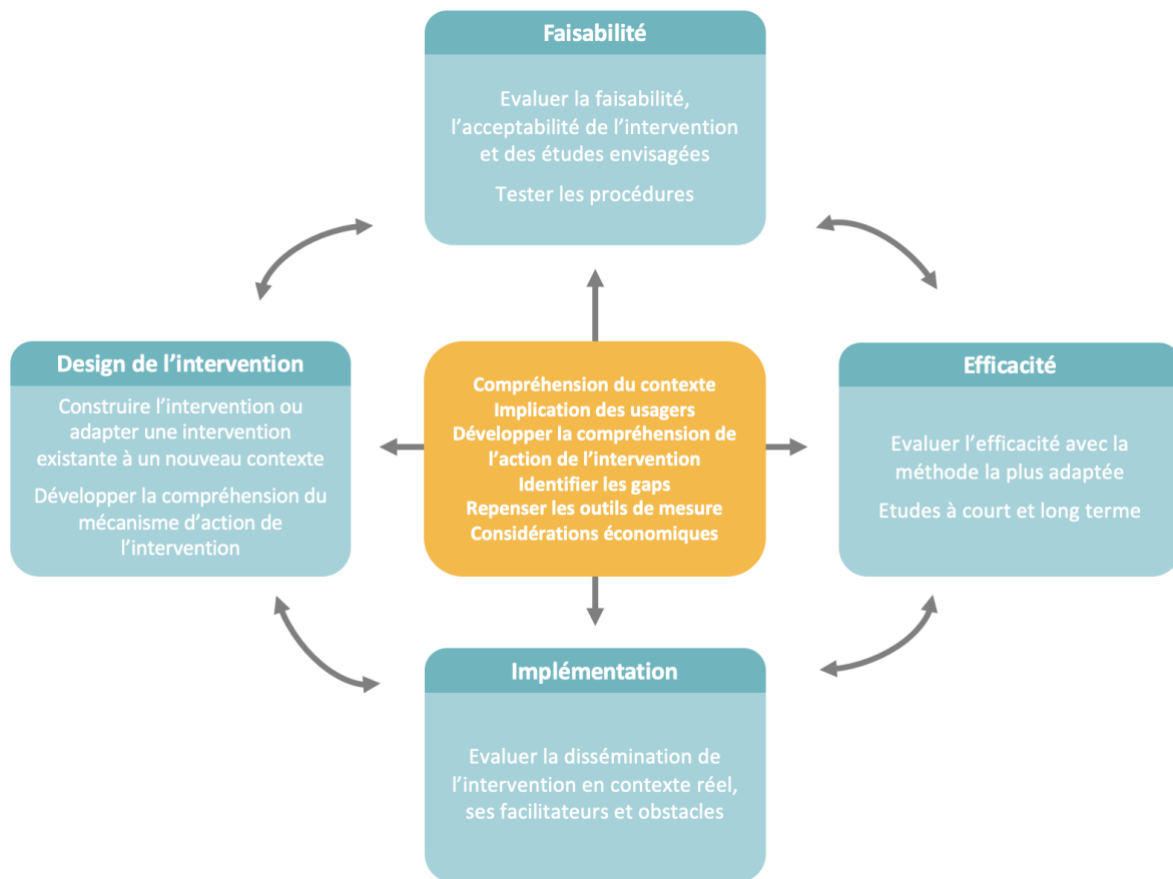


FIGURE 1. PROCESSUS D'EVALUATION D'UNE INTERVENTION COMPLEXE, BASEE SUR LES GUIDELINES MRC (MOORE ET AL. 2015)

## PARTIE 2 : LA RECHERCHE QUALITATIVE, ASPECTS METHODOLOGIQUES

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### 1. DEFINITIONS

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La recherche qualitative est une approche méthodologique fondamentale en sciences sociales et humaines, visant à explorer en profondeur et à comprendre les phénomènes complexes et les questions de recherche à travers une perspective subjective. Souvent définie en opposition à la recherche quantitative, qui s'appuie sur la collecte et l'analyse de données chiffrées, généralement sur de grands échantillons d'individus, la recherche qualitative s'appuie sur des observations de l'enquête et le recueil de verbatims, et accorde la priorité à l'expérience vécue des personnes et aux significations qu'elles attribuent à leurs expériences. Les données sont souvent collectées dans les contextes étudiés et visent à décrire de manière riche des idées ou des processus complexes, bien qu'ils concernent généralement un nombre limité d'individus ou de contextes.

L'objectif de la recherche qualitative va au-delà de la simple description des phénomènes. Elle vise à révéler les motivations, les croyances, les valeurs et les significations profondes qui sous-tendent les comportements humains et les contextes sociaux.

La recherche qualitative a longtemps été rattachée au paradigme constructiviste ou interprétatif (Mays, 2000), reconnaissant que la réalité est construite socialement et interprétée individuellement, en opposition au paradigme positiviste cherchant à établir des lois générales et objectives en se basant sur des méthodes scientifiques quantitatives et des observations mesurables. Si certains auteurs continuent de décrire le besoin d'ancrer les méthodes qualitatives dans un paradigme spécifique (Pernecky, 2016), d'autres proposent de dissocier le paradigme de la méthode. Les méthodes qualitatives pourraient ainsi être considérées comme un outil de recherche, ayant la possibilité de se combiner avec d'autres outils, notamment quantitatifs, sans frontière paradigmatique (Syed & McLean, 2022).

Les méthodes qualitatives ont le plus souvent recours à une démarche inductive, décrite comme opposée à la démarche hypothético-déductive. Aucune hypothèse de recherche n'est ici formulée au préalable. Les hypothèses émergent du matériel collecté, au fur et à mesure de l'analyse des données.

## 2. CADRES THEORIQUES

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La recherche qualitative englobe une variété de méthodes et d'approches, adaptées à différents contextes et objectifs de recherche.

Nous utiliserons les paradigmes de la théorie ancrée, particulièrement adaptée à l'exploration de nouvelles théories émergentes à partir des données, ainsi que le paradigme phénoménologique, qui vise à comprendre l'essence de l'expérience des individus.

La théorie ancrée est une approche méthodologique développée par Glaser et Strauss dans les années 1960 (Glaser & Strauss, 2010). L'objectif principal de la théorie ancrée est de générer des concepts et des théories qui émergent directement des données, plutôt que d'appliquer des cadres théoriques existants.

Concernant la théorie phénoménologique, la phénoménologie est un paradigme philosophique et méthodologique qui se concentre sur l'étude de la conscience humaine et de l'expérience vécue (J. A. Smith et al., 2009). En recherche qualitative, les approches phénoménologiques cherchent à comprendre en profondeur les significations et les essences des expériences vécues par les individus. Cela implique de mettre de côté les préjugés et les présuppositions pour se concentrer sur les expériences telles qu'elles sont ressenties par les participants. Les chercheurs recueillent des données principalement grâce à des entretiens approfondis avec les participants, en leur demandant de décrire leurs expériences et leurs perceptions. L'accent est mis sur la description détaillée des sensations, des émotions et des pensées des participants, dans le but de saisir la signification subjective de leurs expériences.

## 3. METHODES

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### RECUEIL DE DONNEES

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Les méthodes de collecte de données en recherche qualitative comprennent plusieurs types : l'observation, l'entretien, l'entretien de groupe (ou focus group) et l'analyse de documents écrits. En pratique, le choix de la méthode dépend de sa capacité perçue à fournir les informations les plus pertinentes pour répondre à la question de recherche. Plusieurs méthodes de collecte de données peuvent également être combinées pour trianguler les sources de données, un critère de fiabilité de la recherche qualitative, visant à compenser les

avantages et les inconvénients de chaque méthode et à renforcer une interprétation globale (Pope & Mays, 1995).

Les entretiens peuvent être soit libres, ressemblant à des conversations informelles, soit semi-structurés avec une grille de thèmes que l'intervieweur suit en fonction du déroulement de l'entretien. Ces entretiens visent à établir une relation entre l'intervieweur et l'interviewé, positionnant le participant comme expert de son expérience. L'entretien doit être mené de manière à lui permettre de partager son expérience et d'aborder des questions et des thèmes auxquels le chercheur n'aurait pas pensé.

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## DIFFERENTES METHODES D'ANALYSE

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Il existe plusieurs méthodes d'analyse utilisées en recherche qualitative : analyse de contenu, analyse narrative, analyse de discours, analyse de cas, analyse ethnographique, etc... Nous utilisons principalement dans nos travaux l'analyse thématique.

L'analyse thématique est une méthode qualitative qui implique l'identification, la description et l'organisation de thèmes significatifs à partir des données (Guest et al., 2012). Les chercheurs extraient des significations et des patterns récurrents des données pour développer des thèmes qui capturent l'essence des informations recueillies. Les thèmes émergent de manière inductive à partir des données elles-mêmes, et ils sont souvent accompagnés de citations ou d'exemples pour illustrer chaque thème. L'objectif est de créer une compréhension profonde des significations des données brutes.

Plusieurs grandes étapes sont suivies pour réaliser l'analyse thématiques (Guest et al., 2012; Roberts et al., 2019) :

- Familiarisation avec les données : les entretiens, préalablement retranscrits, sont lus attentivement par le chercheur afin de se familiariser avec le contenu et d'acquérir une compréhension globale des informations recueillies.
- Génération des codes : le chercheur identifie des unités de sens dans les données, aussi appelées "unités de codage" qui sont alors résumées à l'aide d'une « étiquette », d'un « code » qui résume le contenu d'une unité de sens spécifique. Ces codes sont générés pour les concepts, les idées, les phrases clés qui se répètent ou qui sont significatifs.
- Regroupement des codes en thèmes : les codes similaires sont regroupés pour former des thèmes. Les thèmes sont des concepts plus larges qui capturent les idées centrales

émergeant des données. Chaque thème doit avoir une signification pertinente pour la question de recherche.

- Développement des thèmes : les thèmes sont développés en sous-thèmes afin de décrire les différentes nuances et facettes du contenu associé à chaque thème. Ce processus est réalisé de façon itérative et les thèmes et sous-thèmes sont comparés aux données originales afin de s'assurer que les thèmes reflètent fidèlement le contenu et les expériences des participants. Les thèmes sont ensuite illustrés par des exemples, issus des entretiens.

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## ÉCHANTILLONNAGE EN RECHERCHE QUALITATIVE

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On ne détermine pas la puissance d'une étude qualitative par la taille de son échantillon mais selon le degré de nouveauté de l'information qu'elle révèle. Contrairement à l'échantillonnage aléatoire fréquemment utilisé dans la recherche quantitative, visant à généraliser les résultats à une population plus large, l'objectif ici est plutôt de comprendre en profondeur les phénomènes et les processus sociaux à travers des cas spécifiques et représentatifs de l'expérience évaluée. Les stratégies de recrutement sont variées : par convenance, par snowballing, raisonnée (purposive sampling) avec variation maximum, raisonnée avec recherche d'homogénéité.... (Patton, 2015; Pope & Mays, 1995)

Le purposive sampling est une technique de recrutement sélective qui sélectionne délibérément des participants ou des sources de données qui sont les plus aptes à fournir des informations pertinentes par rapport aux objectifs de recherche (Patton, 2015). On va donc s'adresser à des personnes pour qui la question de recherche est porteuse de sens, on va choisir à dessein les participants, sélectionnés selon la richesse annoncée du matériel qu'ils peuvent apporter à l'étude du phénomène.

La détermination de la taille de l'échantillon n'est pas prédéfinie. En effet, l'analyse se déroule simultanément avec la collecte des données, et l'inclusion des participants se poursuit tant que l'analyse du matériel continue de fournir des perspectives nouvelles et pertinentes pour l'exploration du phénomène sous enquête. La détermination de la taille de l'échantillon émane de l'atteinte d'une saturation des données, qui se manifeste lorsque l'analyse de nouvelles données atteint un point où la récurrence est évidente et où l'apport d'informations nouvelles est limité (Guest et al., 2020). D'autres auteurs utilisent le concept de « suffisance théorique »

(Saunders et al., 2018) où le recueil de données et l'analyse s'achèvent lorsque les chercheurs jugent que les thèmes obtenus offrent un cadre explicatif suffisant au vu des données collectées.

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## CRITERES DE QUALITE

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S'il est essentiel que la recherche qualitative se soumette à des critères de qualité, les critères de validité des études quantitatives (reproductibilité, validité interne, validité externe, ...) sont difficilement applicables à ce type de recherche. Plusieurs composantes sont attendues pour assurer la rigueur d'une étude qualitative, chacune d'entre elle nécessitant un effort de jugement de la part du chercheur et du lecteur.

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## TRIANGULATION

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La triangulation dans la recherche est l'utilisation de plus d'une approche pour étudier un phénomène. L'objectif est d'accroître la confiance dans les résultats en confirmant une proposition à l'aide d'au moins deux mesures indépendantes (Heale & Forbes, 2013; on behalf of the GRACE INTRO/CHAMP consortium et al., 2015). Quatre types de triangulation ont été identifiés dans la littérature : (1) méthodologique, utilisation de plus d'une méthode de recherche ou de plus d'une technique de collecte de données ; (2) données, utilisation de plusieurs sources de données ; (3) théorique, utilisation de plusieurs théories ; et (4) chercheurs, utilisation de deux chercheurs ou plus pour la réalisation de l'analyse. Le type de triangulation utilisée dans un projet doit refléter la question de recherche individuelle. Le chercheur recherche des modèles de convergence pour développer ou corroborer une interprétation globale. Par ailleurs, s'il existe une faiblesse dans un type de recueil, les autres peuvent la compenser. Il s'agit d'un moyen de garantir l'exhaustivité et d'encourager une analyse plus réfléchie des données. La notion de « triangulation » est au centre des méthodes de recherche mixtes, que nous développerons dans la 3<sup>ème</sup> partie.

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## VALIDATION DES RESULTATS PAR LES REpondants

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Dans un but de réduction des erreurs et de vérification de la crédibilité des résultats, le compte-rendu de la recherche est proposé aux participants de l'étude afin de recueillir leurs réactions et de leur permettre de comparer leur expérience, aux résultats globaux de l'étude. Leurs retours sont intégrés dans les résultats. Le fait que le sujet se reconnaisse dans la description du phénomène est un critère de crédibilité.

## EXPOSITION CLAIRE DES METHODES DE COLLECTE ET D'ANALYSE DES DONNEES

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Étant donné que les méthodes utilisées dans la recherche influencent inévitablement les résultats, il est important d'exposer clairement le processus de collecte et d'analyse des données, de façon que l'étude soit en théorie répliquable. Par ailleurs, bien qu'il augmente la longueur des rapports de recherche, le compte-rendu écrit doit inclure suffisamment de données pour permettre au lecteur de juger si l'interprétation proposée est correctement étayée par les données.

## REFLEXIVITE

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La réflexivité consiste à être sensible à la manière dont le chercheur et le processus de recherche ont façonné les données collectées, y compris le rôle des hypothèses et de l'expérience antérieures, qui peuvent influencer même les enquêtes inductives les plus avouées. Les préjugés personnels et intellectuels doivent être mis en évidence dès le début de tout rapport de recherche afin de renforcer la crédibilité des résultats. Les effets des caractéristiques personnelles telles que l'âge, le sexe, la classe sociale et le statut professionnel (médecin, infirmière, kinésithérapeute, sociologue, etc.) sur les données collectées et sur la "distance" entre le chercheur et les personnes étudiées doivent également être discutés.

## ATTENTION AUX CAS NEGATIFS

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Les éléments qui viennent contredire la théorie émergente ne doivent pas être écartés, mais bien inclus dans l'explication et discutés. Cette "analyse des cas déviants" permet d'affiner l'analyse jusqu'à ce qu'elle puisse rendre compte de la totalité ou la grande majorité des cas examinés.

## PERTINENCE

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Une recherche est considérée comme pertinente lorsqu'elle apporte des connaissances supplémentaires ou qu'elle renforce la confiance dans les connaissances existantes, mais également lorsque les résultats peuvent être généralisés au-delà du cadre dans lequel ils ont été générés. Une façon d'y parvenir est de s'assurer que le rapport de recherche est suffisamment détaillé pour que le lecteur soit en mesure de juger si les résultats s'appliquent ou non à des contextes similaires.

#### 4. OBJECTIFS DE LA THESE

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L'objectif principal de cette thèse est de pouvoir illustrer comment la recherche qualitative peut venir compléter les méthodes quantitatives à différentes étapes du processus d'évaluation d'une intervention dite complexe.

Pour cela nous nous appuierons sur quatre travaux de recherche.

Le premier travail conduit (article en annexe 1), est une étude préliminaire de faisabilité d'une intervention médiée par les parents, formés en visioconférence. Nous chercherons à explorer, grâce à cet exemple, l'apport de la recherche qualitative pour l'évaluation de la faisabilité d'une intervention.

Le deuxième travail (article en annexe 2), est une étude transversale à méthodes mixtes, ayant évalué l'implémentation d'une intervention médiée par les parents, au niveau international. A l'aide d'une méthodologie quantitative et qualitative, les obstacles et facilitateurs de l'implémentation de cette intervention sont explorés. Cette étude nous permettra d'introduire les méthodes mixtes et leur intérêt dans le champ des sciences de l'implémentation.

Le troisième travail (article en annexe 3), est une synthèse des évidences qualitatives (ou métasynthèse) décrivant l'expérience des parents ayant participé à une intervention dite médiée par les parents. Nous poursuivrons, grâce à cette étude, l'analyse des méthodes de synthèse de la littérature qualitative et de leur intérêt plus global dans l'élaboration de recommandations pour la clinique et les recherches futures.

Enfin, le dernier travail (article en annexe 4), est un protocole publié d'une étude contrôlée randomisée multicentrique visant à évaluer l'efficacité de cette même intervention médiée par les parents, formés en visioconférence, sur la sévérité de l'autisme chez de jeunes enfants âgés de 18 à 36 mois. Cette étude nous permettra d'explorer difficultés inhérentes à l'évaluation d'une intervention complexe dans le champ de l'autisme. Nous discuterons de la place de la recherche qualitative et des méthodes mixtes dans ce champ.



# PARTIE 3 : ILLUSTRATION DE L'APPORT DE LA RECHERCHE QUALITATIVE DANS L'ÉVALUATION DES INTERVENTIONS DITES COMPLEXES

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## 1. INTRODUCTION

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Pour illustrer l'apport de la recherche qualitative dans l'évaluation des interventions non médicamenteuses dans l'autisme, et notamment des thérapies médiées par les parents, nous discuterons du modèle de l'intervention PACT que nous souhaitons adapter à la visioconférence.

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### LE PACT EN DETAILS

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Le PACT ou "Preschool Autism Communication Therapy" est une intervention précoce médiée par les parents utilisant l'outil vidéo dans le but d'aider à développer les compétences de communication (développement du langage expressif et réceptif, la compréhension des signaux sociaux, la capacité à interagir avec les autres) des enfants d'âge préscolaire présentant un TSA. Cette intervention, développée par l'équipe de Manchester, a été évaluée dans plusieurs études contrôlées randomisées et a été adaptée en Inde.

Le PACT présente plusieurs caractéristiques, qui en font un modèle développemental naturaliste. Premièrement, le PACT suit une hiérarchie développementale de compétences sociales et de communication, graduellement progressives. Il cherche à consolider le développement social réciproque, pré-linguistique, pragmatique et linguistique, tous altérés dans les TSA. L'objectif de l'intervention est de guider les parents pour offrir un contexte d'interaction adapté, où les réponses de communication et le langage s'ajustent à la compétence de communication de l'enfant. Les parents apprennent à identifier les occasions propices pour améliorer la communication émergente, susciter l'intentionnalité de l'enfant et faciliter les interactions conjointes, dans le but d'améliorer les trajectoires développementales de la communication.

Le PACT met l'accent sur les interactions dyadiques naturelles entre parent et enfant. Le thérapeute observe initialement et propose des suggestions sur la manière d'étendre l'attention partagée pour développer des ouvertures sociales et l'initiation à la communication

de l'enfant. Les opportunités de communication sont établies en examinant en détail les styles d'interaction du parent et de l'enfant, et en analysant la contribution de chaque partenaire à la dyade. La thérapie aide les parents à reconnaître et à répondre aux signaux contextuels, non verbaux et verbaux de l'enfant, ainsi qu'à interpréter ses intentions.

L'intervention se base sur un partenariat avec les parents, en capitalisant sur leurs compétences et en développant leur sentiment de compétence. Si les objectifs sont anticipés en suivant la logique développementale de la communication, le rythme est adapté de façon individuelle en suivant les progrès de l'enfant et des parents. La thérapie est adaptée au style parental pour optimiser l'efficacité. Les parents sont encouragés à identifier leur style d'apprentissage préféré et à réfléchir sur les stratégies qui fonctionnent bien pour eux.

Le programme utilise le video-feedback et un style réflexif de thérapie. Des sessions de jeu parent-enfant sont enregistrées en vidéo, puis les parents sont encouragés à identifier les épisodes positifs d'interaction réciproque et à réfléchir à leur contribution. Cette rétroaction permet également de discuter de l'interaction entre les parents et l'enfant et d'explorer comment les signaux d'initiatives de l'enfant sont déterminés par des indices contextuels, des comportements non verbaux et verbaux.

En pratique, les séances d'intervention individuelles ont lieu une semaine sur deux pendant six mois puis à un rythme mensuel pendant six mois supplémentaires. Dans l'intervention telle qu'initialement décrite par l'équipe de Manchester, une session dure environ deux heures, le parent est présent avec l'enfant. À chaque séance, les parents sont invités à jouer avec l'enfant comme ils le font normalement à la maison et à montrer comment ils appliquent le programme. Pendant ce temps, le thérapeute réalise un enregistrement vidéo de 10 minutes. Les parents et le thérapeute visionnent ensuite l'ensemble de la vidéo et identifient, analysent et discutent des séquences spécifiques qui démontrent la réalisation des objectifs thérapeutiques pour chaque étape du programme. Le thérapeute aide les parents à identifier leurs stratégies, les réponses positives de communication de l'enfant, ainsi qu'à réfléchir aux pistes d'amélioration de l'interaction. Des objectifs sont sélectionnés pour la séance suivante et le parent est invité à s'exercer à la maison et à consacrer 30 minutes par jour à la pratique des stratégies.

La thérapie suit une approche en six étapes basées sur la progression du développement de l'enfant et sur des stratégies visant à établir des compétences fondamentales pour le développement socio-communicatif. Les deux premières étapes visent à améliorer

l'identification par les parents de l'attention et de l'intérêt de l'enfant, de la synchronisation, de leur réactivité et de leur sensibilité à l'intérêt et à la communication de l'enfant. Les troisième et quatrième étapes visent à développer l'expression et la compréhension de l'enfant en commentant et en modélisant un langage adapté au niveau de développement de l'enfant. Les initiatives de communication de l'enfant sont également améliorées au cours de la cinquième étape grâce à différentes stratégies telles que l'anticipation et la routine. La dernière étape vise à développer la conversation et l'expansion du langage pour les enfants verbaux. Le passage d'un stade à l'autre dépend de critères prédéfinis.

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#### JUSTIFICATION DE L'UTILISATION DU PACT EN VISIO

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Le PACT (Pre-school Autism Communication Therapy) a montré une efficacité à court et long terme sur les symptômes de l'autisme évalués chez des enfants âgés de 2 à 10 ans dans une étude contrôlée randomisée britannique (N=152), comparant l'intervention PACT aux soins habituels (Green et al., 2010), ayant fait suite à une étude pilote encourageante (Aldred et al., 2004).

Le PACT a montré une réduction de la sévérité des symptômes de l'autisme mesurée par l'Autism diagnostic Observation Schedule version 2 (ADOS-2) (taille de l'effet 0,64 ; 95 % IC 0,07-1,20) ; et une augmentation de la synchronisation de la communication parentale avec l'enfant (ES = 1.22, 0.85 to 1.59) et des initiatives de communication de l'enfant avec le parent (0.41, 0.08 to 0.74) (Green et al., 2010; Pickles et al., 2016). L'étude de suivi a démontré un effet durable sur la sévérité des symptômes de l'autisme six ans après la fin de l'intervention, mesurée par l'ADOS, avec une réduction globale significative de la gravité des symptômes au cours de l'essai et de la période de suivi (taille de l'effet = 0,55, IC à 95 % 0,14 à 0,91,  $p = 0,004$ ) (Pickles et al., 2016).

Des études « mécanistiques » ont également confirmé que l'effet distal de la thérapie PACT sur la sévérité de l'autisme mesurée par l'ADOS était médié par l'amélioration des initiations de communication de l'enfant, elle-même médiée par l'amélioration de la synchronisation parent-enfant, renforçant les hypothèses théoriques initiales du mécanisme du PACT (Aldred et al., 2012; Carruthers et al., 2023; Pickles et al., 2015).

La disponibilité de la thérapie PACT est limitée, surtout dans les zones rurales ou dans les régions éloignées des centres spécialisés. L'utilisation de la vidéoconférence par des

thérapeutes pour former les parents au PACT peut donc être une alternative viable pour rendre ces thérapies accessibles aux familles vivant loin des centres d'autisme. Des études antérieures ont montré qu'il était possible de proposer des thérapies parentales dans l'autisme par vidéoconférence (Parsons et al., 2017; Pickard et al., 2016).

## 2. ETUDE 1 : INTERET DE LA RECHERCHE QUALITATIVE DANS UNE ETUDE DE FAISABILITE (ARTICLE EN ANNEXE 1)

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### OBJECTIF GENERAL

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Avant le démarrage de l'étude contrôlée randomisée visant à évaluer l'efficacité du PACT en visioconférence chez des jeunes enfants autistes de moins de 3 ans, que nous discuterons dans les suites de ce manuscrit, plusieurs professionnels de notre centre, formés à la méthode PACT, avaient proposé des suivis en visioconférence à des familles pour des raisons pratiques (déménagement, confinement pendant la première vague du COVID-19...).

Les professionnels formés sur notre centre, en collaboration avec l'équipe de Manchester, avaient pu préalablement réfléchir à l'adaptation de l'intervention en visioconférence. Plutôt que d'être filmés sur le centre par le thérapeute, les parents étaient invités à filmer, en amont de la session, dix minutes d'interaction (jeu, moment spécifique) avec leur enfant. Cette vidéo était envoyée au thérapeute sur un serveur sécurisé avant l'intervention. Le jour de l'intervention, le parent était présent en visioconférence, sans l'enfant. La vidéo était partagée sur le logiciel de vidéoconférence par le thérapeute et la session se déroulait comme en présentiel.

Nous avons donc décidé d'analyser ces suivis de façon standardisée, afin d'évaluer la faisabilité du modèle PACT en visioconférence, et de mieux comprendre l'influence du contexte sur la réalisation de l'intervention.

### RESUME DE L'ETUDE

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Nous avons utilisé une approche mixte en combinant des mesures quantitatives rétrospectives de faisabilité technique et des entretiens semi-structurés pour évaluer la faisabilité de l'intervention et décrire l'expérience des professionnels concernant le PACT en visioconférence. L'objectif de l'étude était avant tout descriptif, afin de réajuster au besoin l'intervention PACT en visioconférence en vue de son évaluation à plus grande échelle.

Nous avons inclus tous les enfants ayant reçu un diagnostic de TSA sur notre centre spécialisé selon les critères du DSM-5 et ayant suivi l'intervention PACT jusqu'en janvier 2021. Les professionnels formés au PACT et ayant participé au PACT en visioconférence ont été inclus dans l'étude qualitative.

La faisabilité de l'intervention en vidéoconférence était évaluée sur les vidéos des séances de thérapie enregistrées entre les parents et le professionnel. Cinq critères étaient évalués : qualité de l'audio, qualité de la vidéo, stabilité du réseau internet, possibilité de partager et de regarder simultanément la vidéo parent-enfant avec le parent, et durée totale de la séance.

Pour l'étude qualitative, des entretiens semi-structurés étaient réalisés avec les thérapeutes PACT. Une approche thématique, basée sur la théorie ancrée, était utilisée pour analyser les données.

L'étude a permis d'observer une bonne **faisabilité technique** du PACT en visio sur les critères quantitatifs. Les principales difficultés rencontrées concernaient la qualité audio de la visioconférence et le partage direct de la vidéo parent-enfant sur le logiciel de visio. Ces éléments ont été confirmés par l'étude qualitative.

L'étude qualitative a permis de mettre en lumière la faisabilité du PACT en visioconférence, malgré les difficultés techniques. Les professionnels interrogés ont pu approfondir leur description de l'expérience du PACT en visioconférence et souligner l'aspect écologique de l'intervention en visioconférence, c'est-à-dire dans le milieu naturel de l'enfant et non pas dans le cadre plus inhabituel du cabinet de consultation. Ces entretiens ont également permis de confirmer la **faisabilité théorique** du PACT et d'observer, malgré le contexte visio, les composantes importantes de l'intervention PACT : une relation thérapeutique forte entre le parent et le thérapeute, l'importance du videofeedback, l'augmentation du sentiment de compétence parentale et le changement de position du parent dans l'interaction.

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## DISCUSSION DE L'INTERET DE L'ETUDE QUALITATIVE POUR LES ETUDES DE FAISABILITE

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Le terme étude de faisabilité, regroupe un grand nombre d'études aux objectifs variés, tel qu'évaluer la faisabilité du recrutement, de la randomisation, de la rétention, des procédures d'évaluation, des nouvelles méthodes et/ou de l'implémentation de la nouvelle intervention, etc... (Arain et al., 2010; Leon et al., 2011). Nous avons choisi dans notre étude de nous concentrer sur l'évaluation des procédures de l'intervention et notamment, la faisabilité du

PACT en visioconférence, d'un point de vue technique, mais également sur le maintien des « ingrédients théoriques » du PACT. Notre étude était par ailleurs limitée par son design rétrospectif. Les autres indices de faisabilité pour notre ECR ont été évalués à l'aide des expériences de l'équipe de Manchester et de l'expérience de notre centre, en matière d'étude contrôlée randomisée chez de très jeunes enfants dans le champ de l'autisme (étude IDEA, publication à venir).

Concernant l'intérêt de la recherche qualitative au stade de l'étude de faisabilité, celle-ci a déjà été étudiée concernant la randomisation. Une méta-synthèse de 2022 a regroupé 31 études qualitatives, publiées entre 2010 et 2017, visant à évaluer les obstacles et facilitateurs à la mise en place d'une randomisation dans diverses études. Cette méta-synthèse a permis aux auteurs de proposer 13 obstacles à la randomisation (tels que la forte préférence d'un patient pour un traitement, la difficulté à communiquer autour de l'étude et du principe de randomisation) ainsi que 3 facilitateurs (par exemple, la participation à un réseau de recherche). Néanmoins, les auteurs ont pu mettre en évidence le manque d'impact des résultats des études qualitatives individuelles sur les études contrôlées randomisées de grande ampleur subséquentes, argumentant pour une réflexion plus profonde dans le champ de l'étude de faisabilité, non seulement aux obstacles à la randomisation, mais surtout aux proposition d'adaptation pour le bon déroulé de la randomisation (Elfeky et al., 2022).

L'utilisation de la recherche qualitative pour l'évaluation de l'acceptation de l'intervention et de sa mise en œuvre est également chose courante (Clery et al., 2021; Mishra et al., 2022).

Par ailleurs, la recherche qualitative est une méthode de choix pour transposer une intervention à un nouveau contexte. Pour l'adaptation du PACT en Inde (Divan et al., 2015), l'équipe de Divan et al. a proposé, en collaboration avec l'équipe anglaise initiatrice du PACT, de réaliser une étude préliminaire s'appuyant sur le cadre des interventions complexes proposé par le MRC (Craig et al., 2008), à l'aide d'une méthodologie qualitative. Leurs questions de recherche étaient :

- Comment adapter le contenu d'une intervention préalablement élaborée et évaluée dans un pays à revenus élevés, pour qu'elle réponde aux besoins des communautés dans des contextes à faibles ressources ?
- Comment la délivrance de cette intervention peut-elle être rendue acceptable et réalisable par un professionnel non spécialisé dans l'autisme ?

Des entretiens et groupes de travail ont été proposés et analysés grâce à une méthode qualitative (« framework analysis »).

Plusieurs adaptations au contexte ont été proposées : la possibilité que l'adulte ciblé par l'intervention soit le grand-parent, la possibilité que les sessions plus spécifiques sur le langage puissent être coanimées avec un spécialiste, les « devoirs » au domicile ont été renommé « pratique a la maison », un accompagnement plus renforcé des parents pour trouver des moments propices à l'utilisation des stratégies vues en séance est proposé, le manuel et ses exemples ont été retravaillé pour permettre à des non-spécialistes de proposer l'intervention, etc...

Une série de cas a également permis de souligner une acceptabilité de l'intervention, même lorsque réalisée par des non-spécialistes.

Une ECR a été menée par cette même équipe, et a mis en évidence un effet positif de cette intervention adaptée, versus traitement habituel dans la communauté, sur la synchronie parentale utilisée comme critère de jugement principal (différence moyenne ajustée 0,25 [IC95 % 0,14 à 0,36] ; taille de l'effet 1,61 [IC 95 % 0,90 à 2,32]) (Rahman et al., 2016).

Notre étude aurait pu bénéficier de la complémentarité d'entretiens avec les familles pour évaluer leur perception de l'intervention et la faisabilité technique de leur côté. Une évaluation à plus grande échelle aurait également pu nous permettre d'envisager des différences d'applicabilité entre les différents centres recruteurs pour l'étude IFPAD. Notre étude nous a néanmoins permis de nous assurer d'un maintien des composantes principales du PACT en visioconférence, et d'une faisabilité de l'intervention en contexte de visioconférence.

Une analyse de framework aurait pu apporter une compréhension axée sur les résultats de faisabilité et d'acceptabilité, en utilisant un cadre théorique préconçu. Néanmoins, nous avons choisi une analyse plus inductive, pour nous assurer de n'oublier aucune facette de l'expérience des thérapeutes.

### 3. ETUDE 2 : INTERET DES METHODES MIXTES POUR L'EVALUATION DE L'IMPLEMENTATION (ARTICLE EN ANNEXE 2)

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#### RESUME DE L'ARTICLE

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Comme évoqué précédemment, les interventions et notamment les interventions médiées par les parents sont souvent évaluées sous le prisme de l'efficacité. Les facteurs influençant l'implémentation de ces interventions sont insuffisamment étudiés et la perspective des parents est rarement prise en compte. Dans ce sens nous avons souhaité évaluer les obstacles et les facilitateurs de la mise en œuvre dans la pratique clinique à l'échelle mondiale du PACT, que nous détaillons plus en amont dans ce manuscrit.

Nous avons réalisé une étude transversale sur la base d'une enquête en ligne, analysée grâce à une méthodologie mixte. Des professionnels ayant suivi une formation PACT et des parents ayant reçu au moins trois séances d'intervention PACT via ces professionnels ont été interrogés. Les professionnels (N=460) inscrits dans la base de données de formation IMPACT, et répondant aux critères d'éligibilité, ont été sollicités de janvier à mars 2020 pour participer à l'enquête et étaient invités à leur tour à solliciter les parents avec lesquels ils avaient réalisé les séances de PACT pour partager le lien vers le questionnaire. Les questionnaires étaient disponibles en anglais, italien, français, cantonais et espagnol.

Une analyse concomitante quantitative et qualitative a été réalisée, les résultats ont ensuite été intégrés selon la méthodologie de triangulation des données de Tonkin-Crine et al. (2015).

Sur 460 professionnels ayant suivi une formation formelle au PACT avec IMPACT CiC entre mars 2017 et janvier 2020, 231 réponses ont été obtenues. La majorité des répondants étaient basés en France (45,5%), suivis par l'Italie (15,5%), le Royaume-Uni (11,2%) et Hong Kong, Chine (10,7%). Parmi les 1036 parents que ces professionnels ont déclaré avoir rencontrés en pratique, 94 parents ont répondu à l'enquête.

Les professionnels ont indiqué que le PACT était très acceptable et approprié dans leur pratique, et environ 70% ont déclaré essayer de l'implémenter. En tenant compte de l'intervalle de temps entre la formation et l'enquête, la mise en œuvre post-formation du PACT était facilitée chez les professionnels dont une large proportion de la charge clinique était consacrée

aux enfants autistes et à leurs familles, suggérant une pratique plus spécialisée (OR=4,94 ; [1,47-16,60] ; p=0,01). Parmi les autres facilitateurs clés de la mise en œuvre figuraient le soutien entre pairs de la part des collègues et des responsables, ainsi que la flexibilité de la planification. Il n'y avait pas de preuve d'un effet sur la mise en œuvre en fonction du pays d'origine ou des attitudes envers la pratique basée sur des preuves. Les principaux obstacles étaient le temps (sessions d'intervention > 1 heure), le matériel vidéo adéquat, les faibles capacités d'autoréflexion des parents, les conditions familiales ou de soutien stressantes, ainsi que des préoccupations importantes concernant le comportement de l'enfant.

Les parents ont enregistré des niveaux élevés d'acceptabilité et de faisabilité pour apprendre et implémenter le PACT dans la vie quotidienne, et ont effectué en moyenne 4 (IQR [2, 7]) des 7 sessions de 30 minutes quotidiennes de pratique à domicile prévues pendant l'intervention reçue. Les facilitateurs de la mise en œuvre pour les parents étaient des caractéristiques bien perçues de la méthode PACT (réflexion vidéo, autonomisation des parents, pratique basée sur le jeu). Les obstacles étaient la disponibilité de temps (par exemple, pour la pratique à domicile) et les difficultés liées à l'enregistrement vidéo.

Cette étude à méthodologie mixte est la première à examiner les facteurs déterminant la mise en œuvre post-formation du PACT en pratique clinique, une thérapie médiée par les parents, à diffusion internationale et fondée sur des preuves.

L'étude comprend certaines limites, notamment en raison du taux de réponse des parents et des professionnels. Toutefois, les résultats suggèrent que si le PACT est considéré comme acceptable et faisable par les parents et les professionnels, les facteurs familiaux et contextuels du système de soin sont essentiels dans la réussite de l'implémentation.

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## DISCUSSION

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### APPORT DES METHODES MIXTES

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La recherche mixte est une approche méthodologique qui intègre à la fois des méthodes qualitatives et quantitatives dans une même étude pour aborder un problème de recherche sous plusieurs angles. Cette approche permet de combiner les avantages de ces deux approches et d'obtenir une compréhension plus complète et approfondie du sujet étudié. Elle permet de trianguler les résultats, en observant la convergence ou la divergence des résultats obtenus par différentes méthodes, afin de valider et enrichir les conclusions. Un protocole de

triangulation peut renforcer la validité des résultats et évaluer si les données concordent (convergence, permettant d'accroître la crédibilité du résultat), se complètent (complémentarité, permet d'apporter de la nuance ou du contexte au résultat) ou se contredisent (dissonance). La dissonance n'indique pas un échec de l'étude, mais peut être considérée comme constructive si elle conduit à une compréhension plus riche. Elle peut permettre de donner une nouvelle orientation pour de futures études, afin de mieux cibler le phénomène d'intérêt (Heale & Forbes, 2013; O'Cathain et al., 2010; on behalf of the GRACE INTRO/CHAMP consortium et al., 2015).

Des précautions sont néanmoins nécessaires dans l'interprétation des résultats. Si des résultats convergents sont en faveur d'une crédibilité des données, ils doivent être sous-tendus par une bonne méthodologie. En effet, ces résultats peuvent être convergents du fait d'un manque de rigueur (par exemple une sélection des participants non adéquates) et la convergence à elle seule n'indique pas la qualité méthodologique de l'étude (Heale & Forbes, 2013).

Dans notre étude, plusieurs informations étaient convergentes. Plusieurs facilitateurs de l'implémentation ont ainsi été décrits, notamment la méthode réflexive du PACT (avec le videofeedback) et l'importance de l'alliance avec le thérapeute. Ces éléments font écho aux résultats de notre étude pilote (annexe 1) et aux résultats de notre métasynthèse (annexe 4), renforçant ainsi leur validité interne (convergence au sein de l'étude) et externe (convergence avec la littérature). Ces éléments semblent immuables, d'un contexte à l'autre. Certaines barrières, convergentes dans notre étude, et retrouvées dans d'autres travaux sont également observées, telle que le besoin d'occuper l'enfant pendant le débriefing, ou la nécessité d'être à l'aise avec le matériel (vidéo). L'importance du soutien du groupe professionnel (collègues formés dans la même équipe et collègues favorables à la mise en œuvre du PACT) était également soulignée et convergente entre plusieurs méthodes de recueil.

Des éléments dissonants étaient également retrouvés. L'impact de la disponibilité et de l'organisation des parents sur l'implémentation de l'intervention était rapporté par les professionnels et les parents eux-même en réponse aux questions ouvertes. Cependant, ces résultats n'apparaissent pas sur les réponses aux questions fermées (avec une majorité de réponses négatives à la question « le travail impacte la mise en œuvre du PACT a la maison »). Il est possible que cette dissonance soit en lien avec la formulation de la question relative au

travail dans le questionnaire, alors que les réponses ouvertes évoquent un impact des difficultés d'organisation.

Par ailleurs, une forte dissonance entre les réponses ouvertes des professionnels, des parents, et de la réponse aux questions fermées des parents était observée concernant la population de Hong Kong. Les professionnels ont pu évoquer une difficulté de cette population avec les approches réflexives, les parents ont pu répondre avoir peu de temps pour mettre en œuvre l'intervention, évoquant une dissonance dans la perception des obstacles à l'implémentation. Par ailleurs, les familles ont pu évoquer le besoin d'un suivi plus renforcé, malgré le peu de temps disponible évoqué. Nous pourrions faire ici l'hypothèse d'un besoin de suivi, autre que le PACT et son approche réflexive, évoqué par les parents. Cette dissonance pose les bases de futures études pour une meilleure compréhension des barrières à l'implémentation dans cette population.

## APPORT DE L'ETUDE SUR LE PLAN DE L'IMPLEMENTATION

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Comme évoqué dans le processus de l'évaluation des interventions complexes (Moore et al., 2015; Skivington et al., 2021), l'étude de l'implémentation des interventions reconnues comme efficaces est indispensable. Ce type d'étude vise à évaluer à quelle point une intervention a pu être intégrée en pratique courante et quel impact a pu avoir cette intégration. Ces études ont également pour objectif de comprendre les obstacles et leviers de l'implémentation en pratique courante.

Le « Consolidated Framework for Implementation Research » (un cadre théorique décrivant les différents aspects de la recherche sur implémentation) évoque plusieurs domaines à prendre en compte pour l'évaluation des facteurs d'implémentation (Damschroder et al., 2022) :

- Caractéristiques de l'Intervention (détails sur l'intervention spécifique et les stratégies de prestation)
- Caractéristiques du Praticien Individuel (compréhension individuelle telle que les attitudes envers la pratique basée sur les preuves, le temps disponible, etc.)
- Caractéristiques du Contexte Interne (« caractéristiques des contextes structurels, politiques et culturels à travers lesquels le processus de mise en œuvre progresse »)

- Caractéristiques du Contexte Externe (« contexte économique, politique et social, y compris les besoins des patients et les ressources au sein desquelles une organisation évolue »)
- Processus d'implémentation (« processus de changement actif visant à obtenir l'utilisation de l'intervention au niveau individuel et organisationnel tel que conçu »)

Une combinaison de ces facteurs peut exercer une influence positive ou négative sur la décision des professionnels d'initier et de continuer à fournir une thérapie aux patients.

De même que pour toutes les autres étapes de l'évaluation des interventions complexes, il s'agit ici d'obtenir des informations utiles, pour pouvoir adapter la thérapie et activer les bons leviers pour une diffusion de l'intervention dans la population cible.

Ici, l'intégration d'informations quantitatives et qualitatives, nous permet de répondre à des questions précises mais également d'observer les facteurs relevant de l'expérience des répondant, non anticipés par le chercheur. Après une première analyse de contenu et une triangulation avec les résultats quantitatifs, les données qualitatives ont été réanalysés en utilisant une analyse de framework, selon le « Consolidated Framework for Implementation Research » présenté ci-dessus.

Cette analyse nous permet de rediscuter des grands facteurs d'implémentation (obstacles et facilitateurs) pour le modèle PACT. Au-delà des caractéristiques des participants (contexte externe) et de l'intervention, que nous avons eu l'occasion d'explorer dans notre étude de faisabilité et qui sont également retrouvés dans la métasynthèse, cette analyse a permis de dégager des points essentiels à l'implémentation. Ceux-ci concernent premièrement le contexte interne, avec la nécessité d'un réseau autour de l'intervention, de leader de l'intervention pour permettre un maintien de l'implémentation sur le plus long terme et une motivation des équipes et des conditions organisationnelles propices à la mise en œuvre du PACT. Par ailleurs, des éléments concernant le processus d'implémentation ont été dégagés de notre étude, avec notamment la nécessité de professionnels ayant du temps dégagé pour l'intervention, avec une motivation à utiliser le PACT dans leur pratique, ces professionnels devenant les « fils rouges » de l'implémentation pour agréger les équipes et faire diffuser l'intervention.

## 4. ETUDE 3 : INTERET DE LA SYNTHÈSE DES EVIDENCES QUALITATIVES (ARTICLE EN ANNEXE 3)

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### SYNTHÈSE DE LA LITTÉRATURE QUALITATIVE, UN OUTIL D'OPTIMISATION DE L'UTILISATION DE LA RECHERCHE QUALITATIVE

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Le nombre de travaux qualitatifs progresse rapidement en médecine et en psychiatrie (Lachal et al., 2017). Cependant la littérature qualitative pose plusieurs problèmes, limitant son applicabilité. Les méthodes sont multiples et peu connues du public de cliniciens à qui elles sont destinées. Les études de médecine sont principalement organisées autour du paradigme positiviste et l'introduction à la recherche qualitative y est rare. De fait, l'évaluation de leur qualité et de leur transférabilité n'est ainsi pas toujours aisée. De plus, les études qualitatives sont souvent isolées, et participent peu au mouvement de l'Evidence-Based Medicine (Evans & Pearson, 2001), notamment par le manque d'intégration dans le développement des recommandations de bonnes pratiques (Finfgeld-Connett, 2016).

Ces limitations peuvent être partiellement surmontées grâce à l'utilisation de synthèses qualitatives, puisque ces méthodes consistent à regrouper plusieurs études et à évaluer leur qualité. La synthèse des évidences qualitative (en anglais, Qualitative Evidence Synthesis) ou métasynthèse est un groupe hétérogène de méthodes permettant la collecte systématique et la synthèse d'études qualitatives autour d'un sujet d'intérêt (Hannes & Lockwood, 2011b; J. L. Harris et al., 2018; Lockwood et al., 2015; Noyes et al., 2018; Thomas & Harden, 2008). Elles résument les idées fondamentales de chaque étude dans leurs résultats, ajoutent un niveau d'interprétation supplémentaire ou global, et facilitent la transférabilité des données (Finfgeld-Connett, 2010). Cette approche permet de dépasser les limites des études individuelles en tirant parti de la richesse des données qualitatives existantes et en identifiant des thèmes, des modèles ou des concepts émergents à travers différentes études.

Sur le plan de la terminologie, le Cochrane Qualitative Implementation Methods Group (CQIM group) a choisi d'utiliser le terme Qualitative Evidence Synthesis (synthèse de preuves qualitatives) (Booth et al., 2016), mais les termes « revues d'études qualitatives », « métasynthèses qualitatives », « synthèses d'études qualitatives », « synthèses thématiques », « méta-ethnographies », sont employés comme synonyme de métasynthèse,

bien qu'ils expriment parfois des soubassements théoriques spécifiques. Par souci de simplicité, nous emploierons le terme « métasynthèse » dans la suite de ce chapitre.

La métasynthèse comprend deux grandes étapes qui, selon l'orientation théorique, seront effectuées en parallèle ou de façon simultanée.

La première étape est celle de la revue systématique de la littérature sur le sujet d'intérêt. Pour cette démarche, le chercheur peut choisir une approche classique de revue systématique qui comprendra une recherche dans plusieurs bases de données, avec des mots clés, un screening des articles avec des critères d'inclusion et d'exclusions précis, définis a priori, et une sélection de tous les articles correspondant à ces critères. Certains auteurs adoptent une position différente pour cette recherche, plus inspirée par la recherche qualitative avec une sélection raisonnée d'articles (purposive sampling) et des critères d'inclusion évoluant au fil de l'analyse et de la compréhension du sujet (Booth, 2016; Noyes et al., 2018).

La deuxième étape concerne l'analyse des données et notamment la synthèse des données récoltées. L'objectif de cette étape n'est pas simplement de résumer les résultats des articles inclus, mais de proposer une interprétation de niveau supérieur afin d'atteindre une généralisabilité théorique plus grande, relative à l'expérience explorée. Plusieurs approches et méthodes sont utilisées pour mener à bien la synthèse des évidences qualitatives. De la même façon que pour la recherche qualitative en général, elles s'appuient sur différentes orientations théoriques et leur utilisation dépend de la question de recherche posée. Le Tableau 2, inspiré par les travaux du « Cochrane Collaboration Qualitative Methods Group » décrit les méthodes principales en fonction de l'objectif (Noyes J & Lewin S, 2011).

**TABLEAU 2. DIFFERENTES METHODES DE METASYNTHESSES.**

Agréger, synthétiser et/ou décrire des informations qualitatives autour d'un phénomène	
Analyse thématique :	Identification des thèmes majeurs / récurrents dans les études sélectionnées (réinterprétation des données); résumé des résultats des études primaires sous ces thématiques (Thomas & Harden, 2008)
Méta-agrégation :	L'évaluateur ne réinterprète pas les études incluses, mais présente les résultats des études incluses de manière précise (chaque résultat ou « findings » de l'étude initiale est décrit). Ces résultats sont ensuite agrégés pour former des catégories, qui sont regroupés sous des thèmes de niveau supérieur (Lockwood et al., 2015).

## Interpréter un ensemble de données qualitatives pour développer une théorie explicative ou un modèle de compréhension

Méta-ethnographie :	<p>Une première étape d'analyse translationnelle réciproque identifie les thèmes clés de chaque étude, puis cherche à les interpréter dans le contexte de chacune des autres études. Les thèmes les plus adaptés/explicatifs sont retenus.</p> <p>L'approche peut ensuite proposer une interprétation d'ordre supérieur, qui peut être une image globale du phénomène, ou une synthèse des contradictions entre les résultats des études individuelles (Dixon-Woods et al., 2005).</p>
Théorie ancrée :	<p>La théorie ancrée considère que la préoccupation première de la recherche qualitative est la production de théories (explications généralisables des phénomènes sociaux).</p> <p>Une méthode de comparaison constante permet d'identifier des modèles et des interrelations dans les données primaires des études incluses afin de développer une nouvelle théorie (Dixon-Woods et al., 2005).</p>

## Développer un modèle unique en intégrant des données qualitatives et quantitatives

Synthèses réalistes :	<p>Il s'agit d'une méthode de synthèse de données de recherche qui privilégie la prise en compte des contextes et des réalités pratiques dans lesquels les interventions médicales sont mises en œuvre. Plutôt que de se limiter à une analyse théorique des résultats, une revue ou une synthèse réaliste peut chercher à comprendre comment une intervention fonctionne dans la vraie vie, en prenant en compte les facteurs contextuels, les obstacles potentiels et les considérations pratiques. Les preuves qualitatives et quantitatives sont intégrées (Wong et al., 2013).</p>
“Metanarrative reviews” :	<p>Les « metanarratives reviews » examinent de façon historique comment des traditions de recherche (ou approches) particulières se sont développées au fil du temps et ont façonné le type de questions posées et les méthodes utilisées pour répondre à une thématique de recherche. Elles examinent l'éventail des approches adoptées pour étudier une question, synthétisent les résultats pour chaque approche, et analyse les contrastes entre ces approches pour proposer une synthèse globale. (Greenhalgh et al., 2005)</p>

L'utilisation des métasynthèses dans le champ de l'évaluation d'interventions complexes a été décrit par Flemming et al. La métasynthèse pourrait ainsi permettre de répondre à plusieurs questions, en lien avec les mécanismes de l'intervention (Comment les composantes se combinent-elles pour produire des effets ?), l'évaluation des bénéfices et risques (Pourquoi les résultats n'ont-ils pas été ceux escomptés ? Quelle importance les personnes concernées accordent-elles aux différents résultats ?), de l'acceptabilité (Quels sont les points de vue des parties prenantes sur l'acceptabilité, les préférences ou l'adéquation de l'intervention ?), le contexte d'implémentation des interventions (Comment et pourquoi la mise en œuvre d'une intervention varie-t-elle selon les contextes ?) (Flemming et al., 2019).

Comme discuté précédemment, l'utilisation des interventions médiées par les parents est aujourd'hui incontournable dans le champ de l'autisme. Néanmoins, comme pour toutes interventions, il est nécessaire de s'interroger sur les effets bénéfiques (avec des études d'efficacité mais également en analysant le vécu des individus) et sur les effets potentiellement indésirables de l'intervention. Nous avons donc cherché à décrire l'expérience des parents ayant participé à une intervention médiée par les parents pour leur enfant autiste. La finalité était de pouvoir proposer des recommandations pour l'implémentation de ce type d'intervention en pratique clinique mais également pour les recherches futures.

Si nos précédentes études (étude pilote et étude d'implémentation) avaient également pour objectif de comprendre l'expérience des parents dans ce type d'intervention et de réfléchir aux facteurs d'implémentation, la réalisation d'une métasynthèse sur ce même sujet nous paraissait intéressante pour plusieurs raisons. Premièrement, il s'agissait d'un moyen de confirmer la généralisabilité de nos résultats, via la triangulation avec la littérature scientifique. Par ailleurs, ce travail n'avait pas été réalisé, il nous apparaissait utile, pour la communauté scientifique, d'avoir un travail résumant l'ensemble des études dans ce domaines, pour de futurs recommandations ou développements d'intervention. Dans notre centre particulièrement, ce travail avait pour but de compléter les informations obtenues par l'étude pilote qui n'interrogeait qu'un groupe restreint de professionnels et qui n'interrogeait pas spécifiquement les parents sur leur expérience de l'intervention, pour des raisons de faisabilité de l'étude pilote.

Nous avons ainsi réalisé une revue systématique avec synthèse des preuves qualitatives pour explorer l'expérience des parents impliqués dans les interventions médiées par les parents auprès d'enfants autistes. Nous avons effectué des recherches dans les bases de données PubMed, Scopus, PsycINFO, CINAHL et Sociology Collection à partir de leur création jusqu'au 25 février 2022. Toutes les études qualitatives rendant compte de l'expérience des parents avec ce type d'intervention ont été incluses. Nous avons choisi d'évaluer l'ensemble de la littérature plutôt qu'un échantillon raisonné d'articles. Comme décrit précédemment dans la méthodologie des métasynthèses, la qualité individuelle des études a été évaluée par deux évaluateurs, et les caractéristiques et résultats de chaque étude ont été extraits, évalués sur le plan de la crédibilité puis synthétisés.

Nous avons choisi de synthétiser ces données à l'aide d'une méthode appelée « méta-agrégation » (voir **Tableau 2**).

Pour s'assurer de l'exhaustivité de notre rapport d'analyse et du report adéquat des critères de qualité propres aux revues systématiques et aux métasynthèses, nous avons suivi les guidelines PRISMA et ENTREQ. Pour évaluer la validité de nos données, nous avons suivi les recommandations GRADE-Cerqual.

Au total, 23 études ont été synthétisées, représentant 345 participants. L'âge moyen des participants (parents ayant répondu aux entretiens) était de 36,7 ans (Ecart Type [ET] = 3,4), et la plupart des parents interrogés étaient des femmes (81%). Le niveau d'éducation était relativement élevé dans les différentes études. Les enfants des parents inclus dans les études avaient un âge moyen de 62,7 mois [15-133] et étaient principalement des garçons (81%). Onze études provenaient d'Amérique du Nord ; six étaient d'Europe, quatre d'Asie et deux d'Australie.

La plupart des interventions (médiées par les parents) proposées aux parents visaient à améliorer les compétences en communication sociale et était réalisées de façon individuelle. Pour la plupart des interventions, les parents étaient invités à pratiquer régulièrement l'intervention à domicile.

Nous avons extrait 208 résultats des 23 études. Ces résultats ont été rassemblés en 34 catégories qui ont été résumées en quatre thèmes synthétisés : obstacles à l'implémentation et problèmes logistiques, sentiment d'être submergé et stressé (besoin de soutien), facilitateurs de l'implémentation, et autonomisation (« empowerment ») des parents et évolution de l'enfant.

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## DISCUSSION

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Pour notre thématique, le travail de métasynthèse permet de proposer, au sein d'une expérience commune, ici l'intervention médiée par les parents, une grille de compréhension des facteurs d'implémentation de l'intervention à l'échelle des parents, des ingrédients essentiels transversaux aux différentes méthodes, et des points d'améliorations pour renforcer l'utilité de ces méthodes d'intervention. Le résultat peut être considéré comme transférable à une population similaire à celle décrite dans l'étude, qui, du fait de la synthèse de multiples études, est plus large, plus hétérogène, et représentatives de multiples contextes.

Néanmoins, la métasynthèse présente des limites.

Premièrement, les articles rapportant des études qualitatives sont particulièrement difficiles à trouver sur les bases de données car encore très mal indexées (Booth, 2016). De multiples méthodes existent et de nombreuses terminologies sont utilisées pour évoquer le recueil d'expérience (expérience, attitudes, croyances, regard, opinion, vécu...) et le mot « qualitative » est plus facilement associé à la notion de qualité qu'à la notion de recherche qualitative.

Dans le cadre de notre étude, pour contourner cette difficulté, nous avons choisi pour notre recherche systématique de séparer les critères d'inclusions entre les phases de screening. Nous avons opté pour un critère d'inclusion plus large pour première phase de screening, sur titre et abstract, et avons décidé de présélectionner, tous les articles évoquant la participation de parents à une intervention médiée par les parents, sans prendre en compte le type d'étude et de méthodologie utilisée. A la phase de sélection sur texte entier, nous avons affiné les critères d'inclusions pour ne récupérer que les articles pertinents au regard de notre question de recherche. Si cette méthode a l'avantage de d'éviter l'oubli d'études qualitatives mal référencées, l'inconvénient consiste en son manque de rentabilité. Pour notre étude, nous avons dû lire un total de 2879 abstract puis de 515 textes complets, pour récupérer 23 études qualitatives.

D'autre part, nous avons choisi la méthode de la méta-agrégation, qui vise à extraire les résultats des différentes études incluses afin de les regrouper en catégories puis en grandes thématiques. « Extraire les résultats » peut cependant relever du casse-tête puisque la définition même de « résultats » dans le cadre d'une étude qualitative n'est pas si claire. Pour certains auteurs, les résultats d'une étude qualitative se trouvent tout simplement dans la partie « résultat » de l'article publié. Pour d'autres, les réflexions présentées par les auteurs dans la discussion, sont également une forme d'analyse et donc de résultat qualitatif (Sandelowski & Barroso, 2002). Par ailleurs, au sein même de la partie résultat et de sa description dans un article, le chercheur réalisant l'extraction peut se retrouver confronté à des résultats divergent entre des présentations synthétiques (tableaux, figure) et les descriptions des résultats dans le texte. Le choix de la méthode d'analyse peut permettre de contourner, en partie, cette difficulté. Une analyse thématique (et non par méta-agrégation) aurait pu permettre de considérer les résultats dans la globalité (texte entier des résultats et de la

discussion) et de s'affranchir de la notion de « résultats précis » donnés par les auteurs. L'avantage de la méta-agrégation réside néanmoins dans le fait de respecter les choix des auteurs sur la sélection des résultats pertinents (Hannes & Lockwood, 2011a).

## 5. ETUDE 4 : INTERET DE LA RECHERCHE QUALITATIVE ET DES METHODES MIXTES DANS UNE ETUDE D'EFFICACITE, ETUDE IFPAD (IMPACT FORMATION DES PARENTS A DISTANCE) (ARTICLE EN ANNEXE 4)

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### SYNTHESE DU PROTOCOLE

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Les travaux présentés précédemment ont été construits autour de l'étude IFPAD. Cette étude a pour objectif principal de tester l'efficacité du PACT, délivré par des thérapeutes à des parents vivant dans des zones mal desservies, via vidéoconférence, sur une période de 12 mois, sur l'ensemble des symptômes autistiques, mesurés à l'aide de l'Autism Diagnostic Observation Schedule 2 (ADOS), chez des enfants autistes âgés de 18 à 36 mois à l'inclusion.

Les objectifs secondaires comprendront une évaluation de l'évolution des interactions socio communicatives de l'enfant, du langage, de l'initiation à la communication et du comportement adaptatif quotidien. Un volet parental est prévu afin d'évaluer les effets de l'intervention sur le stress, la santé et le fonctionnement de la famille.

La mise en œuvre (ou implémentation) de la thérapie sera évaluée à travers l'adhésion des professionnels et des parents au PACT, et l'acceptabilité et la faisabilité des sessions PACT pour les parents et les thérapeutes.

Il s'agit d'une étude mixte (quantitative qualitative) prospective multicentrique, en population française. Elle comprendra une étude contrôlée randomisée avec 2 groupes parallèles, ratio 1:1, en simple aveugle, comparant l'intervention PACT + le traitement habituel (TAU) au TAU seul. Cette étude sera associée à une étude qualitative qui visera à comprendre, via des entretiens semi-structurés avec les parents et les thérapeutes, les obstacles et les facteurs facilitant la mise en œuvre du PACT en visioconférence.

Après une analyse séparée, les données quantitatives et qualitatives seront intégrées pour évaluer leur cohérence et proposer un modèle complet de compréhension des obstacles et facilitateurs de la mise en œuvre du PACT en visioconférence.

### AVANCEE DE L'ETUDE

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Les inclusions pour l'étude IFPAD ont débuté en juillet 2021. La figure 2 présente le rythme des inclusions actuelles en comparaison aux prédictions d'inclusion théoriques.

Le rythme initial de recrutement était peu soutenu. Plusieurs facteurs ont été identifiés pour expliquer les difficultés.

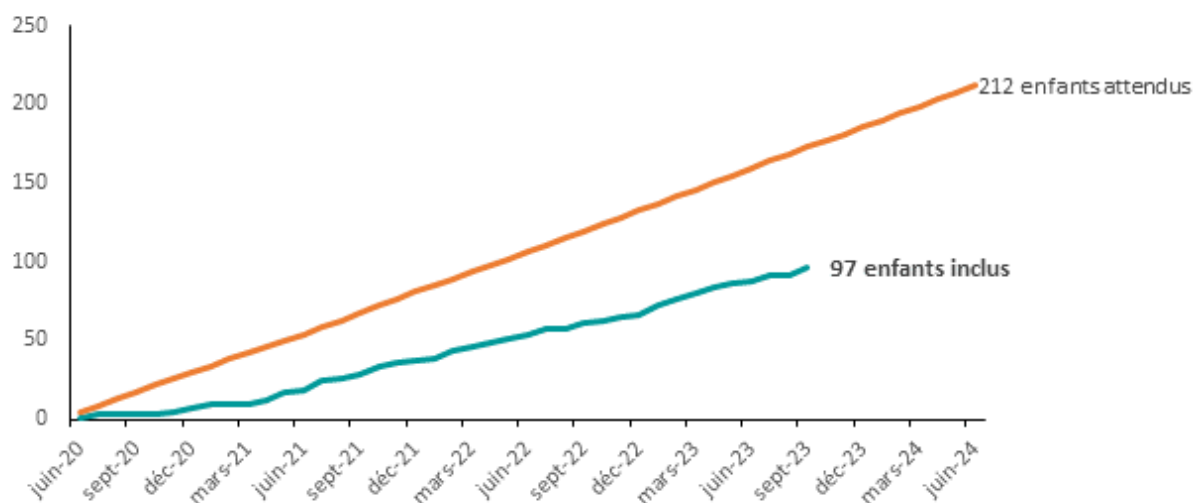


FIGURE 2. NOMBRE D'ENFANTS INCLUS DANS L'ETUDE IFPAD ET RYTHME D'INCLUSION THEORIQUE.

Premièrement, nous avons souhaité cibler une population éloignée des centres spécialisés, pour cibler une population dans des régions plus défavorisées en termes de moyens diagnostiques et thérapeutiques pour l'autisme. A l'époque du premier dépôt du protocole au CPP en 2019, l'intérêt, pour nous, de la visioconférence, était principalement de pouvoir atteindre des régions plus éloignées et des familles souvent avec très peu de prise en charge disponible. Nous avons donc choisi d'inclure des familles résidant à plus de 40 minutes de Lyon. Cependant, l'organisation des soins en psychiatrie, centrée sur le secteur, s'est confrontée à ce critère. Bien qu'une large campagne de communication autour de l'étude ait été réalisée, il ne s'agissait pas de notre territoire d'action habituel, et il était plus difficile de cibler les soignants susceptibles de pouvoir proposer l'étude à des familles. De plus, l'absence de médecins et pédiatres dans de nombreuses régions ciblées ne permettait pas d'avoir un relai, pour proposer l'étude. Par ailleurs, la crise du COVID-19 (et notamment le premier confinement) a eu un impact majeur sur notre utilisation de la visioconférence (Bidmead & Marshall, 2020; Keesara et al., 2020). Devenue un outil du quotidien, la visioconférence est utilisée pour faire des réunions (même avec des collègues d'un bâtiment voisin), est plébiscitée par les patients pour les consultations afin de ne pas s'absenter trop longtemps du travail (ou de l'école pour les enfants). L'intérêt de la visioconférence n'était donc plus uniquement de solutionner l'accès

aux soins pour les familles éloignées, mais également de pouvoir allier « maintien des soins » et « organisation familiale ». Le critère de distance a donc été supprimé. Cette modification a permis d'accélérer le rythme des inclusions.

Dans les obstacles à l'inclusion, nous avons également pointé le critère d'âge des enfants. De nombreux enfants nous étaient adressés après leurs 3 ans, notamment grâce au repérage de signes d'autisme par l'école maternelle. L'étude IFPAD a démarré juste avant la mise en route effective des Plateformes de Coordination et d'Orientation, un projet national visant à améliorer le dépistage et le diagnostic précoce des troubles du neurodéveloppement (TND) et notamment du TSA. Ces plateformes, implémentées suite aux propositions de la Stratégie Nationale pour l'Autisme, au sein des TND (**Stratégie Nationale pour l'Autisme au sein des Troubles du Neurodéveloppement, 2018**), couvrent aujourd'hui une grande partie du territoire avec 78 plateformes identifiées. Elles permettent de renforcer le dépistage systématique des TND chez les médecins généralistes, et de les guider pour avancer sur le diagnostic. Elles étaient, dans leur forme initiale, destinée aux enfants de 0 à 6 ans. Leur mise en place, courant 2021, a également permis un repérage plus précoce des signes d'autisme chez les enfants. Ces plateformes participent activement à la formation des professionnels. De fait, l'orientation des familles vers notre étude s'est faite de façon plus précoce, solutionnant l'obstacle de l'âge que nous avons identifié.

La correction de ces obstacles à l'inclusion et l'ouverture progressive des différents centres a permis, après une période d'adaptation, d'accélérer le rythme des inclusions. Une étude pilote centrée sur l'évaluation de la faisabilité du recrutement aurait pu nous permettre d'identifier ces obstacles et facilitateurs en amont de la mise en place de l'étude (Elfeky et al., 2022).

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#### DISCUSSION : L'ETUDE IFPAD, UN EXEMPLE D'EVALUATION D'UNE INTERVENTION COMPLEXE

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L'étude IFPAD permet d'illustrer l'évaluation d'une intervention complexe, au-delà de la phase pilote de faisabilité. L'objectif principal de l'étude IFPAD est d'évaluer l'efficacité du modèle, cependant, comme décrit précédemment, dans le cadre des interventions dites « complexes », des études multimodales avec des objectifs secondaires plus larges permettent d'informer les décideurs sur l'utilité de l'intervention dans un contexte spécifique, au-delà de la seule notion d'efficacité (Skivington et al., 2021).

Le PACT, et notamment le PACT en visioconférence, peut être considéré comme une intervention complexe pour plusieurs raisons.

Il s'agit d'une intervention avec plusieurs composantes ou « ingrédients », décrits précédemment dans le chapitre sur le PACT. Il est important que ces composantes soient évaluées dans différents contextes afin de comprendre le fondement théorique de l'intervention et, dans la mesure du possible, le modèle causal de l'intervention, c'est-à-dire « quels sont les éléments qui fonctionnent et comment ? ». Comme précisé précédemment, l'équipe à l'initiative du PACT a pu proposer, dans le contexte anglais de l'étude d'efficacité (Pickles et al., 2015) une évaluation des médiateurs de l'intervention. Dans le contexte de notre étude, cette analyse est également prévue. Celle-ci nous permettra de vérifier le modèle causal évoqué par la précédente équipe, dans un contexte différent, et de le confronter aux aspects théoriques de l'intervention. Par ailleurs, l'équipe qualitative peut également apporter des éléments expérientiels sur les composantes utiles du PACT grâce aux entretiens. Les résultats de l'étude qualitative permettront ainsi de renforcer les résultats des analyses de médiation, ou de faire émerger de potentiels facteurs médiateurs ou modérateurs non pris en compte dans le modèle. Le contexte spécifique de l'intervention, ici en visioconférence, pour une population en post-diagnostic immédiat, ne recevant pas ou peu d'autres interventions, sera également pris en compte dans nos analyses.

Notre intervention peut également être considérée comme complexe du fait de l'implication nécessaire du thérapeute et du parent, à des niveaux élevés, pour permettre une bonne réalisation de l'intervention. Concernant le thérapeute, celui-ci nécessite une formation importante, et une adhésion au modèle d'intervention. Ces éléments seront mesurés via l'étude de la fidélité au modèle PACT (des vidéos d'interventions sélectionnées au hasard seront évaluées par un des formateurs PACT, qui s'appuiera sur une grille d'évaluation de la fidélité du PACT pour s'assurer que l'intervention délivrée correspond bien à l'intervention PACT initialement développée) (Green et al., 2010, 2018). Par ailleurs, l'implication des parents et des professionnels sera évaluée via des mesures d'acceptabilité de l'intervention. Les parents seront interrogés en fin d'étude, sur le nombre d'heures d'intervention au domicile ainsi que l'acceptation perçue de la mise en œuvre du PACT au domicile, de façon chiffrée mais également lors de l'étude qualitative. L'évaluation de l'adhésion aux principes du PACT sera évalué grâce à l'outil DCMA (Dyadic Communication Measure In Autism) qui permet une analyse de la contingence de la communication du parent avec l'enfant. Les professionnels

seront également interrogés sur l'acceptabilité au sein de l'entretien semi-dirigé. Le nombre de séance réalisé par famille sera également un indicateur d'acceptabilité de la méthode.

L'autisme est un concept large, multifacette, et le choix des critères de jugement n'est pas aisé pour l'évaluation de l'intervention. L'évaluation de l'efficacité d'une intervention dans l'autisme se fait classiquement sur la sévérité de l'autisme. Des outils cliniques nous permettent l'évaluation la symptomatologie autistique dans son ensemble, tel que l'ADOS-2 (Autism Diagnostic Observation Schedule 2) utilisée comme critère de jugement principal de l'étude IFPAD. Cet outil, largement recommandé dans les guidelines (McConachie et al., 2015), reste néanmoins partiel dans sa capacité à évaluer le concept plus global de l'autisme et du handicap. En fonction du public visé par l'intervention, des critères de jugement évaluant le handicap ou l'autonomie dans son ensemble pourraient être pertinents. Nous pourrions également imaginer l'émergence de pistes de cible d'intervention dans les entretiens qualitatifs avec les parents et les professionnels. La recherche qualitative est une méthode essentielle pour la création d'outils d'évaluation centrés sur les attentes des patients. Dans le champ de l'autisme, le travail de K. Leadbitter (Leadbitter et al., 2018) est un exemple de développement d'un outil visant à évaluer l'expérience des parents dans l'autisme grâce aux méthodologies qualitatives.

Le modèle de l'intervention médiée par les parents est largement plébiscité pour son impact positif sur le développement des jeunes enfants autistes, cependant peu d'étude ne s'intéressent aux effets « indésirables » ou négatifs de ce type d'intervention, alors que l'évaluation de la balance bénéfice-risque reste le point central de décision d'utilisation d'une intervention sur le plan clinique. Ici, nous recueillerons tout évènement indésirable associé à l'intervention, notamment sur le plan de la santé de l'enfant, mais nous avons construit un volet « parent » pour évaluer de façon plus fine l'impact du PACT sur les parents. Des mesures de stress parental et de qualité de vie sont prévues. Ces mesures seront complétées par l'étude qualitative. L'intégration des résultats quantitatifs et qualitatifs permettra d'analyser de façon complète l'expérience des parents suite à l'intervention.

Au total, si l'étude IFPAD est construite pour évaluer l'efficacité du PACT en visioconférence, son design est conçu pour apporter des informations plus globales sur les facteurs de mise en place de l'intervention dans son contexte ainsi que sur l'impact de ce contexte spécifique sur l'intervention et son résultat. Ces éléments seront apportés grâce à la complémentarité des mesures quantitatives anticipées dans ce but, mais également grâce à l'étude qualitative qui

permettra une mise en perspective des résultats quantitatifs, une meilleure compréhension de l'impact de l'intervention sur les participants mais également une triangulation des résultats quantitatifs et qualitatifs permettant d'analyser la validité des résultats.

## DISCUSSION GENERALE

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### 1. INTERET DE LA RECHERCHE QUALITATIVE DANS L'ÉVALUATION DES INTERVENTIONS NON MEDICAMENTEUSES

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Nous avons pu illustrer, avec quatre articles présentant des études d'évaluation d'intervention médiée par les parents, l'importance d'une évaluation complète, au-delà de la notion seule d'efficacité, mais surtout la place de la recherche qualitative dans cette évaluation globale. Ainsi, la recherche qualitative semble avoir toute sa place à différents niveaux du processus d'évaluation, de l'étude de faisabilité à l'étude d'implémentation. Elle peut aider à la compréhension des mécanismes sous-jacents à l'efficacité d'un programme, en explorant les expériences (des patients et des professionnels), la signification des résultats pour les participants, les effets indésirables du programme, les facteurs contextuels ayant pu affecter les résultats. Une étude qualitative peut être génératrice d'hypothèses, et l'interrogation de professionnels ou participants à une intervention pourrait, par exemple, permettre d'envisager a priori les modificateurs d'effet traitement afin de permettre, dans de futures recherches, leur prise en compte.

La recherche qualitative a également une place importante dans l'évaluation des facteurs d'implémentation, et permet notamment au chercheur de ne pas soumettre les informations récoltées dans une étude à la limite de leurs hypothèses a priori, mais de permettre aux experts d'une intervention (professionnels, patients) de s'exprimer et de décrire leur expérience dans sa globalité.

De façon satellite mais essentielle à l'évaluation de l'intervention, la recherche qualitative peut également être une étude préliminaire à la construction d'outils de mesure appropriés aux objectifs de la recherche. De nombreux exemples sont disponibles dans le champ de l'autisme. L'équipe de McConachie (2018) a par exemple proposé de réaliser une revue systématique des études qualitatives ayant interrogé les parents sur les cibles pertinentes pour les traitements dans le champ de l'autisme. Des groupes de travail ont ensuite été réalisés, avec les parents, pour classer par ordre de priorités, les résultats les plus importants à leurs yeux.

Concernant l'impact de l'autisme sur la vie de famille, une étude de 2018, a été réalisée par Kathy Leadbitter et son équipe. Celle-ci a regroupé des experts, décideurs et familles d'enfants

avec autisme, pour construire un outil d'évaluation adapté aux besoins des familles. Des focus-group ont été réalisés et analysés par une méthode qualitative. Les items résultant de cette première étape ont été proposés en ligne à un panel de répondant (parents d'enfants autistes ou experts), invités à noter leur clarté et leur utilité. Les caractéristiques psychométriques de l'outil ont par la suite été évaluées. Cet outil a permis d'évaluer l'expérience et la qualité de vie des familles dans les études précédentes sur le PACT (Leadbitter et al., 2018). Il est également utilisé dans l'étude IFPAD (Jurek et al., 2021).

Nous avons également proposé un travail se rapprochant d'une méthodologie mixte, dans cette même thématique, l'objectif de notre étude était de définir ce que serait un changement minimal cliniquement important des signes autistiques, sur l'échelle CARS (Childhood Autism Rating Scale) (Jurek et al., 2022). La CARS est un outil très répandu d'évaluation des symptômes de l'autisme, permettant d'apprécier le diagnostic mais également la sévérité. Cependant, il n'existe aucun consensus concernant ce que serait « un changement minimal cliniquement important » sur cet outil. Une méthode d'« expert elicitation » a été utilisée (O'Hagan, 2019). Cette méthode consiste à extraire les croyances d'un groupe d'experts sur une quantité inconnue d'intérêt et à représenter ses croyances par une distribution de probabilités. Ainsi, des experts du monde de l'autisme ont été réunis, formant un équivalent de focus group, guidés par le statisticien pour modéliser la quantité attendue. Ce travail a permis d'estimer un changement minimal cliniquement important pour la CARS. Cette estimation est particulièrement importante pour l'évaluation de l'effet des interventions et de leur impact réel sur la symptomatologie autistique, puisque l'affirmation de l'efficacité d'une intervention requièrent non seulement une significativité statistique, mais également une significativité clinique.

## 2. ÉVALUATION D'UNE INTERVENTION COMPLEXE, LE MODELE DU PACT

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Nous avons pu approcher, à l'aide des différents articles présentés, la complexité de l'évaluation des interventions. L'évaluation d'une intervention est un processus long, avec de nombreuses étapes essentielles pour s'assurer, d'un transfert efficace de l'intervention en pratique courante. La place de la recherche qualitative dans ce processus a ainsi pu être illustrée.

Les études autour du PACT (Aldred et al., 2004, 2012; Divan et al., 2015, 2019; Green et al., 2010, 2015, 2017, 2018, 2022; Pickles et al., 2015, 2016; Rahman et al., 2016; the PACT Consortium et al., 2015) sont une bonne illustration du modèle d'évaluation des interventions complexes (figure 1). Ces études aux méthodologies variées, ont permis d'avancer sur la compréhension du modèle d'intervention, dans différents contextes, avec son adaptation et son évaluation sur le plan de la faisabilité, l'efficacité, et l'implémentation (**Tableau 3**). Les résultats sur les obstacles à l'implémentation, ont pu permettre d'affiner les stratégies d'intervention pour les adapter aux contextes dans lesquels elles doivent être mises en œuvre et œuvrer à leur pérennité.

Nos études autour du PACT en visioconférence nous permettront également d'évaluer l'efficacité du PACT dans un nouveau contexte, avec l'adaptation préalable de la méthode, en conservant ses « ingrédients actifs ». Des équipes Danoise et Brésilienne travaillent également sur l'évaluation du modèle PACT, dans leurs populations respectives.

**TABEAU 3. REVUE DES ETUDES SUR LE PACT.**

Auteur, date	Date	Pays	Catégorie	Type d'étude	Populati on	Age en mois
Aldred, 2004	2004	Angleterre	Faisabilité, Efficacité	Etude pilote contrôlée randomisée évaluant le PACT versus TAU	TSA	29 à 60
Green, 2010	2010	Angleterre	Efficacité	ECR d'efficacité du PACT versus TAU	TSA	18 à 60
Aldred, 2012	2012	Angleterre	Mécanisme	Etude des médiateurs de l'efficacité du PACT	TSA	29 à 60
Divan, 2015	2015	Inde	Adaptation	Etude qualitative et serie de cas pour l'adapation du PACT en Inde	TSA	NA
Green, 2015	2015	Angleterre	Efficacité	ECR d'efficacité du PACT contre aucune intervention	A risque de TSA	7 à 10
Pickles, 2015	2015	Angleterre	Mécanisme	Etude des médiateurs de l'efficacité du PACT	TSA	NA
Byford, 2015	2015	Angleterre	Analyse économique	Evaluation économique du PACT versus TAU	TSA	18 à 60
Pickles, 2016	2016	Angleterre	Efficacité	ECR d'efficacité du PACT versus TAU, résultats à long terme	TSA	moyenne 126
Rahman, 2016	2016	Inde	Efficacité	ECR d'efficacité du PACT adapté, délivré par des non-spécialistes versus TAU	TSA	18 à 108
Green, 2017	2017	Angleterre	Efficacité	ECR d'efficacité du PACT contre aucune intervention, suivi à l'age de 3 ans des enfants à risque	A risque de TSA	36

<b>Leadbitter 2018</b>	2018	Angleterre	Validation d'outil	Construction et validation d'un outil d'évaluation de l'expérience des familles dans l'autisme et de leur qualité de vie	TSA	NA
<b>Divan, 2019</b>	2019	Inde	Adaptation, Faisabilité, Efficacité	Etude de faisabilité et d'efficacité d'une méthode PACT adaptée (ajouts de modules pertinents pour le contexte) en Inde	TSA	27 à 105
<b>Leadbitter 2020</b>	2020	Angleterre	Expérience patient	Etude qualitative de l'expérience des parents ayant suivi la thérapie PACT dans le cadre d'une étude contrôlée randomisée	TSA	18 à 60
<b>Jurek, 2021</b>	2021	France	Efficacité, expérience patient	Protocole d'une ECR d'efficacité du PACT en visioconférence versus TAU	TSA	18 à 36
<b>Green, 2022</b>	2022	Angleterre	Efficacité	ECR d'efficacité du PACT délivré à la maison et à l'école versus TAU	TSA	18 à 132
<b>Godoy, 2022</b>	2022	Brésil	Efficacité	Protocole d'une ECR d'efficacité du PACT versus TAU	A risque de TND	18 à 48
<b>Bilenberg, 2023</b>	2023	Danemark	Efficacité	Protocole d'une ECR d'efficacité du PACT versus TAU	TSA	18 à 72
<b>Jurek, annexe 1</b>	2023	France	Faisabilité	Etude de faisabilité du PACT en visioconférence	TSA	moyenne 37,7
<b>Geoffray, annexe 2</b>	2023	Mutiple	Implémentation	Etude de l'implémentation du PACT	TSA	NA
<b>Carruthers 2023</b>	2023	Angleterre	Mécanisme	Etude des médiateurs de l'efficacité à 6 ans du PACT	TSA	moyenne 126

Ce travail collaboratif entre l'équipe à l'initiative du PACT et des équipes de recherche au niveau mondial, a permis d'avancer sur la compréhension de l'intervention et son applicabilité dans différents contextes. Un schéma est proposé en **figure 3** pour illustrer ce propos.

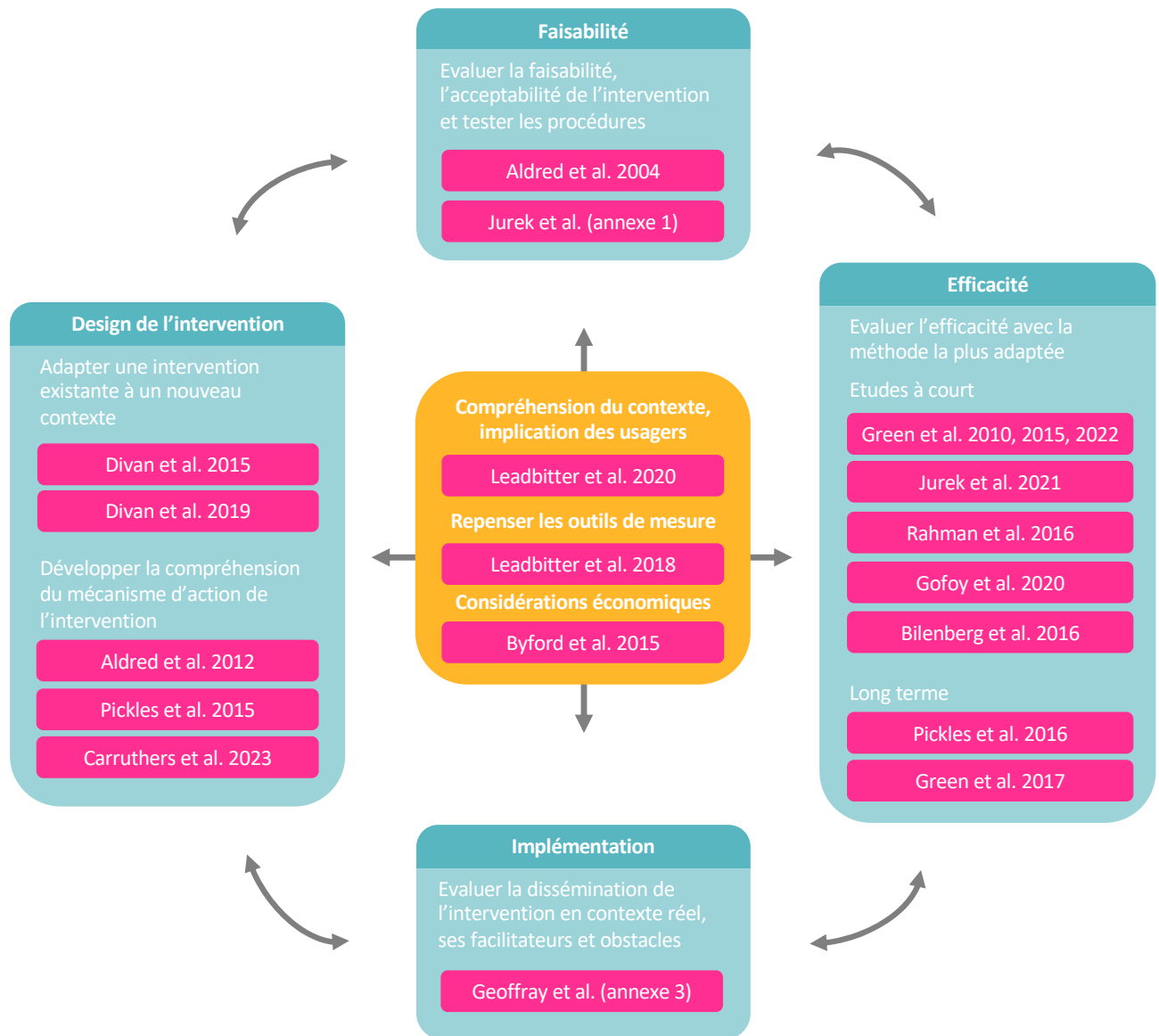


FIGURE 3. LE PACT, AU SEIN DU MODELE DE L'EVALUATION DES INTERVENTIONS COMPLEXES.

### 3. UNE QUESTION DE PARADIGME ?

L'association de la recherche qualitative et la recherche quantitative est parfois difficile. L'association de ces deux « mondes différents » de la recherche peut souffrir d'une difficulté de compréhension entre chercheurs « quantitativistes » et « qualitativistes ». La question des paradigmes de recherche, évoqués brièvement dans la partie introductive sur la recherche qualitative, est probablement à l'origine de cette difficulté.

En recherche, les paradigmes sont des cadres conceptuels ou des perspectives théoriques qui guident la manière dont les chercheurs abordent leurs sujets d'étude, posent des questions de

recherche, collectent des données et interprètent leurs résultats. Chaque paradigme repose sur des croyances fondamentales sur la nature de la réalité, la connaissance, la vérité et la manière dont la recherche devrait être menée. Ces paradigmes sont nombreux et évolutifs sur le plan historique. Sans volonté d'exhaustivité, le tableau 4 présente différents paradigmes (Hall, s. d.). Il est important de noter que ces paradigmes ne sont pas mutuellement exclusifs, et de nombreux chercheurs adoptent une approche interdisciplinaire qui peut combiner des éléments de plusieurs paradigmes. Le choix d'un paradigme dépend de la question de recherche, du domaine d'étude et des croyances philosophiques du chercheur.

Certains paradigmes offrent des conceptions de la recherche diamétralement opposées (par exemple le positivisme et le constructivisme), ces conceptions sont ainsi difficiles à associer au sein d'un travail de recherche global. Dans ce contexte, les recherches sont menées en parallèle, et l'intégration de données issues des différentes recherches peuvent être intégrées par la suite mais n'est pas initialement anticipée lors de la construction de la recherche. La recherche sur l'évaluation de l'efficacité des interventions a longtemps été positionnée dans un paradigme positiviste, tandis que les méthodes qualitatives ont été initialement définies comme constructivistes (Lachal, 2016).

Pour autant, l'objectif d'une recherche avec des méthodes mixtes est de fournir une compréhension plus complexe d'un phénomène qui n'aurait pas été accessible en utilisant une seule approche. Cette position convient que des méthodes de recherches différentes (notamment qualitatives et quantitatives) peuvent être complémentaires, pour pallier les limites de l'autre. Le réalisme critique, par exemple, propose cette intégration. Les réalistes critiques estiment que les théories ne peuvent être que des représentations impartiales de la réalité. L'objectif de la recherche réaliste critique est de mesurer et de vérifier les structures sous-jacentes de la réalité, tout en gardant à l'esprit que l'objectivité ne peut être qu'approximative, car influencée par de nombreux facteurs externes. La prise en compte du paradigme des chercheurs dans la planification d'un projet de recherche mixte, est donc un point important pour faciliter l'intégration des données et éviter un travail mené en parallèle, sans possibilité de réelle collaboration (Hannes & Lockwood, 2011b; O'Cathain et al., 2010).

**TABEAU 4. EXEMPLES DE PARADIGME EN RECHERCHE.**

Paradigme	Définition
Le réalisme	Les chercheurs qui adhèrent au réalisme croient que la réalité existe indépendamment de notre perception et de notre compréhension. Ils visent à découvrir des lois et des faits objectifs, souvent en utilisant des méthodes empiriques pour observer et mesurer la réalité telle qu'elle est. Le réalisme suppose que la vérité est objective et découvrable.
Le réalisme critique	Cette perspective reconnaît que la réalité existe indépendamment, mais elle insiste également sur le fait que notre compréhension de la réalité est influencée par des structures sociales, politiques et économiques.
Le constructivisme	Les chercheurs constructivistes estiment que la réalité est socialement construite et qu'elle dépend des perceptions et des interprétations individuelles et collectives. Ils s'intéressent à la manière dont les individus construisent leur compréhension du monde et comment cette compréhension est influencée par des facteurs culturels, sociaux et historiques.
Le positivisme	Ce paradigme se concentre sur l'objectivité et la mesure précise. Les chercheurs positivistes recherchent des faits empiriques et des lois générales qui peuvent être établis de manière rigoureuse. Ils évitent les spéculations et les jugements de valeur.
L'interprétativisme	Les chercheurs interprétativistes estiment que la réalité est subjective et qu'elle doit être comprise à travers les expériences et les interprétations des individus. Ils utilisent souvent des méthodes qualitatives pour explorer les significations et les perceptions des participants.
Le post-modernisme	Les chercheurs postmodernistes mettent en doute la notion de réalité objective et la vérité universelle. Ils se concentrent sur la diversité des perspectives, la multiplicité des réalités et l'importance de remettre en question les récits dominants.

## CONCLUSION ET PERSPECTIVES

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La recherche qualitative ne prétend pas être le seul outil méthodologique utile dans l'évaluation des interventions. Il s'agit néanmoins d'une méthode complémentaire aux recherches quantitatives, qui permet d'obtenir des informations essentielles, là où les méthodes quantitatives ne sont pas suffisantes. Nous avons vu que dans le processus d'évaluation des nouvelles interventions non médicamenteuses la recherche qualitative a une place de choix, à différents niveaux, tels que la faisabilité, l'implémentation, l'expérience patient, la compréhension des mécanismes de l'intervention...

Nous nous sommes focalisés dans cette thèse, principalement sur l'intervention médiée par les parents, et notamment l'intervention PACT. Le champ de l'intervention dans l'autisme est néanmoins bien plus vaste et de très nombreuses interventions sont proposées. Les chercheurs proposent de nombreuses études sur les différents modèles. Habituellement, les équipes fondatrices d'un modèle proposent une évaluation de l'efficacité, de leur modèle, contre le traitement habituel dans la communauté, mais la réplique dans d'autres contextes n'est pas toujours réalisée. De plus, l'évaluation des interventions contre d'autres types d'intervention (en dehors du traitement dit habituel) n'est jamais réalisée.

La méthodologie des méta-analyses et notamment de la méta-analyse en réseau pourrait permettre de combler cette lacune. Puisqu'il existe une multitude de programmes d'intervention dans le champ de l'autisme, proposés par des équipes différentes, mais au contenu parfois similaire d'un programme à l'autre. Il serait particulièrement utile de synthétiser l'efficacité des interventions selon leur contenu, et de comprendre les composantes utiles de ces interventions ainsi que l'impact de leur interaction. Le modèle de la « component network meta-analysis » (méta-analyse en réseau des composants) permettrait de réaliser ce travail.

D'autres méthodologies auraient un intérêt pour synthétiser, de façon objective, l'efficacité de ces différentes interventions complexes, en conservant la prise en compte du contexte. Des méthodes de revue systématique, avec ou sans méta-analyse, intégrant des données mixtes (revues réalistes, revues systématiques mixtes), permettent également de balayer le large éventail des interventions et d'intégrer de façon systématique l'efficacité des interventions,

l'expérience des patients et des professionnels au sein de ces différentes interventions et ainsi que le contexte spécifique de l'intervention.

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ANNEXE 1 : FEASIBILITY OF A VIDEOCONFERENCING-BASED PARENT-MEDIATED INTERVENTION: A MIXED-METHOD PILOT STUDY

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**Feasibility of a videoconferencing-based parent-mediated intervention: a mixed-method pilot study**

Soumis à **Research in Autism Spectrum Disorder**

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## **ASBTRACT**

**Background:** Autism Spectrum Disorder (ASD) presents early communication and social challenges, necessitating timely and accessible intervention. Preschool Autism Communication Therapy (PACT), a parent-mediated intervention, empowers parents to facilitate their child's development. However, accessibility issues often hinder families from accessing evidence-based intervention. This pilot study assessed the feasibility of videoconferencing-based PACT as a precursor for a multicenter randomized controlled trial.

**Methodology:** A mixed-methods approach integrated quantitative retrospective measures and semi-structured interviews. Participants included children diagnosed with ASD who received PACT, and PACT-trained professionals with videoconferencing experience. Feasibility was assessed through audio and video quality, internet stability, and session length. Professionals' experiences were analyzed using a qualitative thematic analysis. Autism severity, parent-child interaction, and therapeutic changes were also described.

**Results:** Nine parent-child dyads and eight PACT therapists were included in the study. Videoconferencing-based PACT intervention proved feasible, with 95.1% of the 41 sessions rated as feasible on the scale. Technical challenges such as audio quality (7.3%) and screen sharing (19.1%) have emerged, which therapists circumvented to maintain intervention quality. Autism severity and parent-child interaction showed positive trends, supported by qualitative findings reporting increased parental confidence and enhanced synchrony. The core components of PACT were successfully adapted to the remote setting.

**Conclusion:** This pilot study suggests that delivering PACT via videoconferencing is a feasible approach to enhance the accessibility of evidence-based interventions for ASD. Larger-scale research with rigorous controls is required to validate these promising findings. An ongoing multicenter randomized trial aims to address this gap.

## **KEYWORDS**

Autism Spectrum Disorder; parent-mediated intervention; PACT; videofeedback; mixed-methods

## **1. INTRODUCTION**

Early intervention is essential in Autism Spectrum Disorder (Lord et al., 2022). Recent systematic reviews have emphasized the efficacy of developmental methods (Trembath et al., 2023). Because parents are closely connected to their children and play a pivotal role in their development, certain developmental approaches, such as Preschool Autism Communication Therapy (PACT), focus on guiding parents in learning strategies and enacting the approach in daily life (Green et al., 2010; Nevill et al., 2018; Pickles et al., 2016).

Unfortunately, families often lack access to evidence-based interventions, with an average three-year delay in starting early intervention services after diagnosis (Yingling et al., 2018). Several barriers exist, including long waitlists, distant locations from specialized centers, lack of transportation, and parental time constraints (Jurek et al., 2022). These challenges have hindered the implementation of parent-mediated interventions.

Regarding these different barriers, videoconferencing appears to be a promising method for enhancing access to PACT. Nevertheless, it is crucial to assess the feasibility of these programs to ensure that the quality of the intervention is not compromised when it is delivered via videoconferencing. Moreover, given the low attrition rates in intervention programs (Chacko et al., 2016), barriers and facilitators of implementation should also be evaluated.

Therefore, the main objective of the present pilot study was to evaluate the feasibility of videoconferencing PACT in the perspective of undergoing a multicenter randomized controlled study (Jurek et al., 2021).

## **2. METHOD**

The study employed a mixed-methods approach, combining retrospective quantitative measures and semi-structured interviews to assess both the feasibility and the barriers and facilitators of the remote intervention, and its effects on child development and parent-child interactions. If this study did not aim to assess the effectiveness of the intervention, it was important to observe the progress of the parent-child dyad throughout the intervention in this new context of video conferencing (Smith et al., 2007).

### *2.1. Participants*

For the feasibility part, professionals trained in PACT were included if they had at least one experience of PACT using videoconferencing.

For the parent-child observations, children who received a diagnosis of ASD in our specialized unit according to DSM-5 and who received PACT intervention until January 2021 were eligible in our study. They were included if they received at least 12 sessions of PACT and if half of the sessions were proposed remotely.

The study was approved by the University of Lyon Institutional Review Board (IRB 2021-01-12-01) and the parents received written information.

### *2.2. Intervention*

PACT is a parent-mediated therapy in which professionals guide caregivers to identify behaviors that promote communication and reciprocity with their child through short videos of the parent and child playing together (video feedback). Typically, PACT therapists and parents meet each other in person. A video of parent-child play is recorded, and then guidance begins based on the video (in-center setting) (for more details, see Aldred (Aldred et al., 2004) and Green (Green et al., 2010)).

Parents received a total of 12 intervention sessions within this time frame: one session every 15 days for six months and were asked to use PACT strategies in daily life for half an hour.

For the remote PACT, before each session, parents sent a video of a 10-minute playtime with their child to the therapist on a secured cloud. During the remote session, the professional screen shared and used this video to guide the parents through PACT strategies.

### *2.3. Measures and procedures*

#### *2.3.1. Feasibility*

The feasibility of the videoconferencing intervention was rated on videos of the recorded remote therapy sessions between parents and professionals. Six videos were randomly selected during each follow-up session. Five items were evaluated: quality of the audio, quality of the video, stability of the Internet network, possibility of sharing and simultaneously watching the parent-child video with the parent, and total length of the session. An item can receive 1–4 points. A session was considered “feasible” if the score on the scale was equal to or greater than 15.

#### *2.3.2. Autism severity and parent-child interaction assessment*

Every parent-child interaction video sent by the parents to the therapist was saved under a code name with a random number from 1 to 12. Every therapy session was registered (with the parent agreement) and saved on a secured cloud.

To evaluate autism symptoms, the Brief Observation of Social Communication Change (BOSCC)(Grzadzinski et al., 2016) was used and rated by a trained clinician (AJ), and the Clinical Global Impression–Severity (OSU-CGI-S) (OSU Research Unit on Pediatric Psychopharmacology, 2005) was rated by two trained clinicians (MB and GB) blinded to the therapy progression. Clinical Global Impression-Improvement (OSU-CGI-I) (OSU Research Unit on Pediatric Psychopharmacology, 2005) was rated by a trained clinician (LJ) independent of the two other clinicians. The video was rated in order of dates for the rating of the CGI-I. The clinician rating CGI-I was not blinded to therapy progression.

To evaluate changes in parent-child interaction, the Dyadic Communication Measure for Autism (DCMA) (Aldred C. et al., 2012) was rated by a trained research assistant (SD).

#### *2.4. Statistical analysis*

Statistical analyses were conducted using JASP software (JASP Team, 2023). For continuous outcomes, means with standard deviations were calculated. The percentages were calculated for dichotomous outcomes. As this is an exploratory study, no sample size was calculated prior to the study, and the results presented here are only descriptive.

We considered that at least 80% of the recorded debriefing sessions should obtain a score of 15 or more for videoconferencing intervention to be considered feasible.

#### *2.5. Qualitative study*

A qualitative study was conducted to understand the experiences of PACT therapists in delivering PACT through videoconferencing. We aimed to explore their overall experience and identify both barriers and facilitators of videoconferencing-PACT.

Data were collected through semi-structured interviews with the PACT therapists, by a master student, trained in the interview method. The interviews lasted 30–60 minutes and were audio-recorded, transcribed verbatim, and anonymized.

A thematic approach was used to analyze the data (Guest et al., 2012). All original recordings and transcriptions were analyzed in French. First, interview transcripts were read multiple times to allow researchers to familiarize themselves with the data. The transcripts were then hand-coded, which

allowed for the organization of recurring ideas into broader categories. Two researchers (MMB and LJ) independently coded the interviews using NVivo software (version 12) (QSR International Pty Ltd., 2020). Each interview transcript was repeatedly analyzed until no additional categories could be identified by the researcher. The categories created through this process served as the basis for the study's themes and sub-themes. Discussions were conducted whenever there were disagreements in the coding process until a consensus was reached. A third research expert (MMG) was invited to check and revise the extracted codes, categories, and subcategories to ensure data triangulation. The main results were sent to professionals interviewed for their expert opinion. The themes and subthemes were then interpreted to reflect the key ideas underlying the participants' responses in their interviews. The coders selected representative quotations to present the categories or sub-categories.

The French results were translated into English and back-translated into French to ensure consistency.

#### 2.5.1. Reflexivity

The research team for the qualitative analysis consisted of three child and adolescent psychiatrists, two of whom specialize in autism. Two researchers were trained in the qualitative research. The third researcher clarified the results using the scientific literature.

### 3. RESULTS

#### 3.1. *Children population*

Nine children (one girl and eight boys) were eligible and included in the study. The mean age was 37.7 (+/- 8) months at the beginning of the therapy. The time between ASD diagnosis and PACT was between zero and 16 months. Of the nine children, seven were engaged in speech therapy and one in motor therapy. Six children had severe or moderately severe ASD symptoms, two had moderate ASD symptoms and one had minimal ASD symptoms.

#### 3.2. *Therapist population*

Eight therapists were included in the qualitative study. They were all women. Three were working as speech therapists, two as nurses, one as a psychologist, one as a psychomotor therapist, and one as a child and adolescent psychiatrist.

Six of them had been using PACT for two years, and two had 4-years of experience. A mean of 4.25 families by therapist (range 1-9) had been trained in PACT.

### *3.3. Feasibility*

From an expected total of 54 intervention videos, 41 videos were retrieved and rated. A score greater than 15 was obtained for 95.1% of the videos (Figure 1).

Difficulties were observed concerning the quality of the audio in 7.3 % of the videos with an audio quality rated as “poor” and difficulties for the parent and the therapist to hear each other, or delay in the responses. In 19.1% of the sessions, screen sharing of the parent-child video was not possible, and the parents and therapists had to look at the video separately at the same time. Only 2.0% had poor video quality. The PACT sessions were shorter than in center setting with 37% of sessions lasting between 30 and 45 minutes, and 5% of the sessions less than 30 minutes.

### *3.4. Changes during intervention*

Changes in autism severity and child-parent interaction are presented in Table 1.

Concerning the measure of global change with the CGI-I, one child was considered very much improved, two were considered much improved, three were considered minimally improved, and two had no change in global symptomatology.

### *3.5. Barriers and facilitators of videoconferencing intervention: professionals' point of view*

Our thematic analysis yielded four main themes. Themes and subthemes are presented in Table 2 and illustrated with therapists' quotes.

#### *3.5.1. Positive changes on many levels*

Therapists explained that PACT has a multifaceted impact on parents, children, and therapists alike. PACT boosts parental confidence, particularly in their parenting skills. Therapists observed a shift in parents' position during interactions with their children, characterized by increased synchrony and a deeper connection. They described PACT as a means for parents to dedicate a specific time to their children. PACT appeared to enhance parents' ability to adjust their expectations to their child's unique rhythm.

For therapists, PACT serves as a transformative tool that positions them as facilitators of change in a fulfilling manner. Through this therapy, therapists gain insights into better tailoring their approach to

match each parent's individual profile, cultivating a clinical perspective centered on interaction and its evolution.

Remarkably, the impact on child behavior extends beyond mere play, yielding significant and lasting results within a relatively small number of sessions.

### 3.5.2. Intervention may be affected by certain factors

Therapists claim that the success of interventions depends on multiple factors. An increase in flexibility throughout PACT's stages would significantly improve the program's ability to cater to the unique requirements of parents and children. Some stages may not align perfectly with the child's needs and could potentially lead to a decrease in parental motivation.

Similarly, complementary techniques may also be necessary to address unique challenges. Some parents may require assistance with aspects that are not directly addressed by PACT, which can occasionally interfere with the effectiveness of PACT therapy, thus necessitating support from other sources.

The impact of a parent's individual profile on the treatment process should not be underestimated as it requires further adaptation from the therapist. Furthermore, the allocation of homework time may sometimes act as an obstacle to the progress of the intervention, highlighting the importance of considering these practical aspects when implementing PACT.

### 3.5.3. Videoconferencing, a tool that can be implemented under certain conditions

Videoconferencing, while a valuable tool, can be successfully implemented under specific conditions according to therapists' experience.

The advantages of videoconferencing for intervention implementation are clear, as it enables effective therapy delivery. It presents a more ecological approach that allows parents to participate in a familiar environment, where they can feel more secure but also reduces the need for physical travel and can better align with parents' schedules. Therapists have confirmed the possibility of therapeutic relationships using videoconferencing.

However, the success of interventions through videoconferencing is contingent upon addressing technical aspects, ensuring a feasible connection. On the professional side, implementing PACT through videoconferencing demands greater organization but allows the therapist to anticipate and prepare the sessions well.

#### 3.5.4. The essential components of PACT

PACT was described as a useful therapy when the essential components are respected. Implementing a PACT intervention successfully requires a great deal of thought and analysis of the professional's part to maintain fidelity to the PACT model. Therapists have highlighted the importance of the PACT structure and the intervention schedule, which serves as a framework for consistent progress and seems to be acceptable for both parents and professionals. The use of video feedback has also been described as a crucial element in PACT, facilitating parents to progress at their own pace and empowering them to independently identify acceptable solutions to aid in their child's development. Therapists encourage parents to apply the intervention's objectives in their day-to-day lives rather than limit them to the prescribed 30-minute homework sessions, aiming to enhance acceptability while maintaining consistency in the work. They also agreed that PACT requires significant parental investment. Therapists underlined the importance of adequately preparing parents for their participation in the PACT intervention.

## 4. DISCUSSION

Our findings suggest that PACT intervention conducted through video conferencing is feasible even in the face of technical issues reported during feasibility assessments and interviews. The therapist reported some difficulties due to poor audio quality on some computers but also in watching the video together with the parent by sharing their screen. Nevertheless, the qualitative study indicated that therapists found different ways to overcome the technical difficulties in preserving the quality and main components of the intervention. The sessions were shorter than those in the center setting. This result is mainly explained by the organization of PACT in videoconferencing, where the parent-child video was recorded and watched by a professional once before the videoconferencing session.

The main component of PACT was adequately transposed to the remote PACT. The establishment of a therapeutic relationship via video conferencing was successful; therapists and parents worked to improve parent-child synchrony, a main moderator of PACT efficacy, using video feedback. This highlights the adaptability of therapeutic interactions to remote platforms, thereby expanding the accessibility of interventions, such as PACT. The remote format also provided ecological benefits, such as tailoring interventions to parents' needs and minimizing logistical challenges.

Child development and parent-child interactions showed positive changes in various aspects, as supported by the qualitative analysis results. These findings highlight the multiple benefits of PACT, such as enhancing parents' self-assurance in parenting and making professionals' work more impactful. The findings align with previous studies on parent-mediated interventions (Jurek et al., 2022; Leadbitter et al., 2020; Pickard et al., 2016).

However, this study has some limitations. The absence of a control group and the small local sample size are notable constraints that reduce the generalizability of the findings.

Concerning the qualitative study, the use of a convenience sample is a limitation as we had a limited number of participants. Nevertheless, no new themes were found after the 6<sup>th</sup> interview, and our population had sufficient variation to cover multiple experiences with PACT.

## **5. CONCLUSION**

This preliminary study suggests that conducting PACT intervention via video conferencing is feasible despite some technical concerns. The observed enhancements in child development and parent-child interactions and the positive outcomes developed in the semi-structured interviews offer encouraging implications for remote interventions. Further research with larger sample sizes and more rigorous control groups is required to confirm these results. A multicenter randomized trial is planned for this matter (Jurek et al., 2021).

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## **Conflict of Interest and Sponsors' Role**

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**FIGURE 1. Technical feasibility of videoconferencing-PACT.**



**TABLE 1.** Changes in autism severity and in parent interaction during intervention for the nine children included. *SD = Standard Deviation; BOSCC = Brief Observation of Social Communication Change; DCMA = Dyadic Communication Measure in Autism*

<b>Scales</b>	<b>Mean at baseline (SD)</b>	<b>Mean at mid-therapy (SD)</b>	<b>Mean at endpoint (SD)</b>
<b>BOSCC</b>	25.5 (10.4)	27.3 (12.5)	20.5 (13.6)
<b>DCMA parent-child synchrony</b>	27.6 (10.7)	32.9 (18.8)	33.1 (10.5)
<b>DCMA child communicative initiation</b>	7.5 (12.0)	15.8 (12.6)	16.9 (17.2)
<b>DCMA mutual shared attention</b>	76.0 (12.7)	77.3 (19.1)	86.6 (15.1)
<b>CGI-S</b>	4.7 (1.3)	4.7 (1.4)	4.2 (1.4)

**TABLE 2. Results of the qualitative study: Therapists' experience with video-PACT.**

Themes and subthemes	Traduction
<b>Positive changes on many levels</b>	
Increases parents' confidence in their abilities	« It's a valuable support, because it serves beyond the communication goals, which are extremely important, but it also serves parents to increase a sense of effectiveness. »
Change in parent's position in interaction with the child	« I have seen significant behavioral changes in the way parents interact with their children, i.e. being able to adjust more to their child. »
Allows parents to take time specifically for their child	« Understanding autism for their child on a truly individual level, taking time for their child and truly being there for them. »
PACT enables parents to adjust their expectations to the child's rhythm.	« It has also enabled them to understand [...] how their child interacts specifically and to adapt to their child's pace. This is extremely important. And revising their expectations and the pace they set for him, so they can really adapt.»
The therapist's position as a facilitator of change is a satisfying one.	« For me, it has really taught me to question people in a different way, not to tell people what I think is good, but for them to say it and for it to come from them. And I find that empowering as a professional. »
PACT enables the therapist to learn to better adapt to the parent's profile.	« In terms of psychoeducation, it allows for better adaptation to the parents' profile: their communication profile, their interaction profile, and being more attentive. »
PACT enables the therapist to develop a clinical viewpoint more focused on the interaction and its development.	« From a professional perspective, it has taught me a lot in terms of clinical skills, interview management, and sensitivity in observing communication. »
Impact on the child, beyond play	« It unlocked many things [initiation] in these individuals, even though initially, we were just playing games, it had a ripple effect on their whole lives. »
Significant and persistent results for a small number of sessions	« And for the family, I think that for the number of sessions we do, it's very powerful. [...] there are a lot of results for twelve sessions. »
<b>Intervention may be affected by certain factors</b>	

<p>A higher flexibility in the different stages of PACT could allow for better accommodation to parents' and children's needs</p> <p>Complementary techniques may be required</p> <p>Impact of the parent's profile on treatment</p> <p>Homework time as an obstacle to intervention</p>	<p>« In the current method, you are supposed to follow step 1, step 2, step 3, etc., in order. I think we could offer a little more flexibility [based on] what we believe needs to be worked on first. »</p> <p>« For me, as a therapist, PACT cannot address everything. It primarily focuses on the social interactions between the parent and child dyad. So, first of all, a PACT approach is interesting, but it will have to be followed up by other types of intervention. »</p> <p>« And then, there are people who struggle to answer questions. Some find it difficult to talk extensively to express their thoughts. For some, especially those in denial, it can be challenging. »</p> <p>« I find that families still seem to have difficulty integrating it into their daily lives, based on what they tell us. It's more like, 'Oh yes, I played with him, but I didn't necessarily think about it.'»</p>
<p><b>Videoconferencing, a tool that can be implemented under certain conditions</b></p>	
<p>More ecological intervention with video</p> <p>The benefits of videoconferencing for intervention implementation</p> <p>Therapeutic relationship possible in visio</p> <p>Intervention by videoconference is dependent on technical aspects but remains feasible</p> <p>PACT through videoconferencing allows the therapist to anticipate and prepare sessions well</p>	<p>« While here, we are in their living environment, with their parents, and with the games that the parents have selected. So, it's incredibly interesting because it really provides a better understanding. It almost gave me the impression of being more effective. »</p> <p>« In fact, my main point is that it makes people's lives easier. Because they don't have to travel, for people who work, you can squeeze them in between. It only takes an hour, in the end; whereas otherwise, they've got the journey, they've got everything.... »</p> <p>« If the connection works well, these relationships start directly through video. In fact, we manage to create a good therapeutic alliance despite everything. »</p> <p>« Yes, bugs can happen. I've had a few, especially with screen sharing [...] But in the end we always manage to get by. For example, the mom watched the video the minute I told her to. »</p> <p>« And I think it's a good idea to have the time to see the video sent by the parent beforehand, to prepare the session properly, than when it's live, when you film the child straight away and then go over the film with the parent. »</p>
<p><b>The essential components of PACT</b></p>	

Requires a great deal of thought and analysis on the professional's part	« So it's care that's... I was going to say "tiring", but it requires professionals to put a lot, a lot of thought and analysis into getting parents to reflect. »
Importance of PACT structure and intervention schedule	« For parents, it's somewhat the same. That is, it's also structured, they know where they're going, we always do a bit of the same thing, and in this routine, they have the freedom to think about their relationship with their child. »
Importance of video feedback	« There's the impact of the video, which is inherently very objective, so they see themselves in action, and it's very powerful, the video... I was already using it before, but now, showing them the positive moments, it's... yes, it's really strong for the parents, I think. »
Implementing the objectives in day-to-day life rather than in the daily 30-minutes homework	« It's important to emphasize what these thirty minutes should be, in fact. To really provide ideas on how they can integrate it into their daily live. »
Requires significant parental investment	« The family has to invest in it, and people have to want to come, otherwise it doesn't work. »
Importance of preparing parents for PACT intervention	« It has to be care that's well, well explained to parents beforehand, because often what parents want is for us to take care of their child, not them. The introduction of this treatment should also be done in a very specific and well-structured framework, because otherwise, you can quickly find yourself in the first session with parents who haven't understood that we will never intervene directly on their child. »

ANNEXE 2 : PARENT AND PROFESSIONAL PERSPECTIVES ON  
IMPLEMENTING THE PAEDIATRIC AUTISM COMMUNICATION  
THERAPY (PACT): A MIXED-METHODS ANALYSIS.

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**Parent and professional perspectives on implementing the  
Paediatric Autism Communication Therapy (PACT): a mixed-  
methods analysis.**

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## **Abstract**

**Background:** Factors influencing the implementation of parent-mediated intervention are insufficiently studied and often omit the parental perspective. This study examines barriers and enablers of implementation into clinical practice across the world of Paediatric Autism Communication Therapy (PACT), an internationally disseminated, evidence-based, parent-mediated therapy.

**Methods:** Online survey data was obtained separately from (a) professionals who had received PACT training and (b) parents receiving PACT intervention from those professionals. Analysis was cross-sectional and mixed methods, informed by the Consolidated Framework for Implementation Research (CFIR)

**Results:** Over 70% of practitioners recorded intent to implement the therapy after PACT training. Implementation was more likely if the practitioner saw many autistic children within their work ( $OR \approx 5$ ) and was easier with new referrals. Other key facilitators to implementation were peer support from colleagues and managers, allowing scheduling flexibility. The main barriers were time (intervention sessions  $>1$  hour), suitable video equipment, low parent self-reflection abilities, stressed family conditions or support, and significant presenting concerns about child behaviour. Parents recorded high levels (scored 1 to 7) of acceptability (Median 5.7 [IQR 5.1,6.2]) and feasibility to learn and implement PACT in daily life (median 5.4 [4.5, 6.8]) and delivery a median of 4 (IQR [2,7]) of the planned 7 daily home-based practice sessions during received intervention. The facilitators to implementation for parents were positively-regarded features of the PACT method (video reflection, empowerment of parent, play-based practice). Barriers were time availability (e.g., for home practice) and difficulties with video-recording.

**Conclusion:** This study of factors influencing implementation of an evidence-based intervention benefits from the integration of parents' and professionals' views within wider systems context. The results suggest the importance of family- and system-context factors in implementation success.

## **Lay Abstract**

Factors influencing the implementation of parent-mediated intervention are insufficiently studied. This study examines for the first-time factors of implementation into clinical practice across the world of Paediatric Autism Communication Therapy (PACT), an internationally disseminated, evidence-based, parent-mediated therapy.

From both the parents' and the professionals' perspectives, parents attitudes ( e.g :self-reflection abilities, videotaping management) and life-circumstances (e.g: stressed families condition, time) were central factors. In this type of therapy, the target is a child's development; still, this improvement is dependent on the parent's behaviour when interacting with their child.

What's more, PACT method (video reflection, empowerment of parent, play-based) was enabler according to most of the parents.

And more, most professionals report factors linked to the Implementation Process that is planning, execution, reflection, assessment in implementation of a new therapy. Indeed, the professionals underlined barriers related to the population seen in practice, flexibility of schedule, support from colleagues and manager.

All these factors could be improved and addressed with a formal implementation plan including factors related to the parents of each country.

**Keywords:** autism, implementation, parent-mediated therapy, intervention, professional training

## **Background**

There is a pressing need for evidence-based interventions that bring long-term functional improvement in the quality of life of autistic individuals and their families and that are implementable in health systems around the world (Green & Garg, 2018). Parent-mediated interventions (PMI) are increasingly used in this field. Paediatric Autism Communication Therapy (PACT) is one of the few well-evidenced PMIs that have demonstrated sustained effectiveness on blinded distal outcome measures and beyond immediate effect on dyadic interaction (Pickles et al., 2016). Such evidence-based practice (EBP) is recommended by national clinical guidelines (e.g., NICE, 2021) yet is not widely implemented. Optimizing the implementation of such effective treatments is a defining challenge of our time; thus, research into identifying factors that can improve, or impede, implementation of such EBP PMI could provide helpful insights.

There are many steps to achieve a high level of implementation and to ensure that innovations become standard practice that is sustainable over time. Even if an intervention is initially deemed to be appropriate and acceptable, many barriers and enablers can arise at multiple levels of healthcare implementation. The Consolidated Framework for Implementation Research (CFIR) considers such implementation factors at the level of **Intervention Characteristics** (details about the specific intervention and delivery strategies), **Individual Practitioner** (individual understandings such as attitudes toward evidence-based practice, available time etc.), **Inner Setting** (“features of structural, political and cultural contexts through which the implementation process proceeds”);, **Outer Setting** (“economic, political and social context including patient needs and resources within which an organisation resides”) and **Implementation Process** (“active change process to achieve individual and organisational level use of the intervention as designed”) (Damschroder et al., 2009). A combination of these factors can have a positive or negative influence on the decision of professionals to initiate, and continue to deliver a therapy with patients (Tabak et al., 2012, Williams & Beidas, 2019).

**Intervention Characteristics** PACT is a internationally disseminated therapy, and as such, it presents a good model for investigating barriers and enablers of implementation accounting for cultural contexts (Aldred & Green, 2019; French & Kennedy, 2018; Green & Garg, 2018) . PACT uses video-feedback strategies which have shown effectiveness in improving parent awareness and synchronous responses to the child and in turn child communication initiations with the parent (Aldred et al., 2004, 2012; Green et al., 2010; Rahman et al 2015; Divan et al 2017; Pickles et al., 2016). Increased child dyadic initiations of this kind, in turn, improve downstream child communication and social interaction and behaviours in development, sustained from therapy endpoint for next 6 years into middle childhood (Pickles et al 2015, Pickles et al., 2016, Carruthers et al 2023). The therapy lasts six to twelve months, with typically one 1.5-hour therapist session with the parent every fortnight. During this session, the therapist helps the parent to identify their communication behaviours that promote synchrony with their child through short videos of the parent and child playing together. The therapist encourages the parent to use the strategies identified during the sessions to promote parent-child synchrony in daily activities at home outside the sessions.

Videos of the child and the parent playing can be filmed by the therapist during in-person sessions or by the parents themselves when the sessions are delivered by videoconference. The manualised therapy proceeds through sequential, developmentally informed foci, towards optimised child social communication and autistic behavioural outcomes. The PACT intervention supports parents while valuing and affirming neurodiversity in that it promotes parental awareness and their best adaptation to the child's difference, rather than an agenda towards behavioural normalisation. Professional PACT training is delivered worldwide, delivered by a not-for-profit Community Interest Company (IMPACT; Interaction Methods of Paediatric Autism Communication Therapy; <https://www.pacttraining.co.uk/>). Professionals are trained via a preliminary 3-hour e-learning programme and two days of online or face-to-face training plus post-course supervision to competency. They need to follow supervision and send clinical cases (videotape of two parents' training) to become a certified therapist and implement PACT with high fidelity. IMPACT has a database which includes all the contacts of the professionals worldwide who have completed the training. Its professionals come primarily from the health system and can be psychologists, speech therapists, educators, and others.

**Individual practitioner level** Williams and Beidas (2019) found that attitudes toward Evidence Based Practice (EBP) are amongst the most critical implementation factors. Other key factors include: knowledge and beliefs about the intervention; individual openness to change; personal attributes (e.g., tenacity); a sense of self-efficacy and individual identification with the organization (Damschroder et al., 2009).

**Inner setting** the type of culture (norms, values, and basic assumptions of an organisation) can have a significant effect on implementation. Proficient Organizational Culture has been shown to enhance implementation. Proficient Organizational Culture has been described as environments where clinicians have shared norms and expectations emphasising improvement in client well-being, clinician competence in up-to-date and effective treatment practices, and responsiveness to client needs (Williams & Glisson Charles, 2014).

**Outer setting** each country has different health priorities and health systems. Information on this subject, such as ringfenced funding for autism, is essential. There is agreement within the literature on the importance of taking into account of **patient characteristics** (Feldstein & Glasgow, 2008; Graham & Logan, 2004). Parent-mediated therapy involves parents delivering therapeutic strategies with their autistic children on a regular basis; parental engagement is therefore essential. Parent engagement can be divided into parents' learning strategies for the approach and their enactment of the approach in daily life (i.e. the extent to which they carry out the intervention to the intended 'dosage'; McConachie & Fletcher-Watson, 2015). It has already been suggested that parents with difficulties learning or enacting a therapy not sufficiently adapted to their everyday family routine may slow down the implementation of the therapies by new professionals (McConnell et al., 2015).

**Aim:** In this study, we aimed to explore, systematically and through professional and parent survey data, the barriers and enablers to PACT clinical practice implementation following professional training.

## **Methods**

### ***Design***

This study has a cross-sectional mixed-methods design and integrates professional and parents' perspectives to capture a maximum of potential implementation factors.

Eligibility criteria for professionals included: (i) Having attended a two-day PACT training course; (ii) Being fluent in one of the following languages: English, French, Italian, Spanish or Cantonese (to enable completion of the survey in one of these languages).

Eligibility criteria for parents included: (i) Had completed at least three sessions of PACT intervention with a PACT-trained professional; (ii) Were fluent in one of the following languages: English, French, Italian, Spanish or Cantonese.

### ***Recruitment***

Data were collected from professionals and parents. Professionals (N=460) on the IMPACT training database, and who met the eligibility criteria, were approached from January to March 2020 to participate in the survey, by an email invitation explaining the study and including links to the online questionnaires. Consent was obtained via the survey. These same professionals were then invited in turn to approach parents with whom they had delivered PACT - to share the link to the parental questionnaire. Thus, no direct contact was made between the study team and parents (flowchart 1).

This study was approved by the Ethical Review Board CPP Sud EAST IV Clermont Ferrand Réf: 2020/ CE 03.

### ***Measures: Implementation questionnaires***

All data were collected through self-report questionnaires designed for the present study by an expert group of research professionals, clinicians, and parents (JG, CA, KL, MB, MMG) to explore implementation. The first questionnaire was designed for professionals, and the second was intended for parents. Both questionnaires were available in English, Italian, French, Cantonese, and Spanish. They were translated and back-translated by experienced bilingual researchers.

For each questionnaire, qualitative questions explored factors of implementation constructs and were captured through open-ended questions.

Quantitative data was collected through Likert-type questions where the participants chose the most applicable answer. The answers were based on a 1-7 score where 1 is "strongly disagree", and 7 is "strongly agree". In the supplementary file, see professional questionnaires p.1 table S1, parent questionnaire p.7 table S2 and details on the item scoring p.10-11 table S3 and S4.

The questionnaire for professionals included 17 quantitative and nine open-ended questions. The items measured implementation and explored individual characteristics and potential factors influencing PACT implementation, such as practitioner attitudes towards EBP, type of organization and funding, and level of proficient organizational culture. Attitudes toward EBP were measured using scale items designed by Aaron (2004). The items exploring the level of Proficient Organizational Culture were constructed based on the definition of Williams and Glisson (Williams et al., 2017).

This study focused on the measure of implementation with the percentage of positive answers to the item: "I try to implement the PACT" The item "I try to implement" could include professionals who are beginning to think about how to use this therapy in practice (i.e., finding a camera for videotaping, a family to follow with PACT etc.) as well as professionals who are already delivering PACT with one or more families. It excludes professionals who did not have the intention to use PACT in practice.

The questionnaire for parents explored parent experiences about PACT (positive/negative effects) and barriers and facilitators of learning and enacting daily. Five quantitative and 7 open-ended question items generated scores on the impact on the child and parent and the feasibility of learning and implementation at home. Quantitative adherence was measured with the number of times per week parents reported using PACT strategies at home for 15-30 minutes per day.

#### ***Data collection and treatment***

Data were collected via online surveys with the software SelectSurvey.NETv4.075.003. The data was instantly anonymized with the online questionnaire completion, and participants were identified via a unique number. All qualitative data were analyzed in English. Qualitative answers in all languages other than English (French, Italian, Spanish, Cantonese) were translated and back-translated by experienced bilingual researchers, prior to analysis.

#### ***Analyses***

##### ***Part 1- Quantitative analysis***

Questions that assessed the same concept were aggregated. All the variables had the same range (1 to 7), with a higher score indicating a better implementation outcome or a factor positively influencing the implementation. Analyses were done with SPSS version 25 (SPSS Inc, Chicago, USA). Regression models were used to estimate factors influencing implementation; itself measured by the item "I try to implement, on the overall population of professionals. The following explanatory co-variables were added based on CFIR Theory: attitude toward evidence-based practice, level of Proficient Organizational Culture (POC), and percent of autistic children seen in practice and countries. We tested for collinearity (with principal component analysis) and kept the variable with the greater number. We controlled for the time between practitioner training and survey completion. Once the model had been estimated, its goodness of fit was tested with Hosmer and Lemeshow Test. Odds ratio (OR) are presented with 95% confidence intervals.

### *Part 2-Qualitative analysis*

Open-ended questions were analyzed using content analysis. We used inductive category development, whereby we began an analysis of the text with open coding to identify and organize recurring themes. The initial codes served as a basis for developing categories: themes and clusters (similar themes merged). New themes were added when data did not fit existing themes. The frequency rate of the categories was recorded. MB and MMG separately did the preliminary coding and theme identification. Any discrepancies in these initial coding and theme identification were addressed through discussion with a third party (JG, KL) to achieve consensus.

### *Part-3 Integration of result of open-ended question and quantitative data*

The quantitative and qualitative data were integrated following the methodology of data triangulation by Tonkin-Crine et al. (2015). Key findings for each data set were identified, discussed, and agreed upon by consensus between MMG and MB. A matrix displayed the final list of key findings from the four data sets. For each key finding, six paired comparisons were made to compare the data from each data set. The relationship between data was marked as one of four categories: no response, dissonance, agreement and not applicable (Tonkin-Crine et al., 2015). No response was determined for comparisons when data was present in one dataset but not the other, given that there was an opportunity for such data to be reported. Dissonance was determined when data found opposite results and agreement, similar results. The research team then compared and discussed the two individual analyses from each researcher to obtain a consensus about the relationship between the findings. A framework analysis was carried out to summarize and represent all the results (Cf graph 1).

## **RESULTS**

### ***Sample characteristics***

The study had responses from 231 professionals, 50% of the 460 professionals who had completed formal PACT training with IMPACT CiC between March 2017 and January 2020. Most respondents were based in France (45.5%), followed by Italy (15.5%), the UK (11.2%) and Hong Kong, China (10.7%). For more details, see table 1 p.1. The characteristics of professional respondents compared to the overall IMPACT trained population are presented (see supplement file p.12 table S5).

Among the 1036 parents these professionals declared to have seen in practice and to whom they could have potentially sent the questionnaire, 94 parents answered the survey. Twenty-seven from France, 17 from the UK, 26 from China and Hong Kong, 8 from Italy, 3 from Spain, 4 from USA, and 9 from other countries. See details in table 2 p.2.

### ***1 – Quantitative results***

#### *Quantitative results from the professional survey (Table 3)*

Professionals reported that PACT was highly acceptable and appropriate in their practice, and 70.1% (162/231) said they were trying to implement it. Professionals reported to have begun PACT sessions with 1036 families.

Controlling for the time between the training and the survey, the facilitator of significance for post-training implementation of PACT was a higher proportion of the professional's clinical caseload taken up with autistic children and families, suggesting more specialist practice (OR=4.94; [1.47-16.60]; p=0.01). There was no evidence of an effect on implementation of country of origin or attitudes toward evidence-based practice. Full details are in supplement files table S6 p.13.

#### *Quantitative results from the parent's survey (Table 2)*

Parent responders declared acceptability of PACT for themselves and the family with a Median of 5.7/7 [IQR 5.1,6.2] and feasibility to learn and implement PACT in daily life with a median of 5.4/7 [IQR 4.5, 6.8]. As an overall population, parents declared using PACT on the median of 4 times [IQR 2.0,7.0] for half an hour per week at home. Parents from Hong Kong-China reported enacting PACT at home a median of 2 times per week [IQR 1.7,3.2]

## **2– Qualitative results**

Qualitative results are reported in Tables 4, 5 and 6 p.2-6

### Professional Reports of barriers and facilitators

Extracted themes from the professionals' reports (trying or not to implement PACT, N=184) were grouped within three broad clusters framed as "Family", "Therapist", and "Workplace/Organization" (For more details, see table 4 p.2). The cluster "Family" included factors such as family conditions, family adherence to home practice and therapy sessions. Also, delivering PACT with new cases/families instead of families already seen in practice was identified by some as a factor which facilitates PACT implementation. The cluster "Therapist" included factors relating to the background therapist (reflective or directive approaches), the role of the professionals (job responsibilities, functions), the profile of families seen in clinical practice (child age range, autism diagnosis), therapist accessibility to video equipment, time availability and opportunities for support by colleagues and management. The cluster "Workplace/Organization" included implementation factors related to managerial responsibilities like allowing scheduling flexibility and providing training and funding for training for colleagues. Some clinicians also reported that their sessions, according to commissioning agreements, were of a 1-hour duration and therefore not sufficient for PACT implementation (about 1.5 hrs). The most frequently reported barrier was time, with 104/184 (56.5%) of the professionals reporting that lack of time is an obstacle to implementation.

The sub-population of professionals implementing PACT were asked supplementary questions. They reported factors influencing parents' learning and PACT enactment in day-to-day life (for more details, see table 5 p.3), the impact of the identified factors on progress in the initial therapy learning and the daily implementation for parents. Examples include individual skills of the parent, support family system, motivation for play, time and therapist skills and guidance. The "parent individual skillset" factor (good skill set facilitates using PACT and low skillset diminishes using PACT) was 45% more frequently reported in the context of receiving PACT therapy 63/134 (47.01%) when compared with PACT enactment in daily life 27/127 (21.25%). Conversely, 'lack of time' as a factor was twice as frequent in reports as

limiting daily enactment of PACT (85/127 (66.93%) vs 44/134 (32.85%) as taking part in the PACT therapy. Beliefs around “Limited acceptance of autism” 19/134 (14.17%) and “Limited acceptance of PACT” 6/134 (4.47%) were uniquely reported in the context of the therapy process. In comparison, “Child Behaviour Improvement” 19/127 (14.96%) was only reported in the context of daily implementation by parents.

#### Parent Reports of experience of learning and using PACT and barriers and facilitators

This analysis based on the parent’s experience in learning and enacting PACT in everyday life identified four main clusters: “PACT and positive experience for the child”, “PACT and positive experience for the parent and family”, “Facets of the PACT therapy as facilitating factors for learning or using PACT” and “Facets of the PACT therapy as barriers for learning or using PACT” (details in Table 6 p. 5).

The most prevalent reports from the “PACT and positive experience for the child” cluster were improvements in expressive communication, followed by improvements in play, communication comprehension and social engagement. In the “PACT and positive experience for the parent and the family” cluster, the most frequently identified reports were related to direct benefits for the PACT-trained parent (e.g. increased skillset, a better understanding of child), which were followed by family benefits and improvements in the adult-child relationship.

Facilitating factors for parental attitudes and implementation related to positive features of the PACT delivery 42/80 (52.5%) (e.g., feedback from therapist, video reflection with a therapist, empowerment of parents through therapist guidance). Other less frequent facilitating factors reported were related to the play-based setting 9/80 (11.25%) and to some specific PACT techniques (e.g., video feedback) 5/80 (6.25%) acting as facilitators for enacting PACT in daily life.

Barriers to implementation were associated with obstacles directly relating to the parent themselves (e.g., family stress and other children, work) (47/79, 59.49%), with also a lack of time to implement PACT in daily life (28/79, 35.44%). Barriers related to elements of the PACT therapy, mainly videotaping obstacles, were also reported in 19/79 (24.05%). Reports of barriers related directly to the child as lack of child cooperation or lack of motivation to play were less frequent 13/79 (16.45%).

#### ***3-Integrated results across qualitative and quantitative data***

Detailed key findings are in supplement file Table S7 p.14. We identified 29 key independent findings from the qualitative and quantitative data presented across the four datasets: professional quantitative data, professional qualitative data, parent quantitative data, and parents’ qualitative data. Key findings were presented as statements to aid comparison. None of the key findings was present in all four datasets; 10 were only in 1 dataset, 17 were found in 2 datasets, and 2 were found in 3 datasets. For each of the 29 statements, 3 comparisons were made, yielding 87 comparisons. The number of statements within each convergent category resulted from pairwise comparisons (see summary in supplement file S8 p.18). We

report only the findings that added new understanding or were linked to the findings in analysis above.

### *Convergent factors in the integrative data - barriers and facilitators*

The confidence and expertise of therapists in this type of therapy, based on the reflective process and empowerment of parents in more of the traditional therapeutic alliance, were essential to guide parents in learning and using PACT and help them in their reflective process (key findings 6,7,28, 29).

The importance of the professional group's support (trained colleagues in the same team and supportive colleagues about PACT implementation) (key findings 8). Fifty three from 218 professionals answered that they “did this training because my colleague values PACT”. This sub-group of professionals had a median of 4 families seen in practice. Professionals and parents agreed on the fact that implementation of PACT would be facilitated by spreading information about PACT among other professionals and families. Other professionals could be supported in PACT implementation by trained PACT professionals or at least professionals who value this type of therapy. Families could also find other families using the same techniques and share about them.

Parents and professionals also agree that it could conflict with other techniques. However, interestingly some parents also report finding arrangements, stating that the different methods were complementary or, for instance, use PACT themselves at home and let the school use approaches like Applied Behavioural Analysis or Treatment and Education of autistic and related Communications Handicapped Children (TEACCH), for example (key findings 27).

### *Dissonance in integrative data*

Quantitative analysis of parent data did not find that time in employment was a problem for parents; however, parents and professionals' qualitative data reported that availability of parents and organisation with work was an essential factor (key findings 9).

Proficiency of organisation and management of time: Most of the organisations “didn't avoid change” (Median 6/7 at the corresponding survey item). However, professionals reported in qualitative data that they would like the organisation to decide how to fit PACT in practice (e.g., diminish caseloads, implement PACT and no other therapy, etc.). Then, even in organisations which reportedly did not avoid change, more could have been done to allow the shift (Key findings 20)

Parents from Hong Kong, China (HKC) reported less therapy enactment in everyday life, compared to parents in other countries. They reported similar levels of acceptability and feasibility compared to other countries. They reported slightly higher difficulties with time (9/26 (35%) in HKC population versus 17/ 60 (28%) in other countries. There was no difference in the mean number of training sessions among this population compared to the overall population. However, parents of HKC requested more follow-up sessions (5/ 26, i.e. 19.3%

versus 7.7%), suggesting that this type of model may be less easy to adopt by parents from HKC.

**Graph 1** summarises all the findings and the main factors influencing PACT implementation in this study with a framework analysis per level of implementation according to the CFIR. **Table 7** p. 7 presents the **key results** of the study.

### **Discussion**

This mixed-methods study is the first to examine the factors determining post-training implementation of PACT in clinical practice, an internationally disseminated, evidence-based, parent-mediated therapy.

After the 2-day PACT training, more than 70% of the practitioners move from training to 'intent to implement'. We also found a high level of acceptability and feasibility for parents and reported practice in the home, approximately 4 sessions per week of the planned 7 daily home-based practice sessions during received intervention.

Parent attitudes and life circumstances have a prominent place in factor implementation (as shown visually in graph 1) as part of **Outer Setting** according to the CFIR. According to parents, key barriers to implementation are time (for home practice) and, to a lesser extent, videotaping difficulties. And for Professionals, their rating of poor self-reflection abilities of parents, stressed family conditions and support, or a low acceptance of the diagnosis can be an important obstacle to the implementation.

Many studies in parent-mediated interventions address factors related to parents' experience of learning or implementing this type of therapy in everyday life (Jurek et al., 2022; Leadbitter et al., 2020), but they do not address them from an implementation perspective. This study highlights that all the factors (availability, resources, support, culture, skills, motivation, acceptance of the diagnosis and severity of the child's behaviours) that prevent parents from learning and implementing the therapy are likely to slow down the implementation of the therapy, especially among newly trained professionals (McConnell et al., 2015). Our results thus confirmed the importance of considering the needs and resources of the patients as a key factor in implementation, emphasizing that, in this type of therapy, the target is a child's development; still, this improvement is dependent on the parent's behaviour when interacting with their child.

Most professionals report implementation factors which can be linked to the **Implementation Process** of CFIR (planning, execution, reflection, assessment). Indeed, the professionals underlined barriers related to their usual practice and schedule. These results question how the choice of professionals was made (or not) from the perspective of optimizing implementation and how the manager was prepared (or not) to accompany the change. So, implementation was much more likely if the practitioner had a high number of autistic children on their caseload (OR $\approx$ 5) and had ample time dedicated to therapy delivery (as opposed to management, assessment, etc.). These results highlight a perhaps obvious, yet

critical finding that the success of the implementation depends on training the relevant clinical professionals. As such, allocating training resources towards relevant individuals who work directly with autistic children, rather than management personnel, can be one route to improve implementation.

In **Inner Setting from CFIR**, a key facilitator of the study is peer support from colleagues and managers and the manager's engagement and facilitation in allowing flexibility in schedule. The Hong Kong participants, most of whom worked as part of a charitable organization, reported the highest number of families seen per professional. This could be explained by the fact that these organizations are often mainly focused on patient improvement, which is a key factor in effective implementation (Corrigan, 2001).

This study also showed that the **Intervention characteristics** of PACT are an important facilitator, notably for families (video reflection, empowerment of parent, play-based). In contrast, occasionally some families reported that they expected a more directive approach or did not like playing with the child. Such findings about the intervention characteristics can be linked to the preferences and characteristic of the families (Outer Setting) but also with the **characteristics and abilities of the professionals**. When professionals state that one of the obstacles may be the parent's self-reflective abilities, it may also be the therapist's own ability to enable the parent to reflect on their behaviour. Indeed, some professionals declared not feeling comfortable or experienced with the reflective approach. These findings also point that the rate of implementation success also depends on how fitting is the therapy with the clinicians' viewpoints and experience and the family preferences and characteristics.

Another way to consider the results of this study is through the Theory of Planned Behavior (TPB). TPB is a well-known theory for understanding change in behaviour and gaining currency as a tool for understanding clinicians' implementation of evidence-based practices. It also highlights how the different types of factors according to the CFIR can be interrelated. TPB predicts that the implementation is most likely to be acted if the intention is high, the individual can perform the behaviour (viz **Characteristics of the Professionals**), and there are few environmental constraints (viz **Inner and Outer setting**) that prevent the individual from carrying out the implementation, (Armitage & Conner, 2001).

According to this theory, determinants of intention are attitudes toward EBP, but also norms (meaning that EBP is viewed as appropriate by others), and self-efficacy (feeling competent). The attitude towards EBP was high among the professionals in this study and was not discriminatory. However, using an EBP with a precise protocol (manualised) and the certified process could be a source of stress and feeling and diminish self-efficacy. Emotions and stress toward implementing an EBP have known factors influencing the implementation at the practitioner level (Williams et al., 2017).

As suggested by professional and parent reports, norms, as defined by TPB, could also have been an obstacle. Professionals frequently pointed out the necessity for managers and colleagues to support the implementation (even if they don't use this therapy) (Inner setting). According to this, PACT implementation would also improve if PACT was more known by the

network of professionals (outer setting). Some families also reported that knowing other families that use the same therapy and knowing that PACT is widely used is an encouraging factor for them to use PACT.

Such a finding is consistent with another theory, called self-determination theory, in that one of the primary psychological needs is relatedness and then, one of the determinants of behaviour is that the group supports it, i.e., “a need for being connected to the larger social group” (Deci & Ryan, 2002). Such positive feelings could also encourage families to follow through with the therapy and consequently can support and reinforce the implementation process. It is also noteworthy that autistic people and their families should have their differences supported and recognized and while some therapy approaches can be widely endorsed, it may not be the right fit for everyone.

In the same idea, when considering the results across different countries, France was an interesting case of implementation. The number of PACT professionals trained in France was exceptionally high (half of this study's population) because of Outer Setting, which are national guidelines and critical dedicated funds to train professionals in EBP for autistic children (HAS, 2012). However, the number of professionals who try to implement (57.5%) was lower than the mean population (70.8%), even in a population with a high attitude towards EBP and high Proficient Organizational Culture but without an effective implementation process.

#### Strength and limitations

A high proportion of the population trained in PACT responded (50%). The responders to the survey were likely a little more motivated by implementing PACT, as 25% of responders were certified in PACT compared to 20.4% of the overall trained population. (See supplement file 2). Professionals contacted parents themselves to invite them to take part in the study. We had no control over the number of parents to whom this questionnaire was sent among those whom the therapists had seen in therapy. This presents a risk of selection bias towards parents with more positive experiences. Few negative experiences about PACT were reported. Some evidence suggests that parents can be reluctant to express negative views if those views can be linked back to them or if they recognise that therapists have been trying to help them (Freuler et al., 2014). It is reassuring that within the sub-sample, there was a wide range of parent and child characteristics; a variable number of sessions, varying levels of daily use, parental therapeutic alliance, and child response to the therapy (Table 3).

Triangulation methodology was achieved by using different data collection methods, different types of data, and multiple analysts. Despite these strengths, the study would have benefitted from being designed prospectively to allow the design of complementary interview guides and questionnaires and to know which parent was followed by which professional.

Outcomes about implementation obtained with PACT may be generalisable informative to other types of parent-mediated therapy that are similar in philosophy (i.e. socio-

developmental as opposed to behavioural) and delivery methods (e.g. facilitative, use of video feedback).

### **Conclusions**

This paper highlights the dynamic interaction between factors of learning and enacting therapy for parents and the implementation of professionals and how parents must be implicated in improving transfer in the community of evidence-based parent-mediated therapy.

Moreover, this paper points out that even when national policies support evidence base practice and fund training, success may be harder if the health care system is not guided with a proper implementation process (planning, execution, reflection and assessment).

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**Table 1** Characteristics of the sample of professionals who responded to the survey on PACT implementation.

For the Likert items, a higher score means a better outcome.

Professionals' characteristics	Overall professionals	In France	In Italy	In UK	In HK-China	In other countries
<b>N total</b>	<b>231</b>	<b>106</b>	<b>35</b>	<b>26</b>	<b>24</b>	<b>39</b>
<b>Age (range median)</b>	41-50	41-50	41-50	31-40	41-50	41-50
<b>Profession (n, %)</b>						
SLP	60/230 (26.0%)	17 (16.0%)	5 (14.3%)	19 (73.1%)	3 (12%)	16 (41.0%)
Clinical Psychologist	45/230 (19.5%)	29 (27.4%)	7 (20.0%)	3 (11.5%)	0 (0%)	6 (15.4%)
Psychiatrist and CAP	29/230 (12.6%)	20 (19.4%)	8 (22.9%)	0 (0%)	0 (0%)	1 (2.6%)
Educator	19/230 (8.2%)	17 (16.0%)	0 (0%)	0 (0%)	1 (4.0%)	1 (2.6%)
Others	77/230 (33.5%)	23 (21.7%)	15 (42.8%)	4 (15.3%)	20 (80%)	15 (38.5%)
<b>Percent of families with ASD children in clinical practice Mean (SD)</b>	70.50 (21.9)	70.4 (31.8)	69.6 (25.4)	77.6 (19.5)	52.7 (31.3)	79.1 (22.6)
<b>Type of funding of structures</b>						
Publicly funded	142/231 (61.5)	96 (91.0%)	15 (43.0%)	15 (57.7%)	2 (8%)	14 (35.9%)
Private funded	54/231 (23.4%)	9 (8.5%)	18 (51.4%)	6 (23.1%)	1 (4%)	20 (51.3%)
Charity organization	26/231 (11.3%)	1 (0.9%)	0 (0%)	2 (7.7%)	20 (80%)	3 (7.7%)
<b>Proficient organizational culture Range =1-7 Mean (SD)</b>	5.5 (0.9)	5.5 (0.9)	5.2 (1.3)	5.9 (0.5)	5.2 (0.8)	5.8 (0.8)
<b>Attitude toward EBP Range 1 à 7 Mean (SD)</b>	5.9 (0.8)	5.8 (1.0)	6.6 (1.3)	6.5 (1.0)	5.8 (0.05)	6.1 (0.6)

**Table 2** Outcomes of parents who responded to the survey on PACT implementation overall.

Parent's characteristics	N=94	France	In Italia	In UK	HK-China	In others
<b>Age of parent (range median)</b>	31-40	31-40	31-40	31-40	31-40	31-40
<b>Age of the child in years (range median)</b>	3-5	3-5	3-5	3-5	3-5	3-5
<b>Percent of Full-time work Mean (SD)</b>	0.44 (0.5)	0.3 (0.5)	0.63 (0.5)	0.5 (0.5)	0.35 (0.5)	0.63(0.44)
<b>Overall number of therapy sessions with therapist Median (IQR)</b>	10 (7,12)	10 (8,12)	10.5 (8.2, 11.7)	8.5 (5.2,11.7)	12 (10,12)	10 (6,12)

<b>Number of times PACT used per week at home</b> Median (IQR)	4 (2,7)	6.5 (3.2, 7)	5.5 (3.0, 7.7)	7 (5.0,7.0)	2 (1.7, 3.2)	4.0 (3.0, 7.0)
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**Table 3** Outcomes in the population of professionals who responded to the survey.  
For the Likert items, a higher score means a better outcome.

Professionals' implementation	Overall	In France	In Italia	In UK	In HK-China	In others
<b>Time of survey from training</b> Mean in months (SD)	16.2 (10.4)	13.4 (9.3)	18.7 (0.6)	14.0 (11.6)	28.2 (9.8)	15.1 (6.0)
<b>PACT Acceptability and appropriateness</b> Min-Max =1-7	6.1 +/- 1.2	5.2+/-1	5.6+/-2	6.2+/-1.2	6.1+/-0.9	6.2+/-1.2
<b>Intention "I'm trying to implement" PACT"</b>	162/231 (70.1%)	61/106 (57.5%)	24/35 (68.6%)	16/26 (61,5%)	25/25 (100%)	28/39 (71.8%)

**Table 4** Professionals Reports: Factors Influencing PACT Implementation directly for therapists/ professionals (N=184)

Clusters	Definition	Themes	Frequen cy	Sample quote
Family	Previous practice of the professional with family, family conditions, family adherence to practice	New referrals/families	3.3%	<i>I don't want to practice pact with patients I already see with another method</i>
		Therapist and parent alone for reflection without distractions	4.3%	<i>Having only one parent and be constrained to have the child during video feedback/ debriefing</i>
		Parent adherence to practice (learning sessions and enactment at home)	4.3%	<i>Parent's regularity of doing home sessions and attending therapy sessions</i>
Therapist	Background in therapy, Role of the professionals, families seen in practice, accessibilities of material and skills for videotaping, availability, support from colleagues and management	Reflective style (Accustomed to a more directive approach)	7.1%	<i>The teaching format...it still requires a lot of effort to change my way of doing things... I still tend to be too directive.</i>
		Confidence in the techniques	2.7%	<i>Having the trust, it works</i>
		Role of the professionals and families seen in practice	10.3%	<i>My main function is to participate in assessment and diagnosis of children and adolescents with autism. I don't have time dedicated for coaching or therapy / I don't see children with ASD in practice.</i>
		Video use	10.3%	<i>I am not familiar with the use of videos in my practice, Video feedback is a big help for therapist too</i>

		Supportive colleagues	7.6%	<i>I have had good support from more experienced colleague</i>
		Supportive management	6.5%	<i>managerial support</i>
		Accessible equipment	16.8%	<i>Obstacle of not having suitable equipment, particularly video equipment</i>
		Time	56.5%	<i>In my schedule I don't have 2-hour slot time to free per month for 1 patient and his family considering the list</i>
<b>Workplace/Organization</b>	Related with managerial responsibilities like allowing scheduling flexibility and providing training and funding for training for colleagues	Coordination with colleagues	1.6%	<i>facilitating element of colleagues who are also trained in PACT and with whom we can work in pairs</i>
		Funding	7.1%	<i>Seeking funding to support the PACT training</i>
		Allow time flexibility for PACT	33.7%	<i>Allow flexibility in timetabling/appointment length. Allow time for reflective practice</i>
		Provide training and adapt caseload sizes	21.2%	<i>To ensure that other members are trained and that our caseloads can be reduced to facilitate this</i>

**Table 5** Professionals reports of factors influencing the learning and PACT enactment in daily life of parents and indirectly professional implementation (N=127-134)

<b>Cluster</b>	<b>Definition</b>	<b>Themes</b>	<b>Frequency (%)</b>	<b>Sample quote</b>
<b>Factors influencing parents learning</b>	Parent characteristics, support, time and resources, acceptance of this type of therapy or diagnosis of the child, child disability level, therapist quality and PACT techniques	Individual skill set/ ability to self-reflect and communicate about	47.0%	<i>The parent's having capacity to reflect on their own thoughts and behaviours and communicate these openly. Skills of the parents</i>
		Motivation playing	14.2%	<i>The motivation of the parent, difficulty playing and having fun</i>
		Parent with pathology	6.0%	<i>Parent's depressive anxiety episode</i>
		Support system	6.7%	<i>Parent with fairly stable support system</i>
		Time and Resources (room availability, equipment)	32.8%	<i>Time to practice Room for therapy Material for videotaping</i>
		Parent conceptions of therapy	22.1%	<i>Parental expectations/preconceptions how a therapy may be</i>

	Limited Acceptance of PACT	5%	<i>Difficulty in understanding PACT concept/strategy especially if this conflicts with previous advice /strategies recommended by another professional</i>
	Limited Acceptance of ASD diagnosis	14.2%	<i>Limited knowledge/ acceptance of ASD. Sometimes too quick after diagnosis, parents didn't had time to progress towards acceptance</i>
	Diagnosis: child disability level	20.9%	<i>Child disability</i>
	Quality of Therapist guidance	11.2%	<i>Guidance of the therapist</i>
	PACT techniques (Empowerment , video-feedback)	22.4%	<i>techniques with video guidance trying to find their own word and style with their child</i>
<b>Factors influencing enactment of PACT in daily life</b>	Parent skills	21.2%	<i>Parent skills/ I have found that parents don't always generalize what they have found out during the sessions to daily life</i>
	Motivation for play	8.7%	<i>Parental motivation</i>
	Support system	11.8%	<i>Support from partner/ other family members</i>
	Time	66.9%	<i>lack of time... Take time every day to practice</i>
	Following PACT recommendations	19.7%	<i>Some parents find it difficult to follow PACT recommendations</i>
	Clear goals	15.7%	<i>clear goals that make sense to them in everyday life</i>
	Child Skills	7.1%	<i>The skills of their child during sessions</i>
	Child improvement	15.0%	<i>The fact that they can perceive the effects of PACT on the relationship with their child</i>

**Table 6** Parents' perceptions of PACT: Child and parents positive effects, facilitating factors and barriers for learning and enactment of PACT at home (N=80)

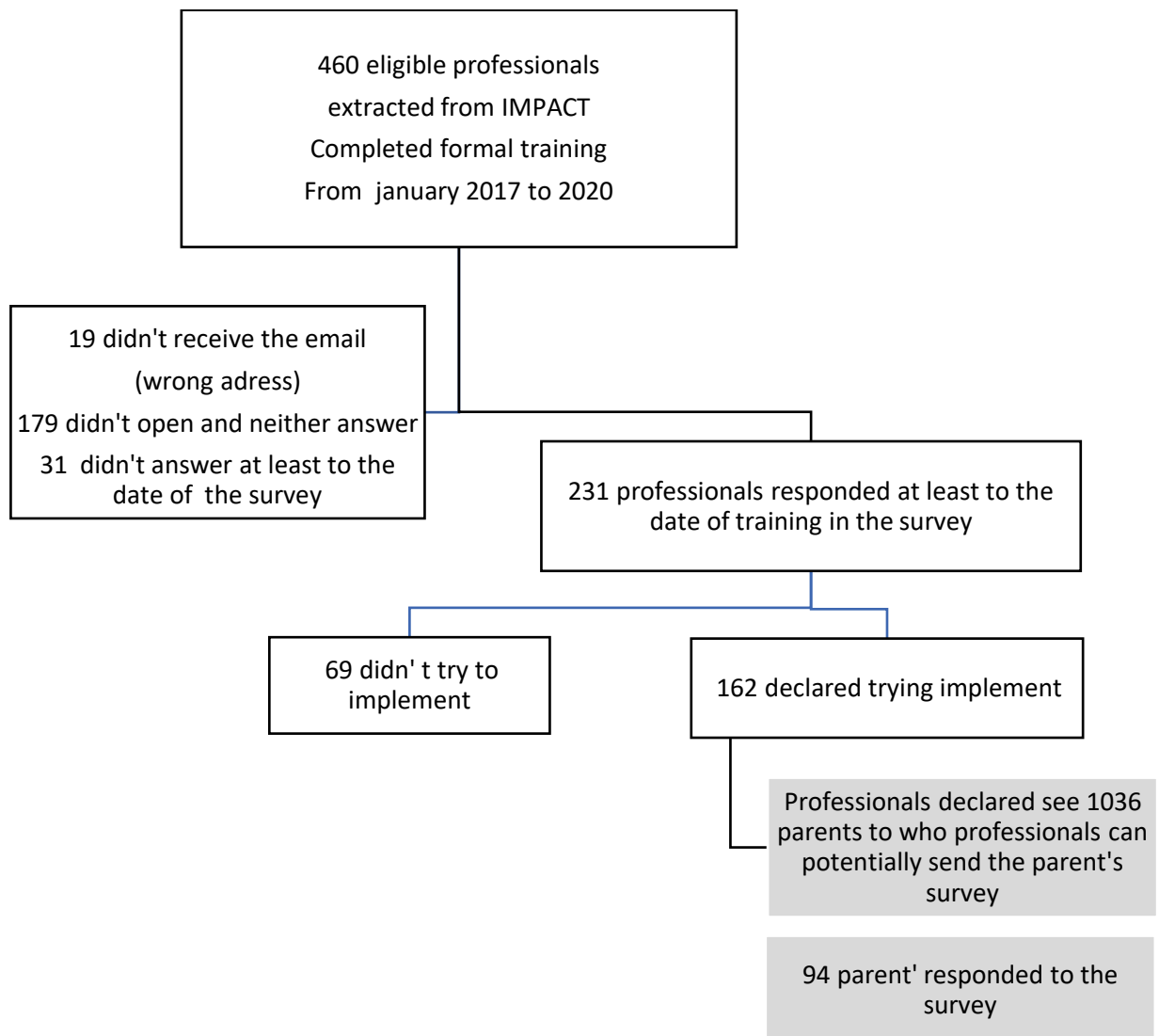
Clusters	Definition	Themes	Frequencies	Sample quote
PACT positive effects on the child	Child play, Social Engagement, expressive and comprehensive, emotional regulation increasing	Child play (interest and duration)	36.2%	<i>My son is able to play for longer periods of time More interest in play</i>
		Child social engagement (sharing, turn taking, enjoyment)	46.2%	<i>He now shares with me when he has my full attention on a one to one Finds pleasure in sharing and playing with others, what he refused before the sessions More turn taking</i>
		Child expressive verbal and nonverbal communication and initiatives in communication	63.75%	<i>My kid initiated more verbal interactions with me/ He smiles at me a lot more now/ He started to use words to express his needs for food, drink, sleep</i>
		Child understanding and following instructions	12.5%	<i>He follows more instructions and understand more words</i>
		Child emotional regulation and temper	10.0%	<i>Her emotion and temper is a lot better</i>
PACT positive effects on the parent	Improvement in self-confidence and skills of parents to understand the child, Parent-Child relationship And Family benefits	Improvements in:  Decrease of parent stress levels, increase of Confidence, understanding of the child and his play	82.5%	<i>I am much less frustrated. I enjoy the game much more with my daughter  I can understand my son more and try a more careful approach to him  I feel more confident and less stressed.</i>
		Better bond/relationship	25.2%	<i>Relation with my child/better relation</i>
		Family joy, Initiate communication/play with other adults, skills for the other adult/parent	35.0%	<i>More family play time at home, and the process was interesting for me and my partner/There is more contact with his sister, even with his father</i>
	Delivery (empowerment of parents/ videobase/ positive),	Feedback from therapist, video reflection with therapist, Rapport with therapist, establishing clear goals and progress steps follow up	52.5%	<i>Trust and good relationship with therapist.  The best part is watching the videos with the therapist  I feel I have an idea of the goal and how we are heading toward it.</i>

<b>Facilitating factors of PACT learning and enactment at home</b>	Techniques and Play- based	Techniques specific to the PACT therapy (e.g play-based, videofeedback)	6.2%	<i>Skills and techniques learnt can be incorporated into everyday life</i>	
		Naturalistic setting, no specific toy demand	11.2%	<i>That play doesn't have to include toys</i>	
	Lack of child social engagement, parents timing, stress, adaptation of the child focus in the play, videotaping	Lack of child social engagement		16.4%	<i>She only likes to play in her own way.</i>
		Time, stress		59.5%	<i>What prevents me from using pact every day is the lack of time, the stress of everyday life</i>
		Difficult to follow child's lead		5%	<i>difficult to let go of the training goals during our play time</i>
		Difficult to videotape		24.0%	<i>It is difficult to make a video when you are tired or anxious It is difficult to shoot a video when I play with my child, because sometimes we change the place of playing (rooms/ table/ floor/ bed).</i>

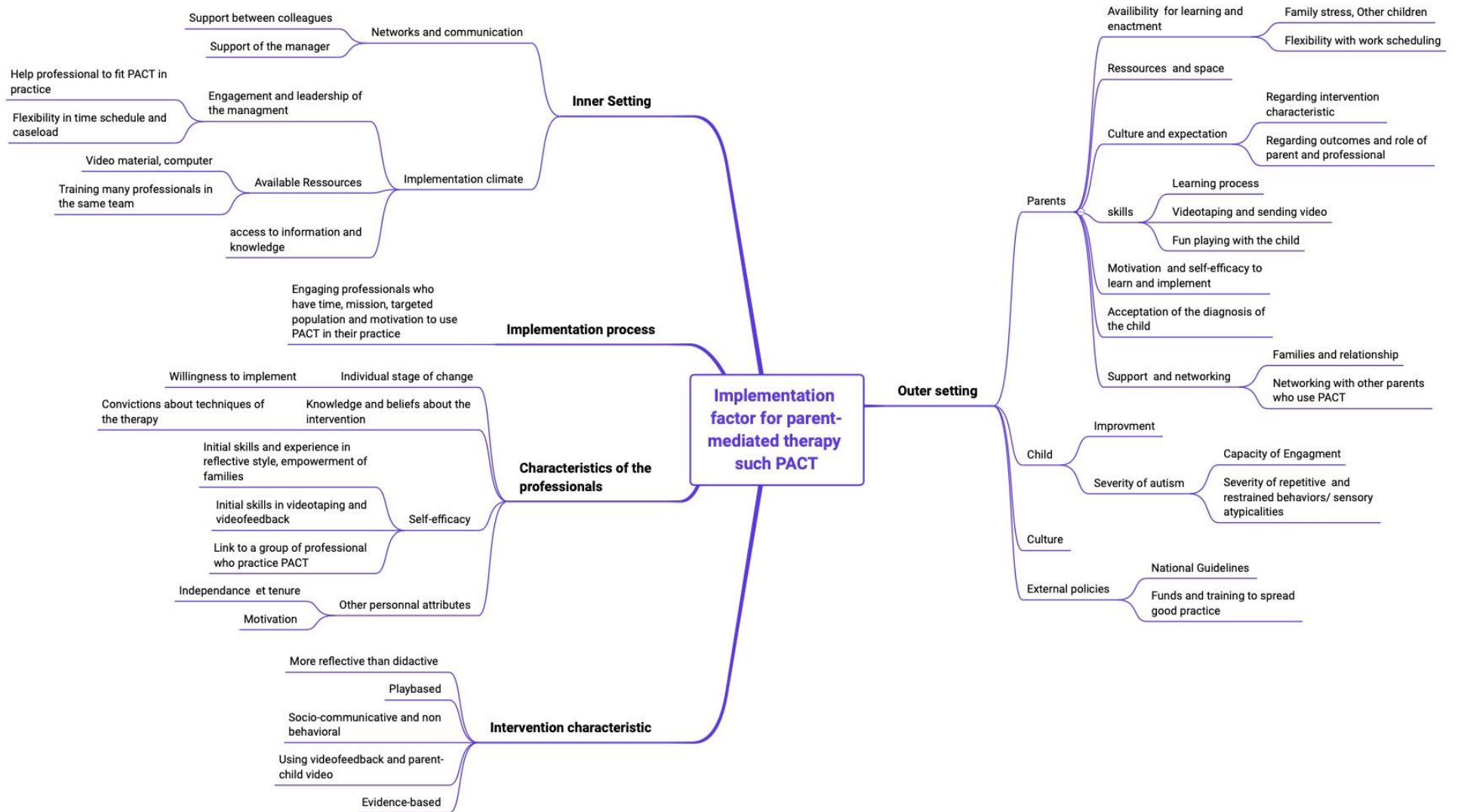
**Table 7 Key results overall**

<p><b>Key implementation data overall</b></p> <ol style="list-style-type: none"> <li>1. 70% of practitioners move from training to 'intent to implement'</li> <li>2. High levels of acceptability and feasibility for parents</li> <li>3. High levels of reported practice in the home - x4 sessions/wk</li> </ol> <p><b>Facilitators to implementation - practitioners</b></p> <ol style="list-style-type: none"> <li>4. Implementation is very much more likely if the practitioner sees a high % of ASD children (OR≈5)</li> <li>5. New cases/families</li> <li>6. Confidence and expertise in reflective therapeutic approach (≠directive)</li> <li>7. Peer support from colleagues</li> <li>8. Managers allow scheduling flexibility</li> </ol> <p><b>Facilitators of implementation - parents</b></p> <ol style="list-style-type: none"> <li>9. Features of the PACT method (video reflection, empowerment of parent, play-based)</li> </ol> <p><b>Barriers to implementation - practitioners</b></p> <ol style="list-style-type: none"> <li>10. Low time in the clinic for practitioners - (needs &gt;1hr)</li> <li>11. Low parent's self-reflection abilities, stressed families condition/ support and adherence to therapy, low acceptance of ASD</li> <li>12. Low child behavior improvement</li> <li>13. Videotaping difficulties</li> </ol> <p><b>Barriers for parents</b></p> <ol style="list-style-type: none"> <li>14. Time for parents (for home practice)</li> <li>15. Videotaping difficulties (25%)</li> </ol>
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## Flow-chart of the 2 populations



Graph 1 : Summary of the results of the post-training PACT implementation survey with framework analysis following the 5 main CFIR categories



## Supplement file

**Table S1** Questionnaire for professionals. *It is a Word copy of what the software survey displayed.*

### PACT Implementation

Dear colleagues,

Thank you for reading and answering the following questions as best you can.

Please follow the order of questions and avoid going back.

If you want to go back, please click on the key “BACK” at the bottom of your screen. Do not use the arrow at the top left of your screen.

If you choose not to answer a question, please skip and move on to the next question. You can't skip the question in yes/no.

#### About your professional profile

1. What is your profession?

- |   |  |  |
|---|--|--|
| <input type="checkbox"/> SLP (SLT)                | <input type="checkbox"/> Clinical psychologist | <input type="checkbox"/> Paediatrician |
| <input type="checkbox"/> Occupational therapist   | <input type="checkbox"/> Neuropsychologist     | <input type="checkbox"/> Social Worker |
| <input type="checkbox"/> Nurse                    | <input type="checkbox"/> Psychiatrist          | <input type="checkbox"/> Other Details |
| <input type="checkbox"/> Teacher                  | <input type="checkbox"/> Child and Adolescent  | .....                                  |
| <input type="checkbox"/> Educator                 | <input type="checkbox"/> Psychiatrist          | .....                                  |
| <input type="checkbox"/> Educational Psychologist |  |  |

2. What is your highest qualification

- Bachelor       Master       PhD       Other: .....

3. You are working in (you can tick as many as needed)

- |   |   |  |                                  |
|---|---|--|----------------------------------|
| <input type="checkbox"/> Public health Center                   | <input type="checkbox"/> IME ( Institut Medico-educatif)        | <input type="checkbox"/> CMPP                      | .....                            |
| <input type="checkbox"/> Private health practice                | <input type="checkbox"/> CAMHS/ HYMS                            | <input type="checkbox"/> CATT                      | .....                            |
| <input type="checkbox"/> CAMSP (Centre d'action Medico-sociale) | <input type="checkbox"/> CRA or Center for assessment of autism | <input type="checkbox"/> National education system | <input type="checkbox"/> Others: |
| <input type="checkbox"/> SESSAD                                 | <input type="checkbox"/> Day hospital                           | Precise.....                                       | .....                            |
|   | <input type="checkbox"/> CMP                                    | .....  | .....                            |
|   |   | <input type="checkbox"/> Network of professionals  | Details                          |

4. What are the best word/s to describe your job? (Tick as many as needed)

- |   |                                    |                                |
|---|------------------------------------|--------------------------------|
| <input type="checkbox"/> Manager/ leader                    | <input type="checkbox"/> Clinician | <input type="checkbox"/> Other |
| <input type="checkbox"/> Independent worker / Self-employed | <input type="checkbox"/> Educator  | details:.....                  |
|   |                                    | .....                          |

5.What is your age ?

.....

6.Please tell us, in which country do you practise?

.....

**About your PACT training**

7.When did you do the PACT training? .....

8.Please indicate the extent to which you agree with each statement

1=Strongly disagree

4=Neither agree or agree

7=Strongly agree

PACT training is relevant to my professional practice	1	2	3	4	5	6	7
PACT fits well with my professional values	1	2	3	4	5	6	7
PACT doesn't fit well with my professional practice	1	2	3	4	5	6	7
I did the PACT training because it was required by my manager	1	2	3	4	5	6	7
I did this training because my colleagues valued PACT	1	2	3	4	5	6	7
I did this training because PACT "made sense" for me	1	2	3	4	5	6	7
I did this training because it is evidence based practice	1	2	3	4	5	6	7
I will not recommend the PACT training to colleagues	1	2	3	4	5	6	7

9.Did you already try to implement or are you implementing PACT in your practice as it was taught in the training?

Yes/no

**About PACT implementation in your practice**

10. With how many families have you already implemented PACT (complete therapy, i.e between 6 and 12 sessions)? .....

11. With how many families have you started to implement PACT (incomplete therapy i.e between 1 to 5 sessions included)

12. With how many families did you start implementing PACT but for who the therapy was interrupted before the end of the therapy (incomplete interrupted therapy, i. 1 to 5 sessions included)

The total number of families you have seen for a complete/ incomplete/ interrupted PACT therapy, this is the total number of families from question 10,11 and 12.

13 .Please indicate the extent to which you agree with each statement

1=Strongly disagree

4=Neither agree or agree

7=Strongly agree

PACT is easy for parents to learn 1 2 3 4 5 6 7

14. According to your experience, what are the elements that help/facilitate for parents to learn PACT? (techniques, your skills, skills of the parents, of the child, time etc...)

.....

15. On the contrary what are the elements that make it difficult to learn PACT? (techniques, your skills, skills of the parents, of the child, time etc...)

.....

16. Please indicate the extent to which you agree with each statement

1=Strongly disagree

4=Neither agree or agree

7=Strongly agree

PACT is easy for parents to use into the daily life 1 2 3 4 5 6 7

17. What helps parents implement PACT in their daily lives? (Techniques, your skills, skills of the parents, of the child, time etc...)

.....

.....

18. What are the barriers if any in parents implementing PACT in their daily lives? (Techniques, your skills, skills of the parents, of the child, time etc...)

19. Please indicate the extent to which you agree with each statement

1=Strongly disagree

4=Neither agree or agree

7=Strongly agree

I am planning to deliver PACT with 1 2 3 4 5 6 7  
families following the PACT training in  
the coming weeks

I use or will use ONLY some principles of 1 2 3 4 5 6 7  
the PACT therapy but not the PACT  
therapy as taught in PACT training

20. Are there principles you learned from the training that have influenced your practice? If so can you say what they are?

21. Please indicate the extent to which you agree with each statement

1=Strongly disagree

4=Neither agree or agree

7=Strongly agree

The PACT therapy as taught is easy to implement in my practice 1 2 3 4 5 6 7

22. What are the main obstacles (if any) in you implementing PACT therapy (as taught in the training) in your practice?

.....

23. What are the main facilitator(s) (if any) in you implementing PACT therapy (complete therapy as taught in the training) in your practice?



I am willing to try new types of therapy / intervention even if I have to follow a treatment manual	1	2	3	4	5	6	7
I know better than academic researchers how to care for the children with ASD and their families	1	2	3	4	5	6	7
I am willing to use new and different types of therapy / interventions developed by researchers	1	2	3	4	5	6	7
Research based treatments / interventions are not clinically useful	1	2	3	4	5	6	7
Clinical experience is more important than using manualized therapy / treatment	1	2	3	4	5	6	7
I would not use manualized therapy/ intervention	1	2	3	4	5	6	7
I would try a new therapy/ intervention even if it were different from what I am used to doing	1	2	3	4	5	6	7

32.Are you working within an organization that provides intervention for children with ASD?

Yes/no

**About your Organizational unit**

33. Please answer the questions based on your observations at the level of your organizational unit, that is, your immediate team, the team you work with daily.

Please indicate the extent to which you agree with each statement

1=Strongly disagree

4=Neither agree or agree

7=Strongly agree

Members of my organizational unit are expected to have up-to-date practices	1	2	3	4	5	6	7
Members of my organizational unit are expected to use evidence-based practices when they are available	1	2	3	4	5	6	7
My organizational unit facilitates implementation of new therapies which require time to read the manual and to follow detailed protocols	1	2	3	4	5	6	7
Members of my organizational unit are expected and encouraged to be formally certified in any therapy they practice	1	2	3	4	5	6	7
My organizational unit expects that each clinician/ member self-evaluate the benefit of his/her work for the children and families.	1	2	3	4	5	6	7
Members of my organizational unit are mostly expected to measure work in a quantitative manner and follow the administratives rules	1	2	3	4	5	6	7

There are no clear expectations from my organizational unit about the type of therapy chosen for the children and families	1	2	3	4	5	6	7
My organizational unit generally avoids change in the treatment practices.	1	2	3	4	5	6	7
I can easily find in my organizational unit other members with whom I can discuss the successes and difficulties in implementing PACT or other new therapies.	1	2	3	4	5	6	7

34.What could managment do to best support PACT therapy become embedded in clinical practice ?

**Table S2** Questionnaire for parents. *It is a Word copy of what the software survey displayed.*

**PACT implementation**

**Some general informations**

Dear parent/carer,

Thank you for reading and answering the following questions as best you can.

Please follow the order of questions and avoid going back.

If you want to go back, please click on the key “BACK” at the bottom of your screen. Do not use the arrow at the top left of your screen.

If you choose not to answer a question, please skip and move on to the next question.

1.What is your age?

2.What is your occupation (please also specify if you are a full-time parent or carer, student or currently unemployed)?

.....

3.In which country do you live?

.....

4.How old is/ was your child when you received PACT from a professional ? .....

5.How many PACT sessions have you received from a professional ? .....

6. Please indicate the extent to which you agree with each statement

1=Strongly disagree

4=Neither agree or agree

7=Strongly agree

PACT suits me and my child well	1	2	3	4	5	6	7
---------------------------------	---	---	---	---	---	---	---

PACT doesn't improve the communication and social skills of my child	1	2	3	4	5	6	7
--	---	---	---	---	---	---	---

PACT improves the language of my child	1	2	3	4	5	6	7
PACT helps my child play in lots of different ways	1	2	3	4	5	6	7
PACT helps my child to be more curious	1	2	3	4	5	6	7
PACT isn't Helpful for my child	1	2	3	4	5	6	7

7. Please describe some of the particular positive effects (if any) of PACT for your child

.....

8. Please describe some of the particular negative effects (if any) of PACT for your child

.....

9. Please indicate the extent to which you agree with each statement

1=Strongly disagree

4=Neither agree or agree

7=Strongly agree

PACT improves my understanding of my child	1	2	3	4	5	6	7
PACT improves the communication between me and my child.	1	2	3	4	5	6	7
PACT improves my well-being	1	2	3	4	5	6	7
PACT can be shared with the other important adults in my child's life	1	2	3	4	5	6	7
PACT improves my confidence in my skills to communicate and interact with my child	1	2	3	4	5	6	7
PACT is helpful for myself as a parent	1						7
PACT is helpful for my family	1						7

10. Please describe some particular positive effects (if any) of PACT for yourself and your family

.....

11. Please describe some particular negative effects (if any) of PACT for yourself and your family

.....

Learning and using PACT

12. Please indicate the extent to which you agree with each statement

1=Strongly disagree

4=Neither agree or agree

7=Strongly agree

PACT is easy to learn and to use with my child 1 2 3 4 5 6 7

13. What are the things that help you learn and use PACT ?

What, on the contrary, are the things that makes difficult learning and using PACT ?

(i.e : techniques, skills of your child, skills of yourself, therapist, time, etc..)

14. PACT is easy to incorporate into my everyday life 1 2 3 4 5 6 7

15. What Helps set up/ you use PACT in this way on a daily basis ? What, on the contrary, are the things that get in the way of using the PACT in your daily life ?

16. If your child is receiving other interventions, are there components/techniques of PACT that work against what other professionals advice for your child? Can you say which ones? How do you manage the different expectations of therapies/interventions while you are doing PACT?

.....

Final questions

17. How many times a week do you find 15 to 30 minutes to do PACT with your child (in the absence of the professional)?

.....

18. What is the best time for you in the day or the week to do the home practice for the PACT therapy?

.....

19. Do you want to add additional information about your PACT experience?

If yes, please specify bellow

.....

**Table S3:** Description of the variables in the professional questionnaire, N and internal consistency (Cronbach's Alpha) per measures

<b>Variables based on bundled items</b>	<b>N</b>	<b>Internal consistency</b>
<i>Level of Acceptability and appropriateness (5 items)</i>	226	0.88
PACT fits well with my professional values I did this training because PACT "made sense" for me I will not recommend the PACT training to colleagues* PACT training is relevant to my professional practice PACT doesn't fit well with my professional practice *		
Level of feasibility to learn and implement for parents according to the professional's implementers (2 items)	139	0.78
PACT is easy for parents to learn PACT is easy for parents to use into the daily life		
<i>Attitude toward evidence-based practice ( 6 items)</i>	184	0.80
I like to use new types of techniques / interventions to help the children and their families I am willing to try new types of therapy / intervention even if I have to follow a treatment manual I am willing to use new and different types of therapy / interventions developed by researchers Research based treatments / interventions are not clinically useful * I would try a new therapy/ intervention even if it were different from what I am used to doing I did this training because it is evidence-based practice		
<i>Proficiency of the organization ( 8 items)</i>	162	0.77
Members of my organizational unit are expected to have up-to-date practices Members of my organizational unit are expected to use evidence-based practices when they are available My organizational unit facilitates implementation of new therapies which require time to read the manual and to follow detailed protocols Members of my organizational unit are expected and encouraged to be formally certified in any therapy they practice My organizational unit expects that each clinician/ member self-evaluate the benefit of his/her work for the children and families.		

There are no clear expectations from my organizational unit about the type of therapy chosen for the children and families		
My organizational unit generally avoids change in the treatment practices*		
I can easily find in my organizational unit other members with whom I can discuss the successes and difficulties in implementing PACT or other new therapies.		

\*Items inversely scored

**Table S4** Description of the variables based on many items in the parent questionnaire, N and internal consistency (Cronbach's Alpha) per measures

<b>Measure</b>	<b>N</b>	<b>Internal consistency</b>
<i>Acceptability and appropriateness of PACT for the child (according to the parent) (7 Items)</i>	78	.913
PACT helps my child be more curious PACT suits me and my child well PACT doesn't improve the communication and social skills of my child * PACT improves language of my child PACT helps my child play in lots of different ways PACT improves the communication between me and my child PACT is not helpful for my child (reverse question)*		
<i>Acceptability and appropriateness for the parents (N=6)</i>	77	.974
PACT improves my understanding of my child PACT improves my wellbeing PACT can be shared with other important adults PACT improves my confidence in my skills to communicate with my child PACT is helpful for me as a parent PACT is helpful for my family		
<i>Feasibility to learn and implement PACT (N=2)</i>	78	0.94
PACT is easy to learn and to use with my child PACT is easy to use and incorporate in daily life		

\*Items inversely scored

**Table S5 Overall IMPACT population compared to the population of professionals who answered to the survey**

	Professionals trained by IMPACT (e-learning and 2 days training)	Survey respondents
<b>N total</b>	460	232
<b>Country</b>		
France	214 (46.5%)	106 (45.7%)
Italy	45 (9.7%)	36 (15.5%)
UK	64 (13.9%)	26 (11.2%)
Hong-Kong	44 (9.6%)	25 (10.7%)
USA	22 (4.8%)	14 (6.0%)
Spain	30 (6.5%)	8 (3.4%)
Others	41 (8.9%)	17 (7.3%)
<b>Profession</b>		
Psychologist	110 (23.9%)	52 (25,4%)
SLT/ SLP/	60 (13,0%)	60 (25.8%)
Psychiatrist and CAP	33 (7.2%)	29 (12.5%)
Nurse	9 (2.0%)	13 (5.6%)
Psychomotricien	18 (4.0%)	11 (4.7%)
TNPEE	9 (2.0%)	6 (2.6%)
Teacher	46 (10%)	13 (5.6%)
Social Worker	14 (3%)	9 (3.9%)
Educator	0	19 (8.2%)
Others or missing data	161 (35%)	4 (1.7%)
Percent of professional PACT certified	<b>94 (20.4%)</b>	<b>58 (25%)</b>

**Table S6 Regression of the logistic model used to estimate the factors which influence trying to implement PACT (N=189)**

	Coefficient $\beta$	T-test (Wald)	p	Odds ratio Exp(B)	95% CI for Exp(B) Lower Upper
Constant	-2.79	5.47	0.02	0.06	
Attitude toward EBP	0.04	1.50	0.22	1.04	0.97 1.11
Percent of child with ASD in clinical practice *	1.60	6.65	0.01	<b>4.94</b>	1.47 16.6
Countries		5.45	0.24		
Time *	0.05	4.66	0.03	<b>1.05</b>	1.00 1.10
Likelihood Ratio Chi-Square (df)	41.7 (7)		<0.00		
Hosmer and Lemeshow Test (df)	6.9 (8)		0.54		
% correct predictions	74.1%				
R2 Nagelkerke	0.28				

**Table S7** Key findings of the Factors of implementation across the 4 types of data and 2 populations. Each key finding is written with a sentence to help the comparison.

<b>Key findings</b>	<b>Quantitative results from professional's survey</b>	<b>Qualitative results from professional's survey</b>	<b>Quantitative results from parent's survey</b>	<b>Qualitative results from parent's survey</b>
<b>1</b>	Poorly seeing ASD in practice is a barrier	Poorly seeing children with ASD in practice is a barrier	NA	NA
<b>2</b>	NA	Poor material accessibility, heavy caseload. Video approach and probing questions were more a barrier for professionals working in publicly funded structure	NA	NA
<b>3</b>	NA	Not having majorly a role of therapist in practice (versus assessment mission or management) is a barrier	NA	NA
<b>4</b>	NA	Seeing families for PACT previously implicated in an other therapy with the same therapist is a barrier	NA	Silence
<b>5</b>	NA	Families not available / stable (physically, emotionally and psychologically).  Lower socio-economic status (poor space to practice) are a barrier	NA	Familial stress is a barrier
<b>6</b>	NA	Professionals not accustomed to a reflective style, in guiding/ empowerment of families is a barrier	NA	The therapist is essential in guiding parents to learn and enact PACT at home

				(guide toward the right goals at home)
<b>7</b>	NA	Professionals not familiar videotaping and the process of video reflection is a barrier	NA	Video reflection is helpful, especially video reflection guided by the therapist
<b>8</b>	Doing PACT training has been encouraged by colleagues who value PACT	Supportive team and management is a facilitator in PACT implementation.  More professionals of the team are trained and can work together more it is facilitating for implementation.	NA	NA
<b>9</b>	NA	Poor enactment of parents in daily life is a barrier	Parents working or not was not influencing how many times parents practice PACT per week	Time, especially time and organization of parents who work is essential to enact PACT in practice
<b>10</b>	NA	Poor availability of materials such as videotapes and camera is a barrier	NA	Poor availability of video equipment and poor knowing how to use it is a barrier
<b>11</b>	NA	Poor individual skillset and motivation of parents is a barrier	NA	Poor motivation of parents is a barrier
<b>12</b>	NA	Parents' expectations regarding the therapy not in line with PACT can be a barrier	NA	Silence
<b>13</b>	NA	Poor acceptance of ASD diagnosis is a barrier	NA	Silence

<b>14</b>	NA	Severe disability of the child, or Motor restlessness can challenge enactment in daily life at home	NA	Poor child social engagement and high level of RRB are a barrier
<b>15</b>	NA	Poor support system is a barrier	NA	Silence
<b>16</b>	NA	Hong Kong parents are less familiar with reflective style approaches	Parents from Hong Kong reported that they have less time to implement at home	Parents from Hong Kong have asked for more follow up
<b>17</b>	NA	Silence	NA	Parents who are finding it difficult to follow child's lead , as recommended to enhance synchronization with the child, can find PACT implementation challenging
<b>18</b>	NA	Parents that can't have fun during play may find it difficult to implement PACT	NA	Parents who are too conscious during play time can find it difficult to implement PACT
<b>19</b>	Some professionals did PACT training because it's evidence-based practice	The evidence-base behind PACT was positively influencing the up-take for PACT Implementation	NA	The evidence-base behind PACT was positively influencing the up-take for PACT Implementation
<b>20</b>	Organizational unit generally doesn't avoid change in the treatment practices	The organizational unit needs to decide whether and how to fit PACT in existing practice	NA	NA
<b>21</b>	NA	It's important that the child is being taken care of during the reflection period	NA	It's important that the child is being taken care of during reflection period

<b>22</b>	NA	Extra support for implementing PACT for children who have sensory symptoms	NA	Some parents ask for more attention to sensory issues
<b>23</b>	NA	Written explanations facilitated the process of learning PACT manual for professionals is helpful	NA	Silence
<b>24</b>	NA	Silence	NA	Some parents would appreciate more follow-up and sometimes 12 sessions is not sufficient for the parent
<b>25</b>	NA	Implementation would benefit if management could spread awareness for PACT to families and colleagues/fellow practitioners	NA	PACT is not very well-known and would be good to spread awareness for suitable families
<b>26</b>	NA	Silence	NA	Other interventions with similar approaches generally aid PACT implementation, but in some instances the child may get bored which is impeding implementation
<b>27</b>	NA	Used to practicing therapies with more directive style and there is a learning curve for being less directive towards the parent.	NA	Conflicts with some teaching styles of other therapies as ABA
<b>28</b>	NA	Good alliance between therapist and parent facilitate learning PACT	NA	Good rapport between therapist and parent facilitates learning PACT

29	NA	Parent skills and therapist skills	NA	Therapist skills was important to guide parents and empower them
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**Table S8** Integrative Analysis: The number of statements within each convergence category resulting from pairwise comparisons of findings in Professionals' and parents' quantitative and qualitative.

Agreement	7
Partial agreement	12
Dissonance	2
Silence	8
Not applicable	58
Total	87

## ANNEXE 3 : PARENTAL EXPERIENCE OF PARENT-MEDIATED INTERVENTION FOR CHILDREN WITH ASD. A SYSTEMATIC REVIEW AND QUALITATIVE EVIDENCE SYNTHESIS

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Jurek, L., Leadbitter, K., Falissard, B., Colin, C., Touzet, S., & Geoffroy, M.-M. (2022). **Parental experience of parent-mediated intervention for children with ASD : A systematic review and qualitative evidence synthesis.** *Autism*, <https://doi.org/10.1177/13623613221112204>

### Abstract

**Background:** Whilst the effectiveness of parent-mediated interventions (PMI) in the field of Autism Spectrum Disorder (ASD) is well documented, information on the experience of parents involved in PMI is limited.

**Methods:** We performed a systematic review with qualitative evidence synthesis to explore the experience of PMI in parents of children with ASD. PubMed, Scopus, PsycINFO, CINAHL, and Sociology Collection were searched from the date of their creation until February 25, 2022. Qualitative studies reporting parents' experience of PMI were included. Two independent reviewers assessed the risk of bias. The findings of the selected studies were extracted and synthesized using the meta-aggregation method. The results are reported according to PRISMA and ENTREQ guidelines.

**Results:** 23 studies were synthesized representing 345 participants. We found 34 categories that were summarized in four synthesized themes: barriers to implementation and logistical issues, feeling overwhelmed and stressed (a need for support), facilitators of implementation, and empowerment in the parent and improvement in the child.

**Discussion:** Parents' experience of PMI in ASD is balanced between positive outcomes for them and their child, emotional struggles, and some difficulties in implementing PMI. Based on these results, we propose new ways to improve PMI implementation and research in the field.

**Protocol :** PROSPERO CRD42020178743

### KEYWORDS

Parent-mediated intervention, Autism Spectrum Disorder, Qualitative research



## Introduction

As identification and diagnosis of Autism spectrum disorder (ASD) in early childhood is now more common, several early interventions targeting core impairments of ASD have been developed and evaluated (Dawson, 2008; Pickles et al., 2016).

Boyd et al. (2010) highlighted in their review that the most promising intervention methods involve components of naturalistic interventions for teaching pivotal skills in natural environments and parent-implemented approaches (Boyd et al., 2010). In such interventions, a trained practitioner shares therapeutic strategies with the parent, either in one-to-one sessions or within a group, and the parent is then encouraged to use these strategies with their child outside of the session (Bearss et al., 2015). Since then, many models of parent-mediated intervention (PMI) have been developed and evaluated.

The model of PMI allows intervention to begin early, with the aim that parent interaction strategies help enhance children's earliest social relationships. Increased parental skills allow for continual opportunities for child learning in a range of situations and increase the intensity of the intervention (Koegel et al., 2001; Oono et al., 2013). It is also proposed that these methods could increase parent empowerment and self-efficacy (Schertz et al., 2011) leading to a reduction in parental stress (Oono et al., 2013).

Nevertheless, recent studies have documented a disconnect between parent and professional perspectives and goals concerning children with disabilities, with professionals focusing more on impairment reduction and parents prioritizing their child's well-being (Leadbitter et al., 2018). Increased understanding could improve the implementation of interventions within community settings. Engaging families could begin with examining their individual intervention priorities at this pivotal time in the life of their child and their family (Wainer et al., 2017). Understanding their experiences is critical because it provides a glimpse into daily family realities and the ways that ASD can impact parent-child dynamics (DePape & Lindsay, 2015) and can allow intervention practitioners to be sensitive and responsive to family circumstances and priorities.

In the case of PMI in ASD, randomized controlled trials are a good tool to evaluate the efficacy of the treatment but do not usually assess the impact of the treatment on the family system. Qualitative evidence allows researchers to analyze the human experience and has a particular role in exploring and explaining why interventions are or are not effective from a person-centered perspective (Bölte, 2014). To date, no qualitative evidence synthesis (QES) review has examined this topic.

The primary objective of this review was to identify, appraise and synthesize qualitative research evidence on the barriers and facilitators for parents participating in PMI for children with ASD.

## Method

The review protocol is available on PROSPERO CRD42020178743 and is reported according to PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) (Moher et al., 2009) and ENTREQ (Enhancing Transparency in Reporting the Synthesis of Qualitative Research) (Tong et al., 2012) statements (checklists can be found in **Supplemental Material**).

### - **Search strategy**

As recommended, a combination of electronic databases (PubMed, Scopus, PsycINFO, CINAHL, and Sociology Collection) was searched from the date of their creation until April 7, 2020. An initial limited search of PubMed was undertaken to identify articles on the topic and create the search algorithm. Keywords describing autism and PMI were combined. Filters were used to exclude review articles. The search strategy for the different databases is provided in the supplement (**Supplemental material**). The reference list of each identified record was reviewed to identify additional relevant studies. The searches were updated before the final analysis was performed on February 25, 2022, and any additional studies that were identified were included.

Only studies published in English and in French were considered for inclusion in this review, with no publication date restrictions.

### - **Selection**

We screened titles and abstracts of identified studies for eligibility, followed by full-text review where indicated. For the titles and abstracts screening, studies that presented a PMI for children with ASD were included regardless of the type of study. This first step was time-consuming but necessary to avoid excluded qualitative research that was not clearly indicated in the abstract (Booth, 2016).

For the full-text review, we considered studies as eligible if: (1) the population included caregivers with a child under 18 years of age diagnosed with ASD according to an international classification manual (including a diagnosis of autism, Asperger's syndrome, Pervasive Developmental Disorder (PDD) and PDD Not Otherwise Specified [PDD NOS]); (2) those parents had been invited to a parent-implemented intervention, defined as skill-focused intervention where the parent is the agent of change and the child is the direct beneficiary of treatment (Bearss et al., 2015). These interventions could focus on core features of ASD or maladaptive behavior; (3) a qualitative methodology (based on a semi-structured interview or focus group) was used to describe the experiences of parents concerning the intervention. Experiences could relate to parent expectations about the intervention, their support requirements and involvement (or lack of), but also the perceived barriers and facilitators to their participation in this kind of intervention.

Studies were excluded if: (1) parents underwent a psychoeducational program without any skill learning; (2) no qualitative method was described (even if some quotations were reported, a method of analysis should be described with the different steps); (3) the outcome of the qualitative study was not focused on the experience of parents concerning the intervention but was more about appraisal and development of specific aspect of the program and not about the general parental experience.

- **Appraisal of the methodological limitations**

Relevant studies were critically appraised by two independent reviewers (L.J. and M-M.G) for methodological quality using the Critical Appraisal Skills Programme (CASP) Checklist for Qualitative Research. The 10-item checklist considers three categories: validity (clarity of research aims, appropriateness of qualitative methodology, research design, recruitment strategy, and data collection, appropriate consideration of researcher reflexivity), results (ethical considerations, appropriateness of data analysis, clarity of findings stated), and utility (the value of the research). Disagreements were resolved by consensus or by referral to a third author (K.L.). As proposed by Cochrane guidelines, we decided to exclude studies with high risk of bias from the analyses (Carroll et al., 2012; Hannes K., 2011). A score below 6 on the CASP was considered at high risk of bias (Butler et al., 2016).

- **Data extraction**

One author (LJ) extracted data from each included study on authors, year, title, aim, study design, context, participants, and analytical approach. Each finding, defined as the authors' interpretation of their results, was identified by a single author and entered a spreadsheet (Microsoft Excel, Redmond, Washington, USA) for all studies. If studies included views other than those of parents of children with ASD, only the data relating to parents' views were included in the synthesis. Using the terminology employed in the Joanna Briggs Institute Reviewer's Manual, each finding was assigned a level of credibility based on illustrations in the form of supporting quotations or field observations (Lockwood et al., 2015).

Possible credibility levels were unequivocal (the illustration supported the finding beyond a reasonable doubt), credible (some association was present between the illustration and the finding, but it did not clearly support it, meaning that the finding was open to challenge), and unsupported (no illustration was presented). The credibility levels were assigned by LJ and KL; discrepancies were resolved by MMG.

- **Data synthesis**

A meta-aggregative approach was used to synthesize the data. In this approach, authors' qualitative findings are inductively linked to a specific category that includes findings with similar meaning. Categories are then summarized in order to produce a comprehensive set of "synthesized findings" that provide a basis for identifying recommendations for action. This method does not seek to re-interpret the findings as do some other methods of QES but seeks to enable generalizable statements in the form of recommendations (Booth et al., 2018; Hannes & Lockwood, 2011).

Two authors, LJ and KL, collaboratively derived categories and synthesized findings by inductively grouping similar findings and identifying conceptual categories. To improve external validity, the categories and synthesized findings were discussed with a third author (MMG). Discrepancies between LJ and KL were resolved by MMG. Some categories were renamed to fit the data and improve comprehension, and findings were reclassified if needed until a consensus was reached between the three authors.

Implications of the results and recommendations for future practice and studies were discussed with a group of experts (LJ, KL, BF, CC, ST, MMG).

#### - **Reflexivity**

As quoted below, we chose to work with a multidisciplinary team to provide a synthesis combining different points of view and decrease the risk of personal bias. Four members of the team are child and adolescent psychiatrists (LJ, MMG, KL, BF) and some are trained in PMI (MMG, KL). ST is a senior physician in public health and has been working on RCTs assessing the efficacy of PMI models with LJ and MMG. CC and BF are professors of public health. Most of the members are trained in qualitative methodology (LJ, KL, ST, BF, CC). KL has conducted qualitative research focused on the experience of parents with PMI.

A summary of team members and their implications in this study can be found in the **Supplementary Material**.

#### - **Confidence in the results**

Each category was assessed using the GRADE-CERQual (Confidence in the Evidence from Reviews of Qualitative Research) approach. CERQual is a method to assess how much confidence to place in findings from QES. It assesses to what extent a review finding (here, our categories) reasonably represents a phenomenon of interest (Lewin, Booth, et al., 2018). The CERQual approach includes four elements: (1) methodological limitations of the individual studies that were here assessed with the CASP, (2) coherence of each category (with the primary data and the other findings), (3) adequacy of data (richness of data, number of studies corresponding to the finding, contexts of the different studies) and (4) relevance to the review question and context. We assessed each of these four components as having minor, moderate, or substantial concerns regarding the specific component. Based on an overall assessment of methodological quality, relevance, adequacy, and coherence, the confidence in the evidence for each review finding was assessed as high, moderate, or low (Lewin, Bohren, et al., 2018).

#### - **Community involvement statement**

No community member was involved in this study, but community members (parents) were involved in every reviewed study.

## **Results**

#### - **Description of included studies**

Our search identified 2879 articles after duplicates were removed. After screening on title and abstracts, 515 articles were assessed for eligibility and 23 articles were included in the systematic review. Three additional studies were retrieved from the second search (**Figure 1**).

[insert figure 1 about here]

#### - **Methodological quality**

The methodological quality of the 26 included articles was assessed. Twenty-three were considered at low risk of bias (**Table 1**) and three were considered at high risk of bias and were excluded from the synthesis (Blake JM. et al., 2017; Chandler S. et al., 2002; Zand et al., 2018).

#### - **Studied population**

The experience of 345 parents was described in the studies. The characteristics of each study are presented in **Table 2**. The mean age of parents was 36.7 years old (SD=3.4), and most of the interviewed parents were women (81%). Concerning the educational level, 49% of the parents had a college-level, 33% completed high school, and 8% had a primary school level.

Children of parents included in the studies had a mean age of 62.7 months [range=15-133] and mainly were boys (81%).

Interventions targeted social communication skills in most of the studies (n=17), but also disruptive behavior (n=4) or both (n=2). Individual parent training was most frequent (n=17). The mean number of sessions guided by a professional or an application was 13.5 (SD=7.9). Parents were asked to regularly practice the intervention at home in most of the studies.

Eleven studies were from North America; six were from Europe, four from Asia, and two from Australia.

Concerning qualitative methodology, thematic analysis was used in 19 studies, framework analysis in 3 studies, and Kawakita Jiro-Method (KJ-Method) in 1 study (a qualitative analysis method developed in Japan that pursues the core themes in a dialog and explores them from a new angle by grouping them and labeling the groups according to common themes, then illustrating them by a diagram).

#### - **Synthesis**

We extracted 208 findings from the 23 studies (see **Supplemental Material**). Concerning the level of credibility, 149 findings were considered unequivocal, 32 as credible, and 26 as unsupported (by the data presented in the article).

These findings were grouped into 34 categories summarized in four synthesized findings: barriers to implementation and logistical issues, feeling overwhelmed and stressed (a need for support), facilitators of implementation, empowerment in the parent, and improvement in the child (**Figure 2**).

As a sensitivity analysis, findings rated as unsupported (by the data) were removed. This did not impact the categories and the synthesized findings.

[insert figure 2 about here]

Confidence in the findings is reported in **Table 3**, along with quotes supporting the results. Twenty-one categories were assessed with high confidence, nine with moderate confidence, two with low confidence, and two with very low confidence (advocacy and importance of early intervention).

The details of the GRADE quotation are available in the **Supplemental Material**.

### ***Synthesized finding 1: barriers to implementation and logistical issues***

Among the different studies, parents reported some barriers to implementing the intervention. They emphasized how **challenging finding time for the intervention implementation** was and how the recommended homework was challenging to organize in their planning. This barrier was described in nearly half the included studies.

Along with this barrier, parents had some concerns about **logistics, particularly about the schedule and location of the sessions**. Parents acknowledged that the session had to be close to their home participate. They also described the **need to occupy the child** during the intervention sessions with the therapist. Some other **environmental barriers** to the intervention's implementation were described, such as the presence of the siblings.

For some parents, integrating intervention into routines or daily activities was a challenge. Parents described **challenges in implementing due to child factors**. Because of ASD symptoms, such as a lack of flexibility in the child, parents experienced difficulties implementing the intervention at home. **Parent-related factors in engagement** were also described. Parents indicated that they sometimes felt unconfident about their parenting skills or overwhelmed by the situation, which led to difficulties in implementing the intervention. Conversely, some parents thought that the child could be more responsive to them than to the therapist because of the established child-parent relationship.

ASD is a large spectrum, and parents share concerns about their child's individuality. They reported **the need for personalization of the approach** with different groups based on children's characteristics (verbal or non-verbal, with or without intellectual disability).

Some parents felt **stressed by the therapist and sometimes undervalued** with therapists presenting as experts and being directive, with no space for the parent to express their feelings.

Another limitation to implementation was **parents' ambivalence concerning effectiveness**. They mentioned their doubt about the causal relationship between the intervention and improvements in the child, which could have reduced the motivation to continue the intervention.

***Synthesized finding 2: feeling overwhelmed and stressed (a need for support).***

Even if parents underlined the **importance of early intervention**, they reported feeling overwhelmed and stressed throughout the process and advocated for more support. The difficulty in **accepting the diagnosis** and the fact that the **post-diagnosis phase is difficult** was well described by the parents.

They mentioned **feeling overwhelmed** and emphasized **the need to go slow at the beginning**, as a great deal of new information is presented to them. Parents explained they went through many **conflicting feelings: between hope and concerns**, from the beginning to the end of the training. They described the **need for parents' self-care** to support the burden of their situation. and evoked that **parent-to-parent support**, with the possibility of sharing experiences, was or could be helpful. For group intervention, parents described the benefits of sharing with other parents, as long as the group was not too big.

Along with the emotional struggles, parents described the need for support. First, they explained the **need to include the extended family and social network in the intervention implementation**. Parents, and notably mothers, reported that including the child's siblings, the partner, and/or the grandparents would help them feel supported and make it easier to implement the intervention. They also underlined **the need for external support during and after the intervention process**. Parents evoked that having regular contact with a professional was essential to respond to any questions and to feel supported. In studies where the intervention was self-guided through a tablet application, parents described the importance of having a coach to help them. Parents also pointed out the anxiety they felt at the end of the program, unsure they would be able to continue implementing the intervention without the therapist.

Along with these worries, parents indicated the **need for more of the same intervention**. Parents reported they would have enjoyed an "advanced program" in some situations. They felt the program was not covering every difficulty related to autism and that, for some families, additional content would be necessary. After going through different interventions (individual PMI or self-guided PMI), parents evoked the **need for complementary interventions** to focus on other aspects of ASD.

***Synthesized finding 3: facilitators of implementation.***

Parents described several factors that enhanced the implementation of the PMI in their daily life. They indicated that the **positive parent-therapist relationship**, and notably the trust in the therapist, was an essential factor in their adherence to the therapy. In most studies, the relationship between parents and the therapist was described as strong, personalized, and cheerful, with the feeling of being understood. As seen previously, some parents felt, on the other hand, stressed by the therapist. This variability could be explained by the heterogeneity in the included populations and the type of interventions.

Along with this, the content and tools used in the PMI were also described as facilitators. Parents reported that the **treatment planning process was effective** as it was systematic: **one step at a time**.

They enjoyed using **reflective techniques in the therapy (role-play, feedback)** and the **use of video feedback**. They described the importance of watching themselves interact with their child to later adjust the interaction.

While some parents have described difficulties in finding time to implement the intervention, others considered the **intervention as feasible, and easy to implement in routine**. Parents explained how they implemented the learned tools in their daily life and how they organized their day to be able to practice the intervention.

***Synthesized finding 4: empowerment in the parent and improvement in the child.***

While describing their global experience of PMI, the parents described several positive outcomes.

Parents reported that they had **limited knowledge of ASD and related services before intervention**; this was corrected by the intervention that gave them **understanding and acceptance of their child's impairments and specific skills to support child development**. They understood, for example, that interaction skills are as essential as language or that following the child's interests can lead to improvement in interaction. Parents developed a sense of ASD **advocacy** and used their new knowledge to help other families.

These new skills made them **more confident about their parenting skills** and brought a **feeling of empowerment after the intervention**. Parents acknowledged that their **high involvement during the intervention** was essential and described themselves as **agents of change**.

Even if the intervention was demanding for the parents, it **improved parents' well-being and decreased parental stress**. A general **improvement of family relationships and interactions** was described by the parents.

Concerning intervention outcomes, parents also described that interventions had **direct benefits on their child with an improvement in child behavior**. This improvement was noted in verbal and non-verbal communication, repetitive behavior, and challenging behavior. Unfortunately, for some families, **improvement was not observed; this was sometimes attributed to the child's factors**.

## **Discussion**

### **- Summary of main results and comparison with literature**

By pooling the main findings of 23 studies, our study synthesized the voices of 345 parents who experienced PMI for their child with ASD. Many parents reported a global feeling of empowerment and improvements in their children. They described the PMI as feasible and underlined the facilitators of implementation. The parent-therapist relationship was described as an important factor for obtaining the full participation of parents in the intervention. Systematic intervention with a well-described process and

a step-by-step approach was appreciated by the parents and the reflective techniques used by the therapists.

Besides those positive reports, many parents emphasized the emotional toll of the situation on themselves. They stated that PMI needs to be carried out alongside support for parents' mental health and wellbeing. Parents highlighted some barriers to PMI interventions that could be addressed in practice and future studies.

#### - **Implication for practice**

Recent implementation science studies suggest that evidence-based treatments for ASD often do not get appropriately transported to real-world practice settings (Wainer et al., 2017). Focusing on parent-level variables as meaningful outcomes and/or moderators could increase the effective implementation of evidence-based interventions in the "real world," as demonstrated in the disruptive behavior (K. Pickard et al., 2019). Therefore, parents' experience of PMI in ASD should be used to inform future intervention models, and the main findings of our study could be used to inform future guidelines.

The findings of our study provide evidence that self-guided intervention needs to include an element of coaching from a professional as the relationship with the therapist is described as a key facilitator of intervention. There is a need to consider how PMI should be included in an individualized intervention plan using a stepped-care framework (Green, 2019). PMI should promote parents' self-efficacy and lead to parents' understanding of their abilities. This first step is essential to help them promote child engagement, which is the main objective of PMI.

Parents sometimes report feeling overwhelmed and stressed after the diagnosis of their child but also, during the intervention process, psychoeducation or "post-diagnosis" support could be recommended before the beginning of PMI. Offering systematic parent care or support along with the intervention program could also help decrease parental stress and promote resilience (Zhao et al., 2021). Support during the intervention can also come from peers and parent-to-parent support programs could be developed. It is important to remember the holistic needs of parents in the intervention process.

Balancing work, family, and demands associated with caring for a child with ASD can be challenging. In our study, parents described how difficult it could be to find time for the training and implementation of the intervention. An intervention should not require a parent to give up employment or personal interests. The clinician should propose the PMI when the timing is suitable for the family and subsequently adapt the PMI to the context of the family (McConnell et al., 2015). Personalization in the choice of intervention could lead to better implementation outcomes. Concerning group-based intervention, it seems important to consider the diversity in ASD and create intervention groups in which children share specific features, such as language or intellectual abilities.

Our review has brought to light the feeling of “empowerment” gained by parents participating in PMI. Empowerment is essential to shared decision-making in healthcare which often has the potential to enhance outcomes and to be cost-effective. While shared decision-making generally leads to improved outcomes, some individuals prefer to leave decision-making in the hands of professionals. (Levinson et al., 2005). It is also important to note that empowerment does not mean putting responsibility in parents’ hands but guiding them to make their own decisions.

- **Implications for research**

Most research focuses on the effectiveness of PMI intervention in reducing autistic symptoms. Improving social communication skills is undoubtedly important; however, other intervention goals are also valuable. Parents’ perceptions and experiences of an intervention should be sought using qualitative methods, and this focus should be included alongside effectiveness studies. The ECO-Framework (Wainer et al., 2017) proposes a reflection on important outcomes for families in the field of early intervention in ASD and describes the need to assess different domains: immediate family outcome (knowing their rights and advocacy; understanding the child’s strengths and special needs; helping their child to develop and learn; having support systems and gaining access to desired services and activities), outcome (quality of life) and intermediate outcome (family interaction, parenting, and well-being).

- **Strengths and limitations**

The strengths of our study are that: we used a two-step robust systematic search strategy to identify studies with a sensitive approach conducted by two independent reviewers to ensure the inclusion of all relevant articles; recent guidelines informed our methodology on conducting a meta-synthesis, and the quality of included studies was assessed with a validated tool. We used transparent and reproducible methods, including pre-registration. Qualitative studies do not aim to be representative but can fill “gaps” in our understanding that are not resolved by the quantitative research (Sandelowski, 2004). Within the meta-synthesis approach, we deeply describe the population of the different studies to enable future comparison and external application of our results. An assessment of confidence in our results has been provided. Heterogeneity in the results has also been described.

Our work also has some limitations. The synthesis was limited to interview (semi-structured interview) and focus group studies only and did not include other qualitative methodologies, such as open-ended questions in questionnaires as they are less likely to generate a rich and detailed description of participant experience with context, personal experience, and social meaning... We chose to exclude survey-generated data to avoid great heterogeneity in the selected methodologies. (LaDonna et al., 2018).

Two researchers conducted the synthesis collaboratively and discussed it with a third author rather than being operated independently. The discussion between the three researchers aimed to validate that the categories and synthesized findings were close to the original findings of the included articles and that

findings were appropriately classified in the different categories. If the first authors misunderstood or misclassified findings, the third opinion enabled correction. A meta-synthesis is an analysis based on previously analyzed data and depends on the subjective interpretation of the original study's authors. This limitation was partly addressed by considering good quality qualitative research with the help of the CASP. Thus, it is still possible that the original interpretation was not informed by the context and did not necessarily reflect a deep understanding of the studied phenomenon.

Meta-synthesis results and recommendations are interpretations made by a group of researchers with their assumptions and biases. To enable the reader to understand those interpretations better, a description of the team's background is described in the supplemental materials. Results were discussed with a multidisciplinary team with diverse expertise in PMI.

A frequent criticism of meta-synthesis is the lack of contextual detail and the loss of original diversity of experiences. Meta-synthesis should be considered a tool to recognize the importance of qualitative studies and highlight shared opinions on a shared experience across a diverse population. Our work included studies focusing on different interventions across various countries and periods. It was striking how similar themes were presented across numerous studies, despite the diverse populations and interventions, and this provides confidence in the applicability of the results.

### **Conclusion**

In this review, parental experiences of PMI for their child with ASD are deeply described. Parents highlight how PMI can lead to feelings of empowerment and positive outcomes for their children. They also raise critical questions about the implementation (barriers and facilitators) "adverse effects" of these non-pharmacological interventions. Still, mostly, they are keys to improving their child's care and support in the future.

#### **CONFLICT OF INTEREST:**

None to report.

#### **AUTHOR'S CONTRIBUTIONS:**

Lucie Jurek designed the study, collected the data, conducted the analyses, wrote the manuscript and designed the figures.

Kathy Leadbitter designed the study, conducted the analyses and wrote the manuscript.

Bruno Falissard participated in results interpretation and wrote the manuscript.

Cyrille Colin participated in results interpretation and wrote the manuscript.

Sandrine Touzet participated in results interpretation and wrote the manuscript.

Marie-Maude Geoffray designed the study, collected the data, participated in results interpretation, wrote the manuscript.

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#### **DATA AVAILABILITY**

Data are available in the supplemental material and upon request to the corresponding author.

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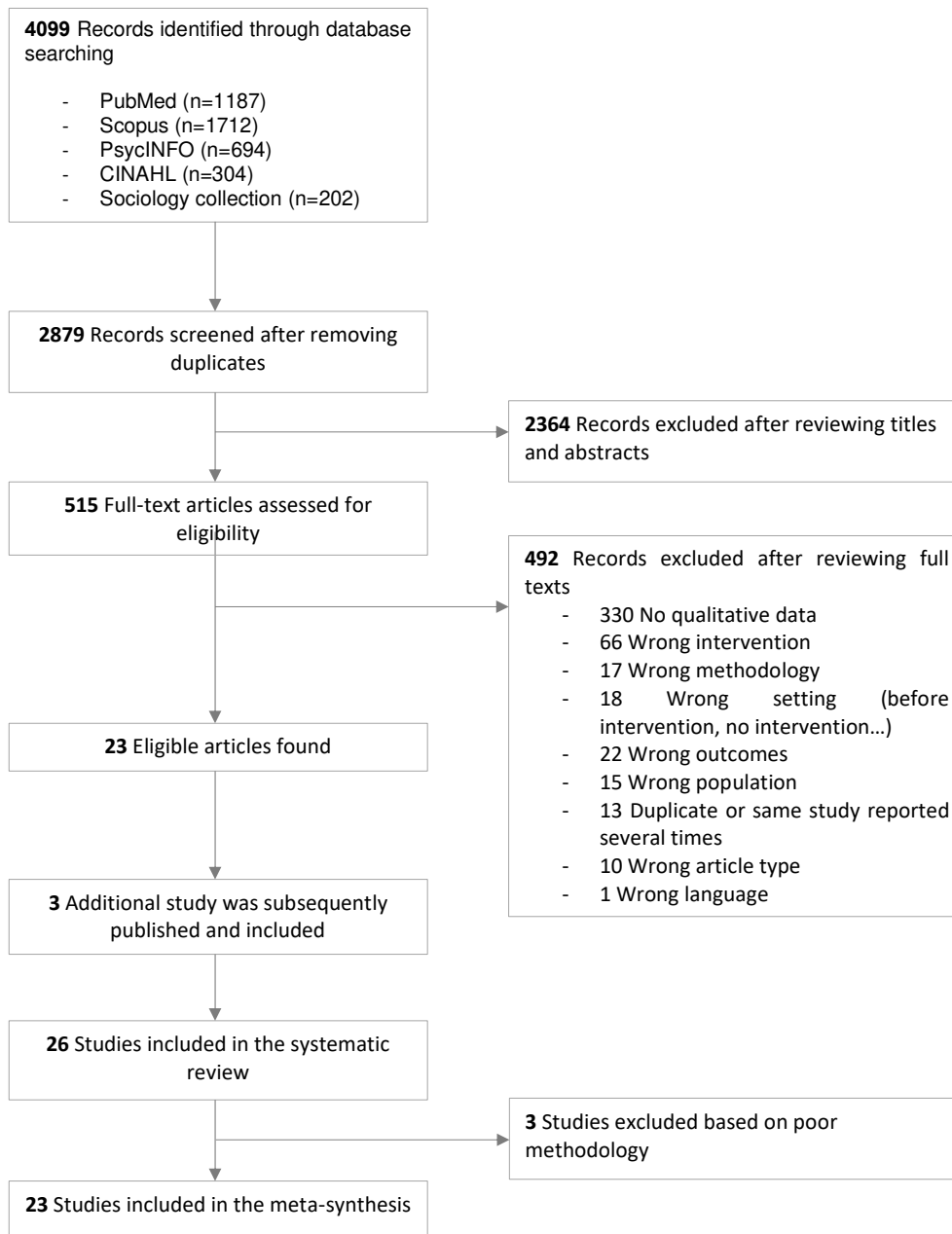
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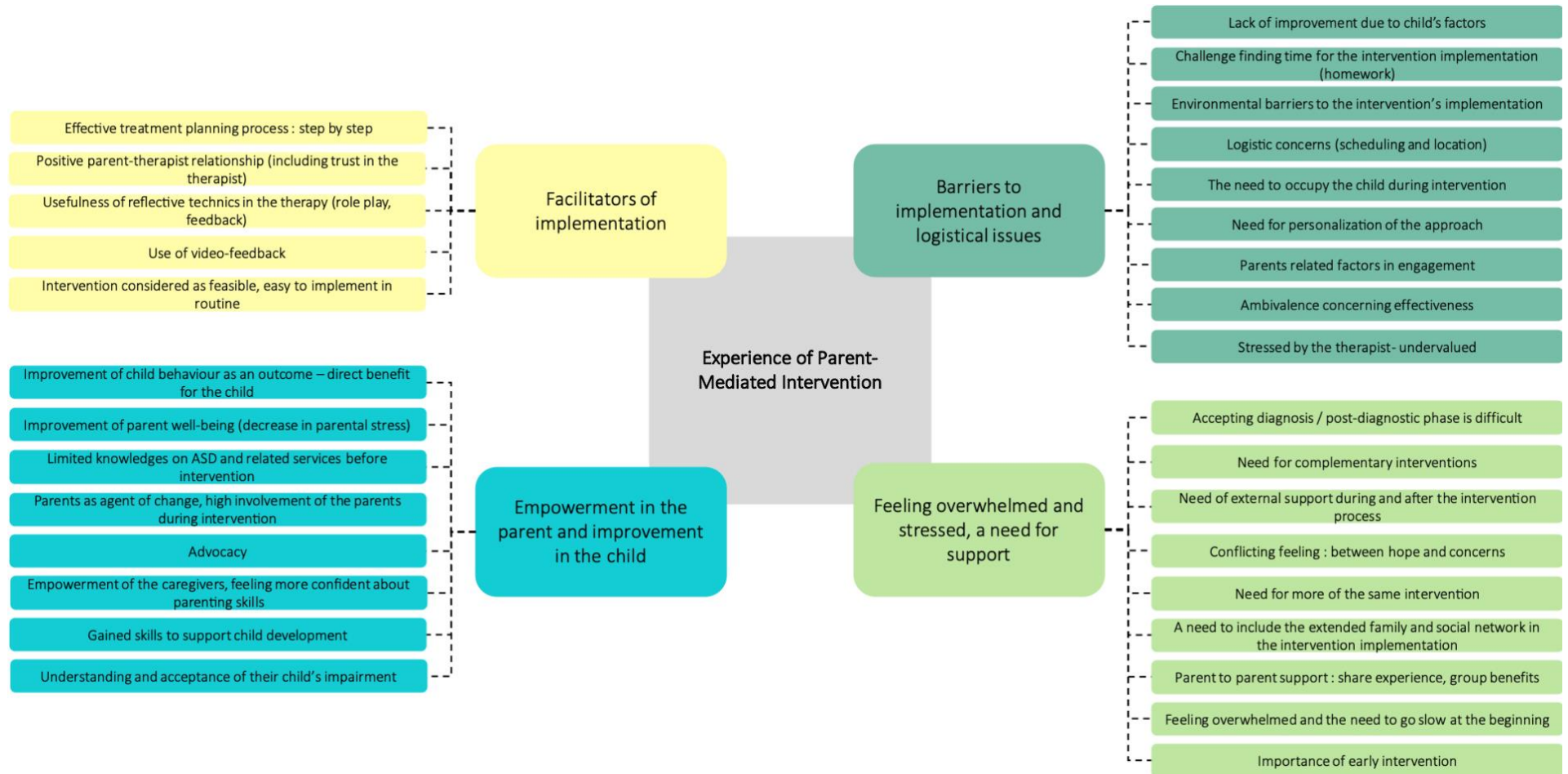
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**FIGURE 1. FLOW CHART**



**FIGURE 2. METASYNTHESIS RESULTS**



**TABLE 1. Assessment of bias using critical appraisal skills program (CASP) tool for quality appraisal in qualitative evidence synthesis.**

*1 = yes, 0 = no or can't/tell, a score below six was considered a high risk of bias.*

	Was there a clear statement of the aims of the research ?	Is a qualitative methodology appropriate ?	Was the research design appropriate to address the aims	Was the recruitment strategy appropriate to	Was the data collected in a way that addressed the research issue	Has the relationship between researcher and	Have ethical issues been taken into consideration ?	Was the data analysis sufficiently rigorous ?	Is there a clear statement of findings ?	How valuable is the research ?	Total	Risk of bias
Amsbary et al. (2020)	1	1	1	1	1	1	1	1	1	1	10	LOW
Blake et al. (2017)	0	1	0	1	0	0	0	0	0	1	3	HIGH
Carr and Lord (2016)	1	1	0	1	0	0	0	1	1	1	6	LOW
Chandler et al. (2002)	1	1	0	0	1	0	0	0	1	1	5	HIGH
Chlebowski et al. (2019)	1	1	1	1	1	1	1	1	1	1	10	LOW
Dababnah and Parish (2016)	1	1	1	1	1	0	1	0	1	1	8	LOW
De Korte et al. (2021)	1	1	1	1	1	1	1	1	1	1	10	LOW
Divan et al. (2015)	1	1	1	1	1	0	1	1	1	1	9	LOW
Donaldson et al. (2011)	1	1	0	1	1	0	1	0	1	1	7	LOW
Furukawa et al. (2018)	0	1	1	0	0	1	1	1	0	1	6	LOW
Hodgetts et al. (2013)	1	1	1	1	1	1	1	1	1	1	10	LOW
Hodgson et al. (2018)	1	1	1	1	0	1	1	1	1	1	9	LOW
Leadbitter et al (2020)	1	1	1	1	1	1	1	0	1	1	9	LOW
Mahonar et al. (2020)	1	1	1	1	1	0	1	0	1	1	8	LOW
Nilses et al. (2019)	1	1	1	1	1	1	1	1	1	1	10	LOW
Ong et al. (2021)	1	1	1	0	1	0	1	0	1	1	7	LOW
Palmer et al. (2020)	1	1	1	1	1	0	1	1	1	1	9	LOW
Parson et al. (2019)	1	1	1	1	1	1	1	1	1	1	10	LOW
Patterson et al. (2011)	1	1	1	0	1	0	0	1	1	1	7	LOW
Peckett et al. (2016)	1	1	1	0	1	1	1	0	1	1	8	LOW
Pickard et al. (2016)	1	1	1	1	1	0	1	1	1	1	9	LOW
Rodgerson	1	1	1	1	1	1	1	1	1	1	10	LOW
Schertz et al. (2020)	1	1	1	1	1	1	1	1	1	1	10	LOW
Stadnick et al. (2012)	1	1	1	0	1	0	1	1	1	1	8	LOW
Stahmer et al. (2017)	1	1	1	1	1	0	1	1	1	1	9	LOW
Zand et al. (2018)	1	1	0	0	1	0	1	0	0	1	5	HIGH

**TABLE 2. Characteristics of the studies included in the metasynthesis.**

Author (date)	Setting and location	Population (N)	Age (SD)	Women ratio	Educational level (N)	Intervention type <i>Name</i>	Intervention frequency (total number of sessions)	Children age (SD)	Children gender (girls)	qualitative method	qualitative analysis framework	Results main themes
Amsbary et al. (2020)	USA	6	35.8 (2.9)	100%	Under high school: 1 High school completion: 0 College level: 5	Individual Therapist-Assisted PMI targeting social skills, using video-feedback  <i>NA</i>	Weekly with home practice between sessions (n=32)	24.5 (4.2)*	20%*	Semi-structured interview	thematic analysis	Effective Training Approach/Process Feeling overwhelmed Daily Implementation Challenges Mixed Feelings Integrating into routines and activities Overall Satisfaction
Carr and Lord (2016)	USA	8	31.4 (4.8)	100%	Under high school: 0 High school completion: 3 College level: 5	Individual Therapist-Assisted PMI targeting social skills  <i>ESI</i>	Twice weekly with home practice between sessions (n=25)	28.4 (5.2)	0%	Semi-structured interview	thematic analysis	Understanding Child's Impairment, Advocacy and Empowerment, Negative Aspects of the Project, and Positive Aspects of the Project
Chlebowski et al. (2019)	USA	29	40 (7.4)	93%	Under high school: 12 High school completion: 9 College level: 8	Individual Therapist-Assisted PMI targeting disruptive behaviors  <i>AIM HI</i>	No info on frequency, home practice between session (n=13)	117.6 (24.7)	10%	Semi-structured interview	thematic analysis	Limited ASD Knowledge Expectations of parental involvement Influences on parent-therapist interactions
Dababnah and Parish (2016)	USA	17	NA	NA	Under high school: 0 High school completion: 2 College level: 15	Group PMI targeting social skills  <i>The Incredible Years</i>	Weekly with home practice between sessions (n=15)	44.4 (11)	30%	Semi-structured interview	thematic analysis	Planning IY Setting the stage: the first session Optimizing parent learning throughout the program Thinking to the future: additional supports for ASD-specific groups
de Korte et al. (2022)	Netherlands	17	36.2 (6.1)	71%	Under high school or high school completion: 5 College level: 11 Unknown : 1	Group PMI Targeting social skills  <i>PRT</i>	Weekly (n=14)	60.12 (14.4)	0%	Semi-structured interview	thematic analysis	Facilitators and barriers Perceived effects

Divan et al. (2015)	India	17	NA	94%	Under high school: 1 High school completion: 16 College level: 0	Individual Therapist-Assisted PMI targeting social skills, using video-feedback	Fortnightly then monthly with home practice between session (n=18)	133.2 (23.5)	NA	Semi-structured interview and focus groups	framework analysis	Adaptation of content Mode of delivery
Donaldson et al. (2011)	USA	10	31 to 51	0%	Under high school: 0 High school completion: 0 College level: 10	Individual Therapist-Assisted PMI targeting social skills, using video-feedback	Twice weekly with home practice between sessions (n=20)	57.6 (8.5)	10%	Semi-structured interview	NA	Sharing Time Having a Close Relationship Accepting the Diagnosis Concerns of, and Hope for, the Future Benefits of the Program
Furukawa et al. (2018)	Japan	21	38.4 (4.3)	100%	Under high school: 0 High school completion: 2 College level: 11	Individual Therapist-Assisted PMI targeting social skills	Weekly (n=8)	62.9 (14.6)	19%	Semi-structured interview	KJ-method	Changes in the Parent-Child interaction Changes in the Child Changes in the Parent Others
Hodgetts et al. (2013)	Canada	10	NA	60%	NA	Individual Therapist-Assisted PMI targeting disruptive behavior	Weekly (n=10)	93.6 (35.2)	33%	Semi-structured interview	thematic analysis	Attribution of cause Who's the boss? Rewarding is rewarding!
Hodgson et al. (2018)	UK	14	NA	93%	Under high school: 6 High school completion: 0 College level: 7	Group and individual Therapist-Assisted PMI targeting disruptive behaviors	Weekly with home practice between sessions (n=8)	60.4 (19.1)	25%	Focus-group	framework analysis	Limited knowledge of RRB Commitment needed to attend and gain from the intervention Benefits of a group-based intervention Homework
Leadbitter et al. (2020)	UK	18	35.6 (6.1)	67%	Under high school: 0 High school completion: 9 College level: 9	Individual Therapist-Assisted PMI targeting social skills, using video-feedback	Fortnightly then monthly with home practice between session (n=18)	44.4 (7)	8%	Semi-structured interview	thematic analysis	The backdrop of the therapy: emotional starting point, hopes and expectations Therapeutic process Practical challenges of the therapy Therapeutic outcomes

Mahonar et al. (2020)	India	54	NA	56%	Under high school completion: 0 High school completion: 27 College level: 27	Individual Therapist-Assisted PMI targeting social skills and disruptive behaviors	Fortnightly with home practice between session (n=5)	41.5 (17.2)	20%	Semi-structured interview	thematic analysis	Impact of the child's disorder Treatment impact and change process Validation and acceptance
Nilses et al. (2019)	Sweden	11	34.5 (7.4)	55%	Under high school completion: 1 High school completion: 8 College level: 3	Individual Therapist-Assisted PMI targeting social skills  <i>P-ESDM or Intensive Learning for Young Children with Autism</i>	Twice weekly with home practice between sessions (n=NA)	54.6 (8.3)	NA	Semi-structured interview	thematic analysis	A overwhelming diagnostic Cooperating with the team Growing as a parent
Ong et al. (2021)	Malaysia	8	NA	75%	NA	Group PMI Targeting social skills  <i>CFT</i>	Weekly with home practice between sessions (n=12)	90 (6.1)	25%	Focus group	thematic analysis	Fear and resistance Awareness, learning, and adjustment Change is hard Identifying support
Palmer et al. (2020)	UK	9	35.3 (6.8)	82%	NA	Group and individual Therapist-Assisted PMI targeting social skills and disruptive behaviors, using video-feedback	Weekly (n=11)	52.1 (9.6)	24%	Semi-structured interview	thematic analysis and framework analysis	Positive aspects of the intervention Challenges to the delivery of EarlyBird
Parson et al. (2019)	Australia	24	37 (5)	100%	Under high school completion: 0 High school completion: 20 College level: 4	Individual Self-Guided PMI targeting social skills (technology based)  <i>TOBY app</i>	No info on frequency, home practice between session (n=NA)	60 (18.9)	25%	Semi-structured interview	thematic analysis	If you have met one child with Autism, you have met one child with autism The Toby App is just one piece of the jigsaw
Patterson et al. (2011)	USA	7	NA	NA	Under high school completion: 0 High school completion: 2 College level: 2	Group and individual Therapist-Assisted PMI targeting social skills  <i>MTW</i>	Weekly (n=11)	31.8 (3.8)	25%	Semi-structured interview	thematic analysis	Program as a good starting point for their learning Unmet emotional and information needs Parent-to-parent contact Individualized coaching and feedback Child-centered factors Special consideration for parents who worked outside the home

Peckett et al. (2016)	UK	5	41 (4.9)	100%	NA	Individual Therapist-Assisted PMI targeting social skills	No info on frequency, home practice between session (n=2)	122.4 (18)	20%	Semi-structured interview	thematic analysis	Family-specific factors Child-specific factors Intervention-specific factors
Pickard et al. (2016)	USA	10	NA	100%	Under high school: 0 High school completion: 1 College level: 8	Individual Self-Guided or Therapist-Assisted PMI targeting social skills (technology based)	No info on frequency, home practice between session (n=12)	46.7 (12.6)	30%	Semi-structured interview	thematic analysis	Perceptions of acceptability Ease of learning ImPACT Online Relative advantage Parent and child outcomes Perceptions of barriers Parent suggestions for dissemination Preparation Support Activities Personal factors
Rodgers et al. (2019)	Australia	17	38.4 (4.9)	77%	Under high school: 0 High school completion: 0 College level: 17	Individual Self-Guided PMI targeting social skills (technology based)	No info on frequency, home practice between session (n=NA)	48 (0)	NA	Semi-structured interview	thematic analysis	Preparation Support Activities Personal factors
Schertz et al. (2020)	USA	11	31.5 (5.3)	100%	NA	Individual Therapist-Assisted PMI targeting social skills, using video-feedback	No info on frequency, home practice between session (n=NA)	27.2 (3.7)	36%	Semi-structured interview	thematic analysis	Autism-related early challenges to parent self-efficacy Parent/professional roles and PSE Manifestations and personal contributors to PSE Child autonomy as an extension of PSE Therapy Process Impression of Therapy Outcomes/Impact
Stadnick et al. (2012)	USA	13	43.4 (6.3)	85%	Under high school: 0 High school completion: 6 College level: 7	Individual Therapist-Assisted PMI targeting disruptive behaviors	No info on frequency, home practice between session (n=11)	123.2 (27)	0%	Semi-structured interview	thematic analysis	Therapy Process Impression of Therapy Outcomes/Impact
Stahmer et al. (2017)	USA	9	34.9 (4)	100%	Under high school: 0 High school completion: 1 College level: 8	Individual Therapist-Assisted PMI targeting social skills	Weekly with home practice between sessions (n=12)	15 (3)	15%	Semi-structured interview	thematic analysis	Parent coaching process Impression of the intervention Treatment planning process Homework Logistics of the intervention format Perceived effectiveness

Improved child social  
communication  
Reduced parent stress

\* data come from the total RCT population and not the subgroup in the qualitative study

N : number ; SD : standard deviation ; PMI : Parent-Mediated Intervention ; ESI : Early Social Interaction (ESI) ; AIM HI : An Individualized Mental Health Intervention for ASD ; PRT : Pivotal Response Treatment ; PACT : Preschool Autism Communication Therapy ; FDIT : Elder's Father-Directed In-Home Training ; PCIT : Parent-Child Interaction Therapy ; SSTP : Stepping Stones Triple P ; MRB : Managing Repetitive Behaviours ; P-ESDM : Parent-Early Start Denver Model ; CFT : Children's Friendship Training ; TOBY App : Therapeutic Outcomes by You application ; MTW : More Than Words ;

**TABLE 3. Summary of findings.**

Categories has been classified by level of confidence inside each synthesized finding.

Categories	CerQUAL confidence	Related quote	Studies
<b>Facilitators of implementation</b>			
<b>Positive parent-therapist relationship (including trust in the therapist)</b>	High confidence	<i>"We had a relationship of trust, it is as if I was talking with a friend, she made me feel as a friend, as someone who I could trust, as someone who listens to me and is not criticizing or watching to see what I'm going to say"</i> (Chlebowski et al., 2018)	Amsbary et al. (2020), Carr and Lord (2015), Chlebowski et al. (2019), Divan et al (2015) Leadbitter et al. (2020), Nilses et al. (2019), Stadnick et al. (2012), de Korte et al. (2021)
<b>Use of video-feedback</b>	High confidence	<i>"You can't think of everything and you don't see all the little things that go right or wrong or anything, so it is so very, very good to watch yourself back on it."</i> (Leadbitter et al., 2020)	Amsbary et al. (2020), Divan et al (2015), Leadbitter et al. (2020), de Korte et al. (2021)
<b>Usefulness of reflective technics in the therapy (role play, feedback)</b>	High confidence	<i>"the time that we spent together working in the therapy setting was great, because I could watch [the therapist] do the strategies and then she gave me time and real-time feedback on how to do things and then she'd always ask me how it was going."</i> (Stahmer AC. et al., 2017)	de Korte et al. (2021), Divan et al (2015), Leadbitter et al. (2020), Mahonar et al. (2020), Stadnick et al. (2012), Stahmer et al. (2017)
<b>Intervention considered as feasible, easy to implement in routine</b>	High confidence	<i>"So, I just did it in different scenarios during the day"</i> (Amsbary et al., 2020)	Amsbary et al. (2020), de Korte et al. (2021), Divan et al (2015), Hodgson et al. (2018), Mahonar et al. (2020), Nilses et al. (2019), Ong et al. (2021), Pickard et al. (2016), Stahmer et al. (2017)
<b>Effective treatment planning process: step by step</b>	Low confidence	<i>"I think the part that worked best for us was breaking things down into the smaller goals"</i>	Amsbary et al. (2020), Stahmer et al. (2017)
<b>Barriers to implementation and logistical issues</b>			
<b>Challenge finding time for the intervention implementation (homework)</b>	High confidence	<i>"Sometimes I am lazy, means the half an hour practice session is the most difficult to tell you the truth."</i> (Divan G. et al., 2015)	Amsbary et al. (2020), Divan et al (2015), Furukawa et al. (2018), Leadbitter et al. (2020), Palmer et al. (2020), Peckett et al. (2016), Pickard et al. (2016), Rodgerson et al. (2019), Stahmer et al. (2017), Ong et al. (2021), de Korte et al. (2021)
<b>Logistic concerns (scheduling and location)</b>	High confidence	<i>"We didn't have to go all the way across the city to see the team. Yeah ... that was better for us. Otherwise it would have been hard, both for us, and for X too"</i> (Nilses et al., 2019).	Carr and Lord (2015), Leadbitter et al. (2020), Nilses et al. (2019), Patterson et al. (2011), Pickard et al. (2016), Stahmer et al. (2017)
<b>Need of personalisation of the approach</b>	High confidence	<i>"For me the PECS stuff wasn't useful because my child's verbal, but obviously it's a very wide spectrum. I don't know in the future if it may be better to group parents in terms of having a non-verbal group and a verbal group."</i> (Palmer M. et al., 2020)	Amsbary et al. (2020), Dababnah and Parish (2016), de Korte et al. (2021), Palmer et al. (2020), Parson et al. (2019), Patterson et al. (2011)
<b>Environmental barriers to the intervention's implementation</b>	Moderate confidence	<i>"[My daughter]'s almost two years older and she was in pre-K and all that so I had to entertain her and then we had homework."</i> (Amsbary et al., 2020)	Amsbary et al. (2020), Ong et al. (2021)
<b>Challenge implementing due to child's factor</b>	Moderate confidence	<i>"It's hard to get him to sit for that long and do blocks. Besides, he likes to line them up and play different."</i> (Carr T. & Lord C., 2016)	Carr and Lord (2015), Peckett et al. (2016), Rodgerson et al. (2019), Ong et al. (2021)

<b>The need to occupy the child during intervention</b>	Moderate confidence	<i>"The only thing I found really hard, [child's name] found it stressful the fact that he couldn't get my attention afterwards, 'cos I had to try and watch the video and talk to [therapist's name] and I found it so hard and I think he did 'cos he mithered [pestered] to death, 'Mummy, mummy, mummy, mummy', and, er, I was torn between the two, trying to concentrate and him."</i> (Leadbitter et al., 2020)	Dababnah and Parish (2016), Leadbitter et al. (2020)
<b>Parent-related factors in engagement</b>	Moderate confidence	<i>"I spend more time with him so . . . maybe what I do sticks with him better just because I'm able to repeat it a lot more throughout the week."</i> (Schertz HH. et al., 2020)	Ong et al. (2021), Rodgerson et al. (2019), Schertz et al. (2020)
<b>Ambivalence concerning effectiveness</b>	Moderate confidence	<i>"I don't know whether that's just the Lego, I think it definitely has helped during the sessions, that we were calm and patient, obviously there was no frustration there, but it's just because Robert generally seems a lot happier, that obviously there is less friction anyway ... it's probably a combination, yeah, probably a combination yeah of him generally being happier and also I think yeah, maybe the Lego."</i>	Parson et al. (2019), Peckett et al. (2016), Stahmer et al. (2017)
<b>Stressed by the therapist - undervalued</b>	Low confidence	<i>"I have never been more intimidated [than] by this team... You have professionals coming in... telling you what you should do... I wish I could be a part of the discussion... I have not felt an equal member of the team. They come over as, "We're the experts." (Rogerson J. et al., 2019)</i>	Carr and Lord (2015), Schertz et al. (2020)

**Empowerment in the parent and improvement in the child**

<b>Improvement of child behavior as an outcome - direct benefits on the child</b>	High confidence	<i>"He's got a lot more eye contact, a lot more communication, like verbally and non-verbally."</i> (Carr T. & Lord C., 2016)	Carr and Lord (2015), de Korte et al. (2021), Donaldson et al. (2011), Furukawa et al. (2018), Hodgson et al. (2018), Leadbitter et al. (2020), Palmer et al. (2020), Peckett et al. (2016), Rodgerson et al. (2019), Stadnick et al. (2012), Stahmer et al. (2017)
<b>Improvement of family relationships and interactions</b>	High confidence	<i>"Our relationship now is... actually very strong... I know how to reach him and how to interact with him and how to have fun with him... he seems to be coming along every day... just something new every day. He's coming out of his shell a little bit more and he does interact with us and it's... its good."</i> (Donaldson SO. et al., 2011)	Amsbary et al. (2020), Carr and Lord (2015), de Korte et al. (2021), Donaldson et al. (2011), Furukawa et al. (2018), Leadbitter et al. (2020), Nilses et al. (2019), Peckett et al. (2016)
<b>Empowerment of the caregivers, feeling more confident about parenting skills</b>	High confidence	<i>"I was very insecure about myself and my parenting skills concern- ing [child's name], because I tried so much and nothing seemed to help... I became more self-confident and now I'm like 'I'm not doing anything wrong, but there are some adjustments possible.'" (de Korte et al., 2022)</i>	Carr and Lord (2015), de Korte et al. (2021), Donaldson et al. (2011), Furukawa et al. (2018), Hodgetts et al. (2013), Hodgson et al. (2018), Pickard et al. (2016), Rodgerson et al. (2019)
<b>Parents as agent of change, high involvement of the parent during intervention</b>	High confidence	<i>"They [traditional early interventionists] write up a plan, they decide... When [the parent-mediated intervention professional] is here... we come up with a plan together... Because I come up with it, I'm more willing to do it"</i> (Schertz HH. et al., 2020)	Chlebowski et al. (2019), Divan et al (2015), Hodgson et al. (2018), Schertz et al. (2020), Stadnick et al. (2012), Stahmer et al. (2017)
<b>Gained skills to support child development</b>	High confidence	<i>"I also learnt. So, not just to bring, let him join. Then, we also have to guide. This is very important. To guide him. Tell him what to do, make, making a friend. That's very important."</i> (Ong et al., 2021)	Amsbary et al. (2020), Ong et al. (2021), Parson et al. (2019), Peckett et al. (2016), Schertz et al. (2020), Stadnick et al. (2012)
<b>Understanding and acceptance of the child's impairments</b>	High confidence	<i>"not only speech, his communication and interaction are equally important"</i> (Manohar et al., 2020)	Carr and Lord (2015), de Korte et al. (2021), Hodgetts et al. (2013), Leadbitter et al. (2020), Mahonar et al.

<b>Limited knowledges on ASD and related services before intervention</b>	High confidence	<i>"At first I thought he has to see a doctor to have an examination to sort out the problem.... It's something he has ... somewhere he's in pain ... or suffers from some kind of disease ... but I was never aware of any autism. [...] That didn't exist in my mind, those kind of diseases ... "</i> (Nilses et al., 2019)	(2020), Ong et al. (2021), Palmer et al. (2020), Peckett et al. (2016), Schertz et al. (2020) Carr and Lord (2015), Chlebowski et al. (2019), Hodgson et al. (2018), Nilses et al. (2019), Ong et al. (2021), Patterson et al. (2011)
<b>Lack of improvement due to child's factors</b>	Moderate confidence	<i>"I was expecting that it would help more with social skills, I guess... He found it too slow, too boring and I guess that's because of his level of functioning compared to other children."</i> (Rodgerson J. et al., 2019)	Ong et al. (2021), Patterson et al. (2011), Rodgerson et al. (2019), Schertz et al. (2020)
<b>Improvement of parent well-being (decrease in parental stress)</b>	Moderate confidence	<i>"I'm a lot calmer and I don't scream at him anymore. More in control"</i> (Palmer M. et al., 2020)	Furukawa et al. (2018), Mahonar et al. (2020), Palmer et al. (2020), Stahmer et al. (2017)
<b>Advocacy</b>	Very low confidence	<i>"I try get awareness out as much as I can... people will ask me questions and I'm like okay, hold on let me make sure I'm giving you the right information."</i> (Carr T. & Lord C., 2016)	Carr and Lord (2015), de Korte et al. (2021)
<b>Feeling overwhelmed and stressed: a need for support</b>			
<b>Need of external support during and after the intervention process</b>	High confidence	<i>"I think we would have been okay up to a point and then at the end it would have been really, really tough putting it together, because that is when I really relied on the coach."</i> (K. E. Pickard et al., 2016)	Dababnah and Parish (2016), Furukawa et al. (2018), Nilses et al. (2019), Parson et al. (2019), Pickard et al. (2016), Rodgerson et al. (2019), Ong et al. (2021)
<b>A need to include the extended family and the social network in the intervention implementation</b>	High confidence	<i>"[My partner attending] would've helped me to feel more supported ... [A] lot of the moms I met are the ones doing the up-front work [on ASD]. I feel like if the other spouse or partner was involved ... it would lend some credence to some of the stuff on the autism front ... I feel that sometimes that exposure to someone other than your spouse relaying all of this information ... can be a nice way to bring couples closer together."</i> (Dababnah S. & Parish SL., 2016)	Amsbary et al. (2020), Dababnah and Parish (2016), de Korte et al. (2021), Hodgson et al. (2018), Ong et al. (2021)
<b>Feeling overwhelmed and the need to go slow at the beginning</b>	High confidence	<i>"Yeah, to me at times it seemed like you go over stuff each session, but then they give you time to ask questions and stuff but then, there were still questions that didn't get answered, sometimes I felt like I left without knowing something... I wanted to ask more about it but we didn't have time so I guess... maybe we need a longer period of time, cover less aspects in one night so you have more time to discuss it..."</i> (Patterson & Smith, 2011)	Amsbary et al. (2020), Dababnah and Parish (2016), Patterson et al. (2011)
<b>Accepting diagnosis / post-diagnostic phase is difficult</b>	High confidence	<i>"I already knew most of it so I wasn't shocked really. It was hard to take at first"</i> (Carr T. & Lord C., 2016)	Carr and Lord (2015), Donaldson et al. (2011), Nilses et al. (2019), Ong et al. (2021)
<b>Conflicting feeling: between hope and concerns</b>	High confidence	<i>"We're not worried any more, never ... We were before, when this whole thing started, but these past six months ... this past year, our feelings have changed."</i> (Nilses et al., 2019)	Carr and Lord (2015), de Korte et al. (2021), Donaldson et al. (2011), Furukawa et al. (2018), Leadbitter et al. (2020), Mahonar et al. (2020), Nilses et al. (2019), Patterson et al. (2011), Rodgerson et al. (2019)
<b>Parent to parent support : share experience, group benefits</b>	High confidence	<i>"It helped me to interact with the other families. It taught me that I wasn't alone, there are many people going through the same struggles. At the beginning, I thought I was basically by myself you know, dealing with this but actually it's nice"</i>	Dababnah and Parish (2016), de Korte et al. (2021), Hodgson et al. (2018), Ong et al. (2021), Palmer et al. (2020), Patterson et al. (2011)

<b>Need for parents self-care</b>	High confidence	<i>to have a group setting and to have other people views and opinions...</i> (Patterson & Smith, 2011) <i>"Market [the program] as how to help your children. You'd have to trick us into [focusing on ourselves]! It feels so selfish, but ... when you get on the scale and you gained 40 pounds, or when you realize you've been collecting records and you haven't listened to them in over a year ... That's pretty depressing ... Then for me, you get resentful of your life ... when you see your partner doing something enjoyable, you're like, "How dare you?" Just all of this stuff, please include it more."</i> (Dababnah S. & Parish SL., 2016)	Dababnah and Parish (2016), Nilses et al. (2019), Patterson et al. (2011)
<b>Need for complementary interventions</b>	Moderate confidence	<i>"Maybe not this particular intervention but the next step maybe, in some kind of social skills groups."</i> (Amsbary et al., 2020)	Amsbary et al. (2020), Carr and Lord (2015), Parson et al. (2019)
<b>Need for more of the same intervention</b>	Moderate confidence	<i>"More on helping to make friends and developing theory of mind. Social skills training would have been really helpful. The social stories were helpful and some of the books but there need to be more. We're working on theory of mind with my child now and there are things that you can do."</i> (Palmer M. et al., 2020)	Dababnah and Parish (2016), Palmer et al. (2020)
<b>Importance of early intervention</b>	Very low confidence	<i>"I read that it's better if you find out and get your child help when they're young because they actually may have signs of it at all time but they can be live to be productive normal people"</i> (Carr T. & Lord C., 2016)	Carr and Lord (2015), Pickard et al. (2016)

Le "supplemental material" est disponible en ligne : <https://journals.sagepub.com/doi/abs/10.1177/13623613221112204>

# ANNEXE 4 : EFFICACY OF PARENT- MEDIATED COMMUNICATION- FOCUSED TREATMENT IN TODDLERS WITH AUTISM (PACT) DELIVERED VIA VIDEOCONFERENCING: A RANDOMISED CONTROLLED TRIAL STUDY PROTOCOL

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Jurek, L., Occelli, P., Denis, A., Amestoy, A., Maffre, T., Dauchez, T., Oreve, M.-J., Baghdadli, A., Schroder, C., Jay, A., Zelmar, A., Revah-Levy, A., Gallifet, N., Aldred, C., Garg, S., Green, J., Touzet, S., & Geoffray, M.-M. (2021). **Efficacy of parent-mediated communication-focused treatment in toddlers with autism (PACT) delivered via videoconferencing : A randomised controlled trial study protocol**. *BMJ Open*, 11(4), e044669.

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## Abstract

**Introduction:** Intervention in the preschool period is currently recommended for Autism Spectrum Disorder (ASD). Therapies delivered by parents are particularly suitable for young children. PACT (Preschool Autism Communication Trial) is a parent-mediated therapy that has shown a significant and sustained impact on autism symptom reduction. However, access to such evidence-based therapies for families is limited due to autism centers located in large urban areas. Using videoconferencing to deliver PACT training to parents may improve accessibility for families living in underserved areas.

**Methods and analysis:** This single-blind randomized controlled trial, involving six sites in France, will investigate the efficacy of a telehealth, videoconferencing-based, parent-mediated PACT therapy on autism symptoms, over a 12-month period. It will compare PACT plus treatment as usual (TAU) against TAU only in a cohort of 238 toddlers (119 per group) aged 18 to 36 months at inclusion and living with their families more than 40 minutes away from the specialist centres for autism. Primary outcome will include change of overall autism score on the Autism Diagnostic Observation Scale (ADOS) at 12 months. Secondary outcomes will measure change in child skills, child functioning, impact on parents (stress, health, priorities) and implementation characteristics.

Repeated measures analyses will be used to test the effect of PACT intervention on the overall ADOS module 1 score over the 12-month study period. Linear mixed models will be used with time, treatment allocation and the interaction between treatment and time as fixed effects and individual variation as random effect.

**Ethics and dissemination:** This protocol (version N°5, date: 10/25/2019) is approved by the French National Review Board (reference No 2018-A02516-49). The results will be disseminated via peer-reviewed journals

# Article Summary

## Strengths and limitations of this study

- Large multicentre randomised controlled trial (RCT) in children with ASD under 3 years old, testing remote delivery of an evidenced intervention
- Assessment partially done by videoconferencing and based on video material sent by parents
- Recruitment targeted to children living in underserved areas
- Short term effect of the intervention will be assessed at 12 months (end of the intervention)
- Owing to the nature of the intervention, parents of the children and PACT therapists will not be blind to the allocation group.

**Keys Words:** Early intervention, Autism spectrum Disorder, PACT, videoconferencing, parent-mediated therapy, video-feedback, Health Services Accessibility

**Trial registration:** ClinicalTrials.gov Identifier: NCT04244721

## Introduction

### Rationale & background

Autism Spectrum disorder (ASD) is a common neurodevelopmental disorder with a population prevalence of at least 1.5% in developed countries and can cause significant lifelong disability [1–3] and burden for families and caregivers [4]. Diagnosis is possible as early as 18 months of age [5]. Current evidence suggests that interventions delivered in the early developmental period before the age of 3 years has the potential for maximal impact on autism symptom severity.

Therapies can be delivered by therapists, teachers and parents [6]. For preschool children with ASD, parent-mediated therapies can guide parents to face challenging social interaction with their children [7,8]. Among the different parent-mediated therapies, PACT (Pre-school Autism Communication Therapy) has shown significant short and long-term efficacy on objectively assessed autism symptoms in children aged from 2 to 10 years in a large UK cohort (N=152); as showed in a recent systematic review [9] and meta-analysis [10]. In PACT, parents are guided by a therapist via video feedback to optimize parent-child dyadic interaction, which in turn impacts on child language, communication and autism symptoms [11]. In a trial of PACT intervention compared to regular care, PACT showed a statistically significant effect at 13 month endpoint to reduce of autism symptom severity measured on Autism diagnostic Observation Schedule version 2 (ADOS-2) (effect size 0.64; 95%CI 0.07-1.20); and an increase in parental communication synchrony with the child and child communication initiations with the parent [12]. The follow-up study showed evidence of sustained effect on autism symptom severity six years after intervention end, with a significant overall reduction in symptom severity over the course of trial and follow-up period (effect size=0.55, 95% CI 0.14 to 0.91, p=0.004)[13]. A mechanistic study also confirmed that the distal effect of PACT therapy on autism severity measured

by ADOS was mediated by the improvement of child communication initiations, which in turn was mediated by improved parent-child synchrony [14].

Availability of PACT therapy is limited; even more so in rural settings or in regions away from specialist centres. Using videoconferencing run by therapists to train parents in PACT, may therefore be a viable alternative to make such therapies available to families living far from autism centers. Previous studies have shown that it is possible to provide parent-mediated therapies in autism by videoconferencing successfully [15,16]. The team who developed the PACT therapy reported positive experiences of parent guidance by videoconferencing (C. Aldred and J. Green, personal communication, June 10, 2020). Indeed, videoconferencing was used to deliver some of the PACT sessions in a recently published RCT [17]. However, PACT has never been evaluated when exclusively delivered by videoconferencing. The barriers and facilitators to delivering such therapies via videoconferencing are not sufficiently well understood, and hence it is essential to investigate and address them adequately [18,19].

Research question: The proposed protocol is for a large RCT in children under 3 years with ASD to evaluate the effectiveness on autistic symptom severity and other measures of PACT therapy delivered to parents by videoconferencing.

A significant effect would justify and facilitate the routine use of videoconferencing therapy in early intervention and improve the dissemination of this evidence-based practice.

The hypothesis is that PACT intervention delivered by videoconferencing + TAU (Treatment As Usual) will have a superior efficacy on child autism symptom severity as compared to TAU alone.

## **Objectives**

Our primary objective will be to test the efficacy of a parent-mediated PACT therapy, delivered by trained therapists to parents living in underserved areas via videoconferencing over a 12-months period, on overall autistic symptoms in children with ASD aged from 18 to 36 months at inclusion, measured using the Autism Diagnostic Observation Schedule 2 (ADOS),

Our secondary objectives will include an evaluation of change in child socio-communicative interactions, language, communication initiation and daily adaptive behaviour. At the parent level, we will evaluate the intervention effects on stress, health and family functioning.

The implementation of the therapy will be evaluated through the adherence of professionals and parents to PACT, and acceptability and feasibility of the PACT sessions to parents and therapists.

## **Method**

### **Study design**

This is a prospective multicentre RCT with 2 parallel group, 1:1 ratio, single blind comparing PACT intervention + Treatment As Usual (TAU) against TAU alone. Evaluation will be carried out using quantitative and qualitative mixed-method approaches [20,21].

Semi-structured interviews with parents and therapists will be conducted to understand the barriers and facilitators of using the videoconferencing approaches to delivering the PACT therapy.

Figure 1 shows consort flow-chart of the study.

## **Setting**

We will run this trial in six academic centres located in child and adolescent public hospitals in France. All centres have a unit for ASD diagnosis and assessment and a distinct unit for intervention where therapists have been trained in PACT and can provide PACT via videoconferencing. The parents receiving intervention come from a French-speaking population including socioeconomically disadvantaged groups.

## **Patient and Public Involvement**

Mrs Alloisio (Association AAA <https://autisme-ambitionavenir.com>) and Mr Belkhat (association <https://bleunetwork.fr/pro>) are parents of a child with autism and represent two different associations. They are part of the steering committee.

## **Population**

### ***Inclusion criteria***

Children will be included if they meet the following criteria:

- i) aged between 18-36 months old at referral
- ii) meet criteria for autism spectrum disorder using the two gold standard instruments ADOS-2 (Autism diagnostic Observation Schedule -2) and ADI-R (Autism Diagnostic Interview-Revised). For inclusion into the study, the severity score comparison (CSS) on ADOS-2 will be greater or equal to 4. The score on the ADI-R algorithm for toddlers will be greater or equal to 11 [22]. The diagnosis will be confirmed by a multidisciplinary team trained in ASD assessment and diagnoses based in the academic departments of the hospitals.
- iii) have a non-verbal age equal to or above 12 months on the Mullen Scale of Early learning ( MSEL) [23].
- iv) live more than 40 minutes away from a Center for Resources in Autism (regional center)

Referred parents will be included if they meet the following criteria:

- ii) speak French with their children

iii) are able to use videoconferencing methods with therapists who will be based at the center (assessed through the conduct of the Vineland Assessment Behavioral Scale by videoconferencing) (see Appendix 1 for details)

### ***Exclusion criteria***

Exclusion criteria for the child will include:

- i) a twin brother or sister with ASD or a brother or sister already included in the study
- ii) diagnosed with epilepsy requiring medication
- iii) severe hearing or visual impairment
- iv) an identification of a genetic anomaly which may impact on their ability to participate in the intervention or on data validity (determined by the principal investigator on a case-by-case basis).

Exclusion criteria for the referred parents (at least one parent with) will include:

- i) severe hearing or visual impairment
- ii) severe psychiatric disorder
- iii) unstable somatic disorders preventing participation in the intervention
- iv) lack of internet provision
- v) not available for regular intervention and follow-up
- vi) opposition of one parent to the child's participation in the study
- vii) currently undertaking PACT therapy

## **Intervention conducted in the experimental group**

### ***Eligibility criteria for PACT therapist and adherence***

Therapists delivering the intervention will include speech language pathologists, occupational therapists, clinical nurses, psychologists or child and adolescent psychiatrists, all specialised in ASD. The therapists have already received formal training and supervision in PACT from the team who developed this training [12,24]. Fidelity to PACT therapy will be maintained by regular meetings between therapists of all centers, with scoring and feedback of videotaped therapy sessions obtained during the study.

### ***PACT treatment principles***

As previously described [17], parents will be trained, via video-feedback, to identify and set key strategies facilitating the socio-communicative interactions between their child and themselves. Parents will also be encouraged to use PACT every day outside the training session at least half an hour a day. The therapy follows a six-staged approach based on child developmental progression and strategies for establishing fundamental skills for the socio-communicative development. The first two stages aim to increase parent's identification of child focus and interest, synchrony, responsiveness and sensitivity to the child interest and communication. The third and fourth stages are targeted towards developing expression and comprehension of the child by commenting and modelling language adapted to the child's developmental level. Child communication initiation is also improved

in the fifth stage through different strategies such as anticipation and routine. The last stage aims to develop conversation and expansion of language for verbal children. Progression from one stage to the next depends on predefined criteria.

Based on the protocol of the first PACT RCT, parents will receive 18 sessions of training in PACT with the therapist over the 12 months [12,13]: 1-hour session every fortnight for the first 6 months to learn PACT strategies followed by 1 hour session per month over the next 6 months to maintain the capacity of parents to deliver the strategies.

Therapist will train only one parent per family and maintain fidelity to the therapy manual. The “referent parent” will have to be designated before the randomisation of the child. If the referent becomes unavailable, the therapy will stop or will continue with the other parent if possible and this change will be reported.

### ***Parent training session with the professional via videoconferencing***

Before each session, the parent will be asked to send a 10-minute video of their interaction with their child to the therapist via a secure cloud link. During the videoconferencing session (as in face-to-face intervention), referent parent will begin with a 5-minute discussion about their progress since the last session. The therapist will then share his/her screen and watch the home-based 10 minutes video together with the parent. Together the parent and therapist will identify, review and discuss specific clips that demonstrate accomplishment of therapy goals for the relevant stage of the PACT programme. The therapist’s role will be to guide parents to identify successful strategies and responses (i.e. episodes of engagement and/ or mutual sharing with their child). Parents will be supported to reflect on their role in enhancing interaction and to identify new intervention goals.

### ***Parent PACT implementation in daily life outside the therapy session***

At the end of each session, the therapist will support the parent in setting 2-3 new goals, based on the strategies identified during the session. The therapist will encourage the parent to practice the strategies for the next session and discuss opportunities to achieve these goals in daily routine at home for at least 30 minutes per day. Parents will be guided to embed PACT strategies in everyday routines across different contexts. As therapy progresses, parents will be asked to send 10-minutes home videos of daily routines in different contexts.

### **Treatment as usual (TAU) and 2 follow-up consultations on ASD and its management**

Regardless of group allocation, parents will receive treatment as usual (TAU) consisting of psychoeducation about ASD, management and educational support for nursery and preschool placement. Parents will be referred to any relevant care available in the community (e.g.: Speech language pathologist (SLP), occupational therapist, educator, behavioural psychologist, psychiatrist). TAU received during the course of the study will be described in both groups.

Regardless of group allocation, a psychiatrist or a psychologist from each autism center will provide two supplementary 45 minutes follow-up consultations conducted by videoconferencing at 3 and 6 months after inclusion. This consultation will be carried out by following an interview guide. Three thematic areas will be systematically discussed with the parents: ASD information, access to treatment in the community, support for school or nursery. These follow-up consultations will ensure that parents of both groups receive homogeneous information on ASD and its management.

### **Avoidance of contamination**

Currently, PACT is not widely implemented in the community in France, particularly in the rural areas. Any families who are currently in receipt of PACT intervention will be excluded from this trial. However, any PACT that might be received in the community as part of TAU will be recorded .[25].

Research assessors will be separate to the therapists and will be located and supervised separately in each center.

Professionals conducting the follow-up consultations will not be trained in PACT therapy or be part of the research assessments.

### **Measures**

#### ***Primary outcome***

#### **To assess autism severity**

Autism Diagnostic Observation Schedule (ADOS-2) is a semi-structured, researcher-child interaction based, standardized observational assessment, in communication, play, imaginative skills, and repetitive behaviours [26,27]. It is a widely used scale in the field of ASD research with good psychometric properties, recommended for the diagnosis of ASD and assessment of core autistic symptoms [28].

At baseline and follow-up assessment after 12 months, we will use only ADOS-2 module 1, for children who are 18 months of age and older children who use no or few words.

There is a good Interrater reliability for Module 1 [27]. Internal consistency Cronbach's alpha coefficients was high in original study [27]. This scale has also shown that it can measure change in autism severity [13,28].

ADOS-2 is composed of different items scored 0 to 3 or 0 to 2. Item A1 codes the level of language, from the severity for 'the child is using regular use of statements with two or more words' (code 0) to 'the child has no spontaneous use of approximate words or words' (code 4). For children with no or limited language ( $A1 \geq 3$ ), the two items measuring language in the algorithm (item A3 speech abnormalities, item A5 stereotyped language) will be scored 3 (worst value) (see Green et al. 2010 [12]). The minimum overall ADOS-2 module 1 raw score will be 0 and the maximum score 42. A higher score means more autistic symptoms.

Our primary outcome will be the change between baseline and 12 months in the overall raw score in reciprocal socio-communicative interactions and repetitive and restrictive behaviours in line with the DSM-V [26,29]

Researchers will be trained to achieve recognized standards. Regular reliability meetings of all researchers will address any discrepant ratings to maintain researcher calibration.

### **Secondary outcomes**

**To assess social communication and interaction in the natural setting of parent-child interaction at home:**

-*Brief Observation of Social Communication Change (BOSCC)*[30].

**To assess dyadic communication in the natural setting of parent-child interaction at home:**

-*Dyadic Communication Measure for Autism (DCMA)* [24].

**To assess child cognitive development:**

-*Mullen Scales of Early Learning (MSEL)* [23]

**To assess child language development:**

-*Development of Expressive Language, (Development du Language de Production In french\_DLPF)*[31].

**To assess Adaptive behavior of the child:**

-*Vineland Adaptive Behaviour Scales second version (VABS-2)* [32]

**To assess Parent's Stress, health, priorities and experience of the family:**

-*Autism Family Experience Questionnaire (AFEQ)* [33]

- *Parental Stress Index (PSI)* [34].

-*General Health Questionnaire (GHQ-28)* [35]

**To assess implementation of the intervention:**

- *PACT Fidelity Rating Scale*

-Number of PACT training sessions undertaken

-Quality of videoconferencing during each session

-Parent's acceptability of videoconferencing and implementation of PACT at home (self-report on Likert-scale)

-Number of hours per day using PACT at home at 12 months

-Parent's qualitative PACT adherence coded with DCMA on a 12-minute home child-parent interaction video

Figure 2 shows schedule of enrolment, interventions and assessments. Appendix 1 shows more detailed about assessment.

### **Participant's timeline**

Toddlers with a suspected ASD will be approached by health professionals with information about this study. The research recruitment team will meet with the family, complete the ADI-R during the first meeting, and confirm eligibility criteria. Complementary assessments, including ADOS, MSEL with the child and Vineland will be conducted by videoconferencing with the parent, to assess the criteria for eligibility. If toddlers and families meet the criteria for participation, parents will be informed about the study and possibility of an intervention using PACT or TAU based on randomisation. A written informed consent will be obtained if the parent/family agrees to participate in the study after one week of reflection (See Consent form in Appendix 2).

The 'referent parent' used to refer to the parents who will engage with the PACT therapy will be decided before randomisation. Children will be subsequently randomised into the intervention or TAU group. Parents will be informed of the result of the randomisation

### **Assignment of intervention**

#### ***Allocation sequence generation and randomisation***

All eligible toddlers, with parental consent, will be assigned to the two study groups in a 1:1 ratio using the minimization method with the following stratification factors: the centre, the children's age, level of language (according to the ADOS2 scale) and gender. To ensure allocation concealment, a minimization algorithm with a .90 random element will be used and known only to the statistician (AD). The randomisation will be centralized.

#### ***Blinding***

Owing to the nature of the intervention, parents and PACT therapists cannot be blind to the allocation group. PACT therapists will not be involved in ASD assessment and diagnosis. An assessor blind to the allocation group will administer every assessment. Data manager and biostatistician will be blinded to the allocation groups.

### **Data collection and management**

#### ***Data collection***

Data will be collected through standardized observations, parental questionnaires and interviews carried out by a researcher blind to the allocation. We will be particularly vigilant about the measurement accuracy of the first criterion of judgement as described in the paragraph on ADOS-2.

### ***Participation retention and follow-up***

Children of both groups will benefit from early diagnosis and assessment and will have the same follow-up evaluations over the 1-year study period in the respective study centres Any discontinuation of study participation will be collected and recorded with the reasons.

### ***Data Management***

The study data will be collected on a secure electronic case report form (eCRF) that will be available at each centre through an internet portal. No personal identifying information will be mentioned on the eCRF. Each subject included in the study will be assigned a unique identification number.

All study data will be stored securely in the Academic Hospital of Lyon. All electronic data will be secured on a password-protected laptop. Paper-based study documents will be stored in a secure filing cabinet at each centre. Access to these files will be limited to research staff involved in the study.

The eCRF will only include the data necessary for the analysis to be reported in a scientific publication.

### **Statistical analysis**

#### ***Simple size calculation***

On the basis of the findings of the PACT Trial (Green, 2010), we have powered the study to be able to detect a difference in overall change on the ADOS score of 2 points. The group difference in mean change between baseline and month 12 was -1 point for ADOS social affect score (mean change=2.9, SD=3.9 in TAU group and mean change=-3.9, SD=4.7 in PACT+TAU group) and -0.5 points for ADOS restricted and repetitive behaviors score (mean change=2.9, SD=3.9 in TAU group and mean change=-3.9, SD=4.7 in PACT+TAU group). The pre- and post-measures were correlated at 0.67. Therefore, the most conservative values were fixed for ADOS standard deviation and for correlation among the repeated measures from a single participant. A target of 238 subjects (119 subjects per treatment arm) was planned to be randomized in the trial. Assuming a 2-point difference in favor of the PACT+TAU compared with TAU, a standard deviation of 5, a correlation between subsequent visits of 0.5, a drop-out rate of 20%, and a two-sided significance level of 0.05, the planned sample size would provide about 80% power for the study.

#### ***Feasibility of recruitment***

A strong partnership with ASD orientation platforms recently implemented in France, a broad communication (meeting, mail, flyers) to healthcare professionals (Speech pathologist, Occupational therapist, therapist, paediatrician, general practitioner) family associations and other stakeholders will allow the trial team to reach the sample size within two years.

### ***Statistical analysis***

A full statistical analysis plan will be finalized prior to database lock. Statistical analysis and results will be reported at the 12-month endpoint in accordance with the CONSORT 2010 statement. No interim analysis will be scheduled. All the statistical analysis will be carried out according to the intention to treat principle using SAS statistical software (SAS Institute Inc., Cary, NC, USA).

Baseline characteristics will be presented in each group.

Summary statistics will be presented for process variables (number of PACT sessions, quality of videoconferencing per session, acceptability and satisfaction of PACT intervention, number of hours declared to be realized with the child) to show the feasibility and acceptability of PACT implementation in the intervention group.

The pattern of missing data will be investigated (number and mechanism of missingness). Missing data strategies can be applied, and sensitivity analysis of different strategies (simple or multiple imputation) will be conducted.

A repeated measures analysis will be used to test the effect of PACT intervention on overall ADOS module 1 score over the 12-month study period. A linear mixed model will be run with the overall ADOS score as the dependent variable and including time (baseline, month 12), treatment (TAU or TAU+PACT) and the interaction between treatment and time as fixed effects and patient as random effect. Model will be adjusted for stratification factors (centre, age, level of language (item A1 ADOS-2) and gender) and baseline variables that show evidence of treatment group imbalance. Time will be represented by dummy variable. Model assumption will be verified according to residual analysis. If most of the assumptions are not met, other alternatives such as transformation of ADOS overall raw score will be examined. Sensitivity analyses like complete case and per-protocol analysis will be performed to assess the robustness of the results to protocol deviations. In complete case analysis, only patient with primary outcome documented will be analyzed. In per-protocol participants who violate the protocol will be excluded from the analysis.

All the secondary outcomes, (overall total score of the BOSCC, communication initiation and synchrony measured with the DCMA, overall raw score of the MSEL in receptive and expressive language, overall score in expressive language of the DLFP, overall raw score of communicative and social of the VABS, Parental Stress Index, Parent General Health score, AFEQ score) will be analyzed in a similar way using with appropriate linear or generalized linear mixed models. Tobit models will be used to address potential floor effects.

We will finally explore the parent's and children's characteristics, moderating the implementation and efficacy of this therapy. We will also test the previously described mediators implicated in the efficacy of this therapy [14].

### **Qualitative analysis of barriers and facilitators of implementation**

Based on parents and of the therapist's reports, we will describe the facilitators and barriers of the implementation of video-conferencing PACT.

The data will be collected through semi-structured interview and will be analysed with the classical technique of Interpretative Phenomenological Analysis (IPA) [36]. Population selection will follow the rules of the purposive sampling and will allow a maximal variation of the sample [37]. An estimation of 30-60 parents will be necessary to reach data saturation based on previous studies [38–40]. The total number of therapists (around 7-8) will be interviewed. During the 40-60 minutes interview, we will explore the barriers and facilitators to implementing PACT by videoconferencing. A guide for the interview will be elaborated in the initial phase of the project based on first interviews. Interviews will be recorded and transcribed before analyses.

Data from the quantitative and qualitative sources in the process evaluation will be analysed separately. The results of the qualitative study will be integrated with the quantitative results to optimize the findings [41].

## **Monitoring**

Dr Marie-Maude Geoffray (IP) Investigators associated, methodologists, statistician, parent representatives and associate researcher composed the trial steering committee (TSC). The TSC is independent of sponsor and funders and have no competing interests. The TSC has developed the study protocol and is responsible for data collection, management, publications and the final data set.

The coordinating center is independent from the centers for investigation.

According to the French law, the study requires formal data monitoring undertaken by the sponsor. Annual reporting will be completed and submitted to the funders.

### ***Adverse events***

Based on results from previous PACT intervention trials, no specific harm from trial participation is anticipated. However, as required by the French law, adverse events will be collected throughout the study and reported in the eCRF section. Description of the event, date of occurrence, intensity, severity, accountability will be reported. Outcomes of this event and action taken after its report will also be concealed.

We anticipate that the early assessment, follow-up consultation on ASD and its management will help and support both groups during the post-diagnosis period. Hence, no post-trial care is planned.

### ***Trial status***

The trial status is currently Recruiting. The study has started the 30th June 30, 2020. The anticipated end date will be 30th June 2023.

Figure 3 shows WHO trial registration data set. Figure 4 shows SPIRIT Checklist.

**Ethics and Dissemination:** This study (protocol version N°5, date: 10/25/2019) is approved by the French National Review Board (reference No 2018-A02516-49). The results will be disseminated via peer-reviewed journals. It will also disseminate via national and international, general and specialist meeting and through the parent association (<https://Bleunetwork.fr>; <https://autisme-ambitionavenir.com>; [desaiespourgrandir.org](https://desaiespourgrandir.org)). An individual feedback to the participant will be done through a regular newsletter. We will adhere to defined authorship criteria as per the International Committee of Medical Journal Editors.

**Author contribution:** MMG and ST, PO, LJ conceived and designed the project, and MMG is leading the coordination of the trial. MMG, LJ and PO drafted the protocol and procured the project funding. LJ and MMG are responsible for study implementation, staff training and supervision. PO, ST, AD, AZ contributed to the sample size calculation, the randomisation procedure and the statistical plan, and are responsible for data management, randomisation and statistical analysis. JG contributed to the protocol and paper writing. ARL contributed to the protocol of the qualitative study. CA and NG to the PACT training and supervision of the team. MMG, MJO, LJ, AA, AJ, AB, CS, TM, TD are responsible for recruitment and evaluation of children. SG contributed to draft the paper. All authors critically reviewed and approved the final version of the manuscript.

**IFPAD Study group:** Mrs Pauline Auphan (psychologist), Mrs Laetitia Bouveret (research assistant), Mrs Laurie Herman (research assistant), Dr Anne-Laure Toureille (PACT trainer), Mrs Lucie Jansen (PACT trainer), Dr Sandrine Sonié (CRA Lyon), Pr. Mario Speranza (CHU Versailles), Pr. Bruno Falissard (Paris), Pr Nicolas Georgieff (Lyon), Dr Matias Winter (HCL), Mrs Nadège Alloisio (parent association), Mr Chams-Ddine BELKHAYAT (parent association).

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**Disclaimer:** The funders and sponsor (CH le Vinatier, 95, boulevard Pinel, France) have no role in study design, data collection, management, data analysis and interpretation of data, in the writing of the report or in the decision to submit the manuscript for publication.

**Competing interests' statement:** None declared.

**Consent:** Obtained from both parents of the child (Please find in Appendix 2).

**Provenance and peer review:** Not commissioned; externally peer reviewed.

**Data sharing statement:** The trial statisticians will have access to the data set for the analysis of trial outcomes. The PI will have access to the data and will take full responsibility for the analysis and publication of the results. Once the main analyses have been undertaken, data will be available to principal and other investigator subject for approval of data analysis plans by the steering committee.

### Figure legends

Figure 1- Consort flow-chart of the study

Figure 2- Schedule of enrolment, interventions and assessments (SPIRIT)

Figure 3- WHO trial registration data set

Figure 4- SPIRIT Checklist

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Figure 1 Consort flow-chart of the study.

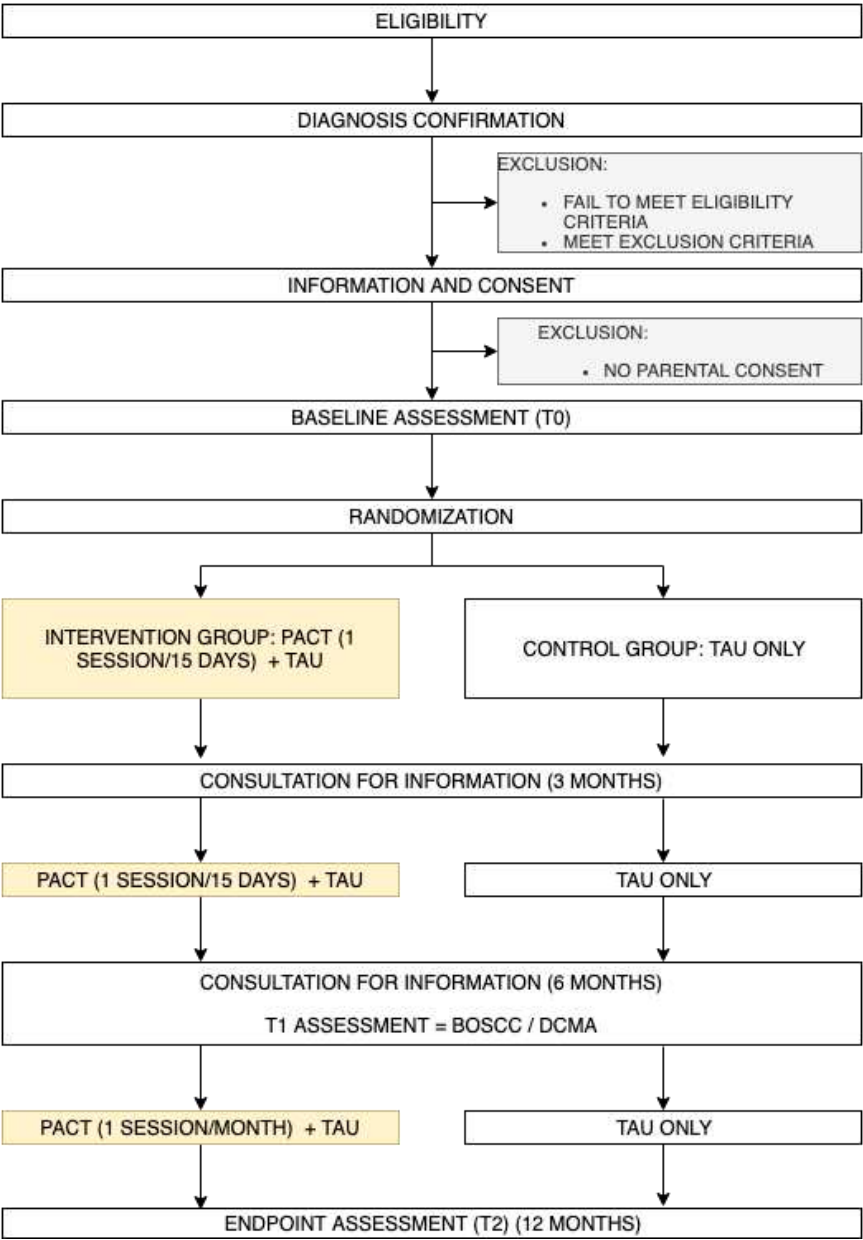


Figure 2. Schedule of enrolment, interventions and assessments (SPIRIT)

Time point	Enrolment	T0 baseline	T1 6 months	T2 12 months
<b>Eligibility screen</b>				
DSM-5 criteria	x			
ADOS-2	x			
ADI-R for toddlers	x			
Nonverbal skills MSEL	x			
Adaptative behavioral level : VABS 2 coded based on an interview done by videoconferencing with parents	x			
Informed consent	x			
Allocation		x		
<b>Intervention</b>				
PACT training (1h/15 days during 6 months then 1h/month during 6 months) + daily practice at home			—————→	
Treatment as usual			—————→	
<b>Assessment</b>				
Sociodemographic data		x		x
Autistic symptoms: ADOS-2 (primary outcome)		x		x
Change in socio-communicative interactions: BOSCC Played-based interaction between carer and child at home		x	x	x
Communication Synchronization and initiatives: DCMA at home		x	x	x
Expressive and receptive language Mullen scale		x		x
Daily language DLFP		x		x
Adaptative behavioral level: VABS 2 by video-conferencing		x		x
Parental stress index: ISP-short form		x		x
General parents health : GHQ-28		x		x
Parents quality of life and priorities. AFEQ		x		x
Acceptability of PACT intervention and TAU. (Linkert scale)				x
Implementation: Number of PACT training session Mean of quality of videoconferencing collected after each session Number of hours declared by parents using PACT at home				x x x

Figure 3 shows WHO trial registration data set

Data category	Information
Primary registry and trial identifying number	clinicaltrials.gov : NCT04244721
Date of registration in primary registry	January 28, 2020
Secondary identifying numbers	2018-A02516-49
Source of monetary or material support	This study is supported by a grant from the Programme Hospitalier de Recherche Clinique inter-régional Rhône Alpes (PHRCI-15-065) from the AURA region and a grant by the Caisse Nationale de solidarité pour l'autonomie (CNSA) as part of the call for projects launched for IReSP (Institut de Recherche en Santé Publique) in 2016 in collaboration with the Institut national de la santé et de la recherche médicale (Inserm) ( IReSP-17-Autisme3-16).
Primary sponsor	Centre Hospitalier Le Vinatier, Bron, France
Secondary sponsor	Not applicable
Contact for public queries	Marie-Maude Geoffray (PI), email: marie-maude.geoffray @ch-le-vinatier.fr ;Lucie Jurek, email : lucie.jurek@ch-le-vinatier.fr
Contact for scientific queries	Marie-Maude Geoffray (PI), email: marie-maude.geoffray @ch-le-vinatier.fr Sandrine Touzet (scientific contact), email: sandrine.touzet@chu-lyon.fr
Public title	Efficacy of a parent- mediated communication-focused treatment in toddler with autism (PACT)when parents trained by videoconference: a randomised controlled trial study protocol
Scientific tittle	Efficacy of a parent- mediated communication-focused treatment in toddler with autism (PACT)when parents trained by videoconference: a randomised controlled trial study protocol
Countries of recrutement	France
Health condition or problem studied	Autism spectrum disorder (ASD)
Interventions	<i>Intervention:</i> PACT for toddlers and young children with ASD <i>Description:</i> PACT delivered by the parent trained remotely by therapists 1 hour per 2-weeks during 6 months and 1 hour per month the following 6 months. <i>Control:</i> care available in the community, i. e. consultations with a psychologist or child and adolescent psychiatrist, speech and language therapy, occupational therapy, individual or group psychotherapy.

Key inclusion and exclusion criteria	<p><i>Age:</i> between 18 and 36 months</p> <p><i>Sex:</i> male or female</p> <p><i>Inclusion criteria:</i> diagnosis of ASD (Diagnosis and Statistical Manual of Mental Disorders, Fifth Edition, and Autism Diagnosis Observation Schedule), child will have a non-verbal age above 12 months on the Mullen Scale of Early learning, family living further than 40 min away from an ASD specialized center.</p> <p><i>Exclusion criteria:</i> serious neurological or physical condition, family unavailable for a regular follow-up.</p>
Study type	<p>Interventional</p> <p><i>Allocation:</i> randomized 1:1; parallel assignment; blinding: assessor blind</p>
Date of first enrolment	15th July 2020
Target sample size	238
Recruitment status	Recruiting
Primary outcome	Change in ADOS-2 module 1 standardized score at 12 months
Key secondary outcomes	Child development, child adaptative behavior, child socio-communicative abilities, quality of life of parents, parental stress, obstacle and facilitators of the intervention model.

FIGURE 4 : SPIRIT 2013 Checklist: Recommended items to address in a clinical trial protocol and related documents\*

Section/item	Item No	Description	Page
<b>Administrative information</b>			
Title	1	Descriptive title identifying the study design, population, interventions, and, if applicable, trial acronym	1
Trial registration	2a	Trial identifier and registry name. If not yet registered, name of intended registry	2
	2b	All items from the World Health Organization Trial Registration Data Set	12 (fig 3)
Protocol version	3	Date and version identifier	13
Funding	4	Sources and types of financial, material, and other support	13
Roles and responsibilities	5a	Names, affiliations, and roles of protocol contributors	13
	5b	Name and contact information for the trial sponsor	13
	5c	Role of study sponsor and funders, if any, in study design; collection, management, analysis, and interpretation of data; writing of the report; and the decision to submit the report for publication, including whether they will have ultimate authority over any of these activities	13
	5d	Composition, roles, and responsibilities of the coordinating centre, steering committee, endpoint adjudication committee, data management team, and other individuals or groups overseeing the trial, if applicable (see Item 21a for data monitoring committee)	13
<b>Introduction</b>			
Background and rationale	6a	Description of research question and justification for undertaking the trial, including summary of relevant studies (published and unpublished) examining benefits and harms for each intervention	3
	6b	Explanation for choice of comparators	3
Objectives	7	Specific objectives or hypotheses	4

Trial design	8	Description of trial design including type of trial (eg, parallel group, crossover, factorial, single group), allocation ratio, and framework (eg, superiority, equivalence, noninferiority, exploratory)	4
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### **Methods: Participants, interventions, and outcomes**

Study setting	9	Description of study settings (eg, community clinic, academic hospital) and list of countries where data will be collected. Reference to where list of study sites can be obtained	4
Eligibility criteria	10	Inclusion and exclusion criteria for participants. If applicable, eligibility criteria for study centres and individuals who will perform the interventions (eg, surgeons, psychotherapists)	5
Interventions	11 a	Interventions for each group with sufficient detail to allow replication, including how and when they will be administered	6
	11 b	Criteria for discontinuing or modifying allocated interventions for a given trial participant (eg, drug dose change in response to harms, participant request, or improving/worsening disease)	10
	11 c	Strategies to improve adherence to intervention protocols, and any procedures for monitoring adherence (eg, drug tablet return, laboratory tests)	6
	11 d	Relevant concomitant care and interventions that are permitted or prohibited during the trial	7
Outcomes	12	Primary, secondary, and other outcomes, including the specific measurement variable (eg, systolic blood pressure), analysis metric (eg, change from baseline, final value, time to event), method of aggregation (eg, median, proportion), and time point for each outcome. Explanation of the clinical relevance of chosen efficacy and harm outcomes is strongly recommended	8
Participant timeline	13	Time schedule of enrolment, interventions (including any run-ins and washouts), assessments, and visits for participants. A schematic diagram is highly recommended (see Figure)	9 (fig 2)
Sample size	14	Estimated number of participants needed to achieve study objectives and how it was determined, including clinical and statistical assumptions supporting any sample size calculations	10
Recruitment	15	Strategies for achieving adequate participant enrolment to reach target sample size	11

### **Methods: Assignment of interventions (for controlled trials)**

Allocation:

Sequence generation	16a	Method of generating the allocation sequence (eg, computer-generated random numbers), and list of any factors for stratification. To reduce predictability of a random sequence, details of any planned restriction (eg, blocking) should be provided in a separate document that is unavailable to those who enrol participants or assign interventions	9
Allocation concealment mechanism	16b	Mechanism of implementing the allocation sequence (eg, central telephone; sequentially numbered, opaque, sealed envelopes), describing any steps to conceal the sequence until interventions are assigned	9
Implementation	16c	Who will generate the allocation sequence, who will enrol participants, and who will assign participants to interventions	10
Blinding (masking)	17a	Who will be blinded after assignment to interventions (eg, trial participants, care providers, outcome assessors, data analysts), and how	10
	17b	If blinded, circumstances under which unblinding is permissible, and procedure for revealing a participant's allocated intervention during the trial	10

**Methods: Data collection, management, and analysis**

Data collection methods	18a	Plans for assessment and collection of outcome, baseline, and other trial data, including any related processes to promote data quality (eg, duplicate measurements, training of assessors) and a description of study instruments (eg, questionnaires, laboratory tests) along with their reliability and validity, if known. Reference to where data collection forms can be found, if not in the protocol	10
	18b	Plans to promote participant retention and complete follow-up, including list of any outcome data to be collected for participants who discontinue or deviate from intervention protocols	10
Data management	19	Plans for data entry, coding, security, and storage, including any related processes to promote data quality (eg, double data entry; range checks for data values). Reference to where details of data management procedures can be found, if not in the protocol	10
Statistical methods	20a	Statistical methods for analysing primary and secondary outcomes. Reference to where other details of the statistical analysis plan can be found, if not in the protocol	11
	20b	Methods for any additional analyses (eg, subgroup and adjusted analyses)	11

	20	Definition of analysis population relating to protocol non-adherence (eg, as randomised analysis), and any statistical methods to handle missing data (eg, multiple imputation)	11
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### **Methods: Monitoring**

Data monitoring	21	Composition of data monitoring committee (DMC); summary of its role and reporting structure; statement of whether it is independent from the sponsor and competing interests; and reference to where further details about its charter can be found, if not in the protocol. Alternatively, an explanation of why a DMC is not needed	12
	21	Description of any interim analyses and stopping guidelines, including who will have access to these interim results and make the final decision to terminate the trial	12
Harms	22	Plans for collecting, assessing, reporting, and managing solicited and spontaneously reported adverse events and other unintended effects of trial interventions or trial conduct	12
Auditing	23	Frequency and procedures for auditing trial conduct, if any, and whether the process will be independent from investigators and the sponsor	13

### **Ethics and dissemination**

Research ethics approval	24	Plans for seeking research ethics committee/institutional review board (REC/IRB) approval	13
Protocol amendments	25	Plans for communicating important protocol modifications (eg, changes to eligibility criteria, outcomes, analyses) to relevant parties (eg, investigators, REC/IRBs, trial participants, trial registries, journals, regulators)	13
Consent or assent	26	Who will obtain informed consent or assent from potential trial participants or authorised surrogates, and how (see Item 32)	13
	26	Additional consent provisions for collection and use of participant data and biological specimens in ancillary studies, if applicable	NA
Confidentiality	27	How personal information about potential and enrolled participants will be collected, shared, and maintained in order to protect confidentiality before, during, and after the trial	13
Declaration of interests	28	Financial and other competing interests for principal investigators for the overall trial and each study site	13
Access to data	29	Statement of who will have access to the final trial dataset, and disclosure of contractual agreements that limit such access for investigators	14

Ancillary and post-trial care	30	Provisions, if any, for ancillary and post-trial care, and for compensation to those who suffer harm from trial participation	12
Dissemination policy	31	Plans for investigators and sponsor to communicate trial results to participants, healthcare professionals, the public, and other relevant groups (eg, via publication, reporting in results databases, or other data sharing arrangements), including any publication restrictions	13
	31	Authorship eligibility guidelines and any intended use of professional writers	13
	31	Plans, if any, for granting public access to the full protocol, participant-level dataset, and statistical code	14

### Appendices

Informed consent materials	32	Model consent form and other related documentation given to participants and authorised surrogates	App 2
Biological specimens	33	Plans for collection, laboratory evaluation, and storage of biological specimens for genetic or molecular analysis in the current trial and for future use in ancillary studies, if applicable	NA

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L'Appendix est disponible ici : <https://bmjopen.bmj.com/content/11/4/e044669.info>

