

Université de Montréal

**Mieux comprendre l'efficacité différentielle de l'intervention comportementale intensive  
auprès des enfants ayant un trouble du spectre de l'autisme**

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*Cette thèse intitulée*

**Mieux comprendre l'efficacité différentielle de l'intervention comportementale intensive  
auprès des enfants ayant un TSA**

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## Résumé

L'intervention comportementale intensive (ICI) est l'intervention offerte par les services publics du Québec aux jeunes enfants diagnostiqués d'un trouble du spectre de l'autisme (TSA). Plusieurs organismes nationaux de santé considèrent l'ICI comme une intervention établie et de nombreuses méta-analyses soutiennent son efficacité. Cependant, les effets varient grandement d'un enfant à l'autre. Alors que certains enfants progressent significativement dans plusieurs sphères du développement, d'autres ne retirent qu'une modeste amélioration. Cette réponse différentielle à l'intervention demeure mal comprise : à l'heure actuelle, il n'y a pas de consensus sur la fiabilité des prédicteurs d'efficacité de l'ICI. L'objectif général de cette thèse est donc d'étudier la réponse différentielle à l'ICI dans un contexte québécois.

Le premier article de cette thèse visait à évaluer les effets de l'ICI sur le fonctionnement adaptatif et les symptômes autistiques des enfants qui la reçoivent, de vérifier si les progrès se maintenaient dans le temps et d'identifier des prédicteurs d'efficacité. Les résultats ont révélé un changement non linéaire du fonctionnement adaptatif, caractérisé par une amélioration significative pendant la période d'intervention et un maintien des gains pendant la période de suivi, ainsi qu'une légère diminution linéaire des symptômes autistiques tout au long de l'étude. L'intensité de l'intervention, l'âge, le QI et les symptômes autistiques étaient associés soit à des progrès pendant la période d'intervention, soit à un maintien pendant la période de suivi.

Le deuxième article investiguait la présence de sous-groupes plus homogènes (c.-à-d. des profils latents) chez les participants sur la base de leurs caractéristiques lors de leur entrée dans les services, examinait les prédicteurs sociodémographiques de l'appartenance à un profil particulier et vérifiait si l'appartenance aux profils était associée à une réponse différentielle à l'ICI. Nous avons trouvé quatre profils dans notre échantillon. Seul le revenu familial annuel prédisait l'appartenance au profil. Tous les profils ont progressé pendant de la période

d'intervention, avec des changements d'ampleur variable. Au cours de la période de suivi, les profils ayant les manifestations les plus sévères ont montré une stabilité ou une amélioration du fonctionnement adaptatif, tandis que les deux profils ayant les manifestations les plus légères ont montré une légère diminution du fonctionnement adaptatif.

Finalement, le dernier article visait à vérifier si l'apprentissage automatique (angl. *machine learning*) pouvait soutenir l'estimation du pronostic des enfants recevant de l'ICI. Pour ce faire, nous avons comparé la précision des prédictions (progrès versus pas de progrès) faites par cinq algorithmes et une assignation aléatoire. Les résultats indiquaient que les prédictions de tous les algorithmes étaient meilleures que l'assignation aléatoire. En conclusion, cette thèse discute des retombées des résultats la recherche, la pratique et pour le domaine de la psychoéducation.

**Mots-clés :** apprentissage automatique, autisme, efficacité différentielle, hétérogénéité, intervention comportementale intensive, prédicteurs d'efficacité

## **Abstract**

Early intensive behavioral intervention (EIBI) is the intervention offered by public services in Quebec to young children diagnosed with an autism spectrum disorder (ASD). Several national health organizations consider EIBI as an established intervention and numerous meta-analyzes support its efficacy. However, its effects vary greatly from one individual to another. While some children progress significantly in several areas of development, others make only modest improvement. This differential response to intervention remains poorly understood: there is no consensus on the reliability of predictors of the efficacy of EIBI. Thus, the main purpose of this thesis is to study the differential response to EIBI in the Quebec context.

The first article of this thesis aimed to assess the effects of EIBI on adaptive functioning and autistic symptoms in children with ASD, to verify whether the changes are maintained over time and to identify predictors of efficacy. The results revealed a non-linear change in adaptive functioning, characterized by a significant improvement during the intervention period and maintenance of gains during the follow-up period, as well as a marginal linear decrease in autistic symptoms from baseline to follow-up. Intervention intensity, age, IQ, and autistic symptoms were associated with either progress during the intervention or maintenance during follow-up.

The second article investigated the presence of more homogeneous subgroups (i.e., latent profiles) among participants based on their characteristics when entering services, examined socio-demographic predictors of profile membership and assessed whether profile membership was associated with a differential response to EIBI. We found four profiles in our sample. Only annual family income predicted profile membership. All profiles progressed during the intervention period, with changes of varying magnitude. During follow-up, the profiles with the most severe manifestations showed stability or improvement in adaptive functioning, while the two profiles with the milder manifestations showed a marginal decrease in adaptive functioning.

Finally, the last article aimed to test whether machine learning could support prognosis estimation for children receiving EIBI. To this end, we compared the accuracy of the predictions (progress versus no progress) made by five algorithms and a random assignment. The results showed that the predictions of all algorithms were better than random assignment. In conclusion, this thesis discusses the implications of the results for research, practice and the field of psychoeducation.

**Keywords:** autism, differential efficacy, early intensive behavioral intervention, heterogeneity, machine learning, predictors of efficacy

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## Liste des sigles et abréviations

AAC : Analyse appliquée du comportement

ABAS-II : Adaptive Behavior Assessment System-II

AIC : Akaike information criterion

ANNs : Artificial neural networks

APA : American Psychological Association

AS: Autistic symptoms

ASD : Autism spectrum disorders

BACB : Behavior analyst certification board

BCBA : Bord certified behavior analyst

BIC : Bayesian information criterion

BLRT : Bootstrap likelihood ratio test

CARS-2 : Childhood Autism Rating Scale – Second edition

CAIC : Consistent AIC

CISSS-MO : Centre intégré de santé et services sociaux de la Montérégie-Ouest

CFI : Comparative fit index

CI : Confidence intervals

CON : Conceptual domain

CRDITED-ME : Centre de réadaptation en déficience intellectuelle et en trouble envahissant du développement de la Montérégie-Est

df: Degrees of freedom

DSM-5 : Diagnostic and statistical manual of mental disorders – 5<sup>th</sup> edition

EBI : Early behavioral intervention

EIBI : Early intensive behavioral intervention

GAC : General adaptative composite

GLC : General language composite

ICI : Intervention comportementale intensive

IQ: Intelligence quotient

LBM : Latent basis model

LGC : Latent growth curve

LPA: Latent profile analysis

MLR : Maximum likelihood estimation robust

OPPQ : Ordre des psychoéducateurs et psychoéducatrices du Québec

PRA : Practical domain

PIQ : Performance intelligence quotient

PWGM: Piecewise growth modeling

QI : Quotient intellectuel

RMSEA : Root mean square error of approximation

SABIC: Sample-size adjusted Bayesian information criterion

SOC : Social domain

SRMR : Standardize Root Mean Square Residual

TSA : Trouble du spectre de l'autisme

TLI : Tucker-Lewis index

VIQ: Verbal intelligence quotient

VLMR : Vuong, Lo, Mendell and Rubin's test

WPSSI-III : Wechsler Preschool and Primary Scale of Intelligence- third edition

*À tous les enfants ayant un trouble du spectre de l'autisme. Vous méritez de recevoir les meilleures interventions pour favoriser la réalisation de votre plein potentiel.*

*À tous les intervenants dévoués qui gravitent autour des enfants ayant un trouble de spectre de l'autisme. L'idée de cette thèse est née de l'espoir de guider (un peu) votre travail.*

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### **Contribution des auteurs aux articles**

Conformément aux règlements de la Faculté des études supérieures et postdoctorales, je certifie avoir contribué de manière « essentielle, majeure et déterminante » aux trois articles qui constituent cet ouvrage. Plus précisément, j'ai procédé à la demande éthique, j'ai élaboré les questions de recherche des trois articles, déterminé et réalisé les analyses statistiques pour répondre aux questions de recherche et j'ai rédigé les trois articles. Marc Lanovaz a supervisé l'ensemble de mes travaux de recherche et a rédigé le code Python qui a permis de conduire les analyses du troisième article. Julien Morizot a co-supervisé l'ensemble de mes travaux de recherche et a soutenu le choix et la réalisation des analyses statistiques des deux premiers articles. Enfin, comme cette thèse repose sur des analyses secondaires, Mélina Rivard a généreusement partagé sa banque de données avec nous, en plus de réviser les trois articles.

## Présentation de la thèse

Cette thèse est rédigée par articles et compte cinq principaux chapitres : l'introduction générale, trois articles empiriques et la discussion générale. Elle contribue à élucider une question primordiale dans le domaine de la recherche évaluative en autisme en tentant de mieux comprendre l'efficacité différentielle de l'intervention comportementale intensive (ICI) auprès des jeunes enfants ayant un trouble du spectre de l'autisme (TSA).

Le premier chapitre présente le contexte théorique dans lequel s'inscrit ma thèse. Après avoir décrit le TSA et ses diverses théories explicatives, il dresse l'état des connaissances concernant l'ICI et ses prédicteurs d'efficacité auprès des jeunes enfants ayant un TSA, précise les particularités du contexte québécois d'intervention et expose les différentes approches statistiques qui ont été utilisées jusqu'à maintenant pour investiguer l'efficacité de l'ICI. Ce chapitre se termine par la présentation des principaux objectifs de la thèse : (a) l'évaluation des effets de l'ICI, telle que dispensée par le Centre intégré de santé et services sociaux de la Montérégie-Ouest (CISSS-MO<sup>1</sup>), sur les symptômes autistiques et le fonctionnement adaptatif des enfants qui la reçoivent, (b) l'exploration de la présence de profils distincts chez les participants, selon leur niveau initial de QI, de fonctionnement adaptatif et de symptômes autistiques et leurs potentielles associations à une réponse différentielle à l'intervention, et (c) l'estimation du pronostic des enfants recevant l'ICI en utilisant l'apprentissage automatique (angl. *machine learning*).

Le deuxième chapitre présente l'article intitulé « Changes in autistic symptoms and adaptive functioning of children receiving early behavioral intervention in a community setting: A latent growth curve analysis », qui a été soumis pour publication au *Journal of Autism and*

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<sup>1</sup> Au moment de la collecte de données, le CISSS-MO était connu sous l'appellation Centre de réadaptation en déficience intellectuelle et en trouble envahissant du développement de la Montérégie-Est (CRDITED-ME).

*Developmental Disorders*. Le premier objectif de cet article était d'évaluer les effets immédiats et à moyen terme de l'ICI dispensée en communauté sur les symptômes autistiques, le fonctionnement adaptatif et ses trois domaines (conceptuel, social et pratique). Le second objectif était d'identifier de potentiels prédicteurs d'efficacité de l'intervention parmi les caractéristiques initiales de l'enfant et certaines caractéristiques sociodémographiques. Pour ce faire, nous avons employé des courbes de croissance latente. Les courbes de croissance latente s'inscrivent dans une approche statistique basée sur les variables et les résultats représentent des associations ou coefficients moyens pour tous les individus de l'échantillon.

Le troisième chapitre présente l'article intitulé « A person-centered perspective of differential efficacy of early behavioral intervention in children with autism : A latent profile analysis » qui a été soumis pour publication à *Research in Autism Spectrum Disorders*. Ce chapitre approfondit les résultats du premier article, en adoptant une approche statistique basée sur les personnes. Les objectifs de ce deuxième article étaient d'identifier des profils distincts d'enfants selon leurs caractéristiques au moment de leur entrée dans les services, d'explorer les prédicteurs d'appartenance aux profils et de vérifier si l'appartenance aux profils était associée aux effets immédiats et/ou à moyen terme de l'ICI. Pour atteindre nos objectifs, nous avons réalisé une analyse de profils latents. La particularité des résultats des analyses centrées sur la personne est que les associations entre les variables ou coefficients moyens peuvent être différentes selon les sous-groupes d'individus identifiés à l'intérieur d'un échantillon.

Le quatrième chapitre présente l'article intitulé « Brief report: Machine learning for estimating prognosis of children with autism receiving early behavioral intervention – A proof of concept », qui a été soumis pour publication au *Journal of Autism and Developmental Disorders*. Cet article avait pour objectif de vérifier si l'apprentissage automatique pouvait soutenir l'estimation du pronostic des enfants recevant de l'ICI. Comparativement aux deux précédents

articles qui découlent des statistiques traditionnelles, l'apprentissage automatique découle de l'apprentissage statistique (angl. *statistical learning*). L'idée était d'utiliser de façon complémentaire une approche novatrice afin de vérifier la potentielle contribution de l'apprentissage automatique à la compréhension de l'efficacité différentielle de l'ICI.

Le cinquième et dernier chapitre constitue la discussion générale de la thèse. D'abord, il résume les principaux résultats des articles empiriques. Puis, nous discutons des implications des résultats de cette thèse sur le plan de la recherche, sur le plan de la pratique, et plus largement, pour le domaine de la psychoéducation. La discussion générale clôt la thèse en présentant les forces et des limites de cette dernière.

## **Chapitre I – Introduction générale**

## Contexte théorique

### Trouble du spectre de l'autisme : Description et prévalence

Selon l'Association américaine de psychiatrie (APA, 2013), le trouble du spectre de l'autisme (TSA) est une condition neurodéveloppementale caractérisée par des symptômes s'inscrivant dans deux critères diagnostiques. Le premier est une altération qualitative de la communication et des interactions sociales. Par exemple, les personnes ayant un TSA peuvent éprouver de la difficulté à utiliser la communication verbale et non verbale, à en décoder le sens chez leurs interlocuteurs, ainsi qu'à établir une réciprocité socioémotionnelle dans leurs relations interpersonnelles. Le second critère diagnostique comprend la présence de comportements stéréotypés, d'intérêts restreints, de particularités sensorielles et l'adhérence à des routines. Les comportements stéréotypés peuvent être verbaux (comme la répétition de sons ou de phrases hors contexte) ou moteurs (comme l'utilisation de partie du corps ou d'objets pour faire des mouvements répétitifs). Les intérêts sont dits restreints lorsque leur nature est circonscrite et leur manifestation intense. Les particularités sensorielles se traduisent par une hypo ou une hypersensibilité à certains stimuli. Finalement, l'adhérence à des routines réfère à la préférence pour la réalisation de certaines tâches selon un ordre établi, prévisible et invariant.

Les symptômes du TSA apparaissent dès la petite enfance et s'expriment de façon hétérogène selon un continuum de gravité, d'où la notion de spectre (Georgiades et al., 2013; Masi et al., 2017; Wiggins et al., 2012). En plus des symptômes autistiques, le fonctionnement intellectuel (Wiggins et al., 2012), les patrons de forces et limites cognitives (Munson et al., 2008) et le fonctionnement adaptatif (Ray-Subramanian et al., 2011) diffèrent dans cette population. Le fonctionnement adaptatif fait référence aux habiletés développées tout au long de la vie qui permettent à une personne de faire face aux situations quotidiennes (Tassé, 2013). Ces habiletés sont divisées en trois domaines de fonctionnement, soient les domaines conceptuel,

social et pratique (Harrions & Oakland, 2003; Schalock et al., 2010). Le domaine conceptuel regroupe les habiletés relatives à la communication, aux acquis préscolaires et à la responsabilité individuelle. Le domaine social regroupe les habiletés relatives aux loisirs et aux aptitudes sociales. Le domaine pratique regroupe les habiletés relatives aux ressources communautaires, la vie domestique, la santé et sécurité et l'autonomie. Pour chaque domaine, le nombre d'habiletés qu'une personne peut réaliser de façon indépendante détermine son niveau de fonctionnement adaptatif et le niveau de soutien requis.

Certaines personnes se trouvant sur le spectre de l'autisme présentent peu de symptômes, éprouvent des difficultés légères, ont des besoins ponctuels et fonctionnent avec un faible niveau de soutien. À l'autre extrémité du spectre se retrouvent des personnes qui présentent des symptômes sévères et éprouvent des difficultés importantes qui affectent plusieurs sphères de fonctionnement, nécessitant un soutien substantiel et continu. Ces symptômes se manifestent au quotidien, influencent le développement, nuisent à l'adaptation de la personne et ont tendance à se chroniciser au cours du développement (Matson & Horovitz, 2010; Simonoff et al., 2020). Sans intervention, le degré d'atteinte de ce trouble reste stable au cours de la vie pour la majorité des personnes diagnostiquées (Bieleninik et al., 2017; Matson & Smith, 2008) et leur niveau de fonctionnement adaptatif n'atteint pas un niveau équivalent à celui des personnes neurotypiques (Kanne et al., 2011).

La prévalence du TSA a considérablement augmenté au cours des dernières années (Centers for Disease Control and Prevention, 2020). Les plus récentes estimations indiquent qu'en 2015, 1,6% des enfants âgés entre 5 et 17 ans avaient un TSA (Agence de santé publique du Canada, 2018), avec un ratio d'une fille pour 4 à 5 garçons (Institut national de santé publique du Québec, 2017). Différentes raisons peuvent expliquer cette croissance de prévalence. D'abord, le changement des critères diagnostiques de cette condition entre la quatrième et la cinquième

édition du Manuel diagnostique et statistique des troubles mentaux a pu mener à des réassignations de diagnostics (Elsabbagh et al., 2012). Ensuite, une plus grande sensibilisation des éducatrices en petite enfance et de la population générale a mené à de plus nombreuses références dans les services de première ligne, ce qui a probablement engendré un meilleur dépistage de la condition. Le nombre grandissant d'enfants diagnostiqués exerce une pression sur le système public qui leur prodigue des services, ce qui souligne l'importance de mettre en place des interventions efficaces auprès de la population ayant un TSA (Wong et al. 2014).

### **Les théories explicatives**

Diverses théories ont tenté d'expliquer la symptomatologie autistique, dont la théorie de la faible cohérence centrale, la théorie du fonctionnement perceptif augmenté, la théorie du cerveau mâle extrême et la théorie de la faible motivation sociale. Les propositions des différents auteurs illustrent des conceptions différentes de l'autisme. La théorie de la faible cohérence centrale (Frith & Happé, 1994) stipule que les personnes autistes auraient un style cognitif axé sur les détails, en raison de leurs difficultés à intégrer l'information en un tout cohérent. La théorie du fonctionnement perceptif augmenté (Mottron et al., 2006; Mottron & Burack, 2001) suggère plutôt que les personnes ayant un TSA auraient de meilleures performances dans le traitement local de l'information, comparativement aux pairs neurotypiques. Ainsi, leurs particularités perceptuelles résulteraient d'une force à traiter l'information locale au lieu d'une incapacité à traiter l'information globale.

Pour sa part, la théorie du cerveau mâle extrême (Baron-Cohen, 2002) hypothétise que les styles cognitifs des individus se situent sur un continuum opposant deux pôles; l'empathisation, davantage associé aux femmes, et la systémisation, davantage associée aux hommes. L'empathisation serait la capacité à comprendre ce que vivent les autres, tandis que la systémisation serait la capacité de déceler les règles et les répétitions. Donc, les symptômes



autistiques seraient le résultat d'un surdéveloppement du pôle de la systémisation au détriment de celui de l'empathisation, d'où l'appellation du cerveau mâle extrême. Finalement, la théorie de la faible motivation sociale suggère que les altérations qualitatives de la communication sociale présentées par les individus ayant un TSA s'expliqueraient par le fait qu'ils n'auraient pas la même propension naturelle que les personnes neurotypiques à s'orienter vers le monde social (Chevallier et al., 2012). Cette absence d'intérêt créerait un cercle vicieux : le manque de motivation sociale limiterait l'attention portée aux stimuli sociaux, ce qui les exposerait à moins d'expériences d'apprentissage social. Leur faible performance sociale maintiendrait leur manque de motivation. Malgré cette myriade de théories, aucune ne fait consensus au sein de la communauté scientifique et aucune n'intègre à elle seule l'ensemble des caractéristiques associées au TSA (Fava & Strauss, 2014).

Même si les causes du TSA demeurent incomprises, de plus en plus de données suggèrent que les difficultés vécues par les personnes ayant un TSA découleraient de l'interaction entre des facteurs génétiques et des facteurs environnementaux qui surviendraient à différents moments critiques du développement (Loke et al., 2015; Mandy & Lai, 2016; Reed, 2016). De récentes études ont identifié plus de 900 associations potentielles entre des gènes et le TSA (Schaafs et al., 2020). Malheureusement, la façon dont ces gènes influenceraient l'expression des manifestations du TSA n'est pas encore comprise, faisant en sorte qu'il n'y ait pas à l'heure actuelle de retombées pour la détection de la condition ou sur le plan thérapeutique (Schaafs et al., 2020). Mandy et Lai (2016) soutiennent qu'en dépit de la composante génétique, la trajectoire développementale d'un individu ayant un TSA n'est pas statique et évoluerait de façon dynamique en fonction des interactions entre le potentiel de la personne et les occasions expérientielles de son environnement. Les caractéristiques associées au TSA prédisposeraient la personne à susciter des expériences spécifiques dans son entourage, ce qui en retour exacerberait

les symptômes autistiques. Par exemple, une personne démontrant de pauvres habiletés sociales est plus à risque de vivre du rejet. Or, ce rejet mènerait à un manque d'occasion de socialisation positive qui exacerberait le déficit des habiletés sociales. Cette perspective est intéressante sur le plan de la pratique, puisqu'elle soutient que (1) développer des compétences chez la personne et (2) moduler les expériences dans son environnement peuvent avoir des répercussions sur sa trajectoire développementale des individus.

### **L'analyse appliquée du comportement et l'intervention comportementale intensive**

Différentes interventions existent pour favoriser le développement des enfants ayant un TSA. Les interventions comportementales, s'appuyant sur l'analyse appliquée du comportement (AAC), comptent parmi les interventions de réadaptation les plus efficaces pour les jeunes enfants ayant un TSA (Makrygianni et al., 2018; Schreibman, 2000; Smith & Iadarola, 2015; Wong et al., 2014). Sept caractéristiques définissent l'AAC ; 1) il s'agit d'une discipline appliquée qui s'intéresse aux comportements importants socialement, 2) elle focalise sur les changements de comportements comme sujet d'étude et d'intervention, 3) son objectif est d'établir une relation fonctionnelle entre l'environnement et les comportements de la personne, 4) la mise en œuvre des procédures est décrite de façon à être reproductible, 5) les procédures utilisées s'appuient sur les principes fondamentaux du conditionnement, 6) l'évaluation de l'efficacité des interventions repose sur le caractère socialement significatif des changements et 7) elle planifie la généralisation et le maintien des acquis (Baer et al., 1968; Cooper et al., 2020). En 1987, la notion de validité sociale a été ajoutée aux caractéristiques de l'AAC (Baer et al., 1987). Au-delà de l'atteinte des objectifs d'intervention, les auteurs précisent que l'évaluation de l'efficacité des interventions basées sur l'AAC devrait prendre en compte les retombées réelles des apprentissages dans la vie de la personne.

L'intervention comportementale intensive (ICI) désigne un ensemble de programmes qui appliquent les principes de l'AAC auprès des jeunes enfants ayant un TSA (Klintwall & Eikeseth, 2014). L'ICI tire ses origines des travaux de Lovaas (1981, 1987) réalisés au département de psychologie de l'Université de Californie à Los Angeles dans le cadre du *Young Autism Project*. En réponse à l'absence de traitements médicaux efficaces pour diminuer les manifestations associées au TSA, ce projet de recherche avait pour but de développer un traitement de modification comportementale pour améliorer le fonctionnement des enfants ayant ce diagnostic. Les travaux reposaient sur l'hypothèse qu'une intervention spécialisée, intensive et visant l'ensemble des sphères développementales permettrait aux très jeunes enfants ayant un TSA de rattraper les retards avant leur entrée en première année du primaire (Lovaas, 1987). Dans le cadre de cette étude déterminante, 47% des enfants avaient réalisé des gains substantiels et ont intégré avec succès l'école régulière, ce qui explique l'engouement de la communauté scientifique pour cette intervention. L'ICI est une intervention globale qui a pour but d'élargir le répertoire comportemental des enfants d'âge préscolaire en leur enseignant des comportements sociaux (par exemple, la communication), cognitifs (par exemple, les acquis préscolaires) et instrumentaux (par exemple, l'attention à la tâche). Les objectifs sont individualisés pour chaque enfant et découlent d'une évaluation initiale. L'ICI est dispensée selon un ratio d'un intervenant pour un enfant de façon intensive (c'est-à-dire entre 30 et 40 heures par semaine). Deux stratégies d'enseignement complémentaires sont utilisées, soient l'enseignement par essais distincts et l'enseignement fortuit (Paquet et al., 2012). L'enseignement par essais distincts consiste à fragmenter un comportement en petites unités d'apprentissage, qui seront enseignées une par une selon la séquence stimulus-réponse-conséquence. L'enseignement fortuit implique de saisir les occasions d'apprentissage qui surviennent dans le quotidien de façon naturelle. L'enseignement

fortuit permet entre autres de favoriser la généralisation des acquis et leur utilisation de façon spontanée.

### **L'état des connaissances sur l'ICI**

Plusieurs organismes nationaux de santé considèrent l'ICI comme une intervention établie (angl. *established intervention*) pour les enfants ayant un TSA (Health Technology Inquiry Service, 2008; INESSS, 2014; Maglione et al., 2012; National Autism Center, 2009; National Institute for Health and Care Excellence, 2013; Prior & Roberts, 2012). De nombreuses études et méta-analyses soutiennent également son efficacité (Eikeseth et al., 2012; Eldevik et al., 2009; Makrygianni & Reed, 2010; Peters-Scheffer et al., 2011; Reichow, 2012; Reichow et al., 2018; Virués-Ortega, 2010; Vismara & Rogers, 2010). La majorité des études rapportent des effets positifs qui se traduisent par des améliorations des habiletés cognitives, de la communication et du fonctionnement adaptatif (Reichow et al., 2018). Certaines études indiquent également une amélioration de la sévérité des symptômes autistiques (Eikeseth et al., 2012). Très peu d'études rapportent des tailles d'effet en raison de l'absence de groupe de contrôle. Cependant, la plus récente méta-analyse sur l'efficacité de l'ICI rapporte des tailles d'effet variant de faibles à modérées (Reichow et al., 2018).

Bien que l'ICI génère des gains positifs pour certains enfants ayant un TSA, plusieurs auteurs soulignent que les effets varient grandement d'un individu à l'autre (Eldevik et al., 2010; Howlin et al., 2009; Magiati et al., 2011; Reichow et al., 2018). Alors que certains enfants progressent significativement dans plusieurs sphères du développement, d'autres ne retirent qu'une modeste amélioration aux tests standardisés (Ben-Itzhak et al., 2014; Dawson et al., 2002; Gabriels et al., 2001; Howlin et al., 2009; Zachor & Ben Itzhak, 2010). Cette réponse différentielle à l'intervention demeure incomprise : les caractéristiques des enfants qui pourraient moduler l'efficacité de l'intervention, les périodes critiques d'intervention, le dosage optimal et

les marqueurs biologiques permettant d'identifier en amont les candidats les plus susceptibles de bénéficier de l'intervention sont à ce jour mal compris (Eapen et al., 2013; Magiati et al., 2011; Reichow et al., 2018). D'ailleurs, plusieurs études ont soulevé qu'à l'heure actuelle, aucun prédicteur fiable des résultats de l'ICI n'a été identifié (Eapen et al., 2013; Magiati et al., 2011; Reichow et al., 2018; Smith et al., 2015; Warren et al., 2011). Très peu d'études sur l'efficacité de l'ICI ont été élaborées de façon à explorer directement les modérateurs des effets de l'intervention (Ben-Itzhak et al., 2014; Eldevik et al., 2010). En raison de l'hétérogénéité du TSA, déterminer qui bénéficiera le plus de l'intervention constitue une question importante (Tiura et al., 2017).

### **Les prédicteurs potentiels de l'efficacité**

Certains chercheurs ont tenté d'identifier des modérateurs ou des médiateurs associés à l'efficacité de l'ICI. Il semble important de souligner que les variables investiguées dans les études portant sur les prédicteurs d'efficacité de l'ICI ont été déterminées sur des bases empiriques et non théoriques (Vivanti et al., 2014). En d'autres mots, il s'agit de variables sur lesquels les groupes d'intervention et les groupes de contrôle se distinguaient. Les variables les plus étudiées sont l'âge au début de l'intervention, l'intensité de l'intervention, le QI, les symptômes autistiques, le fonctionnement adaptatif et les caractéristiques sociodémographiques (Sallows & Graupner, 2005 ; Klintwall et al., 2015 ; Perry et al., 2011 ; Virues- Ortega et al., 2013). L'influence de ces variables a été examinée dans différentes études et méta-analyses, qui obtiennent parfois des résultats contradictoires. Sans prétendre faire une recension systématique, nous résumons ci-dessous les résultats des études ayant investigué l'influence de ces prédicteurs sur l'efficacité de l'ICI pour démontrer le manque de consensus dans la littérature scientifique à ce sujet. Le tableau 1 résume les caractéristiques des études présentées.

### ***Intensité de l'intervention***

L'intensité de l'intervention réfère au nombre d'heures prodiguées par semaine. Une majorité d'études suggère que l'intensité est associée à une meilleure efficacité de l'intervention sur le plan du fonctionnement adaptatif (Eldevik et al., 2010; Linstead et al., 2017; Makrygianni & Reed, 2010; Virués-Ortega, 2010). Cependant, les résultats de Fernell et al. (2011) ne soutiennent pas cette conclusion et suggèrent plutôt que l'intensité n'influencerait pas l'amélioration du fonctionnement adaptatif. En ce qui concerne les effets de l'ICI sur le QI, quelques études soutiennent que l'intensité serait positivement associée à l'amélioration du QI (Eldevik et al., 2010; Linstead et al., 2017; Makrygianni & Reed, 2010), tandis que d'autres n'ont pas trouvé d'association significative entre ces deux variables (Sallows & Graupner; Virués-Ortega, 2010). Nous avons trouvé une seule étude portant sur l'association entre l'intensité de l'intervention et la diminution des symptômes autistiques (Rogers et al., 2021). Les auteurs ont conclu que l'intensité de l'intervention n'influencerait pas la trajectoire des symptômes autistiques.

### ***Âge au début de l'intervention***

Plusieurs chercheurs ont mis en lumière l'importance d'intervenir de façon précoce pour maximiser les effets généraux de l'ICI (Flanagan et al., 2012; Granpeesheh et al., 2009; Makrygianni & Reed, 2010; Perry et al., 2011). Par exemple, un plus jeune âge au début de l'intervention serait associé à de meilleurs progrès sur le plan du fonctionnement adaptatif (Makrygianni & Reed, 2010), du QI (Harris & Handleman, 2000; Flanagan et al., 2012) et des symptômes autistiques (Perry et al., 2011). En revanche, d'autres études n'ont pas identifié de telles associations et soutiennent plutôt que l'âge n'influencerait pas l'efficacité de l'intervention (Bieleninik et al., 2017; Eldevik et al., 2006; Virués-Ortega, 2010; Robain et al., 2020).

## ***Quotient intellectuel***

L'influence du QI sur l'efficacité de l'ICI a été étudiée à de nombreuses reprises en tirant des conclusions contradictoires. Les résultats d'une méta-analyse soutiennent que le QI initial n'influencerait pas l'efficacité de l'intervention, mais serait plutôt fortement corrélé avec le QI post-intervention (Makrygianno & Reed, 2010). En d'autres mots, le QI initial serait associé au QI post-intervention, sans être associé au progrès réalisé par l'enfant. Ainsi, tant les enfants ayant un faible QI initial que ceux ayant un QI initial élevé bénéficieraient de l'ICI et seraient susceptibles de réaliser des gains cognitifs. D'autres études suggèrent plutôt que les enfants avec un QI initial plus faible auraient un plus grand potentiel de progrès, par rapport à ceux qui ont déjà un niveau de QI élevé, qui risquent de plafonner (Reed, 2016; Robain et al., 2020). Inversement, certaines études ont montré que le QI initial élevé était associé à de meilleurs progrès sur le plan cognitif (Harris and Handleman, 2000; Tirua et al., 2017). En ce qui concerne les effets de l'ICI sur fonctionnement adaptatif, le QI initial élevé serait associé à une meilleure efficacité de l'intervention (Ben-Itzchak et al., 2014; Eldevik et al., 2010; Fernell et al., 2011; Robain et al., 2020; Tirua et al., 2017). Très peu d'études ont directement étudié le lien entre le QI initial et l'amélioration des symptômes autistiques. Les résultats de Ben-Itzchak et collègues (2014) suggèrent qu'il n'y aurait pas de différence dans la diminution des symptômes autistiques en fonction du QI initial.

## ***Sévérité des symptômes autistiques***

Les données sont mitigées quant à la possible influence de la sévérité initiale des symptômes autistiques sur l'efficacité de l'ICI (Flanagan et al., 2012; Reed, 2016). Alors que certaines études suggèrent que des symptômes autistiques plus légers seraient associés à une meilleure efficacité de l'ICI (Ben-Itzchak & Zachor, 2007; Sallows & Graupner, 2005; Smith et al., 2000), d'autres suggèrent que des symptômes autistiques plus sévères seraient associés à une

meilleure réponse à l'intervention (Reed & Osborne, 2012; Remington et al., 2007). Il faut également souligner que certains chercheurs n'ont pas trouvé d'association entre ces deux variables (Harris & Handleman, 2000). Même si certaines études individuelles ont investigué l'effet prédictif des symptômes autistiques, aucune méta-analyse n'a encore abordé cette question (Reed, 2016). Une explication potentielle de l'écart observé est le manque d'uniformité dans les instruments utilisés pour mesurer les symptômes autistiques d'une étude à l'autre.

### ***Fonctionnement adaptatif***

L'apport du fonctionnement adaptatif initial dans la réponse à l'intervention est plus consensuel. Plusieurs études soutiennent qu'un fonctionnement adaptatif initial élevé est associé à une meilleure efficacité de l'ICI (Eldevik et al., 2010; Flanagan et al., 2012; Sallows & Graupner, 2005). Plus précisément, le fonctionnement adaptatif initial influencerait positivement les effets de l'intervention sur le plan des habiletés langagières et du fonctionnement adaptatif lui-même (Eldevik et al., 2010; Makrygianni & Reed, 2010) ainsi que sur le QI (Flanagan et al., 2012; Sallows & Graupner, 2005). Le lien entre le fonctionnement adaptatif initial et l'amélioration des symptômes autistiques ne semble pas avoir été directement investigué à ce jour.

### ***Variables sociodémographiques***

Certaines caractéristiques sociodémographiques ont été associées à un plus grand succès de l'intervention. Parmi celles-ci, on compte le statut socio-économique élevé, le niveau d'éducation des parents, le niveau de stress parental bas et le niveau d'implication parentale dans l'intervention (Ben-Itzhack & Zachor, 2011; Gabriels et al., 2001; Osborne et al., 2008; Richards et al., 2009; Shine & Perry, 2010). De plus, certains auteurs reconnaissent qu'un âge plus jeune au début de l'intervention peut être associé à d'autres facteurs liés à l'efficacité de l'intervention, comme les connaissances et les ressources des parents (Perry et al., 2011).



## Contexte d'intervention et transférabilité des résultats

Depuis la publication des orientations ministérielles du ministère de la Santé et des Services sociaux en 2003, les centres régionaux de réadaptation doivent offrir de l'ICI aux enfants de 5 ans et moins ayant un TSA (MSSS, 2003). Une question importante concerne la validité externe des conclusions tirées des études d'efficacité de l'ICI. La mesure dans laquelle ces résultats sont représentatifs des gains qui seraient réalisés par les enfants recevant l'ICI dans les services publics demeure incertaine (Flanagan et al., 2012; Perry et al., 2013; Reichow, 2012; Waters et al., 2020). La plupart des études sur l'efficacité des interventions et leurs prédicteurs ont été menées dans des conditions hautement contrôlées en milieu universitaire. Les études dont l'intervention a été dispensée en communauté obtiennent des résultats variables (Waters et al., 2020) et les effets sont souvent plus faibles (Smith et al., 2015). Les échantillons sont généralement petits (c'est-à-dire 10 à 30 enfants) et les enfants ont souvent moins de 4 ans (Perry et al., 2013), ce qui n'est pas représentatif des enfants recevant des services publics. Les résultats de ces études doivent donc être interprétés avec prudence, d'autant plus que la majorité des programmes d'ICI a été développée aux États-Unis (Jones et al., 2011; Rivard et al., 2013) où des analystes du comportement certifiés supervisent les interventions. Ces programmes peuvent être difficiles à adapter et implanter dans des contextes culturels et linguistiques différents (Jones et al., 2011; Rivard et al., 2013).

Le contexte québécois d'intervention diffère sur certains aspects. La première grande différence réside dans la formation du personnel qui met en place l'intervention. Contrairement aux universités américaines, les universités québécoises n'ont pas de programmes de baccalauréat ou de maîtrise en analyse appliquée du comportement, ce qui limite le nombre d'analystes du comportement disponibles. Selon le *Behavior Analyst Certification Board* (BACB), le Canada compte 1861 analystes du comportement certifiés, dont seulement 56 au Québec (BACB, 2021).

Ce nombre est faible compte tenu du fait que le Québec compte plus de 20 % de la population canadienne. Ainsi, ce sont des techniciens en éducation spécialisée qui mettent en place l'intervention. La plupart d'entre eux ont un diplôme d'études collégiales et sont supervisés par un psychologue (doctorat) ou un psychoéducateur (maîtrise) qui ont rarement une formation formelle en analyse comportementale (voir Mello et al., 2018 pour la description). La deuxième différence importante concerne le dosage. Le nombre d'heures offertes par semaine au Québec est faible à modéré par rapport aux États-Unis et varie selon les différentes régions administratives, car chaque centre régional de réadaptation est responsable d'organiser son offre de services. La dernière particularité du contexte québécois est l'accessibilité. Dans certaines régions, les listes d'attente pour accéder à l'évaluation diagnostique du TSA et à l'ICI sont longues (c.-à-d. plus d'un an), ce qui fait que les enfants sont plus âgés lorsqu'ils commencent à recevoir une intervention. Comme le préscolaire commence à 5 ans, ce délai implique que les enfants reçoivent l'ICI pendant une période de temps relativement courte. Pour l'ensemble de ces raisons, les effets de l'ICI telle qu'implantée au Québec pourraient être différents de ceux rapportés dans les études précédemment citées. La transférabilité des conclusions au contexte québécois reste à établir.

### **Approches statistiques et réponse à l'intervention**

Les protocoles à cas unique dominant la recherche évaluative en autisme (Wong et al., 2014; Steinbrenner et al., 2020). Une récente revue de la littérature sur les pratiques fondées sur des preuves pour les individus ayant un TSA révèle que les devis de groupe ne représentent que 23 % des articles recensés (Steinbrenner et al., 2020). Ni cette revue de la littérature ni les méta-analyses précédemment citées (c.-à-d., Eldevik et al., 2009; Makrygianni & Reed, 2010; Virués-Ortega, 2010; Reichow et al., 2018), ne partagent explicitement d'information sur les analyses statistiques utilisées pour évaluer les effets de l'ICI lorsque des devis de groupe sont employés.

Cela dit, l'inspection des articles inclus suggère que la grande majorité des chercheurs ont opté pour des statistiques traditionnelles, telles que l'ANOVA ou la régression linéaire. Ces analyses s'inscrivent dans une approche centrée sur les variables, qui s'appuie sur la prémisse que les relations entre les variables sont les mêmes à travers tous les individus de la population (Laursen et Hoff, 2006). Cette approche quantifie donc les relations entre les variables en termes d'agrégats statistiques représentant des associations ou coefficients moyens pour tous les individus d'un échantillon. En d'autres mots, leurs résultats représentent le niveau moyen de réponse à l'intervention. Même en utilisant des termes d'interaction dans leur modèle, ces analyses permettent difficilement d'identifier des catégories de personnes, et rendent peu compte de l'interinfluence des variables entre elles (Laursen et Hoff, 2006).

Une explication possible au manque de consensus concernant les prédicteurs potentiels de l'efficacité de l'ICI serait qu'une combinaison de variables (c'est-à-dire les profils des enfants) soit plus importante que la contribution individuelle de ces caractéristiques considérées de façon isolée. Une approche centrée sur la personne pourrait guider l'identification de sous-groupes plus homogènes parmi la population des enfants ayant un TSA. L'approche centrée sur la personne explore les relations entre les variables pour chaque individu et vise ensuite à les regrouper en sous-groupes plus homogènes (Bergman & Trost, 2006 ; Laursen & Hoff, 2006). Dans cette approche, les associations entre les variables peuvent être différentes selon les groupes d'individus identifiés dans l'échantillon. Plusieurs chercheurs recommandent de tenir compte des différences individuelles et d'identifier des sous-groupes plus homogènes dans les échantillons afin de mieux comprendre l'hétérogénéité des symptômes des TSA et la réponse à l'ICI chez les enfants (Eapen et al., 2013; Georgiades et al., 2013; Tiura et al., 2017).

Les exemples d'études portant sur les enfants ayant un TSA adoptant une approche centrée sur la personne sont pourtant encore rares. Certains chercheurs ont tenté d'identifier des

sous-groupes d'enfant ayant un TSA sur la base de leurs symptômes autistiques (Georgiades et al., 2013; Wiggins et al., 2012) ou sur la base d'une combinaison de plusieurs caractéristiques, telles que les symptômes autistiques, les capacités cognitives et le fonctionnement adaptatif (Kim et al., 2016; Zheng et al., 2020). Cependant, les relations entre l'appartenance à des sous-groupes et la réponse à l'intervention demeurent une question peu étudiée. Les travaux de Kim et al. (2016) sont particulièrement intéressants, car ils ont exploré les relations entre les sous-groupes identifiés et la stabilité du diagnostic, la présentation clinique et la réponse à diverses interventions, y compris l'AAC, *Floor Time*, l'orthophonie et l'ergothérapie. Jusqu'à présent, aucune étude ne semble avoir vérifié spécifiquement la relation entre l'appartenance à un sous-groupe et les résultats de l'ICI.

Une troisième approche statistique susceptible d'offrir un éclairage complémentaire à l'efficacité différentielle de l'ICI auprès des enfants ayant un TSA est l'apprentissage automatique (angl. *machine learning*). L'apprentissage automatique est un sous-domaine de l'intelligence artificielle et se taille une place de plus en plus importante dans la recherche psychosociale (Yarkoni & Westfall, 2017). Cette approche repose sur la prémisse qu'un patron (angl. *pattern*) existe dans les données. L'enjeu est d'identifier, parmi l'ensemble des fonctions mathématiques existantes, celle qui définit le mieux ce patron (Bzdok, 2018; Jaeger, 2020). Cette fonction est identifiée empiriquement, à partir de l'analyse des relations entre les données entrantes (angl. *features*) et sortantes (angl. *labels*), et permet de faire des prédictions sur de nouvelles données. L'apprentissage automatique se distingue des statistiques traditionnelles entre autres sur le plan des objectifs et du développement du modèle. Alors que l'objectif principal des statistiques traditionnelles est l'explication des phénomènes observés, notamment par l'identification des mécanismes causaux sous-jacents (Bzdok, 2018; Rajula et al., 2020), l'objectif principal de l'apprentissage automatique est de faire des prédictions les plus justes sur

de nouvelles données (Bzdok et Ioannidis, 2019; Yarkoni et Westfall, 2017). Pour y parvenir, l'algorithme entraîne un modèle sur des exemples afin qu'il reconnaisse les relations entre les données entrantes et sortantes, ce qui lui permet de faire des prédictions au niveau individuel sur de nouveaux cas (Jaeger, 2020). Dans le paradigme des statistiques traditionnelles, le développement du modèle se base sur la théorie (angl. *theory-driven*; Bzdok, 2018). Le choix des variables dépend des connaissances antérieures et découle d'un cadre théorique de référence (Bzdok, 2018; Rajula et al., 2020). De son côté, l'apprentissage automatique est considéré comme étant basé sur les données (angl. *data-driven*; Bzdok et al., 2020). Cela signifie que le choix des algorithmes est fait en fonction de leur capacité empirique et celui produisant les prédictions les plus justes sera retenu (Bzdok, 2018, Bzdok et al., 2020; Orrù et al., 2020). Les variables utilisées par l'algorithme sont celles identifiées comme utiles à la prédiction, qu'elles aient ou non une valeur explicative selon la théorie (Bzdok et Ioannadis, 2019; Bzdok et al., 2020; Jaeger, 2020).

À notre connaissance, seules quelques études ont utilisé l'intelligence artificielle pour étudier les résultats différentiels des interventions comportementales (Linstead et al., 2015, 2017). Par exemple, Linstead et collègues (2015) ont utilisé des réseaux neuronaux artificiels (angl. *artificial neural networks*) de façon complémentaire aux statistiques traditionnelles pour explorer la relation entre l'intensité de l'intervention et le nombre d'objectifs d'intervention atteints. Dans une étude ultérieure, la même équipe de recherche a examiné la relation entre le nombre d'heures d'intervention comportementale (variant de 20,02 à 197,25 par mois) et le nombre d'objectifs d'intervention atteints (Linstead et al., 2017). Les réseaux neuronaux artificiels ont surpassé les modèles de régression linéaire pour prédire le nombre d'objectifs d'intervention atteints en réponse à une intervention comportementale dispensée en communauté à des enfants ayant un TSA. Plus précisément, ces chercheurs ont utilisé les réseaux neuronaux

artificiels pour optimiser la forme de la fonction mathématique qui représente la relation entre l'intensité et le nombre d'objectifs atteints, ce qui a permis d'augmenter la variance expliquée par le modèle, passant de 35% pour la régression linéaire standard à 60% pour le modèle optimisé à l'aide des réseaux neuronaux artificiels. Ces résultats soutiennent le potentiel de l'utilisation de l'apprentissage automatique dans le contexte de la recherche évaluative en autisme.

### **Objectifs de la thèse**

La présente thèse doctorale compte trois principaux objectifs: (a) évaluer les effets de l'ICI, telle que dispensée par le Centre intégré de santé et services sociaux de la Montérégie-Ouest (CISSS-MO<sup>2</sup>), sur les symptômes autistiques et le fonctionnement adaptatif des enfants qui la reçoivent, (b) explorer de la présence de profils distincts chez les participants, selon leur niveau initial de QI, de fonctionnement adaptatif et de symptômes autistiques et leurs potentielles associations à une réponse différentielle à l'intervention, et (c) vérifier si l'apprentissage automatique (angl. *machine learning*) peut contribuer à l'estimation du pronostic des enfants recevant de l'ICI. Ensemble, ces objectifs visaient à mieux comprendre l'efficacité différentielle de l'ICI en utilisant une combinaison de stratégies analytiques complémentaires.

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<sup>2</sup> Au moment de la collecte de données, le CISSS-MO était connu sous l'appellation Centre de réadaptation en déficience intellectuelle et en trouble envahissant du développement de la Montérégie-Est (CRDITED-ME).

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**Tableau 1***Caractéristiques des études sur les prédicteurs d'efficacité*

| Auteurs                   | Devis                       | N    | Variables prédictives                                          | Variables prédites                                                                                                                    |
|---------------------------|-----------------------------|------|----------------------------------------------------------------|---------------------------------------------------------------------------------------------------------------------------------------|
| Ben-Itzhak & Zachor, 2007 | Corrélationnel              | 25   | Symptômes autistiques                                          | Échelle développementale↓                                                                                                             |
| Ben-Itzhak et al., 2014   | Corrélationnel              | 46   | QI                                                             | Fonctionnement adaptatif↑, symptômes autistiques                                                                                      |
| Bielenik et al., 2017     | Méta-analyse (40 études)    | 5771 | Âge                                                            | Symptômes autistiques                                                                                                                 |
| Eldevik et al., 2006      | Groupe de comparaison       | 28   | Âge                                                            | Fonctionnement adaptatif, QI                                                                                                          |
| Eldevik et al., 2010      | Méta-analyse (16 études)    | 453  | Intensité<br>QI                                                | Fonctionnement adaptatif↑, QI↑<br>Fonctionnement adaptatif↑                                                                           |
| Fernell et al., 2011      | Corrélationnel              | 208  | Intensité<br>QI                                                | Fonctionnement adaptatif<br>Fonctionnement adaptatif↑                                                                                 |
| Flanagan et al., 2012     | Groupe avec liste d'attente | 142  | Âge<br>Fonctionnement adaptatif                                | QI↑<br>QI↑                                                                                                                            |
| Harris & Handleman, 2000  | Corrélationnel              | 27   | Âge<br>QI                                                      | QI↑<br>QI↑                                                                                                                            |
| Linstead et al., 2017     | Corrélationnel              | 1468 | Intensité                                                      | Fonctionnement adaptatif↑, QI↑                                                                                                        |
| Makrygianni & Reed, 2010  | Méta-analyse (14 études)    | N/D  | Intensité<br>Âge<br>QI<br>Fonctionnement adaptatif             | Fonctionnement adaptatif↑, QI↑<br>Fonctionnement adaptatif↑<br>Corrélé au QI final (mais pas au progrès)<br>Fonctionnement adaptatif↑ |
| Perry et al., 2011        | Corrélationnel              | 332  | Âge                                                            | Symptômes autistiques↓                                                                                                                |
| Remington et al., 2007    | Groupe de comparaison       | 44   | Symptômes autistiques                                          | QI↑                                                                                                                                   |
| Reed & Osborne, 2012      | Groupe de comparaison       | 66   | Symptômes autistiques                                          | Fonctionnement adaptatif↑                                                                                                             |
| Robain et al., 2020       | Corrélationnel              | 60   | Âge<br>QI                                                      | Symptômes autistiques<br>QI↓                                                                                                          |
| Rogers et al., 202        | Étude randomise-contrôlée   | 87   | Intensité                                                      | Symptômes autistiques                                                                                                                 |
| Sallows & Graupner, 2005  | Étude randomisée-contrôlée  | 24   | Intensité<br>Symptômes autistiques<br>Fonctionnement adaptatif | QI<br>QI↓<br>QI↑                                                                                                                      |
| Smith et al., 2000        | Étude randomisée-contrôlée  | 28   | Symptômes autistiques                                          | Légers = meilleure efficacité                                                                                                         |
| Tiura et al., 2017        | Corrélationnel              | 35   | QI                                                             | QI↑                                                                                                                                   |
| Viruès-Ortega, 2010       | Méta-analyse (22 études)    | N/D  | Âge<br>Intensité                                               | QI, fonctionnement adaptatif<br>QI                                                                                                    |

*Note.* ↑ indique une association positive; ↓ indique une association négative; l'absence de flèche indique l'absence d'association; N/D = non disponible.

**Chapitre II – Article 1**



**Changes in Autistic Symptoms and Adaptive Functioning of Children Receiving Early Behavioral Intervention in a Community Setting: A Latent Growth Curve Analysis**

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## Résumé

Bien que l'intervention comportementale intensive précoce ait démontré des effets dans des études bien contrôlées, la mesure dans laquelle cette intervention produit des changements positifs dans les environnements plus naturels reste incertaine. Ainsi, notre étude a examiné les changements dans les symptômes autistiques et le fonctionnement adaptatif chez 233 enfants autistes recevant une intervention comportementale précoce en communauté. Les résultats ont révélé des changements non linéaires dans le fonctionnement adaptatif, caractérisés par des améliorations significatives pendant la période d'intervention suivis d'une stabilité lors du post-test, et une petite diminution linéaire des symptômes autistiques tout au long de l'étude. L'intensité de l'intervention, l'âge au début de l'intervention, le QI et les symptômes autistiques étaient associés soit à des progrès au cours de la période d'intervention, soit à un maintien au cours de la période de suivi. La prochaine étape pour étendre ce domaine de recherche serait de collecter des données détaillées sur les stratégies d'intervention et la fidélité d'implantation afin de pouvoir faire des recommandations concrètes pour les intervenants.

*Mots-clés* : fonctionnement adaptatif, symptômes autistiques, intervention comportementale précoce, efficacité, courbes de croissance latente

## **Abstract**

Despite showing effects in well-controlled studies, the extent to which early intensive behavioral intervention (EBI) produces positive changes in community-based settings remains uncertain. Thus, our study examined changes in autistic symptoms and adaptive functioning in 233 children with autism receiving EBI in a community setting. The results revealed nonlinear changes in adaptive functioning characterized by significant improvements during the intervention and a small linear decrease in autistic symptoms from baseline to follow-up. The intensity of intervention, initial age, IQ and autistic symptoms were associated either with progress during the intervention or maintenance during the follow-up. The next step to extend this line of research involves collecting detailed data about intervention strategies and implementation fidelity to produce concrete recommendations for practitioners.

*Keywords:* adaptive functioning, autistic symptoms, early behavioral intervention, effectiveness, latent growth curves

## **Changes in Autistic Symptoms and Adaptive Functioning of Children Receiving Early Behavioral Intervention in a Community Setting: A Latent Growth Curve Analysis**

Given that numerous studies and meta-analyses have supported its efficacy (Eldevik et al., 2009; Makrygianni & Reed, 2010; Makrygianni et al., 2018; Peters-Scheffer et al., 2011; Prior et al., 2011; Reichow et al., 2018; Virues-Ortega, 2010; Vismara and Rogers, 2011; Weitlauf et al., 2014), many national health organisations consider early intensive behavioral intervention (EIBI) as an established intervention for children with ASD (Health Technology Inquiry Service, 2008; INESSS, 2014; Maglione et al., 2012; National Autism Center, 2009; National Institute for Health and Care Excellence, 2013; Prior & Roberts, 2012). Researchers have reported positive effects that translate into improvements in cognitive skills, communication abilities, and adaptive functioning (Reichow et al., 2018), but also considered the strength of the evidence as weak, mainly because of nonoptimal study designs and small sample sizes. Some studies have also observed a decrease in autistic symptoms following EIBI (Eikeseth et al., 2012).

Although EIBI generates positive gains for some children with ASD, several researchers point out that the effects vary greatly from one individual to another (Eldevik et al., 2010; Howlin et al., 2009; Magiati et al., 2011; Prior et al., 2011; Reichow et al., 2018). While some children progress significantly in various areas of development, others make only modest, or even no, improvement on standardized tests (Ben-Itzhak et al., 2014; Dawson et al., 2002; Gabriels et al., 2001; Howlin, 2009; Zachor and Ben-Itzhak, 2010). Heterogeneity in outcomes remains poorly understood; the characteristics of the children which could modulate the efficacy of the intervention, the critical period of intervention, the optimal dosage and the biological markers to identify the candidates most likely to benefit from the intervention are still unknown (Eapen et al., 2013; Magiati et al., 2012; Reichow et al., 2018). To date, researchers have not identified reliable predictors of EIBI outcomes (Eapen et al., 2013; Reichow, 2012; Smith et al., 2015;

Warren et al., 2011). Very few studies on the efficacy of EIBI directly explore moderators of the effects of the intervention (Ben-Itzhak et al., 2014; Eldevik et al., 2010). Knowing the predictors of EIBI outcomes is crucial information for customizing the intervention (Tiura et al., 2017). Moreover, knowledge on how to individualize dosage for children with ASD who present various skills, needs, ages, and live in different family contexts is virtually nonexistent (Pellecchia et al., 2019).

### **Predictors of Efficacy**

Some researchers have attempted to identify moderators or mediators associated with the efficacy of EIBI. The most studied variables are age at enrolment, intervention intensity, intellectual quotient (IQ), autistic symptoms, adaptive functioning, and sociodemographic characteristics (Sallows & Graupner, 2005; Klintwall et al., 2015; Perry et al., 2011; Virués-Ortega et al., 2013). The influence of these variables has been examined in different studies and meta-analysis, which sometimes obtain contradictory results. We summarize below the findings on the influence of various predictors on improvement in adaptive functioning, intellectual functioning, language skills and autistic symptoms.

#### ***Intervention Intensity***

Numerous studies have found that a higher intensity is associated with more gains in adaptive functioning (Eldevik et al., 2010; Linstead et al., 2017; Makrygianni & Reed, 2010; Reed, 2016; Virués-Ortega, 2010). However, results from Fernell et al. (2011) do not support this conclusion. In terms of IQ, some studies indicate the intensity of the intervention is positively associated with improvement in IQ (Eldevik et al., 2010; Linstead et al., 2017; Makrygianni & Reed, 2010) while others do not find this association (Sallows and Graupner, 2005; Virués-Ortega, 2010). Similarly, the literature does not show consensus on the association between intervention intensity and language gains. Two studies found a positive association (Linstead et

al., 2017; Virués-Ortega, 2010) whereas another did not (Makrygianni & Reed, 2010). We identified only one study investigating the association between intervention intensity and improvement in autistic symptoms (Rogers et al., 2021). These authors concluded that intervention intensity did not impact the trajectory of autistic symptoms.

### ***Age at Enrollment***

Studies have reported mixed results regarding age at enrollment (Reed, 2016). Some researchers have highlighted the importance of early intervention to maximize the overall effects of the intervention (Granpeesheh et al., 2009; Makrygianni & Reed, 2010; Perry et al., 2013). Results suggest a positive predictive association between age at enrollment and progress in adaptive functioning (Fenske et al., 1985; Makrygianni & Reed, 2010), cognitive gains (Harris and Handleman, 2000; Flanagan et al., 2012; Waters et al., 2020), language (Frazier et al., 2021) and milder autistic symptoms after the intervention (Perry et al., 2011). Despite these results, other studies suggest that age at enrollment does not influence the effects of EIBI (Bieleninik et al., 2017; Eldevik et al., 2006; Virués-Ortega, 2010; Robain et al., 2020).

### ***Intellectual Quotient***

Various researchers have studied the influence of IQ on the efficacy of EIBI, with conflicting conclusions. One meta-analysis suggests IQ was not linked to the efficacy of the intervention but would rather be strongly correlated with the post-intervention IQ (Makrygianni & Reed, 2010). In other words, the initial IQ would be associated with the post-intervention IQ, without being associated with the progress made by the child. Thus, both children with a low initial IQ and high initial IQ would benefit from EIBI and would be likely to achieve cognitive gains. Other evidence rather suggests that children with lower initial IQ would have a greater potential for progress, compared to those who already had high IQ level, which risk peaking (Reed, 2016), while some studies found that children with higher initial IQ benefited more from

the intervention (Fernell et al., 2011; Harris and Handleman, 2000; Tiruas et al., 2017). To our knowledge, only one meta-analysis has examined the influence of initial IQ on adaptive functioning (Eldevik et al., 2010). The results indicate that initial IQ positively predicts gains in adaptive functioning. Very few studies have directly investigated the link between initial IQ and decrease in autistic symptoms. Ben-Itzhak and colleagues (2014) suggest that there would be no difference in the decrease in autistic symptoms based on initial IQ.

Even though some evidence indicates that verbal and non-verbal IQ have different associations with adaptive functioning and autistic symptoms (Black et al., 2009; Munson et al., 2008), the previous meta-analyses have only investigated the full-scale IQ as a predictor of efficacy. The conflicting findings regarding the influence of IQ on the efficacy of EIBI may reflect a concern that full-scale IQ is too general to uncover more specific aspects of IQ that predict child progress. The influence of the different IQ scales is worth being investigated.

### ***Autistic Symptoms***

Again, evidence remains mixed on the possible influence of autistic symptoms on the efficacy of EIBI (Flanagan et al., 2012; Reed, 2016). While some studies suggest that milder autistic symptoms may be associated with better efficacy of EIBI (Ben-Itzhak and Zachor, 2007; Frazier et al., 2021; Sallows and Graupner, 2005; Smith et al., 2000), others did not find this association (Harris and Handleman, 2000), and some even suggest that higher autistic symptoms may be associated with a better response to the intervention (Remington et al., 2007; Reed and Osborne, 2013). Even though some individual studies have investigated the predictive effect of autistic symptoms, no meta-analysis has yet addressed this question (Reed, 2016). One potential explanation for the observed discrepancy is the lack of uniformity in the instruments used to measure autistic symptoms across studies.

### ***Adaptive Functioning***

In contrast, a general consensus has emerged on the influence of initial adaptive functioning. Several studies support that high adaptive functioning in children is associated with improved efficacy of EIBI (Eldevik et al., 2010; Flanagan et al., 2012; Reed, 2016; Reichow, 2012; Sallows and Graupner, 2005; Vivanti, 2014). More specifically, the results of meta-analyses suggest that initial adaptive functioning positively influences the effects of the intervention on language skills and on adaptive functioning itself (Eldevik et al., 2010; Makrygianno and Reed, 2010). A positive association between initial adaptive functioning and post-intervention IQ has also been found (Sallows and Graupner, 2005). That said, research has yet to examine the interaction between initial adaptive functioning and post-intervention severity of autistic symptoms.

### ***Sociodemographic Characteristics***

Certain sociodemographic characteristics were associated with greater success of the intervention, such as high socio-economic status, parental education level, low parental stress, and level of parental involvement in the intervention (Gabriels et al., 2001; Magiati et al., 2011). Moreover, some authors acknowledge that younger age at enrollment (i.e., early entry) may be associated with other factors related to outcome, like parental knowledge and resourcefulness (Perry et al., 2011).

### **Statistical Methods Used to Evaluate Intervention Effects**

When assessing intervention effects in autism, researchers most frequently use single-case experimental designs (Wong et al., 2014; Steinbrenner et al., 2020). A recent literature review on evidence-based practices for people with autism reported that group designs only represent 23% of the articles included (Steinbrenner et al., 2020). Neither this literature review, nor the previously cited meta-analyses (i.e., Eldevik et al., 2009; Makrygianni & Reed, 2010; Virués-Ortega, 2010; Reichow et al., 2018), explicitly share information about the statistical analyses



used to assess intervention effects in group design studies. That said, a brief overview of the included articles indicates that the vast majority used traditional analyses such as ANOVA or linear regression. Despite their usefulness, these traditional analyses rest on a number of assumptions that are either unrealistic or difficult to meet in evaluative research, like the assumptions of compound symmetry, sphericity and homogeneity of variance (Mun et al., 2009; Singer & Willett, 2003).

Recently, researchers in observational studies have begun to employ more contemporary analyses (Caplan et al., 2019; Simonoff and colleges, 2020). One such method is latent growth curve (LGC) analysis. For example, Simonoff and colleges (2020) used LGC in an epidemiological study to estimate the trajectories in autistic symptoms and cognitive ability from childhood to adulthood, while Caplan and colleges (2019) used LGC to assess the relationship between responsive parenting and children's social skills over time. LGC should also be considered for evaluating intervention effectiveness in group designs in the field of early intervention as it has several advantages over traditional methods (Mun et al., 2009): the advantage of handling measurement errors and individual differences in response to intervention as well as avoiding unrealistic assumptions of traditional analyses mentioned above. Moreover, LGC does not require balanced data; that is, each participant does not need to have the same number of time points, allowing for attrition as long as it remains missing at random or missing completely at random (Singer & Willet, 2003). Finally, LGC can effectively assess intervention efficacy in studies utilizing a pre-post-post design (Mun et al, 2009) and researchers should use these analyses more often.

### **Effectiveness of Intervention in Community Setting**

Most studies on the efficacy of interventions and their predictors have been carried out in a university setting, under highly controlled conditions. The extent to which these results are

representative of those that would be achieved by children receiving EIBI in community settings remains uncertain (Flanagan et al., 2012; Perry et al., 2013; Reichow, 2012; Waters et al., 2020). Studies in which the intervention took place in community settings find variable results (Waters et al., 2020), and effects are often smaller (Smith et al., 2015). Samples are generally small (i.e., 10-30 children) and children are often under the age of 4 (Perry et al., 2013), which is unrepresentative of children receiving services in the community and calls for caution in interpreting this body of results. Furthermore, the majority of EIBI programs were developed in the United States (US; Jones et al., 2011; Rivard et al., 2013) where Board Certified Behavioral Analysts (BCBA) supervise the interventions. These programs may be difficult to adapt and implement in different cultural and linguistic settings (Jones et al., 2011; Rivard et al., 2013).

Notably, the intervention context in Québec, Canada, differs in various aspects from those that have been studied in the past. Since the Ministerial orientations published in 2003 by the Québec's Department of Health and Social Services, regional readaptation centers must provide EIBI to children aged from 2 to 5 with ASD (MSSS, 2003). The first major dissimilarity resides in the training of the personnel who implement the intervention. Unlike American universities, Quebec universities do not have bachelor's or master's programs in applied behavior analysis, which limits the number of behavior analysts available. According to the Behavior Analyst Certification Board, Canada counts 1861 certified behavior analysts, with only 56 in Quebec (BACB, 2021). This number is low considering that Québec comprises more than 20% of the Canadian population. Intervention is therefore implemented by special education technicians<sup>3</sup> that received a different training. Most of them have a college degree in special care counselling and

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<sup>3</sup> Special education technicians is a terminology unique to the province of Quebec and refers to college-level technicians.

are supervised by a psychologist (doctoral degree) or a psychoeducator (master's degree) who rarely have formal training in behavior analysis (see Mello et al., 2018 for description).

The second important difference lays in the dosage of the intervention offered. While most studies included in the cited meta-analysis offered high intensity intervention, the number of hours offered weekly in Quebec is low-to-moderate when compared to the United States and varies across different geographic areas, as each regional readaptation center is responsible to organise their service offer. In many cases, the intensity may not qualify the intervention as being “intensive”. Hence, we will use the expression early behavioral intervention (EBI) to refer to their program to prevent misleading the reader. The last particularity of the Quebec context is the accessibility. In some regions, waiting lists to access diagnostic assessment and access EBI are long (more than 1 years), which results in children being older when they start receiving intervention. As preschool starts at 5 years old, this delay implies that children receive the intervention for a relatively short period of time. Therefore, the purpose of our study was to (1) evaluate the effects of a community-based intervention program as offered by a regional readaptation center in Quebec, Canada, (2) determine whether the effects were maintained at the 1-year follow-up, and (3) identify potential predictors of effectiveness.

## **Method**

### **Participants and Procedures**

This study consists of secondary analyses of data from an assessment of the effectiveness of a community-based intervention program conducted among an unselected community sample that took place from 2009 to 2012. The sample included 233 children (78.6% boys) diagnosed with ASD aged between 2.50 to 5.75 years old ( $M = 4.34$ ,  $SD = 0.47$ ) who received one of the two early behavioral intervention options. Participants were divided between the intervention options in the following proportion: 53.9% of participants received low-intensity intervention and

the remaining 46.1% received moderate-intensity intervention (see intervention description below). This study is the first to combine the two intervention options; previous papers only analysed data for the moderate-intensity option.

Given that the detailed procedures were reported elsewhere (Rivard et al., 2014, Rivard et al., 2019), we only provide a summary here. We used a prospective longitudinal design with three annual assessments. Time 1 represents baseline (n = 233), time 2 represents post-intervention (12 months after baseline; n = 219) and time 3 represents 12-months follow-up after the end of the intervention (n = 64). Because the intervention took place the year before entering preschool for the majority of our sample, attrition at time 3 may be the result of children no longer receiving services from the readaptation center, making them harder to reach for the completion of the 12-months follow-up. To be eligible for the intervention and the study, children needed (1) to have a diagnosis of ASD provided by a pediatric psychiatrist and independently confirmed by a multidisciplinary team, (2) to be aged 5 years old or younger, and (3) to live within the geographical area served by the rehabilitation center. In addition, (4) parents had to provide written consent to participate in the study. The Joint Research Ethics Board for [removed for blind review] approved the research project.

### **Early Behavioral Intervention Program**

The study took place in a rehabilitation center providing developmental services to persons with an intellectual disability, ASD, or both in a large catchment area with a suburban and rural population of 847,422 (at the time of the study) located on the South Shore area near Montreal, Quebec, Canada. This public agency provided intervention based on applied behavior analysis. Their program was mostly based on the work of Lovaas and Maurice (Lovaas, 1981; Maurice, Green, & Luce, 1996) and adopted a 1:1 child-to-educator ratio. Generally, two special education technicians alternated working directly with the child. The special education

technicians implemented the intervention in the child's natural environment, usually at home or in a childcare setting.

Every two weeks, a clinical advisor or a psychologist responsible for this service provided supervision to the special education technicians. Intervention objectives were individualized for each child according to their baseline evaluation. These objectives primarily targeted basic (e.g., eye contact, attention to task), social (e.g., communication, social interaction) and cognitive (e.g., school-type tasks) skills. The special education technicians used mostly a combination of two teaching strategies: discrete trial teaching and incidental teaching. Discrete trial teaching refers to the repetitive use of the stimulus-response-consequence sequence to teach a behavior, while incidental teaching implies structuring the environment in order to provide learning opportunities (Paquet et al., 2012). The rehabilitation center offered two intensity intervention options: low-intensity (i.e., between 4 and 12 hours weekly) and moderate-intensity (i.e., between 16 and 20 hours weekly). The intensity option was determined based on the center's evaluation of the children needs at enrollment in the services, and the preferences and availability of the parents (see Rivard et al., 2014, 2019 for more details about the intervention).

## **Measures**

### ***Autistic Symptoms***

The *Childhood Autism Rating Scale 2* (CARS-2; Scholper et Van Bourgondien, 2010) was used to assess the participants' autistic symptoms. The CARS-2 contains 15 items assessing behavior on a 4-point scale from one to four (1 = normal, 4 = severely abnormal) based on direct behavioral observations. Half points can be scored if the child's symptoms are between two anchor points. Items assess different apparent difficulties in children with ASD (i.e., social relations, imitation, emotional responses, use of body, use of objects, adaptation to change, visual responses, auditory responses, taste/smell/touch, fear and anxiety, verbal communication,

nonverbal communication, level of activity, intellectual functioning, and general impression). The scores range between 15 and 60, with higher scores indicating more severe autistic symptoms. The CARS-2 has an excellent internal consistency ( $\alpha = .93$ ) in children aged 0 to 6 years and very good test-retest reliability ( $r = .88$ ; Scholper and Van Bourgondien, 2010). In the present study, the CARS-2 was completed by a parent and a special education technician. As both informants were highly correlated ( $r = .67 - .81$  depending on time point), we used the average score between the two respondents.

### ***Adaptive Functioning***

We used the *Adaptive Behavior Assessment System-II* (ABAS-II; Harrison and Oakland, 2003) to assess adaptive functioning, more precisely the parent/primary caregiver form for children aged 0-5 years old. The ABAS-II contains 241 items rating the performance of various adaptive behaviors on a 4-point scale, from zero to three (0 = never, the child is unable, 3 = always when necessary). The ABAS-II results provide a score for each of the three domains recognized by the American Association for Intellectual and Developmental Disabilities (Schalock et al., 2010) as necessary to assess adaptive behaviors, namely the conceptual, social, and practical domains. The ABAS-II also provides a general adaptive composite score based on the three aforementioned domains. The ABAS-II has demonstrated excellent internal consistency for general adaptive functioning ( $\alpha = .98 - .99$ ) and the three adaptive domains ( $\alpha = .90 - .98$ ), as well as very high test-retest reliability ( $r = .90$ ; Harrison & Oakland, 2003).

### ***Intellectual Functioning***

The *Wechsler Preschool and Primary Scale of Intelligence* (WPPSI-III; Wechsler, 2003) was used to measure intellectual functioning. The WPPSI-III consists of 15 sub-tests, which are in turn grouped into five dimensions: verbal comprehension, visuospatial performance, fluid reasoning, working memory and information processing speed. Depending on the child being

assessed, test administration lasts between 45 and 105 min. The results provide scores for the verbal IQ, the performance IQ, the general language composite, and the full-scale IQ. In the present study, only the verbal IQ, performance IQ and general language composite were considered because they provide more specific information than the full-scale IQ. The scale's internal consistency ranged between .83 and .95 across subtests and .89 and .96 across composite scores. Test-retest reliability coefficients were .87, .81, .88, and .88 for the verbal IQ, performance IQ, full-scale IQ, and general language composite, respectively. Inter-rater reliability ranged between .98 and .99. In this study, the WPPSI-III was administered by research assistants who were psychology graduate students supervised by the head of the research team, a psychologist and a university professor.

### ***Program Intensity***

Program intensity was a dichotomous variable. The response options were: 0 = low-intensity intervention or 1 = moderate-intensity intervention.

### ***Age at Enrollment***

Age was a continuous variable. We used decimals to collect the exact age of the participants. For example, a 2-year-old and 6-month-old would have a score of 2.5 years.

### ***Annual Income***

Annual income was an ordered-categorical variable. We asked the participant's parents the following question: "What is your annual family income?" The response options were: 1 = 10,000 to \$29,999, 2 = \$30,000 to \$49,999, 3 = \$50,000 to \$69,999, 4 = \$70,000 to \$89,999, and 5 = Over \$90,000.

## **Statistical Analyses**

### ***Descriptive Statistics and Attrition Analyses***

We performed preliminary analyses using SPSS 26.0, which involved descriptive (i.e., frequencies, mean, measure of dispersion) and correlational statistics. Considering the high attrition at the third assessment, we made attrition analyses using Little's missing completely at random test, followed by Chi-square tests (for categorical variables) or ANOVAs (for continuous variables) when significant to verify if any of the sociodemographic characteristics were associated with attrition.

### ***Latent Growth Curve Analyses***

Using Mplus 8.3 (Muthén & Muthén, 2017), we conducted LGC analyses within the structural equation modelling framework (Bollen & Curran, 2006) to estimate changes in autistic symptoms, general adaptive functioning, and the conceptual, social, and practical domains of adaptive functioning of the participants. LGC analysis estimates change through latent variables (i.e., unobserved variables; see Bollen & Curran, 2006; Curran et al., 2010). The objective was to determine the shape of the trajectory for the sample that is *a priori* unknown. The analysis begins by estimating an individual growth curve for each child. Then, the program estimates the average growth factor parameters, namely an intercept (i.e., the average score at T1) and a slope (i.e., expected amount of change over time). The variances of these growth factor parameters are also estimated, which represent the differences between individuals. Thus, LGC estimate interindividual differences in intra-individual change (Little, 2013; Stull et al., 2011). Another interesting aspect of LGC within the structural equation modelling framework is that it is straightforward to include categorical or continuous predictors of the intercept and slope parameters (Curran et al., 2010; Little, 2013).

We estimated separate models for five outcome variables (autistic symptoms, general adaptive functioning, conceptual domain, social domain, practical domain). Because of the small sample size and attrition, we followed the recommendation of using the maximum likelihood



robust estimator (MLR; Shi et al., 2021). This estimator essentially corrects the standard errors and chi-square tests for non-normality in the data. We dealt with missing data using full-information maximum likelihood estimation, which allow to use every case in the sample and provide unbiased parameters estimates, even in the presence of large attrition (see Enders, 2010; Little, 2013).

To determine the best growth model for each outcome, we followed a model-building procedure proposed by Preacher et al. (2008), where a series of nested LGC models is specified in a predetermined sequence, starting with an intercept-only without variance model and gradually increasing in complexity, while comparing the model fit. Our analyses sequentially estimated the following models for all outcome variables to determine the best shape of the trajectory in the data: Model 0 = fixed intercept (no variance), Model 1 = a random intercept, Model 2 = a random intercept and a fixed slope, Model 3 = a random intercept and a random slope, Model 4 = a random intercept, a random slope and imposed homoscedasticity of the residuals. We also estimated models with autocorrelations of the time-specific residuals across time, but they were not retained in any models, as either the autocorrelations were not significant, or it resulted in out-of-bound parameters (i.e., negative variance). Even though we only had three time points, it was important to test for potential nonlinear change in the growth curves in order to determine if the nature of the change was different during the year after the intervention has ended. In principle, only a linear model can be estimated with three time points (for instance, a quadratic slope cannot be estimated because it is not statistically identified), but alternative specifications are possible to test for nonlinearity (see Bollen & Curran, 2006).

Consequently, Model 5 tested the nonlinearity of the growth curves by estimating latent basis models (LBM), where the first two loadings of the slope are fixed at 0 and 1, while the last loading was freely estimated (Bollen & Curran, 2006). In such model, a significant increase in

model fit paired with a third estimated loading largely different from the value expected for a linear trajectory (in this case, a loading of 2) suggests presence of nonlinear change. When nonlinearity was detected using LBM, we used a piecewise growth modeling (PWGM; Kamata et al., 2013) in Model 6 to estimate an intercept and two slopes; the first slope captures change between time 1 and time 2 (slope 1), while the second slope captures change between time 2 and time 3 (slope 2). The resulting PWGM is a saturated model (i.e., with no degree of freedom and perfect fit), but it has the distinct advantage of providing an estimate of two different slopes and regressing predictors on these two different slopes to determine whether predictors are associated to different phases of change.

To produce a PWGM that was identified with only three time points, we followed the model specification suggested by Kamata et al. (2013). Finally, we modeled conditional LGC in Model 7 by including time-invariant exogenous predictors of the intercept and slopes. For all conditional models, predictors were age at enrolment, intensity of intervention, family annual income, verbal IQ, performance IQ, general language composite. We added general adaptive functioning as a predictor for the autistic symptoms model, and we added autistic symptoms as a predictor for the general adaptive functioning, conceptual, social and practical models.

**Model Fit.** In the structural equation modelling framework, various statistical tests and fit indices are used to determine to what extent the model-implied covariance-matrix adequately reproduced the observed data (see West et al., 2012). For absolute fit, Mplus report the chi-square test, the root mean square error of approximation (RMSEA) and the Standardize Root Mean Square Residual (SRMR). A non-significant chi-square test, an RMSEA value under .6 and a SRMR value under .08 indicate good fit of the model to the data (Hu & Bentler, 1999). For comparative fit, Mplus reports the comparative fit index (CFI) and the Tucker-Lewis index (TLI). CFI and TLI values under .95 indicate excellent fit to the data (Hu & Bentler, 1999), but values

of at least .90 still indicate acceptable fit (Little, 2013). Three Information criteria used to compare different models are also calculated, namely the Akaike information criterion (AIC), the Bayesian information criterion (BIC), and the sample-size adjusted Bayesian information criterion (SABIC). Lower values of AIC, BIC and SABIC indicate a better fitting model. To compare nested models, we used the Satorra-Bentler adjusted chi-square test suitable for the MLR estimator, as well as the difference in RMSEA ( $\Delta$ RMSEA) and CFI ( $\Delta$ CFI). A significant Satorra-Bentler adjusted chi-square test indicates that the compared models provide significantly different fits to the data (Satorra, 2000). A difference in RMSEA smaller than 0.015 suggests that there is no significant difference between models (Chen, 2007), while a difference in CFI greater than 0.02 indicates a significant difference between models (Cheung and Rensvold, 2002).

## **Results**

### **Descriptive Statistics and Attrition Analyses**

Table 1 presents the descriptive statistics. Given that intervention option was determined based on the evaluation of the children needs, we provide descriptive information for each intensity option in the online supplements (see Table S1). For the attrition analyses, Little's missing completely at random test was nonsignificant for autistic symptoms, but was significant ( $p = .029$ ) for the constructs measured by the ABAS-II, namely general adaptive functioning, conceptual, social and practical domains. This result suggested that attrition was not completely random; however, chi-square tests and ANOVAs comparing the scores of children who were lost to attrition to those who were not were all nonsignificant. Therefore, we approximated a missing at random pattern and the use of the robust maximum likelihood estimator is adequate.

### **LGC Analyses**

#### ***Model Selection***

The various fit indices indicated that, for all outcomes considered in this study, the addition of parameters in the models up to a linear slope with a variance resulted in significant improvement in fit to the data; for lighten the presentation, Table S2 in the online supplements presents the fit indices for Model 0 to 3, while Table 2 presents the fit indices for subsequent Models 4 to 7. For autistic symptoms, the best fitting unconditional model was Model 4 (i.e., random intercept, random slope, and homoscedasticity of residuals). All the fit indices suggest this model provided good fit to the data. Testing nonlinearity of the trajectory in Model 5 (i.e., LBM), inspection of the freed loading clearly suggested linear changes in autistic symptoms.

For three of the variables linked to adaptive functioning (general adaptive functioning, conceptual domain, social domain), Model 5 (i.e., LBM) fitted significantly better than the linear change model. In addition to improved fit, inspection of the freed loadings clearly suggested nonlinearity over time. For the practical domain of adaptive functioning, inspection of the fit indices did not allow distinguishing the linear model from the non-linear model with certainty, but visual inspection of the observed trajectory suggested possibility of nonlinear change, so we estimated Model 6 (i.e., PWGM) to inspect the slopes. For all four variables, Model 6 (i.e., PWGM) confirmed nonlinearity by estimating different values for slope 1 (T1 to T2) and slope 2 (T2 to T3). Model 6 is saturated; therefore, the fit of this model can only be compared to previous models using the information criteria. We observed large reductions in AIC, BIC and SABIC comparing this model to Model 4 (i.e., linear growth), but trivial differences compared to Model 5 (i.e., LBM), which confirmed the adequacy of Model 6 (i.e., PWGM). Figure 1 shows the plotted observed and model-implied (predicted) scores for the four variables of adaptive functioning.<sup>4</sup> Visual inspection reveals a deceleration between T2 and T3, which is consistent

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<sup>4</sup> In order to provide estimates of uncertainty for the mean scores across time, Table S3 available in the online supplements provides residual variances for Model 6 at each time points.

with nonlinearity, similarly to the estimation of two different slopes in the Model 6 (i.e., PWGM). As for the practical domain, it may appear more linear than the other variables' trajectories, but it is mostly because it has the smallest slope, followed by stability like the other domains. Figure 1 also shows that the predicted trajectories of the final selected model followed closely the observed trajectory.

### ***Growth Parameters***

Table 3 presents the growth parameters for all the final models for the four outcome variables. Readers can use the spreadsheet available on the online supplements to compare their data with our average estimated trajectories. For autistic symptoms, the final unconditional model (Model 4) reveals that the intercept and its variance were statistically significant, suggesting that autistic symptoms varied between children at baseline. The slope was also significant, which indicates that autistic symptoms significantly decreased linearly over time. The decrease represents about a quarter of a standard deviation for this variable. The slope variance was not significant, indicating that the autistic symptoms decreased at approximately the same rate for all children in the sample. The intercept and slope were not correlated in this model.

For all the variables linked to adaptive functioning, the final unconditional models (Model 6) indicate the intercepts and their variance were statistically significant. In other words, general adaptive functioning and the conceptual, social, and practical domains varied between children at baseline. Their slope 1 were also significant and suggest that the four variables increased during the intervention period (i.e., T1 to T2). General adaptive functioning and the conceptual and social domains increased for about a third of the standard deviation for those variables, while the practical domain increased for about a fifth of its standard deviation. All slope 1 variances were not significant, meaning that, on average, children progressed at the same rate. For all four variables, their slope 2 were not significant, indicating that, on average, general adaptive

functioning and the conceptual, social, and practical domains remained stable during the period following the intervention (i.e., T2 to T3). For the practical domain, the slope 2 variance was significant, which merely suggests that there was variability between children in the stable level of practical scores between during the follow-up period. Looking at the correlations between the intercepts and the slopes, the only significant correlation is between the intercept and the slope 2 for the conceptual domain. Since slope 2 was not significant (stability over time), this correlation merely suggests that conceptual level at baseline was significantly correlated with conceptual level during the period following the intervention.

### ***Predictors of Growth Parameters***

Table 4 presents the coefficients for the predictive relations estimated from the conditional models (Model 7). Each coefficient represents the simple effect of a given predictor, holding all other variables constant. For autistic symptoms, general adaptive functioning and age at baseline negatively predicted the intercept, meaning that younger children and children with lower general adaptive functioning also had more severe autistic symptoms at baseline. Two other predictors almost reached the typical criteria for statistical significance. First, program intensity was negatively related to the slope, which suggests that autistic symptoms scores tended to decrease more slowly for children who receive less intense intervention. Second, general language composite positively predicted the slope, implying that children with higher general language composite scores at baseline tend to show more rapid decreases in autistic symptoms over time.

For general adaptive functioning, autistic symptoms negatively predicted the intercept and slope 1, meaning that children with higher autistic symptoms tended to have lower general adaptive functioning scores at T1 and to show smaller increases in general adaptive functioning during the intervention period. Program intensity marginally predicted the intercept. The

association is negative, which is not surprising considering that program intensity was determined based on the evaluation of the children's needs at enrollment in the services. Thus, children who were assigned low-intensity intervention were more likely to have higher general adaptive functioning at baseline. Age at enrollment negatively predicted slope 2, which was not significant, implying that younger children tended to have higher stable general adaptive functioning levels during the period following the intervention. Performance IQ also marginally predicted slope 2, which merely suggests that children with higher performance IQ were more likely to maintain their general adaptive functioning level between T2 and T3.

For the conceptual domain of adaptive functioning, autistic symptoms negatively predicted the intercept. Thus, children with higher autistic symptoms were also more likely to have lower conceptual scores at T1. Family income, performance IQ and general language composite positively predicted the intercept. These results suggest that children in families with higher income, higher performance IQ and general language composite were also more likely to have higher conceptual functioning scores at T1. Autistic symptoms also negatively predicted slope 1, meaning that children with higher autistic symptoms at baseline tended to show slower increases in conceptual scores during the intervention period. Age at enrollment negatively predicted slope 2, indicating that younger children tended to have higher stable conceptual levels during the period following the intervention. For the social domain of adaptive functioning, autistic symptoms negatively predicted the intercept, implying that children with higher autistic symptoms also had lower social scores at T1. General language composite negatively predicted slope 2, suggesting that children with lower general language composite at baseline tended to maintain their higher stable social level during the period following the intervention.

Finally, autistic symptoms negatively predicted the intercept and slope 1 for the practical domain, meaning that children with higher autistic symptoms also had lower practical

functioning at T1. Also, the participants made less improvement regarding the practical functioning during the intervention period. Age at enrolment negatively predicted slope 2, which is not significant, which again suggest that younger children tended to show higher stable practical level during the period following the intervention. Lastly, performance IQ predicted slope 2, implying that children with higher performance IQ at baseline also tended to have higher stable practical level during the period following the intervention.

### **Discussion**

Each of our initial objectives produced results that contribute to our knowledge base on early behavioral intervention. All the variables linked to adaptive functioning (general adaptive functioning, conceptual domain, social domain, practical domain) increased during the intervention period (i.e., between T1 and T2), but became stable during the period following the intervention (i.e., between T2 and T3). At this point, it should be noted that the majority of the participants had entered school at the moment of T3. The increase in scores during the intervention period, followed by stability during the period after the intervention, implies that when we interrupt the intervention, the children stop progressing, but maintain their gains. This observation is consistent with a previous finding indicating that EBI has positive effect on adaptive functioning (Reichow et al.,2018). Looking at the three domains of adaptive functioning, our results showed that scores on the practical domain increased at a slower rate during the intervention than the conceptual and social domains. Another peculiarity of the practical domain was that slope 2 was not significant (no change during the follow-up period), but its variance was, which merely suggests that there was variability between children in the level of practical domain during the period following the intervention. Because the latent variable slope is an average, a nonsignificant slope with a significant variance may imply that the practical



domain score increased for some children while decreasing for others. Subsequent studies should use mixture modeling to help uncover this possible phenomenon.

We observed small but steady changes in autistic symptoms across all three time points. As there is no significant difference in the rate of change between the intervention period and the period following the intervention, it is unclear whether the intervention is related to the decrease in autistic symptoms. Nevertheless, our results suggest that adaptive functioning may improve even when the severity of autistic symptoms decreased marginally. This result is consistent with a recent meta-analysis suggesting that autistic symptoms are notably stable over time across childhood and that intervention studies should also consider improvement in adaptive functioning (Bieleninik et al., 2017). We chose to evaluate the effectiveness of EBI using scores on standardized measures. Some authors argue that standardized scores may underestimate the individual progress for some children receiving the intervention (see Klintwall et al., 2015). Those authors have shown that even small improvements on standardized scores may narrow the gap between children with autism and their typically developing peers when looking at age-equivalents rather than standardized scores (Klintwall et al., 2015). As such, the CARS scores may not be the best measure to assess intervention effectiveness on autistic symptoms.

Regarding predictors of effectiveness, various variables were associated with progress during the intervention and maintenance during the follow-up periods. Autistic symptoms decreased more slowly for children who received less intensive intervention, and more rapidly for those who had a higher general language composite at baseline. As program intensity was determined by the evaluation of each child's needs, this association may be the result of attrition to the mean. General adaptive functioning tended to increase more slowly for children with higher autistic symptoms. Younger children and children with higher performance IQ were more likely to maintain their gains in general adaptive functioning. Conceptual domain scores

increased more slowly for children with higher autistic symptoms, and younger children tended to have higher stable conceptual level during the period following the intervention. No variable was associated with increase the in social domain during the intervention period, but children with lower general language composite scores maintained their high stable social level during the period following the intervention. Practical domain scores increased more slowly for children with higher autistic symptoms. Younger children and children with higher performance IQ were more likely to show higher stable practical level during the period following the intervention.

Despite a marginally significant association with reductions in autistic symptoms, intervention intensity did not predict improvements in adaptive functioning (i.e., general adaptive functioning, conceptual domain, social domain, practical domain). One potential explanation is that both intervention options were far below the 40 hr per week Lovaas suggested in his original work (Lovaas, 1981). Therefore, optimal progress may have been more difficult to achieve. Another potential explanation lies in the study design. As we used a correlational design, we did not have a high level of control over each variable. The influence of intervention intensity may not have been detectable. One possible variable that may have interacted with the intervention intensity is intervention fidelity, as suggested by Pellechia et al. (2019). The intervention was individualized according to an initial needs assessment. Children with more severe symptoms of autism received more intensive intervention, while children with less severe symptoms received less intensive intervention. The non-random group attribution between the low-intensity and the moderate-intensity intervention limits the inferences we can make about the influence of intervention intensity on the effectiveness of EBI.

The main limitation of this study is the absence of a control group. We used a correlational design, with only one time point before intervention implementation. Such design limits the causal inferences between the intervention and the observed change, and do not allow

to control for maturation effects. Lack of control group is common in early intervention research, as many consider it would be unethical to assign children with a neurodevelopmental condition to a control group for a long time during a critical developmental period (Matson, 2007). Another limitation concerns the raters assessing outcomes (i.e., adaptive functioning and autistic symptoms), who were the special education technicians implementing the intervention and the parents, which could have induced bias. Future research should opt for more rigorous group designs, such as a regression discontinuity design, waiting list design or randomized control trials and, blind raters to assess intervention outcomes to better demonstrate the impact of the intervention (Steinbrenner et al., 2020; Shadish et al., 2002). Another limitation relates to the absence of a measure of implementation fidelity and of the quality of the supervisions offered to the special education technicians. Those critical aspects can influence intervention effectiveness (DiGennaro et al., 2007; Durlak et DuPre, 2008), especially because the training of the special education technicians who implemented the intervention differed from the training of technicians in other countries. For example, errors in the integrity of the treatment, especially in the way of providing reinforcement, would influence the effectiveness of the intervention (Bottini et al., 2020). Likewise, the quality of supervision received by special education technicians would influence their implementation of the intervention (Davis et al., 2002). Beyond the characteristics of the children, implementation fidelity may be partly responsible for the effects of the intervention (Klintwall et al., 2015). Future large-scale community-based studies should collect detailed data about intervention strategies and implementation fidelity so that results could have practical implications on how to make the best intervention for children with autism.

Our sample size was not ideal as larger sample sizes produce more trustable estimates in the structural equation framework (Little, 2013). Nonetheless, simulation studies evaluating the reduction of standard errors for sample size between 40 and 500 suggest that between 40 and 100,

the standard errors decrease quickly, while the rate of error reduction transitions from rapid to slow between 100 to 150 (Little, 2013). Finally, we had high attrition during the follow-up period. Nonetheless, growth models can be estimated in the presence of partially missing data when data are missing completely at random or missing at random (Curran et al., 2010). Our verifications indicated that the data in our sample were missing at random and that the use of maximum likelihood robust estimator was adequate and allowed us to estimate the models despite the presence of large attrition (Enders, 2010; Little, 2013). Future research should opt for group designs with larger sample sizes and use a retention strategy to prevent high attrition.

Our study contributes to the knowledge base on the effectiveness of EBI. The results revealed nonlinear changes in adaptive functioning characterized by significant improvement during the intervention period and a small linear decrease in autistic symptoms from baseline to follow-up. In addition, intensity of intervention, age at enrolment, IQ and autistic symptoms were either associated with progress during the intervention period or maintenance during the follow-up period. Taken together, these results underline the importance of conducting further replications in community settings. In terms of methodological contribution, this study is one of the few to use more contemporary statistical analyses, which have many advantages over more traditional analyses to assess intervention effectiveness. Our results could thus encourage other researchers to integrate these analyses in their future work.

### **Compliance with Ethical Standards**

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**Ethical Approval:** All procedures performed in this study were in accordance with the ethical standards of [removed for blind review] and with the 1964 Helsinki declaration and its later amendments.

**Informed Consent:** Parents provided informed consent for them and their child.

**Conflict of Interest:** The authors have no conflict of interest to report.

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**Tableau 1***Descriptive Statistics*

| Variable          | <i>N</i> | Min | Max   | M     | SD    | Skewness | Kurtosis |
|-------------------|----------|-----|-------|-------|-------|----------|----------|
| GAC T1            | 229      | 41  | 130   | 64.94 | 14.83 | 0.89     | 1.55     |
| GAC T2            | 219      | 40  | 125   | 70.31 | 18.06 | 0.18     | -0.64    |
| GAC T3            | 64       | 40  | 113   | 70.87 | 18.43 | 0.47     | -0.36    |
| CON T1            | 229      | 45  | 123   | 68.51 | 15.11 | 0.50     | 0.08     |
| CON T2            | 219      | 45  | 121   | 74.78 | 17.41 | 0.01     | -0.87    |
| CON T3            | 64       | 45  | 109   | 74.69 | 16.40 | 0.01     | -0.73    |
| SOC T1            | 229      | 48  | 130   | 70.65 | 16.58 | 0.55     | -0.02    |
| SOC T2            | 219      | 48  | 121   | 75.78 | 18.67 | 0.05     | -0.92    |
| SOC T3            | 64       | 48  | 125   | 78.05 | 17.74 | 0.48     | -0.05    |
| PRA T1            | 229      | 41  | 129   | 65.55 | 14.20 | 0.63     | 1.40     |
| PRA T2            | 29       | 41  | 126   | 67.92 | 16.36 | 0.23     | -0.10    |
| PRA T3            | 64       | 40  | 116   | 68.59 | 19.90 | 0.52     | -0.67    |
| AS T1             | 226      | 17  | 54.75 | 31.25 | 7.77  | 0.36     | -0.47    |
| AS T2             | 152      | 17  | 55    | 29.58 | 7.48  | 0.91     | 1.15     |
| AS T3             | 56       | 15  | 49    | 26.86 | 8.04  | 0.67     | -0.29    |
| Program Intensity | 230      | 0   | 1     | 0.46  | 0.50  | 0.16     | -1.99    |
| PIQ               | 224      | 47  | 130   | 80.08 | 20.65 | 0.18     | -1.00    |
| VIQ               | 223      | 48  | 122   | 72.43 | 17.28 | 0.58     | -0.62    |
| GLC               | 224      | 47  | 117   | 74.29 | 19.33 | 0.15     | -1.07    |
| Annual Income     | 227      | 1   | 5     | 2.91  | 1.45  | 0.15     | -1.07    |
| Age at T1         | 225      | 2.5 | 5.75  | 4.34  | 0.47  | -0.83    | 2.02     |

*Note.* GAC = General Adaptive Composite; CON = Conceptual Domain; SOC = Social Domain; PRA = Practical Domain; AS = Autistic Symptoms; PIQ = Performance Intellectual Quotient; VIQ = Verbal Intellectual Quotient; GLC = Global Language Composite; T1 = Time 1, T2 = Time 2; T3 = Time 3.

**Table 2***Model Fit Indices for the Latent Growth Curve Models*

| Models                               | Mod. Ref. | $\chi^2$      | df | RMSEA | [90%CI]      | SRMR | CFI   | TLI   | AIC  | BIC  | SABIC | $\Delta S\chi^2$ | $\Delta$ dl | $\Delta$ CFI | $\Delta$ RMSEA |  |
|--------------------------------------|-----------|---------------|----|-------|--------------|------|-------|-------|------|------|-------|------------------|-------------|--------------|----------------|--|
| <b>Severity of Autistic Symptoms</b> |           |               |    |       |              |      |       |       |      |      |       |                  |             |              |                |  |
| Model 4                              | 3         | 2.572         | 3  | .000  | [.000, .105] | .073 | 1.000 | 1.003 | 2837 | 2857 | 2838  | +2.384           | 2           | 0            | 0              |  |
| Model 5                              | 4         | 2.041         | 4  | .000  | [.000, .072] | .104 | 1.000 | 1.010 | 2834 | 2851 | 2835  | -                | -           | 0            | 0              |  |
| Model 6                              | -         | -             | -  | -     | -            | -    | -     | -     | -    | -    | -     | -                | -           | -            | -              |  |
| Model 7                              | 4         | 15.099        | 12 | .036  | [.000, .084] | .048 | .990  | .979  | 2355 | 2415 | 2358  | -                | -           | -.021        | -.036          |  |
| <b>General Adaptive Functioning</b>  |           |               |    |       |              |      |       |       |      |      |       |                  |             |              |                |  |
| Model 4                              | 3         | 38.046**<br>* | 3  | .224  | [.164, .291] | .128 | .859  | .859  | 4111 | 4132 | 4113  | +21.383***       | 1           | -.081        | +0.044         |  |
| Model 5                              | 3         | 2.067         | 1  | .068  | [.000, .202] | .096 | .996  | .987  | 4086 | 4113 | 4088  | -12.953***       | 1           | +0.056       | -.112          |  |
| Model 6                              | 5         | -             | 0  | 0     | [-]          | .000 | 1.000 | 1.000 | 4086 | 4117 | 4089  | -                | -           | +0.004       | -.068          |  |
| Model 7                              | 6         | -             | 0  | 0     | [-]          | .000 | 1.000 | 1.000 | 3385 | 3484 | 3389  | -                | -           | .000         | .000           |  |
| <b>Conceptual Domain</b>             |           |               |    |       |              |      |       |       |      |      |       |                  |             |              |                |  |
| Model 4                              | 3         | 26.891**<br>* | 3  | 0.185 | [.125, .252] | .104 | .884  | .884  | 4095 | 4115 | 4096  | -.048            | 1           | +0.011       | -.052          |  |
| Model 5                              | 4         | 10.986**<br>* | 2  | 0.139 | [.067, .224] | .152 | .956  | .934  | 4079 | 4103 | 4081  | -11.972          | 1           | +0.050       | -.046          |  |
| Model 6                              | 5         | -             | -  | 0     | [.000, .000] | .000 | 1.000 | 1.00  | 4076 | 4107 | 4079  | -                | -           | +0.044       | -.139          |  |
| Model 7                              | 6         | -             | -  | 0     | [.000, .000] | .000 | 1.000 | 1.00  | 3366 | 3466 | 3371  | -                | -           | .000         | .000           |  |
| <b>Social Domain</b>                 |           |               |    |       |              |      |       |       |      |      |       |                  |             |              |                |  |
| Model 4                              | 3         | 6.804         | 3  | .074  | [.000, .149] | .052 | .973  | .973  | 4184 | 4205 | 4186  | -.023            | 1           | +0.013       | -.036          |  |
| Model 5                              | 4         | 2.080         | 4  | .000  | [.000, .072] | .136 | 1.000 | 1.010 | 4175 | 4193 | 4177  | -                | -           | +0.027       | -.074          |  |
| Model 6                              | 5         | -             | 0  | .000  | [.000, .000] | .000 | 1.000 | 1.000 | 4181 | 4212 | 4183  | -                | -           | 0            | 0              |  |
| Model 7                              | 6         | -             | -  | .000  | [.000, .000] | .000 | 1.000 | 1.000 | 3515 | 3614 | 3519  | -                | -           | 0            | 0              |  |
| <b>Practical Domain</b>              |           |               |    |       |              |      |       |       |      |      |       |                  |             |              |                |  |
| Model 4                              | 3         | 4.203         | 3  | .042  | [.000, .125] | .067 | .995  | .995  | 4042 | 4063 | 4044  | +270             | 1           | -.001        | -.004          |  |
| Model 5                              | 4         | 5.019         | 2  | .081  | [.000, .172] | .068 | .988  | .981  | 4044 | 4068 | 4046  | +0.001           | 1           | -.007        | +0.039         |  |
| Model 6                              | 5         | -             | 0  | .000  | [.000, .000] | .000 | 1.000 | 1.000 | 4044 | 4075 | 4047  | -                | -           | +0.012       | -.081          |  |
| Model 7                              | 6         | -             | 0  | .000  | [.000, .000] | .000 | 1.000 | 1.000 | 3395 | 3495 | 3400  | -                | -           | .000         | .000           |  |

Note.  $\chi^2$  = chi square; df = degrees of freedom; CFI = Comparative Fit Index; TLI = Tucker-Lewis Index; RMSEA = Root Mean Square Error of Approximation; 90% CI = 90% Confidence Interval of the RMSEA; SRMR = Standardized Root Mean Square Error of Approximation; Ref = Reference Model;  $\Delta S\chi^2$  = Satorra-Bentler Scaled chi-square difference test;  $\Delta$ df = change in degrees of freedom;  $\Delta$ CFI = change in CFI;  $\Delta$ RMSEA = change in RMSEA;

<sup>a</sup> p<.06. \* p<.05. \*\* p < .01. \*\*\*p ≤.001

**Table 3***Growth Parameters for the Final Selected Models*

| Models                                | Intercept                     |            | Slope 1                       |          | Slope 2                   |          | Correlation<br>Intercept<br>/Slope 1 | Correlation<br>Intercept<br>/Slope 2 |
|---------------------------------------|-------------------------------|------------|-------------------------------|----------|---------------------------|----------|--------------------------------------|--------------------------------------|
|                                       | Mean<br>[95%CI]               | Variance   | Mean<br>[95%CI]               | Variance | Mean<br>[95%CI]           | Variance |                                      |                                      |
| <b>Autistic<br/>Symptoms</b>          | 31.189***<br>[30.199, 32.179] | 45.616***  | -1.877***<br>[-2.462, -1.291] | 0.131    | -                         | -        | .625                                 | -                                    |
| <b>General Adaptive<br/>Composite</b> | 64.891***<br>[62.985, 66.796] | 203.603*** | 6.192***<br>[4.657, 7.728]    | 29.861   | -0.726<br>[-3.625, 2.173] | 19.144   | .168                                 | -.427                                |
| <b>Conceptual<br/>Domain</b>          | 68.480***<br>[66.531, 70.428] | 196.284*** | 6.960***<br>[5.469, 8.451]    | 25.776   | -1.401<br>[-4.160, 1.358] | 36.127   | .192                                 | -.605**                              |
| <b>Social Domain</b>                  | 70.580***<br>[68.449, 72.710] | 229.188*** | 5.979***<br>[4.302, 7.656]    | 2.374    | 0.240<br>[-2.837, 3.316]  | 19.652   | .374                                 | -.624                                |
| <b>Practical Domain</b>               | 65.499***<br>[63.675, 67.322] | 178.176*** | 2.985***<br>[1.574, 4.395]    | 35.070   | -0.079<br>[-3.334, 3.175] | 87.492** | .066                                 | .018                                 |

Note. CI = Confidence intervals

\* p<.05. \*\* p < .01. \*\*\*p ≤.001

**Table 4***Predictors of the Growth Parameters for the Final Selected Models*

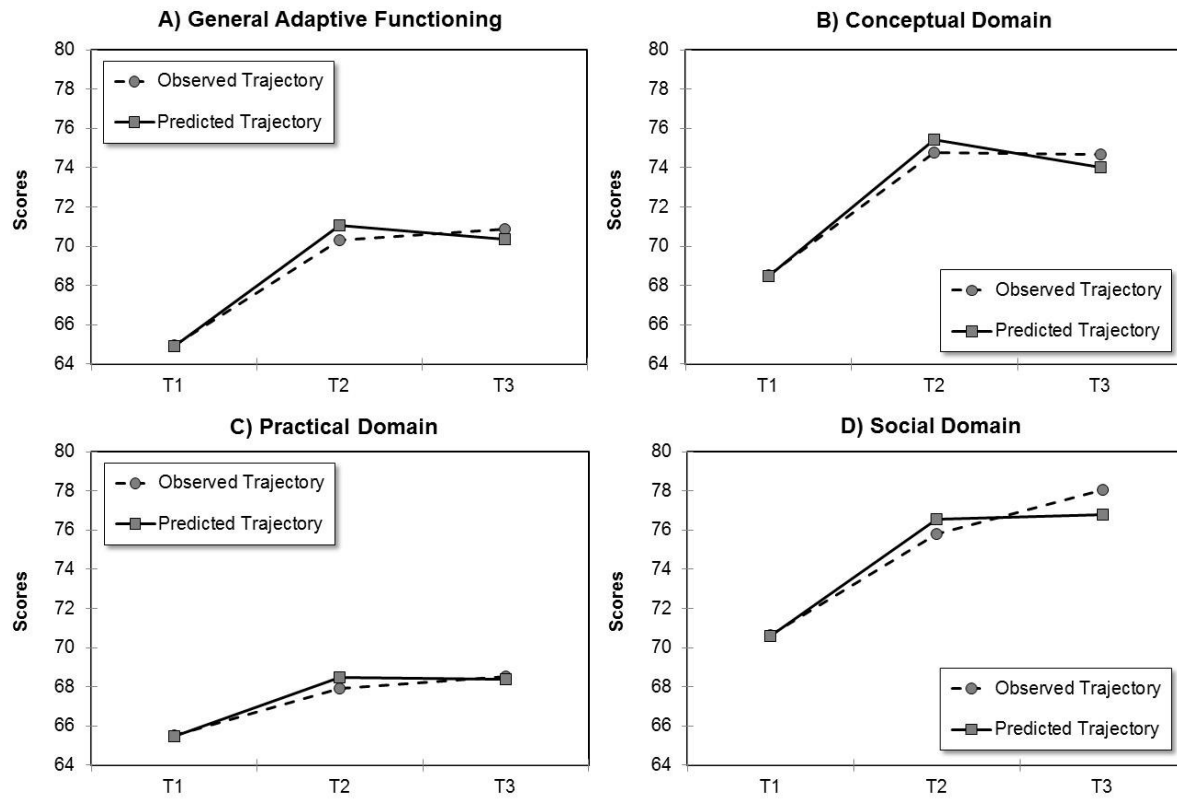
|                                   | Intercept          |                | Slope 1            |                | Slope 2           |                 |
|-----------------------------------|--------------------|----------------|--------------------|----------------|-------------------|-----------------|
|                                   | Estimate           | [95%CI]        | Estimate           | [95%CI]        | Estimate          | [95%CI]         |
| <b>Autistic Symptoms</b>          |                    |                |                    |                |                   |                 |
| GACT1                             | -.584***           | [-.728, -.441] | -.042              | [-.835, .750]  | -                 |                 |
| Age T1                            | -.199***           | [-.331, -.087] | -.263              | [-.975, .449]  | -                 |                 |
| Income                            | -.055              | [-.176, .065]  | .247               | [-.678, 1.172] | -                 |                 |
| Intensity                         | .082               | [-.052, .217]  | -.716 <sup>a</sup> | [-1.457, .025] | -                 |                 |
| .PIQ                              | -.101              | [-.258, .055]  | -.841              | [-1.182, .130] | -                 |                 |
| GLC                               | -.102              | [-.383, .179]  | 1.358 <sup>a</sup> | [-.051, 2.768] | -                 |                 |
| VIQ                               | .035               | [-.215, .285]  | -1.069             | [-2.564, .426] | -                 |                 |
| <b>General Adaptive Composite</b> |                    |                |                    |                |                   |                 |
| AST1                              | -.499***           | [-.600, -.398] | -.366**            | [-.638, -.095] | .136              | [-.466, .738]   |
| Age T1                            | -.028              | [-.114, 0.57]  | .052               | [-.152, .256]  | -.431**           | [-.759, -.104]  |
| Income                            | .084               | [-.012, .179]  | -.137              | [-.336, .061]  | -.105             | [-.508, .299]   |
| Intensity                         | -.085 <sup>a</sup> | [-.172, .003]  | .139               | [-.076, .355]  | .068              | [-.351, .486]   |
| PIQ                               | .105               | [-.040, .250]  | -.008              | [-.318, .301]  | .575 <sup>a</sup> | [-.015, 1.165]  |
| GLC                               | .082               | [-.190, .354]  | .285               | [-.157, .728]  | -.672             | [-1.544, .200]  |
| VIQ                               | .148               | [-.085, .382]  | .044               | [-.411, .499]  | -.672             | [-.490, 1.077]  |
| <b>Conceptual Domain</b>          |                    |                |                    |                |                   |                 |
| AST1                              | -.381***           | [-.494, -.268] | -.288*             | [-.569, -.006] | .070              | [-.600, .740]   |
| Age T1                            | -.005              | [-.092, .083]  | -.008              | [-.248, .233]  | -.421**           | [-.730, -.112]  |
| Income                            | .118*              | [.022, .215]   | -.088              | [-.308, .132]  | -.180             | [-.558, .199]   |
| Intensity                         | -.065              | [-.170, .040]  | .193               | [-.050, .436]  | -.023             | [-.447, .401]   |
| PIQ                               | .176*              | [.036, .317]   | -.002              | [-.324, .321]  | .207              | [-.403, .816]   |
| GLC                               | .319*              | [.020, .618]   | .085               | [-.387, .556]  | -.465             | [-1.332, .392]  |
| VIQ                               | .008               | [-.241, .258]  | .266               | [-.231, .763]  | .184              | [-.543, .912]   |
| <b>Social Domain</b>              |                    |                |                    |                |                   |                 |
| AST1                              | -.603***           | [-.718, -.489] | -.276              | [-.865, .314]  | .008              | [-.695, .711]   |
| Age T1                            | -.068              | [-.166, .030]  | .155               | [-.255, .565]  | -.205             | [-.665, .256]   |
| Income                            | .007               | [-.104, .118]  | .323               | [-.903, .257]  | .073              | [-.410, .557]   |
| Intensity                         | -.084              | [-.191, .023]  | .302               | [-.237, .841]  | .174              | [-.338, .687]   |
| PIQ                               | .068               | [-.093, .229]  | .042               | [-.569, .654]  | .475              | [-.295, 1.244]  |
| GLC                               | .090               | [-.170, .350]  | .396               | [-.625, 1.418] | -1.034*           | [-2.063, -.005] |
| VIQ                               | .112               | [-.135, .359]  | .056               | [-.809, .920]  | .346              | [-.610, 1.301]  |
| <b>Practical Domain</b>           |                    |                |                    |                |                   |                 |
| AST1                              | -.492***           | [-.607, -.377] | -.400*             | [-.758, -.043] | .168              | [-.365, .691]   |
| Age T1                            | -.011              | [-.116, .093]  | -.017              | [-.260, .227]  | -.385*            | [-.681, -.089]  |
| Income                            | .035               | [-.071, .140]  | -.048              | [-.271, .176]  | -.086             | [-.426, .255]   |
| Intensity                         | -.057              | [-.159, .045]  | .039               | [-.206, .284]  | .001              | [-.378, .380]   |
| PIQ                               | .089               | [-.066, .244]  | -.113              | [-.465, .238]  | .557*             | [.022, 1.092]   |
| GLC                               | .059               | [-.218, .336]  | .364               | [-.144, .871]  | -.239             | [-1.109, .630]  |
| VIQ                               | .188               | [-.054, .430]  | -.152              | [-.688, .384]  | .129              | [-.676, .935]   |

Note. The standardized estimates are reported. GAC = General Adaptive Composite; AS = Autistic Symptoms; PIQ = Performance Intellectual Quotient; VIQ = Verbal Intellectual Quotient; GLC = Global Language Composite; T1 = Time 1

<sup>a</sup> p<.06. \* p<.05. \*\*p < .01. \*\*\*p ≤.001.

**Figure 1**

*Mean Scores for the Observed and Estimated Latent Trajectories*





## Supplementary Material

**Table S1**

*Descriptive Statistics for the Two Intervention Intensity Options*

| Variables                    | Low Intensity |          |           | Moderate Intensity |          |           | <i>t-Test</i>       |
|------------------------------|---------------|----------|-----------|--------------------|----------|-----------|---------------------|
|                              | <i>N</i>      | <i>M</i> | <i>SD</i> | <i>N</i>           | <i>M</i> | <i>SD</i> | <i>p</i> (2-tailed) |
| Age                          | 120           | 4.39     | .47       | 102                | 4.30     | .44       | .151                |
| Annual Income                | 119           | 3.03     | 1.41      | 105                | 2.72     | 1.45      | .117                |
| Autistic Symptoms            | 121           | 28.30    | 6.93      | 102                | 34.56    | 7.39      | .000                |
| General adaptive Functioning | 122           | 70.87    | 15.27     | 104                | 58.12    | 11.02     | .000                |
| Verbal IQ                    | 118           | 80.11    | 19.98     | 102                | 63.54    | 12.79     | .000                |
| Performance IQ               | 118           | 88.21    | 17.68     | 103                | 70.83    | 19.77     | .000                |
| Global language Composite    | 118           | 82.69    | 17.01     | 100                | 64.45    | 17.26     | .000                |

**Table S2***Model Fit Indices for the Latent Growth Curve Models*

| Models                               | Mod. Ref. | $\chi^2$   | df | RMSEA | [90%CI]      | SRMR | CFI   | TLI   | AIC  | BIC  | SABIC | $\Delta S\chi^2$ | $\Delta$ dl | $\Delta$ CFI | $\Delta$ RMSEA |
|--------------------------------------|-----------|------------|----|-------|--------------|------|-------|-------|------|------|-------|------------------|-------------|--------------|----------------|
| <b>Severity of Autistic Symptoms</b> |           |            |    |       |              |      |       |       |      |      |       |                  |             |              |                |
| Model 0                              | -         | 186.972*** | 5  | .397  | [.349, .447] | .467 | .000  | .279  | 3022 | 3036 | 3023  | -                | -           | -            | -              |
| Model 1                              | 0         | 36.824***  | 4  | .188  | [.136, .246] | .151 | .783  | .837  | 2870 | 2888 | 2872  | -163.324***      | 1           | + .783       | -.209          |
| Model 2                              | 1         | 1.304      | 3  | 0     | [.000, .079] | .060 | 1.000 | 1.011 | 2835 | 2856 | 2837  | -32.856***       | 1           | + .217       | -.188          |
| Model 3                              | 2         | .199       | 1  | 0     | [.000, .134] | .009 | 1.000 | 1.016 | 2838 | 2866 | 2840  | -1.134           | 2           | 0            | 0              |
| <b>General Adaptive Functioning</b>  |           |            |    |       |              |      |       |       |      |      |       |                  |             |              |                |
| Model 0                              | -         | 325.011*** | 5  | .525  | [.478, .574] | .451 | .000  | .228  | 4347 | 4361 | 4348  | -                | -           | -            | -              |
| Model 1                              | 0         | 71.975***  | 4  | .271  | [.218, .327] | .125 | .727  | .795  | 4145 | 4163 | 4147  | -473.532***      | 1           | + .727       | -.254          |
| Model 2                              | 1         | 17.843***  | 3  | .146  | [.086, .215] | .054 | .940  | .940  | 4096 | 4117 | 4098  | -48.307***       | 1           | + .213       | -.125          |
| Model 3                              | 2         | 17.041***  | 2  | .180  | [.108, .263] | .050 | .940  | .909  | 4096 | 4121 | 4098  | -1.721           | 1           | 0            | + .034         |
| <b>Conceptual Domain</b>             |           |            |    |       |              |      |       |       |      |      |       |                  |             |              |                |
| Model 0                              | -         | 278.671*** | 5  | .486  | [.438, .535] | .440 | .000  | .200  | 4330 | 4344 | 4331  | -                | -           | -            | -              |
| Model 1                              | 0         | 75.013***  | 4  | .277  | [.224, .333] | .167 | .654  | .741  | 4150 | 4167 | 4151  | -439.048***      | 1           | + .654       | -.209          |
| Model 2                              | 1         | 27.512***  | 3  | .188  | [.127, .255] | .156 | .881  | .881  | 4096 | 4117 | 4098  | -36.001***       | 1           | + .227       | -.039          |
| Model 3                              | 2         | 28.038***  | 2  | .237  | [.164, .318] | .117 | .873  | .810  | 4097 | 4121 | 4099  | +0.911           | 1           | + .008       | + .049         |
| <b>Social Domain</b>                 |           |            |    |       |              |      |       |       |      |      |       |                  |             |              |                |
| Model 0                              | -         | 210.160*** | 5  | .421  | [.373, .470] | .429 | .000  | .137  | 4408 | 4422 | 4409  | -                | -           | -            | -              |
| Model 1                              | 0         | 55.682***  | 4  | .236  | [.183, .293] | .204 | .638  | .728  | 4225 | 4243 | 4227  | -97.490***       | 1           | + .638       | -.185          |
| Model 2                              | 1         | 9.077*     | 3  | .093  | [.027, .166] | .115 | .957  | .957  | 4185 | 4206 | 4187  | -92.031***       | 1           | + .319       | -.143          |
| Model 3                              | 2         | 7.661*     | 2  | .110  | [.036, .198] | .061 | .960  | .940  | 4187 | 4211 | 4189  | -0.797           | 1           | + .003       | + .007         |
| <b>Practical Domain</b>              |           |            |    |       |              |      |       |       |      |      |       |                  |             |              |                |
| Model 0                              | -         | 310.484*** | 5  | .513  | [.466, .562] | .424 | .000  | .250  | 4283 | 4297 | 4284  | -                | -           | -            | -              |
| Model 1                              | 0         | 24.862***  | 4  | .150  | [.097, .209] | .055 | .915  | .936  | 4060 | 4077 | 4061  | -634.900***      | 1           | + .915       | -.363          |
| Model 2                              | 1         | 6.636      | 3  | .072  | [.000, .095] | .088 | .985  | .985  | 4045 | 4066 | 4047  | -18.461***       | 1           | -.070        | -.078          |
| Model 3                              | 2         | 2.970      | 2  | .046  | [.000, .146] | .028 | .996  | .994  | 4043 | 4068 | 4045  | -3.297           | 1           | + .011       | -.026          |

Note.  $\chi^2$  = chi square; df = degrees of freedom; CFI = Comparative Fit Index; TLI = Tucker-Lewis Index; RMSEA = Root Mean Square Error of Approximation; 90% CI = 90% Confidence Interval of the RMSEA; SRMR = Standardized Root Mean Square Error of Approximation; Ref = Reference Model;  $\Delta S\chi^2$  = Satorra-Bentler Scaled chi-square difference test;  $\Delta$ df = change in degrees of freedom;  $\Delta$ CFI = change in CFI;  $\Delta$ RMSEA = change in RMSEA;

<sup>a</sup> p<.06. \* p<.05. \*\* p < .01. \*\*\*p ≤ .001

**Table S3***Residual Variances for Model 6 (Piecewise Growth Model)*

|                              | Time 1 | Time 2 | Time 3 |
|------------------------------|--------|--------|--------|
| General Adaptive Functioning | 15.00  | 85.83  | 85.83  |
| Conceptual Domain            | 32.00  | 66.80  | 66.80  |
| Social Domain                | 44.00  | 112.31 | 112.31 |
| Practical Domain             | 22.00  | 54.18  | 54.18  |

**Chapitre III – Article 2**

**A Person-Centered Perspective on Differential Efficacy of Early Behavioral Intervention in  
Children with Autism: A Latent Profile Analysis**

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## Résumé

**Contexte :** Les personnes ayant un trouble du spectre de l'autisme (TSA) présentent des symptômes et une réponse à l'intervention hétérogènes. L'intervention comportementale intensive (ICI) est un exemple d'intervention qui produit des effets variables. S'intéresser aux différences individuelles et à l'identification de sous-groupes plus homogènes dans les échantillons pourrait aider à mieux comprendre l'hétérogénéité des symptômes de TSA et de la réponse à l'ICI.

**Méthode :** Adoptant une perspective centrée sur la personne, nous avons mené des analyses de profils latents pour explorer la présence de sous-groupes homogènes dans notre échantillon de 233 enfants d'âge préscolaire ayant une TSA recevant de l'ICI. Nous avons étudié les prédicteurs de l'appartenance aux profils à l'aide de régressions logistiques multinomiales et les conséquences de l'appartenance aux profils avec l'approche BCH.

**Résultats :** Nous avons identifié quatre profils : un profil de manifestations légères, un profil de manifestations sévères et deux profils intermédiaires ayant des combinaisons de manifestations légères à modérées sur le plan des symptômes autistiques, du fonctionnement adaptatif et du QI. Seul le revenu familial annuel prédisait les profils. Tous les profils ont progressé pendant la période d'intervention, avec des changements d'ampleur variable. Pendant la période de suivi, le fonctionnement adaptatif des deux profils ayant les manifestations les plus sévères est resté stable ou s'est amélioré, tandis qu'il a diminué pour les profils ayant les manifestations plus légères.

**Conclusions :** Notre étude contribue à la littérature en suggérant la présence de profils distincts avec des différences dans leur réponse à l'ICI. Les profils associés à de meilleurs résultats à court terme étaient différents des profils qui maintiennent davantage leurs gains. Cette constatation peut guider à la fois les praticiens et les chercheurs dans l'évaluation des effets de l'intervention.

*Mots-clés* : analyse de profils latents, approche centrée sur la personne, intervention comportementale intensive, hétérogénéité, réponse différentielle, trouble du spectre de l'autisme

## Abstract

**Background:** Individuals with autism spectrum disorders (ASD) present heterogeneous symptom manifestations and responses to intervention. Despite being well-established, early intensive behavioral intervention (EIBI) is an example of intervention that has produced inconsistent responding across studies. Investigating individual differences and identifying more homogenous subgroups in samples may lead to a better understanding of symptom heterogeneity in ASD and response to EIBI.

**Method:** Adopting a person-centered perspective, we conducted latent profile analyses (LPA) to explore the presence of homogenous subgroups in our sample of 233 preschoolers with ASD receiving early behavioral intervention services. We investigated predictors of group membership using logistic multinomial regressions and outcomes of membership with the BCH approach.

**Results:** We found four latent profiles in our sample: a mild impairment profile, a severe impairment profile, and two intermediate profiles with combinations of mild to moderate autistic symptoms, adaptive functioning, and intellectual functioning. Only the annual family income predicted profile membership. All profiles made progress during the intervention period, with varying magnitudes of change. During the follow-up period, the moderate impairment and the severe impairment profiles showed stability or improvement in adaptive functioning, while the two mild impairment profiles showed a slight decrease.

**Conclusions:** Our study contributes to the literature by suggesting the presence of distinct profiles with differences in their response to EIBI. The profiles associated with better short-term outcomes were different than the profiles who maintain their gains more consistently over time. This finding may guide both practitioners and researchers assessing the effects of intervention.

*Keywords:* autism spectrum disorders, differential response, EIBI, heterogeneity, latent profile analysis, person-centered.



## **A Person-Centered Perspective on Differential Efficacy of Early Behavioral Intervention in Children with Autism: A Latent Profile Analysis**

According to the Diagnostic and statistical manual of mental disorders – 5<sup>th</sup> edition (DSM-5), autism spectrum disorder (ASD) is a neurodevelopmental condition that is characterized by persistent deficits in social communication and interaction across various contexts, and by the presence of restricted, repetitive patterns of behaviors and interests (American Psychiatric Association, 2013). In addition, persons with ASD differ in terms of intellectual functioning (Wiggins et al., 2012), patterns of cognitive strengths and weaknesses (Munson et al., 2008), and levels of adaptive functioning (Ray-Subramanian et al., 2011; Szatmari et al., 2002). On one end of the autism spectrum, individuals have mild difficulties, occasional needs, and function with a low level of support, while on the other end of the spectrum, individuals experience serious difficulties that affect many areas of activities and require significant and ongoing support. Without intervention, manifestations of ASD remain stable across the lifetime of the majority of diagnosed individuals (Bieleninik et al., 2017).

Besides heterogeneity in symptom presentation, individuals with ASD tend to respond differently to intervention (Sherer & Schreibman, 2005). Even though early intensive behavioral intervention (EIBI) is considered one of the most effective interventions for children with ASD (Health Technology Inquiry Service, 2008; INESSS, 2014; Maglione et al., 2012; National Autism Center, 2009; National Institute for Health and Care Excellence, 2013; Prior & Roberts, 2012), some children only make modest, or even no improvement (Reichow et al., 2018). Such variability in response to treatment complicates attempts at predicting clinical outcomes, and individualizing treatment targets and strategies (Kim et al., 2016; Masi et al., 2017; Zheng et al., 2020). Heterogeneity in outcomes continues to be poorly understood and the characteristics of

children that may influence efficacy of EIBI are still debated (Eapen et al., 2013; Reichow et al., 2018).

Studies examining predictors of EIBI outcomes have produced conflicting results on some individual characteristics, such as age at enrollment (Bieleninik et al., 2017; Makrygianni & Reed, 2010), intellectual functioning (Makrygianni & Reed, 2010; Reed, 2016; Tiura et al., 2017), and autistic symptoms (Flanagan et al., 2012; Reed, 2016). The influence of adaptive functioning is more consensual, and numerous studies support that high adaptive functioning in children is associated with improved efficacy of EIBI (Eldevik et al., 2010; Flanagan et al., 2012; Reed, 2016; Reichow, 2012; Sallows & Graupner, 2005; Vivanti et al., 2014). A possible explanation for the disagreement concerning the potential predictors of efficacy is that combination of variables (i.e., the profiles of the children) may be more important than the individual contribution of these characteristics considered in isolation. Several researchers recommend accounting for individual differences and identifying more homogenous subgroups in samples to better understand symptom heterogeneity in ASD and response to EIBI across children (Eapen et al., 2013; Georgiades et al., 2013; Tiura et al., 2017).

A person-centered perspective could guide the identification of more homogenous subgroups in the population of autistic children. Contrary to the variable-centered approach (the most commonly used in social and psychological sciences) that investigates the relationships between variables, the person-centered approach explores the relationships within individuals and aims to group individuals into subgroups (Bergman & Trost, 2006; Laursen & Hoff, 2006). In the person-centered approach, the associations between the variables may therefore be different depending on the groups of individuals identified within a sample. Adopting a person-centered approach may help inform evaluative research in ASD, as one of its advantages is that the

generalization of findings applies to persons and not variables (Magnusson, 1998). Hence, the results of person-centered studies are likely to translate to clinical applicability.

In this context, a crucial question is on what basis should we identify the subgroups when considering the autistic population? Previous studies have adopted two different approaches. Some researchers have attempted to identify subgroups relying solely on autistic symptoms. For example, Wiggins et al. (2012) investigated subgroups based on the Childhood Autism Rating Scale (Schopler et al., 1980), and Georgiades et al. (2013) used the Autism Diagnostic Interview Revised (Rutter et al., 2003). Both studies found three subgroups in their sample of autistic children, with varying severity of social communication deficits, and fixated interests and repetitive behaviors. The purpose of these studies was related to the conceptualisation of the diagnostic criteria for ASD. Some authors have pointed out that focusing only on measures designed to screen for diagnosis of ASD (i.e., distinguish ASD from non-ASD) to identify subgroups is problematic, because such tools were not meant to describe variability within the autistic population (Zheng et al., 2020).

More recently, researchers combined multiple features, such as autistic symptoms, cognitive abilities, and adaptive functioning, to investigate the presence of subpopulations in autistic children (Kim et al., 2016; Zheng et al., 2020). Zheng et al. (2020) identified three subgroups in a sample of 188 preschoolers with ASD. Children in the first cluster (51%) displayed relatively high cognitive, language and adaptive abilities, and relatively low levels of social symptoms, repetitive behaviors, and sensory issues. Children in the second cluster (24.5%) presented cognitive, language and adaptive abilities similar to first cluster, but more severe social deficits as well as repetitive and sensory behaviors. Children in the third cluster (24.5%) showed lower cognitive, language and adaptive abilities, and more severe social, repetitive, and sensory symptoms.

In another study, Kim et al., (2016) found four clusters among 95 toddlers with ASD. The first cluster (36%) constituted the highest functioning group and was characterized by moderate impairments in social communication and repetitive behaviors. The second cluster (16%) grouped relatively high-functioning children that had similar cognitive skills, but less severe social affective symptoms and lower adaptive functioning than the first cluster. The third cluster (31%) constituted a relatively low-functioning group, with severe autistic symptoms and some delays in adaptive skills, but better verbal and nonverbal skills than the fourth cluster. The fourth cluster (17%) represented the very low-functioning group and was characterized by severe autistic symptoms, and significant delays in all areas of functioning. The study conducted by Kim et al. (2016) is particularly relevant because it explored the relationships between the identified subgroups and diagnosis stability, clinical presentation, and intervention outcomes. Children in the sample received various type of intervention, including applied behavior analysis, Floor Time, speech therapy, and occupational therapy. To our knowledge, no study has directly verified the relation between membership to a subgroup and EIBI outcomes.

Expanding on the findings of a previous study which evaluated the effectiveness of EBI<sup>5</sup> using a variable-centered approach (Préfontaine et al., 2021), the purpose of the current study was to (1) use a person-centered approach to identify distinct profiles of children receiving EBI based on measures of autistic symptoms, three domains of adaptive functioning and three subscales of IQ, (2) examine whether sociodemographic characteristics predicted profile membership, and (3) assess whether profile membership was associated with different response to EBI. Together, these questions sought to better understand the differential efficacy of EBI in children diagnosed with ASD.

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<sup>5</sup> Because the intensity offered to the participants may not qualify the intervention as being “intensive”, we will use the expression early behavioral intervention (EBI) to refer to their program to prevent misleading the reader.

## **Method**

### **Participants and Procedure**

The Joint Research Ethics Board for Public Rehabilitation Centers for Persons with Intellectual Disabilities and ASD in Quebec approved the research project. Parents had to provide written consent for their children to participate in the study. The sample comes from a larger research project (see Rivard et al., 2014; 2019) and consisted of 233 children (79% boys) diagnosed with ASD aged between 2.50 to 5.75 years old ( $M = 4.34$ ,  $SD = 0.47$ ) who received one year of low-intensity intervention (53.9% of participants) or moderate-intensity intervention (46.1% of participants). Regardless of the intensity option, the intervention was based on applied behavior analysis and qualified for early behavioral intervention. Children who participated in the study had to meet the following inclusion criteria: (a) have a diagnosis of ASD provided by a pediatric psychiatrist and independently confirmed by a multidisciplinary team, (2) be aged 5 years old or younger, and (3) live within the geographical area served by the rehabilitation center.

The larger research project used a prospective longitudinal design with three annual assessments, where time 1 represents baseline ( $n = 233$ ), time 2 represents post-intervention (12 months after baseline;  $n = 219$ ) and time 3 represents 12-months follow-up after the end of the intervention ( $n = 64$ ). Since most of the sample had entered preschool at time 3 and were no longer receiving services from the readaptation center, they may have been harder to reach for the completion of the 12-months follow-up. This challenge may explain high attrition at time 3. To explore the presence of distinct profiles on the children, the current study used the data from time 1.

### **Measures**

#### ***Demographics***

Parents of participants completed a sociodemographic questionnaire, and their responses were then coded in the database. Gender of the participants was a dichotomous variable (code 1 = female and 2 = male). Age was a continuous variable, where decimals allowed to record the exact age of the participants (i.e., 2-year-old and 6-month-old is coded 2.5). Annual family income was an ordered-categorical variable (code 1 = \$10,000 to \$29,999, 2 = \$30,000 to \$49,999, 3 = \$50,000 to \$69,999, 4 = \$70,000 to \$89,999, and 5 = Over \$90,000) and were reported in Canadian dollars.

### ***Autistic Symptoms***

A parent and a special education technician<sup>6</sup> completed the *Childhood Autism Rating Scale – Second Edition* (CARS-2; Scholper et Van Bourgondien, 2010) to measure autistic symptoms. The CARS-2 contains 15 items assessing different apparent difficulties in children with ASD on a 4-point scale from one to four (1 = normal, 4 = severely abnormal). Higher scores indicate more severe autistic symptoms. We used the average score between the two respondents because they were highly correlated ( $r = .81$ ).

### ***Adaptive Functioning***

Using the parent/primary caregiver form for young children (0 to 5 years old), a parent completed the *Adaptive Behavior Assessment System-II* (ABAS-II; Harrison and Oakland, 2003) to measure adaptive functioning. This form of the ABAS-II contains 241 items rating the performance of various adaptive behaviors on a 4-point scale, from zero to three (0 = never, the child is unable, 3 = always when necessary). The results provide a general adaptive composite score and a score for each of the three domains of adaptive functioning, namely the conceptual,

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<sup>6</sup> Special education technicians is a terminology unique to the province of Quebec and refers to college-level technicians.

social, and practical domains. In the present study, we used the scores of the three adaptive domains.

### ***Intellectual Functioning***

Research assistants supervised by a psychologist administered the *Wechsler Preschool and Primary Scale of Intelligence* (WPPSI-III; Wechsler, 2003) to measure intellectual functioning. The WPPSI-III totalizes 15 sub-tests capturing five dimensions (i.e., verbal comprehension, visuospatial performance, fluid reasoning, working memory and information processing speed). In addition to the full-scale IQ, results provide three subscale scores, namely verbal IQ, performance IQ and general language composite. In the present study, we considered the verbal IQ, performance IQ and general language composite because they provide more specific information on cognitive abilities than the full-scale IQ.

### ***Response to EBI***

In a previous study adopting a variable-centered approach (Préfontaine et al., 2021), we conducted latent growth curves (LGC) analyses to capture changes in autistic symptoms and adaptive functioning (general adaptive functioning and the conceptual, social, and practical domains of adaptive functioning) of children receiving EBI in a community setting. The results showed a linear decrease in autistic symptoms from baseline to follow-up (T1 to T3), and nonlinear changes in adaptive functioning characterized by improvement during the intervention period (T1 to T2), followed by stability in adaptive functioning during the follow-up period (T2 to T3). To better understand the impact of profile membership on response to EBI, the individual estimates from these LGC analyses were saved and used as the outcome variable, response to EBI, in the current study. For autistic symptoms, response to EBI refers to slope 1, that estimates the expected change between each time point from T1 to T3, as the change detected was linear across the entire study. For the three domains of adaptive functioning, response to EBI refers to

two slopes because the LGC analyses detected nonlinear changes. Slope 1 is the estimated expected change during the intervention period (T1 to T2) while slope 2 is the estimated expected change during the follow-up period (T2 to T3). Using the individual estimates of LGC instead of the scores at T3 allowed us to examine association between the profile membership and the change (i.e., progress) made by the children receiving EBI.

### **Analytical Strategy**

First, we performed descriptive statistics using SPSS 26.0. and then used Mplus 8.3 (Muthén & Muthén, 2017) to conduct Latent Profile Analyses (LPA; Muthén & Muthén, 2000) with the maximum likelihood estimation robust (MLR; Shi et al., 2021), which corrects the standard errors for non-normality in the data. Missing data were handled using full-information maximum likelihood estimation, which allow to use every case in the sample (Enders, 2010). We followed the recommendations of Masyn (2013) to test four different parametrizations of the means and variance-covariance matrix: 1) conditional independence with equal variance across profiles model (i.e., Mplus default), 2) conditional independence with unequal variance across profiles model, 3) conditional dependence with variance and covariance equal across profile, and 4) conditional dependence with unequal variance and covariance across profiles. For each parametrization, the analyses involved iteratively specifying the LPA models, starting with one profile up to six profiles. In order to avoid convergence to a local solution (i.e., false maximum likelihood; Hipp & Bauer, 2006), we estimated the models with 5000 random sets of starts values, with the 50 best retrained for the final optimization.

A critical question when conducting LPA is determining the number of profiles in the data. Various statistical tests and indices can support decision-making (Masyn, 2013; McLachlan & Peel, 2000). As log likelihood in mixture models are not distributed according to the chi-square distribution, the regular likelihood ratio test cannot be used to compare models. The



Vuong, Lo, Mendell and Rubin's test (VLMR) and the Bootstrap likelihood ratio test (BLRT) are adjusted likelihood ratio tests that allow for the comparison of the current model to a model with  $k-1$  profiles. A nonsignificant VLMR or BLRT indicates that the more parsimonious model is better fitting (Ferguson et al., 2019). Information criteria can also be used to determine the best model in the data. The Akaike information criterion (AIC), Consistent AIC (CAIC), Bayesian information criterion (BIC) and the sample-size adjusted Bayesian information criterion (SABIC) are such indices that allow comparing model with different number of classes, with lower values indicating a better fit. The magnitude of the differences between models is also important for interpretation, because in some samples, AIC, CAIC, BIC and SABIC values tend to continuously decrease as the number of profiles increase (Ferguson et al., 2020; Masyn, 2013). Consequently, making an elbow graph reporting these values for each model can support interpretation.

In LPA, entropy estimates the degrees of classification uncertainty (or precision with which individuals are classified) for each profile (Ferguson et al., 2019). Lower values of the entropy statistic indicate more uncertainty and values greater than 0.80 indicate that the profile classification is adequate (Tein et al., 2013). Entropy should not be used for profile enumeration, but it can support the comparison of models. Finally, given our use of a small sample, the number of individuals in the smallest class should also be taken into account as it could influence replicability of the model (Ferguson et al., 2020; Muthén & Muthén, 2000). Beyond the statistic tests and indices, the retained model should be meaningful conceptually (Ferguson et al., 2020; Morin & Litalien, 2019)

Once the final unconditional model was determined, we examined whether a number of variables would predict group membership (i.e., covariates having an impact on profile membership) and we evaluated group differences on outcomes (i.e., adjusted mean differences on

outcomes between latent class). Our analyses directly incorporated predictors to the final model to predict class membership through a multinomial logistic regression (Morin & Litalien, 2019). For the outcome, we opted for the BCH approach available in Mplus, which has the advantage of avoiding shifts in the profiles from the unconditional model (Asparouhov & Muthén, 2014; Bakk & Vermunt, 2016).

## **Results**

Table 1 reports descriptive statistics of the sample. The sample included children with varying characteristics, as shown by the large variances. Table 2 reports the fit indices of the LPA models; the upper panel shows results for conditional independence with equal variance across profiles, while the lower panel displays results for conditional independence with unequal variance across profiles. Models with the two other parametrizations led to convergence problems. Looking at the information criteria, AIC, CAIC, BIC and SABIC were constantly lower in the conditional independence with unequal variance models than in the conditional independence with equal variance models when considering the same number of profiles. Therefore, we retained conditional independence with unequal variance across profiles as the optimal parametrization.

As the information criteria continuously decreased as the number of profiles increased, we looked at the elbow graph (see Figure 1) to inform the selection of the best model. Visual inspection suggested that the optimal solution was between three and four profiles. To help with the final model selection, we created histograms with the characteristics of the latent profiles to see how profiles were distinct from one another and verify conceptual relevance. Considering all information, we opted for the four profiles model, as this solution had good entropy, reasonable number of children in the smallest class and each profile was qualitatively different from each other. We did not retain the solution with five profiles, because two profiles seemed to result

from a quantitative division of the same profile, rather than being qualitatively different from each other. Table 3 presents the estimated posterior probabilities for the final latent profile model. Values were excellent and suggest high precision in the classification of children between the different profiles.

Table 4 presents the latent profile means and variances on the mixture indicators. Figure 2 depicts these characteristics in *Z*-scores; the results were standardized to help with the interpretation of this histogram. Children in the first latent profile had low autistic symptoms combined with the highest adaptive and intellectual functioning. This *mild impairment with average IQ profile* described 27.47% of the children ( $n = 64$ ). Children in the second latent profile had the lowest autistic symptoms, similar adaptive functioning than the mild impairment with average IQ profile, but presented lower average IQ. This *mild impairment with lower average IQ profile* also described 27.47% of the children ( $n = 64$ ). Children in the third latent profile had moderate autistic symptoms, and low intellectual and adaptive functioning. This *moderate impairment profile* described 24.46% of the children ( $n = 57$ ). Finally, children in the fourth latent profile had the most severe autistics symptoms combined with the poorer intellectual and adaptive functioning. This *severe impairment profile* described 20.60% ( $n = 48$ ) of the children in our sample.

Table 5 reports the multinomial logistic regression of the various predictors of group membership, using the “mild impairment with average IQ profile” as the reference group. Lower annual income predicted membership to the moderate impairment profile and the severe impairment profile. Younger age marginally predicted membership to the severe impairment profile. Gender does not influence membership to the different profiles.

Table 6 reports the distinctions between profiles’ response to EBI, and Figure 3 depicts the estimated trajectories of each profile for all outcomes across the three time points. All profiles

showed a small reduction in autistic symptoms from baseline to follow-up, with the biggest improvement made by the children in the mild impairment with lower average IQ. As stated in the measures section, results of the effectiveness study from which the outcome variables come from showed a linear decrease in autistic symptoms from baseline to follow-up. Thus, response to EBI pertaining to autistic symptoms constituted only one outcome variable. Regarding the three domains of adaptive functioning, results revealed that the different profiles responded differentially to the intervention. During the intervention period (slope 1), all profiles made progress on the three adaptive domains, with the greatest improvement made by children in the mild impairment with average IQ and mild impairment with lower average IQ profiles for all outcomes. Looking at the follow-up period (slope 2), profiles were quite different in the way they evolved. For the conceptual and social domains of adaptive functioning, the moderate impairment and severe impairment profiles continued to progress, while the mild impairment with average IQ and mild impairment with lower average IQ profiles showed a small decrease. For the practical domain, the mild impairment with average IQ profile continued to slightly improve, while the mild impairment with lower average IQ, the moderate impairment and the severe impairment profiles stayed relatively stable.

### **Discussion**

Our study contributes to the literature on the heterogeneity of symptom presentation and response to intervention in ASD. Similar to Kim et al. (2016), we identified four profiles, including a mild impairment profile, a severe impairment profile, and two intermediate profiles with combinations of mild to moderate alterations for autistic symptoms, adaptive functioning and intellectual functioning. Even though we labeled profiles by qualifying the severity of impairment, it should be noted that even in the mild impairment with average IQ and mild impairment with lower average IQ profiles, children were far under the level of adaptive

functioning of their typically developing peers. Only children in the mild impairment with average IQ profile had IQs in the normal range. Within a given profile, the three domains of adaptive functioning are relatively homogeneous, as are the three subscales of IQ. Concerning the potential predictors of profile membership, only lower annual income predicted membership to the moderate and severe impairment profiles, which underlines the possibility that these family had less resources and support to alleviate (or intervene on) the symptoms before receiving formal support from public agencies. Another possible explanation is that parents with less income are less likely to seek services in the presence of mild symptoms. Thus, the differential use of services could also explain the overrepresentation of high-income parents in the mild impairment profiles. Gender did not influence profile membership in our sample, which was unexpected considering previous studies had shown behavioral and cognitive differences across genders (Frazier et al., 2014; Hull et al., 2017). Because our sample was considerably younger, this disparity may be the result of the specific age group studied (Zheng et al., 2020).

Profiles showed distinction in their response to EBI. During the intervention period, the mild impairment with average IQ and mild impairment with lower average IQ profiles achieved the largest gains on all outcome variables, which is consistent with the findings from Kim et al. (2016). Despite the magnitude of the change being smaller, the moderate impairment and severe impairment profiles also improved on all outcome variables. At the follow-up period, the severe impairment and moderate impairment continued to improve or maintained their gains in the conceptual and social domains, while the two mild impairment profiles showed small reductions. These results suggest that although their progress is smaller and slower, the improvements are sustainable over time for children who present more impairment when they enter services. Hence, evaluative studies should consider long-term effects when assessing intervention. As children were no longer receiving services from the readaptation center and had started school during the

follow-up period, we have no information on the type of support each child received in class at school. One hypothesis is that the children with mild impairments received less support from their school than the children in the moderate impairment and severe impairment profiles. This observation highlights the need to conduct more research on the factors that influence maintenance of improvements after receiving EBI. In addition, these results show that the person-centered perspective may help uncover differential patterns of change that would remain unknown in a variable-centered perspective.

Our study has some limitations that should be discussed. Because we had a limited number of variables, the external validators (i.e., outcome variables used for the external validation of the profiles) were changes on the initial level of some of the variables used to identify the profiles (i.e., mixture indicators). This method is unconventional, but it allowed us to identify profiles of children from their pre-intervention characteristics, and to associate them with their response to EBI during the intervention and follow-up periods. Thus, results of cross-sectional profiles at time 1 may support practitioners in evaluating the prognosis regarding response to intervention of the children with whom they intervene. To expand these findings, future research should use latent transition analysis, which identify profiles at different time points and investigate the probability of a transition from a profile to another across time (Muthén & Muthén, 2000). An additional possibility involves using latent growth mixture modeling (Morin & Litalien, 2019), but these analyses typically require large samples. Another limitation concerns the response to EBI variables, which were computed from an effectiveness assessment that used a correlational design. The absence of control group limits the causal inferences between the intervention and the observed changes, and does not control for maturation effects. In addition, results regarding the association between profile membership and the follow-up period (T2 to T3) must be interpreted with caution, given the high attrition in our

sample between these time points. Future research should attempt to limit attrition by using retention strategies. It should be noted that, because the profiles we made from cross-sectional data at T1, this limit does not jeopardize the result of the LPA analysis.

### **Implications**

Our study is one of the few to explore the presence of subgroups in preschoolers with ASD, and to investigate predictors and outcomes of membership. To our knowledge, our results are the first to show that the profiles associated with better short-term outcomes of EBI are different than the profiles who maintain their gains better. This finding could guide both practitioners and researchers assessing the effects of EBI. Future research should try to replicate those findings with a larger sample and consider using designs with more time points during the follow-up to better understand the factors associated with the maintenance of outcomes.

## **CRedit authorship contribution statement**

**Isabelle Préfontaine:** Conceptualization, Methodology, Formal analysis, Writing – Original Draft. **Julien Morizot:** Supervision of analysis, Writing – Review and editing. **Marc J. Lanovaz:** Supervision, Writing – Review and editing. **Mélina Rivard:** Methodology, Data Curation, Review.

## **Declaration of Competing Interest**

The authors of this paper declare no conflicts of interest in the completion of the study or the preparation of this manuscript. Authors have no relevant financial relationships to disclose.

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**Table 1***Descriptive Statistics of the Sample*

| Variables                    | <i>N</i> | Min  | Max   | <i>M</i> | <i>SD</i> | Variance | Skewness | Kurtosis |
|------------------------------|----------|------|-------|----------|-----------|----------|----------|----------|
| Autistic symptoms            | 226      | 17   | 54.75 | 31.25    | 7.77      | 60.42    | 0.36     | -0.47    |
| General Adaptive Functioning | 229      | 41   | 130   | 64.94    | 14.83     | 219.82   | 0.89     | 1.55     |
| Conceptual Domain            | 229      | 45   | 123   | 68.51    | 15.11     | 228.44   | 0.50     | 0.08     |
| Social Domain                | 229      | 48   | 130   | 70.65    | 16.58     | 247.87   | 0.55     | -0.02    |
| Practical Domain             | 229      | 41   | 129   | 65.55    | 14.20     | 201.60   | 0.63     | 1.40     |
| Performance IQ               | 224      | 47   | 130   | 80.08    | 20.65     | 426.31   | 0.18     | -1.00    |
| Verbal IQ                    | 223      | 48   | 122   | 72.43    | 17.28     | 298.61   | 0.58     | -0.62    |
| Global Language Composite    | 224      | 47   | 117   | 74.29    | 19.33     | 373.73   | 0.15     | -1.07    |
| Annual Income                | 227      | 1    | 5     | 2.91     | 1.45      | 2.09     | 0.15     | -1.07    |
| Age                          | 225      | 2.50 | 5.75  | 4.34     | 0.47      | 0.21     | -0.83    | 2.02     |

**Table 2***Summary of Fit Statistics for Different Latent Profile Models*

| Model                                                 | LL       | #fp | Scaling | AIC   | CAIC  | BIC   | SABIC | Entropy | Smallest class n (%) | VLMR | BLRT |
|-------------------------------------------------------|----------|-----|---------|-------|-------|-------|-------|---------|----------------------|------|------|
| <b>Conditional independence with equal variance</b>   |          |     |         |       |       |       |       |         |                      |      |      |
| 1-Class                                               | -6543.97 | 14  | 0.933   | 13115 | 13129 | 13164 | 13119 | -       | -                    |      |      |
| 2-Class                                               | -6134.85 | 22  | 1.254   | 12313 | 12335 | 12389 | 12319 | .932    | 41%                  | .000 | .000 |
| 3-Class                                               | -6036.63 | 30  | 1.298   | 12133 | 12163 | 12236 | 12141 | .880    | 26.87%               | .035 | .000 |
| 4-Class                                               | -5966.09 | 38  | 1.645   | 12008 | 12046 | 12139 | 12018 | .884    | 15 (6.44%)           | .561 | .000 |
| 5-Class                                               | -5890.94 | 46  | 1.320   | 11873 | 11919 | 12032 | 11886 | .899    | 15 (6.44%)           | .042 | .000 |
| <b>Conditional independence with unequal variance</b> |          |     |         |       |       |       |       |         |                      |      |      |
| 1-Class                                               | -6543.97 | 14  | 0.933   | 13115 | 13178 | 13164 | 13119 | -       | -                    |      |      |
| 2-Class                                               | -6032.21 | 29  | 1.272   | 12122 | 12251 | 12222 | 12130 | .989    | 54 (23.17%)          | .000 | .000 |
| 3-Class                                               | -5844.06 | 44  | 1.182   | 11776 | 11971 | 11927 | 11788 | .942    | 48 (20.60%)          | .000 | .000 |
| 4-Class                                               | -5773.13 | 59  | 1.094   | 11664 | 11926 | 11867 | 11680 | .911    | 48 (20.60%)          | .000 | .000 |
| 5-Class                                               | -5718.25 | 74  | 1.079   | 11584 | 11913 | 11839 | 11605 | .897    | 21 (9.01%)           | .069 | .000 |

*Note.* LL: Model LogLikelihood; #fp: Number of free parameters; Scaling = scaling factor associated with MLR loglikelihood estimates; The VLMR test and the BLRT compare the current model to a model with k-1 profiles. LPA = latent profile analysis; AIC = Akaike's Information Criterion; CAIC: Constant AIC; BIC = Bayesian Information Criterion; SABIC = Sample-Adjusted BIC; VLMR = Vuong- Lo-Mendell Ruben; BLRT = bootstrap likelihood ratio test.



**Table 3***Classification Table Based on Estimated Posterior Probabilities for the Final Latent Profile Model*

|                                          | 1          | 2          | 3          | 4          |
|------------------------------------------|------------|------------|------------|------------|
| 1. Mild Impairment with Average IQ       | <b>.94</b> | .06        | .00        | .00        |
| 2. Mild Impairment with Lower Average IQ | .05        | <b>.94</b> | .02        | .00        |
| 3. Moderate Impairment                   | .00        | .02        | <b>.96</b> | .01        |
| 4. Severe Impairment                     | .00        | .00        | .01        | <b>.99</b> |

**Table 4***Latent Profile Means and Variances on the Mixture Indicators*

|                           | Mild Impairment with<br>Average IQ |          | Mild Impairment with<br>Lower Average IQ |          | Moderate Impairment |          | Severe Impairment |          |
|---------------------------|------------------------------------|----------|------------------------------------------|----------|---------------------|----------|-------------------|----------|
|                           | Mean                               | Variance | Mean                                     | Variance | Mean                | Variance | Mean              | Variance |
|                           | Autistic symptoms                  | 27.65    | 40.50                                    | 25.62    | 20.87               | 33.84    | 23.53             | 40.14    |
| Conceptual domain         | 81.07                              | 173.61   | 76.72                                    | 71.60    | 60.24               | 43.31    | 50.33             | 14.36    |
| Social domain             | 80.78                              | 283.27   | 81.54                                    | 82.53    | 62.76               | 79.73    | 51.78             | 21.52    |
| Practical domain          | 74.43                              | 208.67   | 73.74                                    | 55.99    | 60.92               | 58.97    | 48.01             | 25.14    |
| Verbal IQ                 | 94.45                              | 104.81   | 72.75                                    | 74.36    | 63.41               | 49.66    | 52.65             | 2.12     |
| Performance IQ            | 101.43                             | 160.19   | 83.25                                    | 145.02   | 73.44               | 255.80   | 55.04             | 38.03    |
| Global language composite | 96.14                              | 98.55    | 78.53                                    | 121.10   | 64.93               | 91.68    | 48.89             | 5.17     |

**Table 5***Results from the Multinomial Logistic Regression Evaluating the Effects of Predictors on Latent Profile Membership*

| Predictors | Mild Impairment with Average IQ          |      |              |                        |      |             |                      |      |              |
|------------|------------------------------------------|------|--------------|------------------------|------|-------------|----------------------|------|--------------|
|            |                                          |      |              | vs.                    |      |             |                      |      |              |
|            | Mild Impairment<br>with Lower Average IQ |      |              | Moderate<br>Impairment |      |             | Severe<br>Impairment |      |              |
|            | Coef. (SE)                               | OR   | 95% IC       | Coef. (SE)             | OR   | 95% IC      | Coef. (SE)           | OR   | 95% IC       |
| Gender     | .84 (.73)                                | 2.31 | [.55, 9.75 ] | .30 (.53)              | 1.35 | [.48, 3.83] | .42 (.56)            | 1.52 | [.51, 4.56 ] |
| Age        | .76 (.67)                                | 2.14 | [.58, 7.88 ] | -.39 (.45)             | .68  | [.28, 1.64] | -.87 (.52)           | 0.42 | [.15, 1.15 ] |
| Income     | -.25 (.16)                               | .78  | [.56, 1.07 ] | -.49 (.17)**           | .61  | [.44, 0.85] | -.56 (.17)***        | 0.57 | [.41, 0.80 ] |

*Note.* Coef. = Coefficient; SE = Standard error.

\*p &lt; .05, \*\*p &lt; .01. \*\*\*p &lt; .001

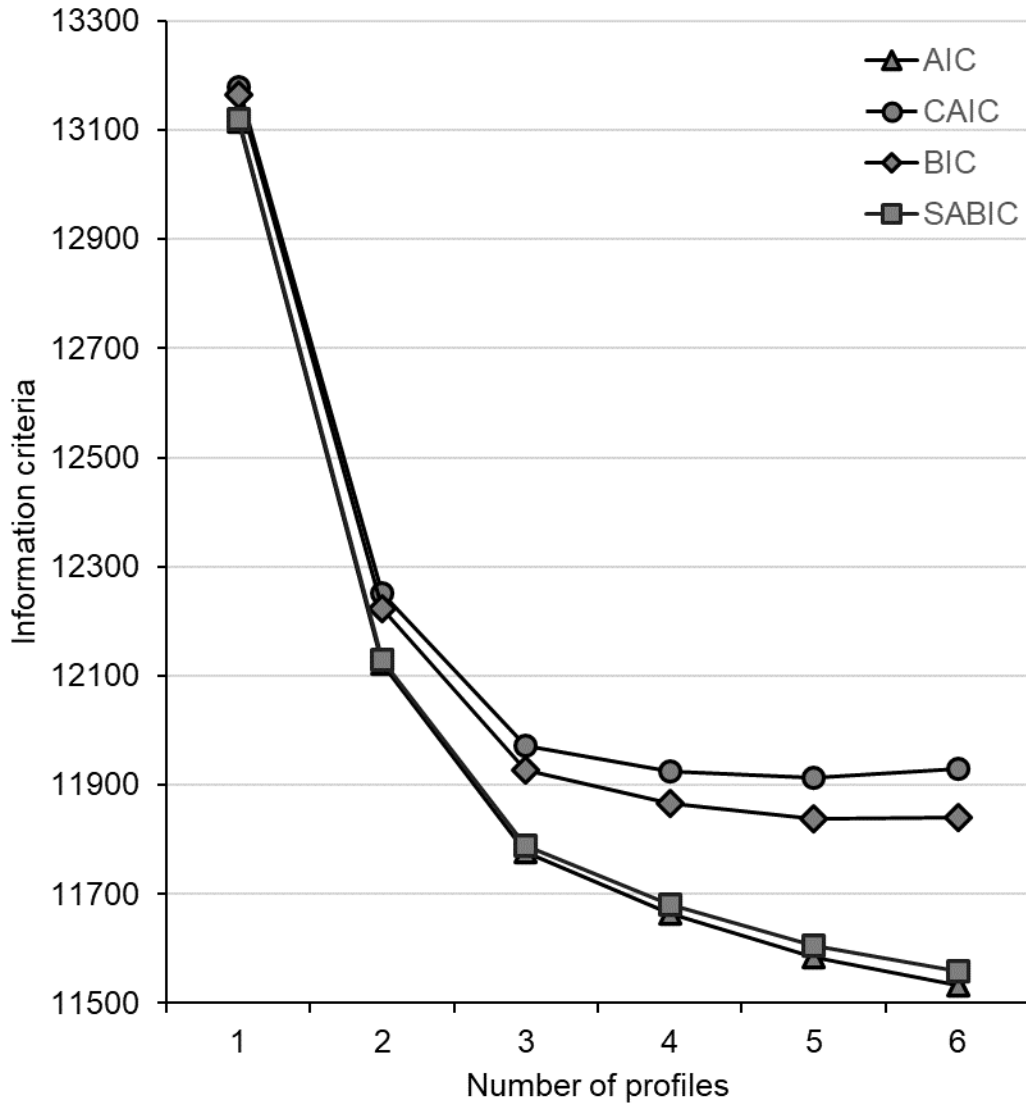
**Table 6***Profiles' Distinction on Response to EBI*

| Outcomes          | Mild Impairment<br>with Average IQ | Mild Impairment with<br>Lower Average IQ | Moderate Impairment | Severe Impairment | Summary of<br>significant<br>differences |
|-------------------|------------------------------------|------------------------------------------|---------------------|-------------------|------------------------------------------|
|                   | Coef. (SE)                         | Coef. (SE)                               | Coef. (SE)          | Coef. (SE)        |                                          |
| Autistic symptoms | -1.97 (.02)                        | -2.03 (.02)                              | -1.81 (.02)         | -1.62 (.03)       | 2 > 1 > 3 > 4                            |
| Slope 1           |                                    |                                          |                     |                   |                                          |
| Conceptual domain | 8.17 (.34)                         | 7.75 (.32)                               | 6.79 (.31)          | 4.48 (.29)        | 1 = 2 > 3 > 4                            |
| Social domain     | 6.33 (.07)                         | 6.38 (.05)                               | 5.79 (.06)          | 5.21 (.04)        | 2 = 1 > 3 > 4                            |
| Practical domain  | 3.76 (.46)                         | 3.53 (.48)                               | 3.18 (.44)          | 1.00 (.36)        | 1 = 2 = 3 > 4                            |
| Slope 2           |                                    |                                          |                     |                   |                                          |
| Conceptual domain | -4.26 (.42)                        | -3.29 (.34)                              | 0.50 (.21)          | 2.66(.18)         | 4 > 3 > 2 = 1                            |
| Social domain     | -1.26 (.33)                        | -1.69 (.19)                              | 1.48 (.20)          | 3.30 (.13)        | 4 > 3 > 1 = 2                            |
| Practical domain  | 0.90 (.51)                         | -0.84 (.60)                              | 0.08 (.41)          | -0.62 (.27)       | 1 > 3 = 4 = 3                            |

*Note.* SE = Standard error; Significant differences are determined by an alpha level of .05.

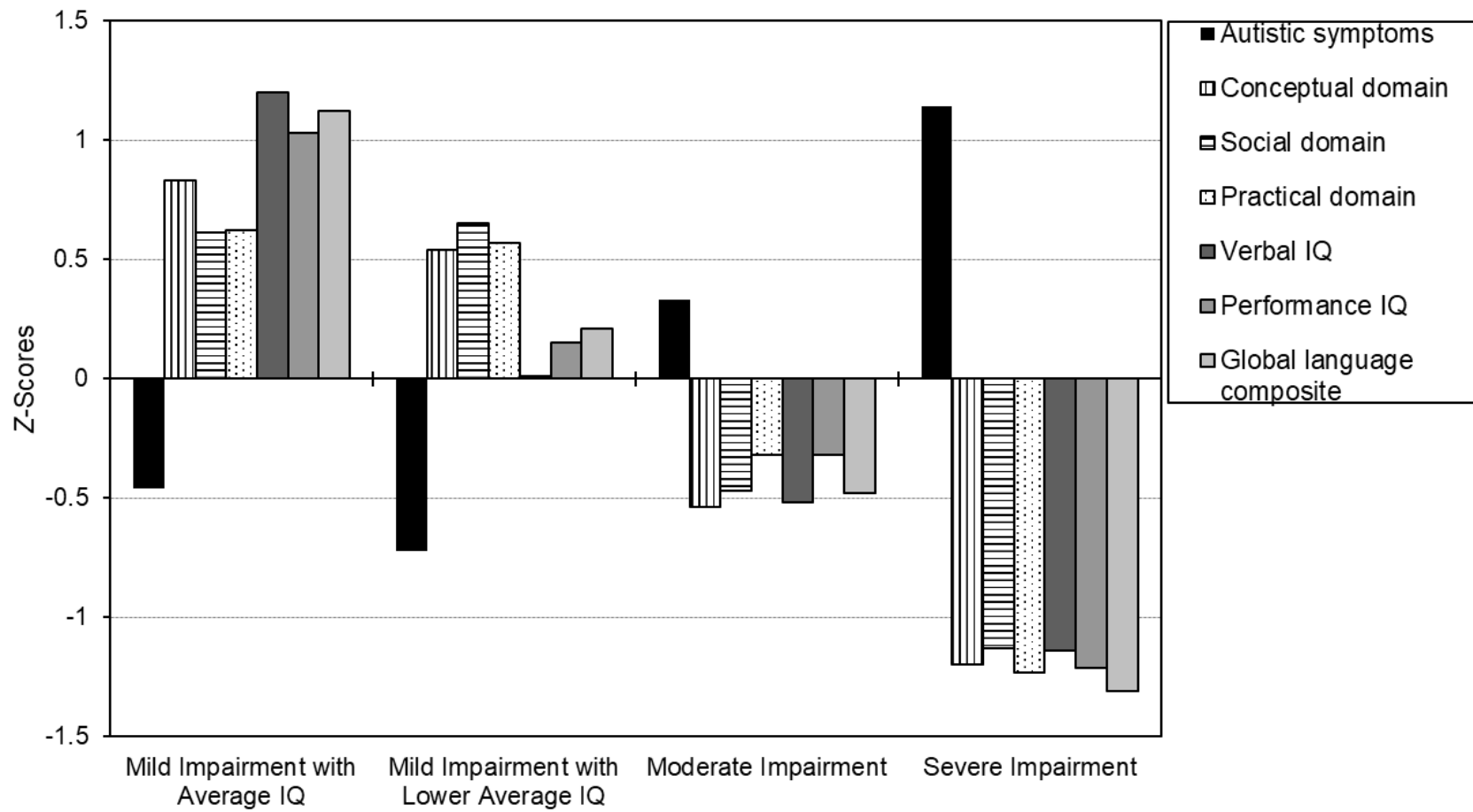
**Figure 1**

*Elbow Graph of the Information Criteria*



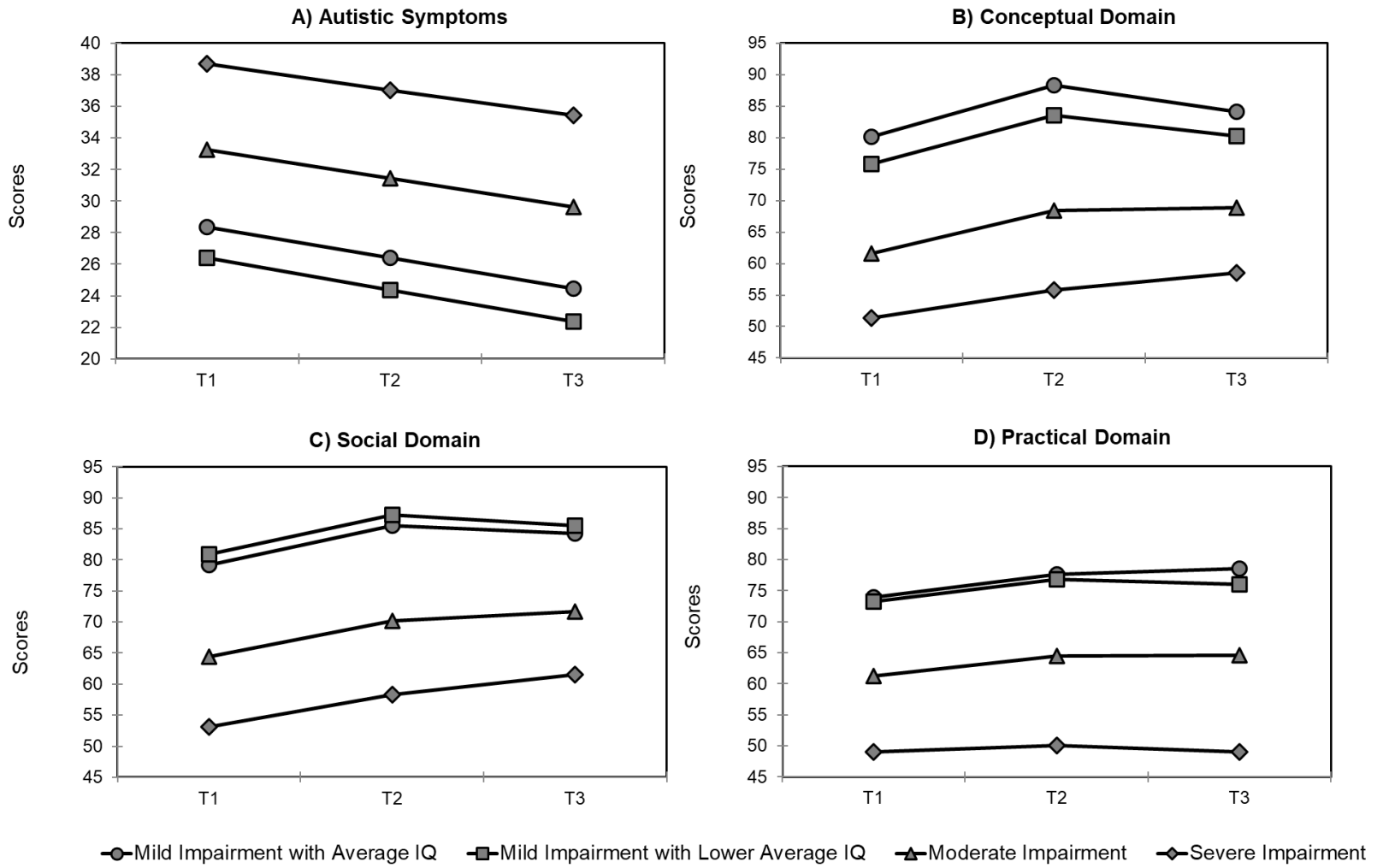
**Figure 2**

*Latent Profile Standard Scores on the Mixture Indicators.*



**Figure 3**

*Changes in Autistic Symptoms and the Three Domains of Adaptive Functioning for Each Latent Profile.*



**Chapitre IV – Article 3**



**Brief Report: Machine Learning for Estimating Prognosis of Children with Autism  
Receiving Early Behavioral Intervention – A Proof of Concept**

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## Résumé

Bien que l'intervention comportementale intensive soit considérée comme soutenue empiriquement pour les enfants autistes, l'estimation du pronostic en réponse à une intervention est un défi pour les praticiens. Une solution potentielle consiste à utiliser l'apprentissage automatique (angl. *machine learning*) pour guider l'estimation de la réponse à l'intervention. Ainsi, notre étude a comparé cinq algorithmes d'apprentissage automatique pour estimer la réponse à l'intervention pour deux variables (c.-à-d. le fonctionnement adaptatif et les symptômes autistiques). Tous les algorithmes d'apprentissage automatique ont produit de meilleures prédictions pour les deux variables qu'une prédiction aléatoire. Ces résultats indiquent que l'apprentissage automatique est une approche prometteuse pour estimer le pronostic chez les enfants autistes, mais des études comparant ces prédictions avec celles produites par des praticiens qualifiés restent nécessaires.

*Mots-clés* : apprentissage automatique, autisme, intervention comportementale intensive, pronostic, réponse différentielle

## **Abstract**

Although early behavioral intervention is considered as empirically-supported for children with autism, estimating treatment prognosis is a challenge for practitioners. One potential solution is to use machine learning to guide the prediction of the response to intervention. Thus, our study compared five machine algorithms in estimating treatment prognosis on two outcomes (i.e., adaptive functioning and autistic symptoms) in children with autism receiving early behavioral intervention in a community setting. Each machine learning algorithm produced better predictions than random sampling on both outcomes. Those results indicate that machine learning is a promising approach to estimating prognosis in children with autism, but studies comparing such predictions with those produced by qualified practitioners remain necessary.

*Keywords:* autism, differential response, early behavioral intervention, machine learning, prognosis

## **Brief Report: Machine Learning for Estimating Prognosis of Children with Autism Receiving Early Behavioral Intervention – A Proof of Concept**

One challenge faced by practitioners who intervene with individuals with mental health and developmental disorders involves estimating prognosis given a specific treatment (Cearns et al., 2019; Dwyer et al., 2018). Even though the number of evidence-based interventions is increasing (Cuijpers, 2017; Steinbrenner et al., 2020), a considerable percentage of individuals make little or no progress with these interventions (Bzdok & Meyer-Lindenberg, 2018; Schwartz et al., 2021). Early intensive behavioral intervention (EIBI) is no exception to this phenomenon. Based on the principles of applied behavior analysis, EIBI, is considered one of the most effective interventions for young children with autism spectrum disorders (ASD; Eldevik et al., 2009; Makrygianni et al., 2018; Makrygianni & Reed, 2010; Peters-Scheffer et al., 2011; Reichow et al., 2018). Despite showing positive effects on adaptive functioning, autistic symptoms, cognitive skills, and communication abilities (Eikeseth et al., 2012; Reichow et al., 2018), the results of EIBI outcome studies consistently report differential response to intervention among children (Fava & Strauss, 2014; Howlin et al., 2009; Magiati et al., 2011; Makrygianni et al., 2018; Reichow et al., 2018). Hence, some children show considerable improvements in many areas of functioning, but others make minimal to no progress on norm-referenced measures (Ben-Itzhak et al., 2014; Gabriels et al., 2001; Howlin et al., 2009; Zachor & Ben Itzhak, 2010).

Although researchers have identified some predictors of EIBI outcomes (e.g., intervention intensity, age at enrollment, IQ, adaptive functioning, autistics symptoms and sociodemographic characteristics), these predictors differ across studies, which questions their reliability (Eapen et al., 2013; Reichow et al., 2018; Smith et al., 2015; Warren et al., 2011). Moreover, studies directly exploring moderators of intervention effects are scarce (Ben-Itzhak et al., 2014; Eldevik et al., 2010; Warren et al., 2011). Therefore, practitioners may find it difficult to estimate how a

specific child with their combination of individual characteristics will respond to EIBI. Given the challenge associated with estimating prognosis for children receiving EIBI, one possible solution may be to use machine learning to guide the prediction of the response to intervention.

Machine learning is a subdomain of artificial intelligence that involves using algorithms to “train” a model to recognize patterns in data in order to make predictions (Turgeon & Lanovaz, 2020). To our knowledge, only a couple of studies have used artificial intelligence to investigate differential outcomes of behavioral interventions in children with autism (Linstead et al., 2015, 2017). For example, Linstead et al. (2015) have used artificial neural networks (ANNs) to train non-linear models on historical clinical data and to explore the relationship of intervention and supervision intensity on acquisition of learning outcomes. In a subsequent study, the same research team examined the relationship between the number of hours of behavioral intervention (ranging from 20.02 to 197.25 per month) and mastery of learning objectives (Linstead et al., 2017). In both studies, ANNs outperformed the linear regression models at the task of predicting mastery of learning outcomes. These results support the potential of using machine learning to estimate treatment prognosis in this population. However, ANNs are only one type of machine learning algorithm among many others and more research is needed to better understand how, and to what extent, machine learning can contribute to the field of evaluative research. The purpose of our study was to extend the prior studies by examining whether a selection of machine learning algorithms could estimate prognosis of children receiving early behavioral intervention (EBI)<sup>7</sup> in a community setting and comparing their relative prediction accuracy regarding improvement on adaptive functioning and autistic symptoms.

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<sup>7</sup> Because the intensity of the intervention provided to our sample may not qualify the intervention as being “intensive”, we will use the expression EBI when referring to the program that we evaluated to avoid misleading the reader.

## **Method**

### **Participants**

Our dataset originates from a study assessing the effects of a community-based intervention program conducted with 233 unselected children that took place from 2009 to 2012 (see Préfontaine et al., 2021). Children received a low to moderate intensity intervention based on the principles of applied behavior analysis that qualifies for EBI in our province (Quebec, Canada). Participants obtained a diagnosis of ASD from an independent multidisciplinary team. Parents provided written consent for their child prior their participation in the study. The original project (see Rivard et al., 2014; 2019) used a prospective longitudinal design with annual assessments. We used the data from time 1 and time 2 to train and test the machine learning algorithms to estimate the prognosis of short-term outcomes. Our analyses involved a subsample for which we had complete data for the outcome variables (i.e., adaptive functioning,  $n = 216$ ; autistic symptoms,  $n = 149$ ). Table 1 presents descriptive statistics for each sample.

### **Machine Learning**

Machine learning algorithms involve training a model to recognize patterns in data to make predictions (Turgeon & Lanovaz, 2020). Two types of data are required for supervised learning: features and labels. Features are the input data that the algorithms use to make predictions. The features represent measurable aspects of the studied phenomenon. Labels are the output data, or the results of the prediction. The algorithm trains a model to recognize the patterns between features and labels in a subsample (i.e., training set) to make predictions on the remaining subsample (i.e., test set).

### ***Features***

We used individual characteristics that are considered potential predictors of EIBI effectiveness as features: age at enrollment (Bieleninik et al., 2017; Makrygianni & Reed, 2010),

intellectual functioning (Makrygianni & Reed, 2010; Reed, 2016; Tiura et al., 2017), autistic symptoms (Flanagan et al., 2012; Reed, 2016), and adaptive functioning (Eldevik et al., 2010; Flanagan et al., 2012; Reed, 2016; Reichow, 2012; Sallows & Graupner, 2005; Vivanti et al., 2014). In addition, previous research has shown that behavioral and cognitive difference across gender (Frazier et al., 2014; Hull et al., 2017), and some evidence has suggested that high socioeconomic status is associated with better outcomes for the intervention (Gabriels et al., 2001; Magiati et al., 2011). Consequently, we also included gender and annual income among the features.

We briefly describe how features were measured, but readers can see Préfontaine et al. (2021) for comprehensive descriptions. Age was a continuous variable, where decimals allowed to record the exact age of the participants (i.e., 2-year-old and 6-month-old was coded as 2.5). Research assistants supervised by a psychologist administered the *Wechsler Preschool and Primary Scale of Intelligence* (WPPSI-III; Wechsler, 2003) to measure intellectual functioning. A parent and a special education technician<sup>8</sup> completed the *Childhood Autism Rating Scale – Second Edition* (CARS-2; Scholper et Van Bourgondien, 2010). We used the average score between the two respondents because they were highly correlated ( $r = .81$ ). A parent completed the *Adaptive Behavior Assessment System-II* (ABAS-II; Harrison and Oakland, 2003) to assess adaptive functioning. Gender of the participants was a dichotomous variable (code 1 = female and 2 = male). Annual family income was an ordered-categorical variable (code 1 = \$10,000 to \$29,999, 2 = \$30,000 to \$49,999, 3 = \$50,000 to \$69,999, 4 = \$70,000 to \$89,999, and 5 = Over \$90,000) and were reported in Canadian dollars.

### ***Labels***

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<sup>8</sup> Special education technicians is a terminology unique to the province of Quebec and refers to college-level technicians.

We used two labels: improvement of autistic symptoms and improvement in adaptive functioning. We chose those labels to represent response to intervention because a recent systematic review has observed reductions in autistic symptoms and improvements in adaptive function as primary outcomes of EIBI (Reichow et al., 2018). For both variables, change scores were computed by subtracting the score at time 1 from the score at time 2. Then, we transformed the labels values to binary outcomes. For autistic symptoms, a change score of 0 or more represented no improvement, and a change score of lower than 0 represented improvement. For adaptive functioning, a change score of 0 or less represented no improvement and a change score higher than 0 represented improvement. According to these definitions, 64% of children improved their adaptive functioning and 65% improved their autistic symptoms.

### *Algorithms*

One of the important aspects when building a machine learning model is determining the appropriate algorithm for the task of interest (Yang & Shami, 2020). Different algorithms make predictions using the features in different ways. We compared the prediction of five algorithms that can solve classification problems (i.e., logistic regression,  $k$  nearest neighbors, Gaussian process, random forest, and support vector classifier). Logistic regression is a linear model that identifies a cut-off (or threshold) to separate label values and make classification according to this cut-off (Yang & Shami, 2020).  $k$  nearest neighbors uses the  $k$  closest cases to identify the appropriate classification (Yang & Shami, 2020). Data in the training sets are placed on a map (or a graph) according to their features and data in the test set obtain the same label value as the majority of its  $k$  closest cases. Gaussian process consists of tracing the Gaussian curves for each feature and each label in the training set and predicting the label for the test set according to the relative position of the novel data on the Gaussian curves (Daemi et al., 2019). Random forest involves building numerous decision trees to resolve classification problems (Jiang et al., 2020).



Each tree in the forest makes a prediction, and this forest selects the classification supported by the largest number of trees. The support vector classifier projects the data in a higher dimension and then separates the classes using a hyperplane (Yang & Shami, 2020). Projecting the data into a higher dimension allows their separation in classes, solving the overlapping problem in the lower dimension.

### **Analyses**

To limit the risk of overfitting and considering our small sample size, we used the  $k$ -fold cross-validation methodology (Yarkoni & Westfall, 2017) to train the models and test the accuracy of the prediction for each algorithm. The  $k$ -fold cross validation consists of randomly splitting the dataset in  $k$  groups (here  $k = 5$ ); the first group is treated as the test set and the model is trained on the remaining groups. The procedure is repeated  $k$  times, so that each group forms the test set once. In this context, accuracy represented the average percentage of agreement (i.e., number of agreements divided by total of participants) between the true values and the predicted label for each fold. To be able to qualify the performance of the different algorithm, we also computed the accuracy of random sampling for the two labels over 10,000 iterations.

For each algorithm, we conducted the analyses twice with two different sets of features to identify which one best predicts improvements. The first set contained gender, age, family annual income, autistic symptoms, full-scale IQ and general adaptive functioning score. The second set contained gender, age, family annual income, autistic symptoms, the three subscales of IQ (namely, verbal IQ, performance IQ and general language composite score) and the three domains of adaptive functioning (namely, the conceptual, social and practical domains). We chose to test these two sets to examine whether having more precise features (i.e., subscales vs. global scores) would lead to more accurate predictions.

## **Results and Discussion**

Table 2 presents the accuracy for each algorithm on the test set. The upper panel shows the results for the first set of features (global) and the lower panel shows the results for the second set of features (subscales). For both sets of features, the machine learning algorithms produced better predictions than random sampling. The mean difference between the random sampling and the predictions of the algorithms was 11.2% for adaptive functioning and 6.5% for autistic symptoms. Comparing the two sets of features, the prediction accuracy for the same algorithm was similar, with an average difference of 1.4% for adaptive functioning and 1.7% for autistic symptoms. This result indicates that the predictions were similar regardless of whether we used the full-scale scores (i.e., full scale of IQ and the general adaptive composite score) or the subscales scores (i.e., verbal IQ, performance IQ, global language composite score, and the conceptual, social and practical domains of adaptive functioning) as input. In both sets, the algorithms predicted adaptive functioning with better accuracy than autistic symptoms. Comparing algorithms with each other, the Gaussian process performed best on adaptive functioning, and support vector classifier achieved the most accurate predictions for autistic symptoms. None of the algorithms produced systematically worst predictions than all the others.

Our results are consistent with previous work that used machine learning to predict treatment outcomes in depression (Chekroud et al., 2016). Their machine learning models produced predictions with an accuracy ranging from 59.6% to 64.6%, while clinicians had an average accuracy of 49.3%. In the same vein, the next important step in this line of work would be to assess whether the algorithms can estimate prognosis better than qualified practitioners in EIBI who intervene with children with ASD. To this end, the goal of machine learning should not be to aim for absolute accuracy, but rather to demonstrate its incremental utility by improving current practices (Cearns et al., 2019). Improving the estimation of prognosis would allow practitioners to adjust their intervention when a child is not showing expected progress.

Our study has some limitations that should be noted. The small sample size limited the cross-validation techniques that we could use and probably had an impact on accuracy. To address this issue, future research should replicate our study with larger sample sizes. A larger sample size would also allow researchers to tune the hyperparameters in order to optimize (i.e., improve) the models. Overlooking hyperparameters tuning is a frequent limitation in current machine learning work in psychiatry, and tuning hyperparameters may help discriminate between algorithms by improving their prediction accuracy in a differential manner (Cearns et al., 2019). Finally, an interesting avenue for future research would be to diversify the features used, as machine learning has the capacity to integrate a large amount of data from different sources (e.g., behavioral, genetic, neuroimaging) in the same model (Miotto et al., 2018; Sengupta et Shrestha, 2019; Shatte et al., 2020). This interdisciplinary approach has the potential to further improve the accuracy of models used to predict response to intervention.

### **Compliance with Ethical Standards**

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**Ethical Approval:** All procedures performed in this study were in accordance with the ethical standards of Joint Research Ethics Board for Public Rehabilitation Centers for Persons with Intellectual Disabilities and ASD in Quebec and with the 1964 Helsinki declaration and its later amendments.

**Informed Consent:** Parents provided informed consent for them and their child.

**Conflict of Interest:** The authors have no conflict of interest to report.

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**Table 1***Descriptive Statistics of Each Sample at Enrolment*

| Characteristics              | Adaptive functioning<br>(n = 216) |           | Autistic symptoms<br>(n = 149) |           |
|------------------------------|-----------------------------------|-----------|--------------------------------|-----------|
|                              | <i>M</i>                          | <i>SD</i> | <i>M</i>                       | <i>SD</i> |
| Autistic symptoms            | 31.61                             | 7.70      | 31.66                          | 7.35      |
| General adaptive functioning | 64.14                             | 14.08     | 63.96                          | 14.14     |
| Conceptual domain            | 67.77                             | 14.52     | 67.65                          | 14.80     |
| Social domain                | 69.74                             | 15.99     | 69.49                          | 15.99     |
| Practical domain             | 64.89                             | 13.70     | 64.87                          | 13.68     |
| Full-scale IQ                | 70.85                             | 20.17     | 70.94                          | 19.30     |
| Verbal IQ                    | 72.12                             | 17.26     | 71.43                          | 16.04     |
| Performance IQ               | 79.59                             | 20.49     | 80.44                          | 20.43     |
| General language composite   | 73.73                             | 19.46     | 73.57                          | 18.62     |
| Age                          | 4.32                              | 0.47      | 4.26                           | 0.44      |
|                              | N                                 | %         | N                              | %         |
| Gender                       |                                   |           |                                |           |
| Male                         | 171                               | 79.17     | 122                            | 81.88     |
| Female                       | 45                                | 20.83     | 27                             | 18.12     |
| Annual income                |                                   |           |                                |           |
| \$10,000 to \$29,999         | 44                                | 20.37     | 29                             | 19.46     |
| \$30,000 to \$49,999         | 50                                | 23.15     | 38                             | 25.50     |
| \$50,000 to \$69,999         | 41                                | 18.98     | 31                             | 20.81     |
| \$70,000 to \$89,999         | 32                                | 14.81     | 24                             | 16.11     |
| Over \$90,000                | 43                                | 19.90     | 24                             | 16.11     |

**Table 2***Average Prediction Accuracy of Each Algorithm on the Test Set*

|                                                        | <b>Random<br/>Sampling</b> | <b>Logistic<br/>Regression</b> | <b><i>k</i> Nearest<br/>Neighbors</b> | <b>Gaussian<br/>Process</b> | <b>Random<br/>Forest</b> | <b>Support<br/>Vector<br/>Classifier</b> |
|--------------------------------------------------------|----------------------------|--------------------------------|---------------------------------------|-----------------------------|--------------------------|------------------------------------------|
| <b>First set of features (using full-scale scores)</b> |                            |                                |                                       |                             |                          |                                          |
| Adaptive Functioning                                   | .542                       | .639                           | .648                                  | .684                        | .662                     | .662                                     |
| Autistic Symptoms                                      | .545                       | .631                           | .603                                  | .610                        | .597                     | .638                                     |
| <b>Second set of features (using subscales scores)</b> |                            |                                |                                       |                             |                          |                                          |
| Adaptive Functioning                                   | .542                       | .648                           | .625                                  | .671                        | .648                     | .653                                     |
| Autistic Symptoms                                      | .545                       | .598                           | .583                                  | .591                        | .611                     | .638                                     |

## **Chapitre V – Discussion générale et conclusion**

## Résumé des principaux résultats empiriques

Cette thèse doctorale comptait trois principaux objectifs : (a) évaluer les effets de l'ICI, telle que dispensée par le CISSS-MO, sur les symptômes autistiques et le fonctionnement adaptatif des enfants qui la reçoivent, (b) explorer la présence de profils distincts chez les participants, selon leur niveau initial de QI, de fonctionnement adaptatif et de symptômes autistiques et leurs potentielles associations à une réponse différentielle à l'intervention, et (c) vérifier si l'apprentissage automatique peut contribuer à l'estimation du pronostic des enfants recevant de l'ICI. Le premier article présentait la trajectoire des symptômes autistiques et du fonctionnement adaptatif chez des enfants qui recevaient de l'ICI. Les résultats révèlent que les symptômes autistiques diminuaient légèrement de façon linéaire tout au long de l'étude. Pour leur part, le fonctionnement adaptatif général et ses trois domaines (conceptuel, social et pratique) montraient un changement non linéaire, caractérisé par une amélioration pendant la période d'intervention et une stabilité pendant la période de suivi. Le domaine pratique progressait cependant moins que les domaines conceptuel et social. Compte tenu de la grande attrition entre le temps 2 et le temps 3, les résultats qui concernaient la période de suivi doivent être interprétés avec prudence. Les symptômes autistiques étaient associés à des améliorations au fonctionnement adaptatif général et aux domaines conceptuel et pratique pendant la période d'intervention, tandis que l'âge au début des services était associé au maintien de ces mêmes acquis. La composante langage du QI était associée au maintien des gains dans le domaine social, alors que le QI de performance était associé au maintien des gains dans le domaine pratique. Les trajectoires du fonctionnement adaptatif et des symptômes autistiques étaient issues d'analyses centrées sur les variables et représentaient les changements moyens dans l'ensemble de l'échantillon. Puisque les résultats des courbes de croissance latente révélaient des variances significatives pour les interceptes (c.-à-d. le niveau initial) pour les symptômes autistiques, le fonctionnement adaptatif

et ses trois domaines, nous avons voulu savoir si des sous-groupes se distinguaient dans notre échantillon.

Le deuxième article explorait la présence de profils distincts chez les participants lors de leur entrée dans les services, basés sur une combinaison de symptômes autistiques, des trois domaines du fonctionnement adaptatif et des trois sous-échelles du QI. Les résultats révèlent la présence de quatre profils : un profil de manifestations légères, deux profils de manifestations intermédiaires et un profil de manifestations sévères. Ces résultats étaient comparables à ceux de Kim et collègues (2016) qui avaient également trouvé quatre sous-groupes avec des combinaisons d'indicateurs similaires. Parmi les variables sociodémographiques, seul le revenu annuel familial prédisait l'appartenance aux profils. En général, nous avons observé des progrès chez tous les profils pendant la période d'intervention, avec des changements d'ampleur variable. Les deux profils présentant les manifestations les plus légères ont réalisé les meilleurs gains sur le plan des symptômes autistiques et des domaines conceptuel, social et pratique du fonctionnement adaptatif. Alors que le premier article révélait que les niveaux initiaux de symptômes autistiques et de fonctionnement adaptatif prédisaient l'amélioration sur certaines variables, les résultats issus des analyses centrées sur la personne suggèrent que c'est la combinaison de ces niveaux initiaux favorables qui est associée à une meilleure efficacité pendant la période d'intervention.

Pendant la période de suivi, les deux profils ayant les manifestations les plus sévères ont maintenu leurs gains ou ont continué de progresser alors que les deux profils ayant les manifestations les plus légères ont présenté une légère diminution des acquis. Ces résultats apportent un éclairage différent à ceux obtenus dans le premier article, adoptant une approche centrée sur les variables, qui révélaient une stabilité du fonctionnement adaptatif pendant la période de suivi. Les résultats du deuxième article permettent de constater que certains enfants

perdent leurs gains, alors que d'autres continuent de progresser pendant la période de suivi. Ce phénomène n'aurait pu être observé sans le recours à un approche centrée sur les personnes. Rappelons que pendant la période de suivi, les enfants ne recevaient plus de services du CISSS-MO et avaient commencé la maternelle. Une hypothèse qui pourrait expliquer cette différence dans le maintien des acquis est que les enfants appartenant aux profils de manifestations plus sévères recevaient davantage de services dans leur classe. Comme nous n'avons pas de données contextuelles pendant la période de suivi, nous ne pouvons vérifier cette hypothèse. De plus, les associations entre les profils et le maintien (ou non) des gains pendant la période de suivi doivent être interprétées avec prudence en raison de la forte attrition entre les temps 2 et 3. En nous appuyant sur l'observation que les quatre profils identifiés dans notre échantillon répondaient de façon différentielle à l'ICI, nous nous sommes demandé s'il était possible d'estimer la réponse à l'intervention en fonction des caractéristiques initiale des enfants, ce qui nous a menés au troisième article.

Le troisième article vérifiait dans quelle mesure les algorithmes d'apprentissage automatique pouvaient soutenir l'estimation du pronostic de réponse à l'intervention chez les enfants recevant de l'ICI, c'est-à-dire distinguer ceux qui progresseront de ceux qui ne progresseront pas, sur la base de leurs caractéristiques initiales. Utiliser les scores globaux des échelles de QI et de fonctionnement adaptatif ou utiliser les scores de leurs sous-échelles comme données entrantes engendrait une précision de prédiction similaire. Tous les algorithmes ont fait de meilleures prédictions que le hasard. La précision de prédiction de nos algorithmes était semblable à celle d'une étude ayant utilisé l'apprentissage automatique pour estimer le pronostic des participants suivant un traitement pour la dépression (Chekroud et al., 2016). Nos résultats soutiennent que l'apprentissage automatique est une avenue prometteuse pour estimer le pronostic des enfants ayant un TSA recevant de l'ICI. Puisque nous sommes à un stade de preuve



de concept, l'apprentissage automatique doit encore démontrer sa validité incrémentielle, notamment en vérifiant si les algorithmes représentent un avantage comparativement aux prédictions faites par des intervenants formés en ICI.

### **Retombées sur la recherche**

Les trois articles empiriques qui forment cette thèse ont permis de faire avancer les connaissances dans le domaine de la recherche évaluative en autisme. Comme mentionné dans l'introduction générale, le contexte québécois d'intervention se distingue sur plusieurs plans de celui des États-Unis, d'où proviennent la majorité des recherches portant sur l'ICI. De plus, la mesure dans laquelle les résultats des études menées en milieu universitaire dans des conditions hautement contrôlées sont comparables aux effets qui seraient observés en communauté reste à établir.

Dans ces circonstances, nos résultats nous informent sur les effets moyens de l'ICI, telle que dispensée par le CISSS-MO. L'intervention semble davantage améliorer le fonctionnement adaptatif que les symptômes autistiques. Les résultats rendent compte que les effets de l'ICI dispensée en contexte québécois sont cohérents avec la récente méta-analyse de la *Cochrane Library* qui conclut que l'ICI améliore le fonctionnement adaptatif et que les preuves actuelles ne permettent pas de soutenir qu'elle améliore les symptômes autistiques (Reichow et al., 2018). À la lumière de ces résultats, nous pensons qu'une réflexion quant à la façon d'évaluer les effets de l'ICI dans la recherche serait bénéfique. Certains auteurs argumentent qu'utiliser des scores bruts provenant d'instruments standardisés pourrait ne pas rendre compte des progrès réalisés par les enfants qui reçoivent l'intervention et suggèrent plutôt d'utiliser des scores d'équivalence d'âge (angl. *age-equivalents*) pour mesurer les effets de l'intervention (Klintwall et al., 2015). Cet aspect méthodologique pourrait expliquer pourquoi certaines études peinent à déceler des effets

sur les symptômes autistiques. D'autres auteurs avancent plutôt que les symptômes autistiques demeurent particulièrement stables tout au long de l'enfance en dépit des services reçus et que les interventions devraient plutôt viser à améliorer le fonctionnement adaptatif (Bieleninik et al., 2017). L'usage des variables latentes pourrait également être une avenue intéressante pour évaluer l'ICI, car elles ont l'avantage de corriger l'erreur de mesure (McArdle, 2009). Une telle réflexion sur la façon d'évaluer les effets de l'ICI pourrait aider à comprendre les résultats contradictoires de certaines études.

Nos résultats représentent également une avancée dans le développement des connaissances concernant l'hétérogénéité des manifestations autistiques et de la réponse à l'ICI. En plus d'identifier des profils similaires à ceux de Kim et collègues (2016) auprès d'une population différente (c.-à-d., des enfants ayant un TSA vivant au Québec), nos résultats sont les premiers à montrer que l'appartenance à des profils particuliers semblent engendrer une réponse différentielle à l'ICI. Les retombées de ces résultats sont importantes sur le plan de la recherche, puisqu'ils confirment la pertinence de considérer l'hétérogénéité des manifestations et l'appartenance à des sous-groupes plus homogènes lors de l'évaluation d'une intervention. Étudier plus précisément les différents profils pourrait éventuellement guider l'individualisation des interventions. Sur le plan du maintien des acquis, nos résultats indiquent qu'en moyenne, les enfants cessent de progresser lorsque l'intervention prend fin. Certains enfants continuent de progresser tandis que d'autres perdent des acquis pendant l'année qui suit la fin de l'intervention. Pour la recherche, cette différence de trajectoire pendant la période de suivi implique de se pencher sur la transition entre les services de réadaptation et le système scolaire pour identifier les facteurs qui favorisent le maintien des acquis. D'autres chercheurs ont également identifié cet enjeu (Starr et al., 2016). Selon une récente revue narrative, la littérature scientifique portant sur la transition vers la maternelle des enfants ayant un TSA s'est surtout concentrée sur la

perception des parents et des enseignants à propos de cette période transition (Girard et al., 2019). Il n'existe cependant pas de recommandations claires pour guider les professionnels quant aux pratiques à mettre en place pour favoriser le maintien des gains réalisés dans le cadre de l'ICI lors de la transition vers le système scolaire (Rivard et al., 2015).

Même si nous sommes seulement à l'étape de la preuve de concepts, nos résultats soulignent le potentiel de l'apprentissage automatique dans la recherche évaluative en autisme. La précision des prédictions effectuées par les différents algorithmes est encourageante, surtout considérant la taille de notre échantillon. Il est possible d'imaginer que dans un futur proche, l'apprentissage automatique pourrait être utilisé de façon complémentaire aux statistiques traditionnelles pour évaluer l'efficacité de l'ICI. Par exemple, l'apprentissage automatique faciliterait en amont l'estimation du pronostic de réponse à l'intervention des participants, ce qui permettrait d'étudier de façon plus précise ceux dont le pronostic est défavorable dans le but de comprendre pourquoi ils répondent moins bien à l'intervention et guider l'individualisation de l'intervention. Dans le même ordre d'idée, il serait possible d'étudier les caractéristiques des enfants qui défient le pronostic de l'algorithme en répondant mieux ou moins bien à l'intervention que ce qui avait été prédit afin de raffiner le modèle de décision.

Au-delà des résultats des trois articles, les stratégies analytiques choisies sont susceptibles d'avoir des retombées positives sur la recherche évaluative en autisme par leur caractère novateur. Les courbes de croissance latente, les analyses de profils latents et l'apprentissage automatique sont des méthodes peu représentées dans ce domaine. Les articles de cette thèse pourraient possiblement encourager les chercheurs à utiliser ces méthodes à leur tour. Diversifier les méthodes permet d'investiguer un sujet de recherche sous divers angles et peut donc aider cumulativement à mieux comprendre le phénomène d'intérêt.

## **Retombées pour la pratique**

Dans l'introduction générale de cette thèse, nous avons expliqué que le contexte québécois d'intervention est particulier, ce qui limite la représentativité des résultats des études d'efficacité de l'ICI aux services publics du Québec. Cette thèse soutient que l'ICI, telle que dispensée par le CISSS-MO, permet d'améliorer le fonctionnement adaptatif et de diminuer légèrement les symptômes autistiques des enfants qui la reçoivent. Ce résultat est encourageant sur le plan clinique, car certaines études avancent que le fonctionnement adaptatif des personnes ayant un TSA peut être significativement altéré tout au long de la vie, et ce, indépendamment de la sévérité des symptômes autistiques et du fonctionnement intellectuel (Kanne et al., 2011). Une intervention précoce permettant d'augmenter le fonctionnement adaptatif a donc le potentiel d'influencer positivement la trajectoire développementale des enfants qui la reçoivent.

En plus de soutenir la pertinence d'offrir des services publics d'ICI, nos résultats fournissent aux praticiens un comparatif pour les aider à situer les progrès des enfants auprès desquels ils interviennent par rapport à la trajectoire moyenne estimée. En complément, nos résultats précisent le changement attendu en fonction du profil initial des enfants. Puisque plusieurs composantes de l'ICI sont individualisées (notamment, les procédures d'enseignement, le type de renforçateur, la procédure de correction de l'erreur), savoir qu'un enfant n'atteint pas le changement attendu pourrait indiquer aux intervenants et aux superviseurs du programme d'ICI de revoir la mise en œuvre de l'intervention pour s'assurer de ne pas garder en place des procédures inefficaces.

Une autre retombée pratique de nos résultats est la possibilité d'offrir une intervention différentielle en fonction de l'appartenance à un profil particulier au moment de l'entrée dans les services. L'ICI est d'emblée une intervention individualisée, mais comme expliquée en introduction, les connaissances pouvant guider cette individualisation sont rares (Pellecchia et al.,

2019; Tirua et al., 2017). L'appartenance à un profil préintervention est un point de départ intéressant. Dans un autre ordre d'idée, le revenu annuel familial est la seule variable, parmi les caractéristiques sociodémographiques investiguées, qui prédisait l'appartenance aux profils. Il est possible que les familles ayant un plus faible revenu aient eu moins de ressources et de soutien pour gérer les manifestations autistiques et favoriser le développement de leur enfant avant leur entrée dans les services publics. Ce résultat met en lumière l'importance de soutenir les familles des enfants en attente d'une évaluation et de services pour ne pas exacerber leurs difficultés.

En lien avec le maintien des acquis, nos résultats suggèrent que certains enfants cessent de progresser, et même perdent des acquis, lorsque l'intervention prend fin. Sachant que la majorité des enfants avait intégré la maternelle pendant la période de suivi, il est possible que le type de soutien offert à l'école influence le maintien ou la perte des acquis. Malgré que nous n'ayons pu vérifier cette hypothèse empiriquement, elle souligne l'importance d'investir dans la transition entre les services de réadaptation et le système scolaire. Considérant que le réseau de la santé et des services sociaux ainsi que le système scolaire sont les milieux qui comptent le plus de psychoéducateurs actifs (respectivement 46,7% et 32,2%; OPPQ, 2020a), il serait souhaitable d'établir des ponts entre ces deux milieux. Une telle collaboration pourrait favoriser la généralisation des acquis de l'enfant à son nouveau milieu, le partage des stratégies d'intervention efficaces et le maintien des gains. La collaboration entre le réseau de la santé et des services sociaux et le milieu scolaire fait d'ailleurs partie des recommandations formulées par l'Ordre des psychoéducateurs et des psychoéducatrices du Québec (OPPQ) dans son Mémoire sur la trajectoire de services destinés aux enfants vulnérables et à leurs familles (OPPQ, 2020b). L'absence de mécanismes officiels et obligatoires pourrait expliquer la faible collaboration entre les différents services lors de la transition (Therrien et Goupil, 2009; Ruel et al., 2016). Une solution pour favoriser la collaboration entre les services de réadaptation et le système scolaire

serait le recours systématique à des plans de transition individualisés et intersectoriels pour les élèves handicapés ou en difficultés d'adaptation et d'apprentissage (Ruel et al., 2015).

Un autre enjeu en lien avec la perte des gains lors de l'entrée à l'école concerne le déploiement graduel de la maternelle 4 ans dans le système scolaire québécois (Gouvernement du Québec, 2021). À l'heure actuelle, la maternelle 4 ans n'est pas obligatoire et le Gouvernement du Québec la considère comme une option qui bonifie les services éducatifs. Sachant que l'âge moyen d'entrée dans les services dans notre échantillon était de 4 ans 4 mois, les enfants dont les parents opteraient pour la maternelle 4 ans n'auraient pas accès à l'ICI, puisque les services de réadaptation ne sont pas typiquement donnés de façon concomitante aux services du système scolaire. Comme nos résultats suggèrent que l'ICI engendre davantage de progrès que la fréquentation scolaire, les parents devraient en tenir compte dans leur prise de décision quant à la maternelle 4 ans.

Une dernière retombée pratique concerne la possibilité d'estimer le pronostic de réponse à l'ICI. Bien que nous sommes encore à un stade préliminaire du développement d'un algorithme utilisable dans la pratique, les résultats soutiennent le potentiel de l'apprentissage automatique pour atteindre cet objectif. Compte tenu du manque de consensus sur les prédicteurs fiables d'efficacité de l'ICI, le développement d'un outil concret qui permettrait de soutenir les intervenants dans l'estimation du pronostic représente une retombée pratique intéressante.

### **Retombées pour la psychoéducation**

De façon générale, le champ d'exercice de la psychoéducation inclut l'évaluation des capacités adaptatives et des difficultés d'adaptation, ainsi que l'élaboration d'un plan d'intervention qui vise à rétablir et développer le fonctionnement adaptatif de la personne (OPPQ, 2021). Bien que les psychoéducateurs soient amenés à intervenir auprès de multiples

clientèles, ils sont largement impliqués auprès des personnes ayant un TSA. En effet, 11,45% d'entre eux travaillent directement au sein de Centre de réadaptation en déficience intellectuelle et en trouble du spectre de l'autisme (OPPQ, 2020a), sans compter tous ceux qui interviennent auprès de personnes ayant un TSA dans d'autres milieux (par exemple, les services éducatifs à l'enfance, le milieu scolaire, les Centres locaux de services communautaires). Étant donné qu'une des cibles d'intervention de l'ICI est l'amélioration du fonctionnement adaptatif, la psychoéducation est une profession de choix pour assurer la mise en œuvre de l'ICI en absence d'analystes du comportement certifiés. Dans le cadre de l'ICI, les psychoéducateurs sont responsables d'évaluer le développement des enfants ayant un TSA, de déterminer le plan d'intervention, de superviser la mise en œuvre de l'intervention et d'en évaluer les effets (Abouzeid & Poirier, 2014). Les résultats de cette thèse ont diverses retombées sur l'exercice de la psychoéducation auprès des enfants ayant un TSA, notamment sur le plan des opérations professionnelles. Huit opérations professionnelles définissent la pratique psychoéducative (Gendreau, 2001), mais les implications de nos résultats concernent majoritairement l'évaluation préintervention et l'évaluation post-situationnelle.

L'évaluation préintervention, telle que définie par Gendreau (2001), comprend l'évaluation en cours d'intervention (ou sur-le-champ) et l'évaluation en retrait. Nos résultats pourraient soutenir l'évaluation préintervention, plus précisément l'évaluation en retrait, qui consiste à analyser les informations préalablement recueillies. Cette opération permet d'établir le potentiel adaptatif de l'individu, c'est-à-dire le portrait de ses capacités et ses difficultés adaptatives, et le potentiel expérientiel de l'environnement, c'est-à-dire les possibilités d'apprentissage offertes par l'environnement à l'individu. Le potentiel adaptatif de l'individu et le potentiel expérientiel interagissent ensemble. Le psychoéducateur proposera une intervention adaptée sur les bases de l'intervention préintervention; elle est donc primordiale. La description

des profils des enfants ayant un TSA, qui sont basés sur une combinaison de caractéristiques individuelles, pourrait aider le psychoéducateur à faire sens des capacités et difficultés adaptatives de l'enfant qui interagissent entre elles, afin d'en tenir compte dans l'établissement du potentiel adaptatif. Dans le même ordre d'idée, les algorithmes d'apprentissage automatique, qui pourraient éventuellement mener au développement d'un outil concret pour estimer le pronostic de réponse à l'ICI, constituent une opportunité d'objectiver l'évaluation du potentiel adaptatif et de fournir une appréciation de la capacité de la personne à répondre aux interventions qui seront proposées. Tant la description des profils que le recours à l'apprentissage automatique pourraient contribuer à l'évaluation du potentiel adaptatif de l'individu, dans l'optique où l'OPPQ (2020c) préconise l'utilisation d'une approche multiméthodes en évaluation. Nos résultats informent également l'évaluation du potentiel expérientiel de l'environnement en ce qui concerne le milieu familial et le milieu scolaire. Ils révèlent que le revenu familial faible est la seule variable investiguée qui était associée aux deux profils d'enfants ayant des manifestations plus sévères et que l'environnement scolaire ne permettait pas à certains enfants de continuer de progresser ou de maintenir les gains réalisés lors de l'ICI. Ces éléments devraient être pris en compte lors de l'évaluation du potentiel expérientiel de l'environnement.

La combinaison des résultats de nos analyses centrées sur les variables et de nos analyses centrées sur la personne a des retombées sur l'évaluation post-situationnelle. L'évaluation post-situationnelle comprend l'évaluation formative, qui consiste à évaluer la pertinence des moyens d'intervention, et l'évaluation sommative, qui consiste à évaluer l'efficacité des interventions de manière objective. Dans le contexte d'une condition neurodéveloppementale comme le que TSA, où les manifestations initiales sont hétérogènes et où la réponse à l'intervention est difficilement prévisible, compléter l'évaluation post-situationnelle peut représenter un défi. Contrairement à d'autres difficultés d'adaptation, le TSA ne se guérit pas et la personne vivra avec certaines



manifestations qui y sont associées tout au long de sa vie. Compte tenu de la grande hétérogénéité qui caractérise la condition, il peut également être difficile de quantifier les progrès réalisés par un individu donné par rapport à son potentiel de changement. Connaître la trajectoire moyenne pour le fonctionnement adaptatif et les symptômes autistiques et savoir comment le profil initial est associé à la réponse à l'intervention offrent des critères plus objectifs pour guider le psychoéducateur dans son exercice de l'opération professionnelle de l'évaluation post-situationnelle.

Nous aurions aimé que cette thèse ait également des retombées sur l'opération professionnelle de la planification. La planification réfère entre autres aux choix des objectifs et des moyens d'intervention en fonction des résultats de l'évaluation préintervention (Gendreau, 2001). Ainsi, offrir une intervention différentielle en fonction des prédicteurs d'efficacité relève de la planification. En absence de données précises et détaillées sur l'ensemble des stratégies employées auprès de chaque enfant dans la cadre de l'ICI et de leur mise en œuvre, il nous est malheureusement impossible d'identifier comment individualiser l'ICI en fonction de l'appartenance à un profil particulier. Dans le même ordre d'idée, nous n'avons pas assez d'information pour comprendre pourquoi certains enfants maintiennent leurs gains lors de leur entrée à l'école, alors que d'autres perdent leurs acquis. L'individualisation de l'ICI et la mise en place de stratégies qui favorisent le maintien des effets de l'intervention en se basant sur des données probantes devraient être des priorités. La poursuite des recherches sur l'efficacité différentielle de l'ICI pourrait éventuellement avoir de retombées sur l'opération professionnelle de la planification.

## Forces et limites de la thèse

Cette thèse compte certaines forces. Plusieurs auteurs identifient la question de l'efficacité différentielle de l'ICI comme primordiale (Eapen et al., 2013; Tiura et al., 2017) : la pertinence du sujet de la thèse est donc sa première force. Pour répondre à cette question, nous avons combiné des approches conceptuelles et statistiques à la fois différentes, complémentaires et novatrices dans le domaine de la recherche évaluative en autisme. Notre premier article, qui adopte une approche centrée sur les variables, présente les résultats de courbes de croissance latente. À notre connaissance, l'usage de courbes de croissance latente auprès d'un échantillon de personnes ayant un TSA se limite à deux études épidémiologiques (Caplan et al., 2019; Simonoff et al., 2020). Comme ces analyses peuvent évaluer efficacement les effets d'une intervention dans les études utilisant des devis pré-post (Mun et al., 2009), elles constituent un outil analytique intéressant pour la recherche évaluative. Le deuxième article adopte une approche centrée sur la personne, qui est particulièrement appropriée pour approfondir les connaissances quant à l'hétérogénéité de la condition. Contrairement aux analyses centrées sur les variables, les analyses centrées sur les personnes permettent de tenir compte des interactions entre les variables (Laursen & Hoff, 2006). Puisque les résultats concernent des personnes (et non des variables), ils sont plus facilement généralisables à la pratique (Magnusson, 1998). Pour sa part, le troisième article recourt à l'apprentissage automatique, qui permet de faire des prédictions sur des données inconnues. La prédiction est un objectif de recherche complémentaire à l'explication des phénomènes qui a été largement sous-investie dans le domaine psychosocial (Yarkoni et Westfall, 2017). Les résultats issus de l'apprentissage automatique montrent néanmoins son potentiel de contribution à la recherche évaluative. Une autre force de cette thèse réside dans la collecte de données. Plusieurs informateurs ont été impliqués : le QI a été mesuré par un assistant

de recherche, la sévérité des symptômes autistiques a été mesurée à la fois par l'éducateur de référence et un parent et le fonctionnement adaptatif a été mesuré par un parent.

La taille de l'échantillon est à la fois une force et une limite de la thèse. Les recherches évaluatives auprès des jeunes enfants ayant un TSA ont souvent de très petites tailles d'échantillon, ce qui limite la portée des résultats. Compter 233 participants est donc une force qui distingue notre thèse des autres études dans le domaine. Cependant, la taille de l'échantillon n'était pas idéale au regard des analyses choisies, notamment parce que des échantillons plus grands produisent des estimations plus fiables pour les courbes de croissance latente (Little, 2013). Néanmoins, des études de simulation évaluant la réduction des erreurs standards pour une taille d'échantillon entre 40 et 500 suggèrent qu'entre 40 et 100, ces erreurs diminuent rapidement, tandis que le taux de réduction des erreurs ralentit passablement entre 100 et 150 (Little, 2013). Nos résultats concernant la période de suivi sont également limités par la grande attrition entre le temps 2 et le temps 3. Cette attrition s'explique probablement par le fait que la grande majorité des enfants de l'échantillon avait intégré le système scolaire lors du troisième temps de mesure. Ces enfants étaient possiblement plus difficiles à retracer pour la passation des outils psychométriques à ce moment. Nos vérifications ont toutefois indiqué que les données manquaient de façon aléatoire (angl. *missing at random*) et donc, que l'utilisation de l'estimateur robuste de vraisemblance maximale était adéquate et pour compléter nos analyses malgré la présence d'une forte attrition (Enders, 2010 ; Little, 2013). La taille d'échantillon a également limité le choix de la méthode de validation des algorithmes d'apprentissage automatique et la précision des prédictions du troisième article. Les recherches futures devraient viser des tailles d'échantillon plus grandes et mettre en place des stratégies de rétention pour limiter l'attrition.

Le nombre de temps de mesure a également limité la portée de nos analyses. Bien que trois temps de mesure permettent de réaliser des analyses de courbes de croissance latente, un

plus grand nombre de temps de mesure aurait été préférable. Par exemple, avoir au moins quatre temps de mesure aurait permis d'estimer une pente quadratique, plutôt que d'avoir recours au modelage par période (angl. *piecewise growth*; Kamata et al., 2013). De plus, avoir plusieurs temps de mesure avant l'intervention aurait permis de mieux contrôler certaines sources d'invalidité, comme la maturation. Les recherches futures devraient prévoir plus de temps de mesure, idéalement plusieurs temps avant, pendant et après la période d'intervention, afin de mieux décrire la trajectoire des symptômes autistiques et du fonctionnement adaptatif.

Le devis de recherche ne prévoyait pas de groupe de contrôle, faisant en sorte que certaines sources d'invalidité ne sont pas contrôlées. Rappelons que le Gouvernement du Québec mandate depuis 2003 les Centres de réadaptation régionaux à offrir l'ICI aux enfants ayant un TSA et que les données utilisées pour ce projet de thèse proviennent directement des services offerts par le CISSS-MO. Malheureusement, rares sont les études d'efficacité de l'ICI qui ont recours à un devis à répartition aléatoire, notamment parce que ces devis sont très demandant à mettre en place et parce que plusieurs considèrent qu'il ne serait pas éthique d'assigner des enfants à un groupe de contrôle pendant une période développementale critique (Matson, 2007). En raison de cette limite, les inférences découlant des résultats de cette thèse doivent être interprétées avec prudence. Dans le même sens, le manque de contrôle sur certaines variables prédictives investiguées limite la portée des inférences sur celles-ci. Par exemple, il est difficile de se prononcer sur le rôle de l'intensité de l'intervention dans l'efficacité différentielle de l'ICI, car l'option d'intensité a été déterminée sur la base des besoins des enfants et des préférences des parents. Les futures recherches devraient tenter de pallier cette limite en utilisant des devis plus rigoureux, tels que le devis avec liste d'attente et le devis régression-discontinuité (Steinbrenner et al., 2020). Ces deux devis seraient cohérents avec le contexte québécois d'intervention, qui accuse malheureusement de longs moins d'attente avant que les enfants accèdent aux services. Le

recours à des devis plus rigoureux permettraient de mieux isoler les variables prédictives, et donc, de mieux comprendre leur influence sur l'efficacité de l'intervention.

Une autre limite réside dans le fait que ni la fidélité de l'implantation de l'intervention ni la qualité des supervisions reçues par les éducateurs n'ont été mesurées. Or, il a été démontré que la fidélité de l'implantation de l'intervention peut influencer son efficacité (DiGennaro et al., 2007; Durlak et DuPre, 2008). Une étude suggère que des erreurs dans l'intégrité du traitement, notamment dans la façon de présenter le renforcement, influenceraient l'efficacité de l'intervention (Bottini et al., 2020). Cette limite est fréquente dans le domaine; une récente revue de la littérature a identifié qu'une seule étude utilisant un devis de groupe a examiné l'impact de la fidélité d'implantation (Brand et al., 2019). Dans le même sens, la qualité de la supervision reçue par les éducateurs influencerait leur implantation de l'intervention (David et al., 2002). Il est très probable qu'au-delà des caractéristiques des enfants que nous avons investiguées, la fidélité de l'implantation soit en partie responsable de l'efficacité différentielle de l'ICI. Les recherches futures devraient prévoir dans leur méthodologie des façons de mesurer la fidélité d'implantation, ce qui permettrait non seulement de comprendre son influence sur l'efficacité de l'ICI, mais aussi de dégager des pistes pour guider l'individualisation de l'intervention. Finalement, il semble important de souligner que le fait de détecter des effets différentiels empiriquement signifie que les différences trouvées sont guidées par les données et non par une théorie. Ainsi, les conclusions issues de telles analyses exploratoires devraient faire l'objet de plus de recherches et la reproduction des résultats sur d'autres échantillons est encouragée (Van Horn et al., 2009).

## Conclusion

Cette thèse visait à mieux comprendre l'efficacité différentielle de l'ICI, telle que dispensée par le CISSS-MO, auprès des jeunes enfants ayant un TSA. Pour atteindre cet objectif, nous avons réalisé plusieurs analyses complémentaires. D'abord, nous avons évalué les trajectoires de changement des symptômes autistiques, du fonctionnement adaptatif et ses trois domaines. Puis, nous avons exploré la présence de sous-groupes plus homogènes dans l'échantillon et leurs associations avec la réponse à l'intervention. Finalement, nous avons vérifié dans quelle mesure l'apprentissage automatique pouvait estimer le pronostic de réponse à l'ICI chez nos participants. Les résultats qui en découlent contribuent au développement des connaissances concernant l'efficacité de l'ICI dispensée par les services publics au Québec et la réponse différentielle à l'intervention. Cette thèse identifie des pistes concernant les prédicteurs d'efficacité de l'ICI, la réponse différentielle à l'intervention en fonction du profil de l'enfant lors de son entrée dans les services et l'estimation du pronostic quant à la réponse à l'ICI. Ces connaissances sont susceptibles de contribuer à l'exercice de la psychoéducation, plus particulièrement les opérations de l'évaluation préintervention, l'évaluation post-situationnelle et éventuellement la planification. Les travaux futurs devraient approfondir ces connaissances en colligeant des données précises et détaillées sur les stratégies d'intervention employées et leur fidélité d'implantation afin d'en comprendre les impacts sur l'efficacité de l'ICI et de guider l'individualisation de l'intervention.

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