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Réorganisation cérébrale chez l'adulte sourd : de la privation à la restauration auditive

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Réorganisation cérébrale chez l'adulte sourd : de la privation à la restauration auditive

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Résumé

On estime que 5 % de la population dans le monde souffre d'une perte auditive handicapante, dont 34 millions d'enfants. Ce déficit perceptif, lorsqu'il survient dès la naissance ou lors des premières années de vie, a de multiples répercussions sur le développement cérébral et neurocognitif. La réorganisation cérébrale ayant cours dans le cerveau des individus privés de l'audition précocement constitue un sujet d'étude très prisé par la communauté scientifique, mais pour laquelle de nombreuses questions restent en suspens. Ainsi, les articles qui composent cette thèse ont pour objectif principal d'améliorer nos connaissances portant sur les mécanismes de réorganisation cérébrale, tant au niveau fonctionnel que structurel afin de mieux comprendre leur implication comportementale chez les individus sourds.

Pour ce faire, nous avons souhaité investiguer, par le biais de l'imagerie par résonance magnétique fonctionnelle, quel était le lien entre les activations cérébrales et les performances comportementales lors d'une tâche portant sur les mouvements biologiques chez des adultes sourds congénitaux, en comparaison à des pairs neurotypiques. L'article 1 révèle que les individus sourds présentent une sensibilité accrue à la reconnaissance du mouvement biologique, et notamment des emblèmes, en comparaison à des individus neurotypique. De plus, cette spécificité comportementale observée uniquement chez les individus sourds, s'accompagne d'un recrutement extensif des régions comprises dans le gyrus temporal supérieur, et tout particulièrement le cortex auditif primaire ainsi que le planum temporale. Nos résultats supportent la présence d'une réorganisation intermodale qui s'exprime par le

recrutement cérébral des régions auditives lors de stimulations visuelles complexes, entraînant une amélioration de la reconnaissance des mouvements biologiques chez les adultes sourds.

Par la suite, nous avons souhaité préciser les mécanismes de réorganisation cérébrale de type structurel. En raison de l'hétérogénéité des résultats rapportés précédemment dans la littérature à propos des changements de matière grise et de matière blanche chez les enfants, les adolescents et les adultes sourds privés de l'audition précocement, la réalisation d'une revue systématique a permis de répertorier l'ensemble des changements structurels obtenus par le biais de diverses techniques d'analyse en imagerie par résonance magnétique. L'article 2 de la présente thèse offre une généralisation des altérations structurelles et intègre une visée clinique à la compréhension de ces changements anatomiques et notamment leur impact sur le développement langagier et neurocognitif.

Mis ensemble, ces résultats contribuent à une meilleure appréciation des changements cérébraux à la suite d'une privation précoce de l'audition. En outre, ils offrent une perspective développementale à ces changements par la description de comportements adaptatifs à la situation de handicap auditif, ainsi que du profil neurocognitif de ces individus, dans le but d'apporter de nouvelles pistes aux stratégies de restauration de l'audition et du langage.

Mots-clés : Surdité, neuroplasticité, neuroimagerie, développement cérébral, langage

Abstract

It is estimated that 5% of the world's population suffers from a disabling hearing loss, including 34 million children. This sensory deficit, when it occurs at birth or in the first years of life, has multiple repercussions on the brain and neurocognitive development. The brain reorganization taking place in the brain of early-deaf individuals is an area of research highly valued by the scientific community but for which many questions remain unanswered. Thus, the main objective of the articles in this thesis is to improve our knowledge of brain reorganization mechanisms, both at the functional and structural levels, in deaf individuals. This will allow a better understanding of their impact on the behavioural adaptations of deaf individuals.

To do this, we investigated, through functional magnetic resonance imaging, the relationship between brain activation and behavioural performance in a task involving biological motions in early-deaf adults, compared to hearing peers. Article 1 reveals that deaf individuals are more sensitive to the recognition of biological motion, including emblems, than hearing individuals. In addition, this behavioural specificity, observed only in deaf individuals, is accompanied by extensive recruitment of the regions included in the superior temporal gyrus, such as the primary auditory cortex but more particularly, the planum temporale. Our results support the presence of intermodal reorganization, which is expressed by brain recruitment of auditory regions during complex visual stimuli, leading to improved recognition of the biological motion in early deaf adults.

On the other hand, we wanted to specify the mechanisms of structural brain reorganization. Due to the heterogeneity of the results previously reported in the literature on changes in grey matter and white matter in early-deaf children, adolescents, and adults, the completion of a systematic review identified all the structural changes obtained through various magnetic resonance imaging analysis techniques. The second article of this thesis offers a generalization of structural alterations. It also integrates a clinical frame to the understanding of these anatomical changes to optimize the language and neurocognitive development of these individuals.

Together, these results contribute to a better appreciation of brain changes following an early hearing loss at both the functional and structural levels. Besides, they offer a developmental perspective to these changes by describing adaptive behaviours and the neurocognitive profile of these individuals, with providing new insights into hearing and language restoration strategies.

Keywords: Deafness, neuroplasticity, neuroimaging, brain development, language

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Chapitre IV

Liste des sigles et abréviations

AAF	*Anterior Auditory Field*
AD	*Axial Diffusivity*
ADHD	*Attention Deficit and Hyperactivity Disorder*
ANOVA	*Analysis of variance*
BA	*Broadmann Area*
BOLD	*Blood-Oxygen-Level-Dependent Imaging*
BRIEF	*Behaviour Rating Inventory of Executive Function*
CI	*Cochlear Implant*
CPT	*Continuous Performance Task*
CT	*Cortical Thickness*
DKI	*Diffusion Kurtosis Imaging*
DTI	*Diffusion Tensor Imaging*
DZ	*Dorsal Zone*
EF	*Executive Functions*
FA	*Fractional Anisotropy*
fMRI	*Functional magnetic resonance imaging*
IFG	*Inferior Frontal Gyrus*
IPL	*Inferior Parietal Lobule*
IQ	*Intelligence Quotient*
MNI	*Montreal Neurological Institute*
MK	*Mean Kurtosis*
ONU	Organisation des Nations Unies
PAF	*Posterior Auditory Field*
pMTG/MT+	*posterior Median Temporal Gyrus*
PRISMA	*Preferred Reporting Items for Systematic Reviews and Meta-Analyses statement*
pSTS	*posterior Superior Temporal Sulcus*
RD	*Radial Diffusivity*

ROI	*Region-of-Interest*
TBM	*Tensor-Based Morphometry*
TEP	Topographie par Émission de Positons
VBM	*Voxel-Based Morphometry*
WISC	*Weschler Intelligence Scale for Children*
WHO	*World Health Organization*

Tous les hommes pensent que le bonheur
se trouve au sommet de la montagne
alors qu'il réside dans la façon de la gravir.

Confucius

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Cette aventure débute en 2012 dans un temple dédié à la déesse des marins sur l'île d'Enoshima, au Japon. J'y fais un vœu, celui de réaliser un doctorat dans le domaine de la surdité. Me voici à présent aux dernières lignes de ce projet sur le point de se concrétiser.

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Introduction générale

La perte auditive qui survient précocement, soit au cours du développement intra-utérin ou de la période prélinguale, perturbe la maturation du système auditif et son efficience dans le traitement, la discrimination et la reconnaissance des sons de l'environnement. En raison de la proximité anatomique entre les régions cérébrales impliquées dans le traitement de l'audition et du langage, la perte auditive précoce a des répercussions sur l'acquisition et, conséquemment, sur la maîtrise des habiletés langagières des individus sourds. Selon le célèbre neurologue Oliver Sacks (1990) : « La surdité en tant que telle n'est pas une catastrophe ; les désastres ne commencent que lorsque les échanges communicationnels et langagiers sont entravés. Si aucune communication ne peut être établie, si l'enfant n'est pas exposé à un langage ou à un dialogue satisfaisant, on verra se succéder toutes sortes de difficultés d'ordre linguistique, intellectuel, psychoaffectif et culturel qui assaillent à des degrés divers la majorité des sourds de naissance ». Dès lors, la restauration auditive et la rééducation de la parole sont deux entités indissociables l'une de l'autre, et constituent le cœur de ce champ d'études. En outre, elles font l'objet de débats incessants au sein des communautés scientifiques, médicales, pédagogiques, et de la communauté sourde.

Les enjeux reliant étroitement la perte auditive et le langage trouvent leur origine au fil d'évènements historiques notables. En premier lieu, la création au XVIII^{e} siècle d'un langage signé par l'abbé Charles-Michel de l'Épée influence le sort des individus sourds qui étaient auparavant considérés comme retardés cognitivement, inaptes à acquérir le langage et à s'intégrer dans la société. À partir de cette découverte, les langues des signes connaissent un essor mondial fulgurant puisque maints pays instaurent ce moyen de communication dans les pratiques pédagogiques des écoles spécialisées pour enfants sourds. En 1880, lors du Congrès de Milan, des centaines de professionnels spécialisés dans l'enseignement auprès d'enfants

sourds débattent des meilleures pratiques, la méthode orale exclusive est alors imposée, les professeurs de langue des signes sont renvoyés des écoles et la langue des signes est proscrite. Ce fait majeur a marqué des générations d'individus sourds qui, privés d'une langue qu'ils considèrent naturelle, se sont retrouvés soudainement sans moyen optimal de communication. À cette époque, l'instauration de la méthode orale est influencée par les grandes avancées scientifiques dans le domaine de la médecine et de la linguistique du XIX^{e} siècle. La quantification et la classification des pertes auditives à l'aide de mesures audiométriques médicalisent la condition de surdité. De plus, les études des localisations cérébrales par Franz Joseph Gall, ou encore des lésions par Paul Broca, font basculer la surdité dans la catégorie des pathologies dites du langage (Virole, 2000). À la suite des travaux pionniers sur l'aphasie, les gestes et les mimiques, qui composent en partie les langues des signes, sont distingués de toutes habiletés langagières puisque les individus cérébrolésés au niveau des aires du langage demeuraient capables de produire des gestes. La méthode orale dominera tous les principes pédagogiques et rééducatifs des enfants sourds pendant plus de cent ans. Vers les années 1980, un nouveau virage s'effectue et les langues des signes sont progressivement revalorisées, notamment par les travaux de William Stokoe sur *l'American Sign Language* aux États-Unis (Ducharme & Mayberry, 2005). En 2006, l'Organisation des Nations Unies (ONU) reconnaît aux langues des signes le même statut que les langues parlées et souligne l'importance de la langue des signes pour l'éducation des enfants sourds. De nos jours, les méthodes de rééducation privilégiées pour les enfants sourds reposent sur un dépistage précoce de la surdité, une restauration de l'audition par le port de prothèses auditives ou de l'implant cochléaire et l'apprentissage du langage selon la méthode auditivo-verbale, complétée

éventuellement par le recours à la langue des signes ou à des systèmes visuomanuels d'aide à la lecture labiale (Transler, Leybaert, & Gombert, 2005).

En 2019, l'ONU estime que 5 % de la population mondiale présente une perte auditive handicapante, comprenant 34 millions d'enfants. Le programme universel de dépistage de l'audition chez le nouveau-né constitue une mesure sensible et spécifique pour détecter les déficiences auditives congénitales chez l'enfant et présente une influence positive sur le développement des habiletés langagières (par ex. Nelson, Bougatsos, & Nygren, 2008). Toutefois, l'absence de dépistage systématique dans certains pays, et ce, même dans certains pays développés (d'après la Société Pédiatrique Canadienne, 2019 : seuls 30% des nouveau-nés québécois bénéficient du dépistage, contre 94% en Ontario) ou le manque d'examens complémentaires puis de suivi après un dépistage positif, représentent encore des faiblesses majeures de ce programme (Wroblewska-Seniuk, Dabrowski, Szyfter, & Mazela, 2017). Une fois la perte auditive dépistée, seuls 10% des besoins de prothèses auditives dans le monde sont comblés. En contrepartie, le nombre d'enfants sourds ayant reçu un implant cochléaire a connu une nette augmentation. Après quatre décennies, l'implant cochléaire est reconnu internationalement comme le moyen le plus efficace et le plus fiable dans la restauration auditive des déficiences auditives sévères à profondes survenant avant l'âge de trois ans (Sharma & Campbell, 2011; Yawn, Hunter, Sweeney, & Bennett, 2015). L'implant cochléaire présente des bénéfices certains pour le développement des habiletés auditives et langagières, mais également en termes de réussite académique, de qualité de vie et enfin, d'insertion professionnelle des individus sourds (Vincenti et al., 2014). En 2007, des enquêtes statistiques démontraient que 4200 Canadiens sourds étaient porteurs d'un implant cochléaire, dont 1710 enfants (Fitzpatrick & Brewster, 2010). Notons toutefois que ces chiffres représentent

seulement 33 personnes implantées par million au Canada, pour 61 personnes par million en Angleterre et 180 personnes par million pour l'Australie (Fitzpatrick & Brewster, 2010). De plus, entre 20 à 40% des enfants sourds présentent des comorbidités associées à la déficience auditive (Berrettini et al., 2008), dont de multiples troubles neurodéveloppementaux (par ex. : trouble du spectre de l'autisme : 4 %, trouble des apprentissages : 7.20 %, retard global du développement : 8.80 %, trouble déficitaire de l'attention : 5.4 %, trouble primaire du langage : 4 à 6 %) (Cejas, Hoffman, & Quittner, 2015; Hawker et al., 2008). Ces doubles conditions retentissent sur l'efficacité de la restauration auditive, tout particulièrement avec un implant cochléaire, et complexifient les interventions auprès de ces enfants (Birman, Elliott, & Gibson, 2012; Young, Weil, & Tournis, 2016).

Cet aperçu statistique sur les individus, et notamment les enfants, souffrant d'une perte auditive permet d'illustrer les défis cliniques persistants autour de la prise en charge de la déficience auditive, allant du dépistage à l'intervention spécifique. Depuis quelques années, un consensus émerge dans la littérature et supporte l'hypothèse que la réorganisation cérébrale à la suite d'une privation sensorielle précoce serait le mécanisme à l'origine des contraintes actuelles de la restauration auditive, notamment par l'implant cochléaire. Par exemple, Lazard, Giraud, Truy, & Lee (2011) explicitent que « l'organisation fonctionnelle du cerveau pourrait prédire plus efficacement le niveau de récupération auditive obtenu post-implantation, et ce par rapport aux variables cliniques connues telles que la durée de la surdité, l'étiologie de la surdité, les facteurs liés à la chirurgie et bien d'autres encore ». Par conséquent, il apparaît crucial de comprendre les changements qui surviennent dans le cerveau de ses individus et de ses enfants afin de contribuer au développement de nouvelles stratégies d'intervention plus appropriées et efficaces.

Le présent projet de thèse s'inscrit dans cette voie et vise une meilleure compréhension des mécanismes de plasticité cérébrale à la suite d'une perte auditive. Par le biais de l'imagerie par résonance magnétique fonctionnelle (IRMf), le premier objectif sera d'examiner la réorganisation cérébrale sur le plan fonctionnel. Pour s'adapter à ce handicap sensoriel, les individus sourds démontrent d'une plus grande sensibilité visuelle en comparaison à des pairs neurotypiques, notamment sur le plan de la détection du mouvement. Cette sensibilité est d'autant plus essentielle pour les personnes sourdes dont le moyen de communication préférentiel est la langue des signes. Par conséquent, nous souhaitons savoir si les individus sourds se montrent également plus sensibles à la détection du mouvement biologique, soit une forme simplifiée des gestes humains. La relation entre les performances comportementales et les régions cérébrales activées par le mouvement biologique sera discutée entre les personnes sourdes et leurs pairs neurotypiques, ainsi qu'entre les individus sourds signeurs et oralistes.

Le second objectif vise à détailler les mécanismes de réorganisation cérébrale sur le plan structurel. Le cerveau est un organe sensible à l'expérience, par conséquent l'absence d'afférence auditive entraîne des modifications anatomiques. Une revue systématique de la littérature nous permettra de répertorier les principales modifications de matière blanche et de matière grise associées à la privation auditive. Ces altérations cérébrales seront discutées par la suite sous l'angle de la restauration auditive, du développement neurocognitif, de la rééducation du langage et de l'intervention.

Afin d'introduire les articles qui composent cette thèse, nous commencerons par une mise en contexte théorique de la plasticité cérébrale à la suite d'une perte sensorielle. Nous

poursuivrons par un état des lieux de la littérature portant sur la plasticité cérébrale appliquée à la surdité et son impact sur la réorganisation cérébrale de type fonctionnel puis structurel.

Chapitre I : Contexte théorique

1. Influence de l'expérience sur la plasticité cérébrale

Le cerveau de l'homme est vital, pour en comprendre son rôle et son fonctionnement, nos ancêtres préhistoriques effectuaient des trépanations de la boîte crânienne sur des pairs encore vivants (Bear, Connors, & Paradiso, 2007). De nos jours, l'émulation des neurosciences pour percer le mystère de cet organe ne cesse de s'accroître au fil des avancées scientifiques. Les neurosciences qui allient entre autres, la neuropsychologie, science qui étudie la relation entre le cerveau neurotypique ou atypique et le comportement, avec des techniques de neuroimagerie, rendent possible l'observation du cerveau *in vivo.* Les neurosciences ont ainsi révélé la fascinante capacité du cerveau humain, de la naissance à la fin de sa vie, à se modifier sous l'effet de l'expérience par le biais de changements morphologiques et fonctionnels (Pascual-Leone, Amedi, Fregni, & Merabet, 2005). Ces changements sont classiquement définis comme de la plasticité cérébrale, soit la capacité du cerveau à se réorganiser par lui-même à la suite d'évènements, qu'ils soient adverses (par ex. : lésion cérébrale, traumatisme craniocérébral, malformation congénitale, dégénérescence) ou favorables (par ex. : expertise musicale, entraînement, bilinguisme).

Les sens, et notamment la vision et l'audition, constituent les fonctions de base du cerveau, se développent prioritairement en raison de leurs rôles contributifs à toutes les autres fonctions de plus haut niveau et sont les premières à atteindre la maturité corticale (Gogtay et al., 2004; Grantham-McGregor et al., 2007). Par conséquent, l'impact du développement sensoriel, et tout particulièrement, l'absence d'afférence sensorielle sur la maturation du cerveau, thème crucial de la présente thèse, apparaît une voie privilégiée pour rendre compte de la réorganisation cérébrale. Toutefois, l'étude isolée de la réorganisation cérébrale à la suite

d'une privation sensorielle ne peut suffire à en comprendre sa fonction ou les processus qui la sous-tendent, il importe plutôt de mettre en relation les données d'imagerie fonctionnelle et structurelle avec des mesures comportementales (Voss & Zatorre, 2012). Dans les prochaines sections, nous dresserons un bref résumé des données probantes en termes de plasticité cérébrale et de comportements chez les individus privés de la vision et chez les animaux, puisqu'elles représentent le socle théorique et méthodologique du champ de la privation auditive.

1.1 Application de la plasticité cérébrale à la privation sensorielle

La vision populaire de la privation sensorielle est que les individus privés d'un sens compensent leur handicap par la présence de capacités sensorielles affinées dans les modalités sensorielles préservées. Ces capacités sensorielles améliorées sont magnifiées chez les individus aveugles congénitaux, qui pour certains, possèdent par exemple des talents musicaux exceptionnels. En outre, l'hypothèse communément admise pour expliquer la présence de ces capacités sensorielles améliorées est que la région cérébrale privée de ses afférences sensorielles primaires est récupérée par une ou plusieurs autres modalités sensorielles préservées, soit la plasticité intermodale (Pascual-Leone et al., 2005). De plus, l'étendue de cette récupération est directement liée aux comportements dits compensatoires ou adaptatifs à la situation de handicap sensoriel (Voss, Pike, & Zatorre, 2014).

Ainsi, les évidences scientifiques en faveur de capacités sensorielles améliorées dans les modalités sensorielles préservées des personnes souffrant d'une cécité congénitale sont abondantes et robustes. Citons à titre d'exemples, l'observation de meilleures habiletés comportementales chez les individus aveugles comparativement aux individus neurotypiques

au sein de la modalité tactile, en termes d'acuité tactile fine (Alary et al., 2009) et de la modalité auditive, en ce qui a trait à la discrimination du pitch (Gougoux et al., 2004) et à la localisation spatiale du son (pour une revue exhaustive de la littérature, voir Silva et al., 2018). De plus, de nombreuses études en IRMf ont démontré un lien de causalité entre une augmentation de l'activation cérébrale du cortex occipital lors de stimulations auditives et tactiles chez les individus aveugles et leurs capacités sensorielles améliorées (par ex. Collignon, Lassonde, Lepore, Bastien, & Veraart, 2007; Gougoux, Zatorre, Lassonde, Voss, & Lepore, 2005; Voss, Gougoux, Zatorre, Lassonde, & Lepore, 2008). Au-delà des modalités sensorielles, des auteurs ont également démontré un recrutement du cortex occipital lors de tâches cognitives de haut niveau chez les individus privés précocement de la vision. Ainsi, les individus aveugles obtenaient de meilleurs résultats lors de tâches évaluant la mémoire verbale (Amedi, Raz, Pianka, Malach, & Zohary, 2003) ou encore, la fluidité verbale (Burton et al., 2002) en comparaison à des pairs neurotypiques. D'importance pour le second objectif de cette thèse, il a également été démontré que la réorganisation structurelle du cortex occipital qui survient chez les individus aveugles, c'est-à-dire les modifications en termes de matières blanches et grises, sont prédictives des habiletés comportementales obtenues lors de tâches non visuelles (Voss et al., 2014).

Sur le plan clinique et notamment en ce qui concerne la restauration et la rééducation à la suite d'une privation sensorielle, le champ de la cécité s'avère, sur ce plan, plus restreint que celui de la surdité. Ceci peut s'expliquer par l'absence de procédure systématique de restauration visuelle, notamment en raison de l'organisation particulièrement complexe de la rétine en comparaison à celle de la cochlée (Heimler, Weisz, & Collignon, 2014). Toutefois, la restauration visuelle a connu de très grands progrès dans les dernières années avec

l'émergence des prothèses rétiniennes (pour une revue récente, voir Beyeler & Fine, 2017). Néanmoins, l'absence de stimulations visuelles précoces demeure une contre-indication à l'implantation (Gabel, 2017) supportant cette fois-ci l'hypothèse que la réorganisation cérébrale peut se définir, non plus comme étant adaptée mais inadaptée (Heimler et al., 2014). Le caractère inadapté de la plasticité cérébrale dans le contexte de la privation sensorielle fait référence au fait que l'absence de stimulations sensorielles précoces entraîne une réorganisation qui nuit aux possibilités de restauration puisque les afférences sensorielles normalement dédiées à la fonction atteinte sont réallouées aux autres modalités sensorielles.

En guise de conclusion à cette section, Heimler et collaborateur (2014) explicitent que les études comportementales et de neuroimagerie chez les aveugles précoces ont permis d'établir plusieurs postulats portant sur le caractère adaptatif de la plasticité intermodale à la suite d'une privation sensorielle : 1) la plasticité intermodale est intrinsèquement liée aux comportements ; 2) la plasticité intermodale soutient les comportements dits adaptatifs, ou les capacités sensorielles améliorées de ces individus dans les modalités sensorielles préservées, tant au niveau d'une réorganisation fonctionnelle que structurelle ; 3) la plasticité intermodale dans le cortex occipital est fonctionnellement organisée selon les principes qui régissent le système visuel (pour détails sur le recrutement différencié des voies visuelles ventrale et dorsale par les autres modalités sensorielles et selon les tâches, voir Bola et al., 2017). En dépit de ces postulats communément acceptés par la communauté scientifique, de nombreuses énigmes demeurent à ce jour. Nous pouvons citer la présence d'incohérences sur le plan des répercussions comportementales, soit l'obtention par les individus aveugles d'habiletés comparables ou moins bonnes que les individus neurotypiques lors de certaines tâches auditives ou somesthésiques (pour une revue de la littérature, voir Singh, Phillips, Merabet, &

Sinha, 2018), ou encore par le fait que certains des individus aveugles ne présentent pas tous la même trajectoire sur le plan de la réorganisation cérébrale (par ex. une absence d'activation intermodale, Gougoux et al., 2005). Finalement, de nombreuses questions demeurent concernant le caractère inadapté de la réorganisation cérébrale pour la restauration optimale de la fonction atteinte. L'ensemble de ces éléments seront repris dans le cadre de la discussion générale de cette thèse.

1.2 Le cas spécifique de la perte auditive : réorganisation cérébrale fonctionnelle

Comme il a été démontré chez les individus souffrant d'une cécité congénitale, l'adaptation quotidienne à leur handicap sensoriel repose, en partie, sur leurs modalités sensorielles préservées. Par conséquent, de nombreux chercheurs se sont intéressés à l'identification de capacités sensorielles accrues chez les individus sourds en comparaison à des individus neurotypiques. Les données accumulées restent plus inconsistantes que celles obtenues chez les personnes aveugles (Pavani & Röder, 2012). Nous discuterons des contraintes spécifiquement liées à la recherche auprès des individus sourds, qui sont parmi les explications très souvent rapportées à ces divergences comportementales lors de la discussion.

Depuis une trentaine d'années, les évidences les plus consistantes à travers la littérature s'accumulent et concernent la modalité visuelle. Les protocoles comportementaux et d'imagerie cérébrale chez les individus sourds ont été largement influencés par la recherche chez l'animal, et notamment chez le chat, il apparaît donc nécessaire à ce stade d'en réaliser un survol. Ainsi, le traitement visuel chez des chats sourds congénitaux a été comparé à celui de chats neurotypiques lors de diverses tâches (Lomber, Meredith, & Kral, 2010). Les résultats

de Lomber et collaborateurs démontrent un avantage comportemental des chats sourds lors de la détection du mouvement visuel et de la localisation spatiale périphérique. En revanche, aucune différence n'était retrouvée entre les deux groupes de chats lorsque ceux-ci devaient discriminer le mouvement selon la direction et la vélocité, ou encore sur le plan de l'acuité visuelle. De plus, les résultats de cette étude établissent un lien de causalité entre les avantages comportementaux mentionnés ci-haut et une réorganisation intermodale de certaines régions auditives spécifiques chez le chat (PAF : *posterior auditory field* ; DZ : *dorsal zone*) ne comprenant toutefois pas le cortex auditif primaire. Une étude subséquente démontre que chez le chat sourd, la région nommée *anterior auditory field* (AAF) est largement recrutée par des stimulations non auditives de type somatosensoriel et dans une moindre mesure par des stimuli visuels (mouvement et vélocité) (Meredith & Lomber, 2011). Par conséquent, le recrutement des régions normalement impliquées dans le traitement auditif par des stimuli non auditifs questionne sur l'influence respective des régions sensorielles primaires. Pour tenter de répondre à ces questions, des chercheurs se sont intéressés à la nature des connexions entre les régions sensorielles primaires (auditive : A1, visuelle : V1 et somatosensorielle : S1). Dans une récente revue de la littérature, Meredith et Lomber (2017) rapportent une absence de connexion anatomique directe entre les régions sensorielles primaires, mais la présence de projections corticales de plus haut niveau sur ces régions. Toutefois, le modèle animal souffre encore de faiblesses alors même que Meredith et Lomber (2017) déterminent des divergences en termes de connectivité entre différentes espèces animales, notamment entre les rongeurs et les mammifères (chats, furets). Il apparaît donc encore complexe d'extrapoler les données animales sur le modèle humain.

1.2.1 Les habiletés visuelles primaires

Chez l'homme, les régions cérébrales associées à l'audition incluent le cortex auditif primaire (BA 41), qui comprend le gyrus de Heschl, le cortex auditif secondaire (BA 42) et le cortex auditif associatif, qui inclut le planum temporale (BA 22) (pour une représentation visuelle et une comparaison interespèces, voir Alencar et al., 2019). D'un point de vue fonctionnel, le cortex auditif primaire a pour rôle d'analyser tous les types de sons alors que les cortex secondaire et associatif, et notamment le planum temporale sont impliqués dans le traitement catégoriel et serviraient de centre computationnel permettant le transfert des informations auditives aux aires cérébrales de plus haut niveau. Bien que ces régions soient principalement impliquées dans le traitement de l'audition, de nombreuses études réalisées chez l'homme ont démontré une influence des autres sens sur le cortex auditif primaire et associatif, notamment en termes d'intégration multisensorielle (par ex. Bizley, Nodal, Bajo, Nelken, & King, 2006).

À l'instar des études préalablement décrites chez les chats, les travaux de recherche se sont majoritairement concentrés sur la plasticité intermodale des régions auditives en réponse à des stimuli visuels, à l'exception de quelques études portant sur la modalité somatosensorielle (Auer, Bernstein, Sungkarat, & Singh, 2007; Karns, Dow, & Neville, 2012; Levänen & Hamdorf, 2001). Ainsi, les individus sourds congénitaux ont été soumis à de nombreuses tâches de traitement visuel primaire afin de déterminer des convergences comportementales. Les résultats hautement hétérogènes obtenus, s'expliquant notamment par la multitude de procédures méthodologiques différentes, suggèrent néanmoins une absence de différence comportementale entre les individus sourds et neurotypiques en termes de discrimination (orientation, brillance), de sensibilité aux contrastes, d'acuité visuelle et sur le

plan du mouvement (direction, vélocité) (pour une revue de la littérature détaillée, Alencar, Butler, & Lomber, 2019). Le processus visuel présentant le plus grand consensus correspond, quant à lui, à la diminution du seuil de détection du mouvement visuel chez les sourds de points en mouvement (Bosworth & Dobkins, 2002; Hauthal, Sandmann, Debener, & Thome, 2013; Neville & Lawson, 1987; Shiell, Champoux, & Zatorre, 2014a). En ce qui a trait à la réorganisation cérébrale, des activations dans les régions du cortex auditif chez les sourds sont principalement rapportées à la suite de stimuli visuels en mouvement, supportant l'hypothèse compensatoire d'une réorganisation cérébrale de type intermodal (Fine, Finney, Boynton, & Dobkins, 2005; Finney, Clementz, Hickok, & Dobkins, 2003; Sadato et al., 2005; Vachon et al., 2013). De plus, deux récentes études ont démontré des corrélations entre ces compétences comportementales sensorielles améliorées en termes de détection du mouvement avec l'épaisseur corticale du planum temporale droit (Shiell, Champoux, & Zatorre, 2016) ainsi que l'intégrité de la matière blanche de cette région auditive (Shiell & Zatorre, 2016). Ces résultats sont importants puisqu'ils confirment l'importance d'étudier les changements de réorganisation fonctionnelle à la lumière de la réorganisation structurelle. Finalement, une réorganisation de type intramodal est également rapportée dans quelques études (Bottari et al., 2014; Hauthal et al., 2013), citons par exemple une amélioration comportementale liée à une plus grande activation du complexe MT+ (V5) chez les individus sourds, soit l'une des régions cérébrales spécialisées dans le traitement visuel du mouvement (Bavelier et al., 2001).

À l'instar des chats sourds, l'ensemble des régions appartenant au cortex auditif semble concerné par la réorganisation intermodale lors d'une perte auditive, sans qu'il soit néanmoins possible de les associer fonctionnellement à des comportements (Bola et al., 2017). En effet, des activations corticales sont retrouvées à la suite de stimulations visuelles de bas niveau au

sein du cortex auditif primaire (Fine, Finney, Boynton, & Dobkins, 2005; Finney & Dobkins, 2001), du cortex auditif secondaire et du cortex auditif associatif (Cardin et al., 2013; Fine et al., 2005; Finney & Dobkins, 2001; Vachon et al., 2013).

1.2.2 La langue des signes

Considérant les propriétés visuelles, et notamment le mouvement, qui composent la langue des signes, de nombreuses études se sont naturellement intéressées aux régions cérébrales impliquées dans des tâches de perception de la langue signe chez les individus sourds. Bien que la langue des signes repose sur la modalité visuelle et la motricité manuelle, il est admis que son acquisition en termes de jalons développementaux ne diffère pas des langues auditivo-verbales chez les sourds natifs (Petitto, Holowka, Sergio, & Ostry, 2001; Petitto & Marentette, 1991), c'est-à-dire ayant grandi au sein d'une famille dont les parents étaient sourds et qui avaient pour langue première la langue des signes. Les langues des signes possèdent des propriétés linguistiques complexes à l'instar des langues auditivo-verbales, telles que la phonologie, la syntaxe et la sémantique (Sandler & Lillo-martin, 1999). Par conséquent, les régions impliquées dans le traitement de la langue des signes recouvrent largement celles des langues auditivo-verbales (MacSweeney, Capek, Campbell, & Woll, 2008; Newman, Supalla, Fernandez, Newport, & Bevelier, 2015). Une récente méta-analyse démontre que la compréhension de la langue des signes chez les individus sourds est associée à des activations bilatérales du gyrus frontal inférieur (BA 44, 45), du gyrus temporal supérieur postérieur (BA 22, planum temporale), du cortex prémoteur (BA 6 et 8) (Trettenbrein, Papitto, & Zaccarella, 2019). Toujours selon ces mêmes auteurs, des activations sont également rapportées dans l'insula et en lien avec la modalité visuelle, notamment au sein du gyrus fusiforme (BA 37) et du gyrus occipital médian (BA 19).

En lien avec la réorganisation cérébrale, plusieurs auteurs ont démontré une activation des régions auditives par la langue des signes chez les individus sourds en l'absence d'activation similaire chez des pairs neurotypiques (par ex. Fine et al., 2005; Nishimura et al., 1999; Petitto et al., 2000). À notre connaissance, seule une étude en IRMf rapporte une activation du cortex auditif primaire par la langue des signes chez les individus sourds (Finney et al., 2003). En ce qui a trait au planum temporale, soit le cortex auditif associatif, les activations cérébrales retrouvées dans cette région lors de tâches en langue des signes seraient spécifiques aux individus sourds et sont interprétées comme étant un phénomène de plasticité intermodale, soit le traitement fonctionnel des propriétés visuelles de la langue des signes par cette région (Cardin et al., 2013; MacSweeney et al., 2002; Petitto et al., 2000; Sadato et al., 2005). Néanmoins, les interprétations quant à la fonction de cette activation cérébrale souffrent de divergences. Pour certains auteurs, le planum temporale deviendrait une région unimodale spécifique au traitement de la vision sous l'influence de la privation auditive (MacSweeney et al., 2002), alors que pour d'autres, il s'agirait plutôt d'un rôle d'intégration multimodale lié à l'influence du mode de communication et tout particulièrement de la langue des signes (Sadato et al., 2005). Plus récemment, Cardin et collaborateurs (2013) suggèrent une latéralisation de cette activation corticale, ainsi le planum temporale au sein de l'hémisphère droit desservirait un traitement des propriétés visuelles (activation corticale tant chez les sourds signeurs natifs de la langue des signes que les sourds dits oralistes) alors que le planum temporale gauche desservirait le traitement linguistique de la langue des signes (activation uniquement chez les sourds signeurs).

1.2.3 Le mouvement biologique

L'un des champs d'études chez les individus sourds en lien avec leurs habiletés visuelles correspond à la perception du mouvement biologique. Cet intérêt trouve son origine, car les individus sourds, comme nous l'avons vu précédemment, dépendent de façon accrue sur leur capacité de perception visuelle du mouvement pour encoder leur environnement. Le mouvement biologique s'avère ainsi un sujet de recherche pertinent à explorer chez les personnes souffrant d'une perte auditive.

En 1973, le terme de mouvement biologique a été théorisé pour la première fois par Johansson. Il définit le mouvement biologique comme la perception visuelle de séquences gestuelles qui caractérisent les êtres vivants (homme ou animal, par ex. la marche). La particularité du mouvement biologique est qu'il peut être facilement identifiable sur la base de quelques points lumineux placés aux articulations principales du corps humain (Johansson, 1973), permettant ainsi d'isoler les informations visuelles minimales nécessaires à la reconnaissance de l'action humaine (Zaini, Fawcett, White, & Newman, 2013). De nombreux chercheurs se sont prioritairement intéressés au mouvement biologique de la marche (par ex. Giese & Poggio, 2003) puis, les mouvements biologiques se sont considérablement diversifiés et complexifiés au fil des études. Parmi la diversité des mouvements biologiques existants, nous nous intéresserons particulièrement à deux types d'entre eux : les pantomimes et les emblèmes. Les pantomimes se définissent comme des gestes transitifs dont l'objectif est orienté vers un objet, une action ou un évènement (Arbib, 2004). Les emblèmes, quant à eux, se définissent comme des gestes intransitifs et correspondent à des actions non verbales dont l'objectif est de transmettre une information à une autre personne (Goldin-Meadow, 1999).

Les emblèmes sont des gestes conventionnels (Ozyurek, 2012) qui sont influencés culturellement (Molnar-Szakacs, Wu, Robles, & Iacoboni, 2007), l'un des exemples les plus connus est « le pouce levé » voulant signifier « Ok ». Ces deux mouvements sont d'intérêt puisque la distinction entre les deux permet de comparer les gestes selon qu'ils aient pour objectif de transmettre une information ou non (Zaini et al., 2013).

Du point de vue neuro-anatomique, les régions cérébrales impliquées dans l'observation du mouvement biologique sur la base de points lumineux sont semblables à l'observation d'actions humaines (Saygin, Wilson, Hagler, Bates, & Sereno, 2004). Historiquement, le réseau cérébral dédié au traitement de l'action (mouvement biologique, action humaine) faisait référence au réseau des neurones miroirs (pour en revue récente voir, Rizzolatti & Sinigaglia, 2016). Ces neurones miroirs découverts initialement chez le macaque ont la particularité de s'activer simultanément lors de la réalisation motrice de l'action, mais aussi, lors de la simple observation d'un geste par un pair. Ce réseau chez l'homme comprenait le lobule pariétal inférieur (Inferior Parietal Lobule, IPL), le cortex prémoteur ventral (Ventral Premotor Cortex, PMv) ainsi que le gyrus inférieur frontal (Inferior Frontal Gyrus, IFG), régions correspondant aux homologues cérébraux du macaque (Fabbri-Destro & Rizzolatti, 2008). Depuis, il est communément admis que le réseau cérébral de l'observation de l'action chez l'homme est largement plus étendu que les régions précédemment citées (Caspers, Zilles, Laird, & Eickhoff, 2010). Dans une méta-analyse reprenant 139 études de neuroimagerie sur le réseau de l'action humaine, plusieurs régions cérébrales ont été additionnées à ce réseau : le gyrus inférieur frontral (IFG, BA 44/45), le cortex prémoteur dorso-latéral et l'aire motrice supplémentaire (BA 6), le lobule pariétal inférieur (IPL), le cortex somatosensoriel primaire (BA 1/2), le sillon temporal supérieur postérieur (pSTS), le cortex intrapariétal (IPS), la région

qui associe le gyrus temporal médian postérieur (pMTG) à l'aire visuelle supplémentaire V5 et les aires du gyrus fusiforme impliquées dans le traitement des visages et du corps (Caspers et al., 2010).

L'étude du mouvement biologique chez l'individu sourd a un intérêt sur le plan adaptatif. Comme nous l'avons vu précédemment, les individus sourds présentent une sensibilité accrue pour la détection de cibles visuelles en mouvement (Bosworth & Dobkins, 2002; Hauthal et al., 2013; Neville & Lawson, 1987; Shiell et al., 2014a). Cette sensibilité visuelle est expliquée par un phénomène de plasticité intermodale soit l'activation du cortex auditif primaire et secondaire par des stimuli visuels (Fine, Finney, Boynton, & Dobkins, 2005; Finney, Clementz, Hickok, & Dobkins, 2003; Sadato et al., 2005; Vachon et al., 2013). Il semble alors intéressant d'explorer si les individus sourds présentent un avantage comportemental lors de la reconnaissance de cibles visuelles en mouvements plus complexes, tels que le mouvement biologique et si, cet avantage implique l'activation cérébrale des régions du cortex auditif. Ainsi, dans les prochaines lignes, nous résumerons les travaux précédemment réalisés chez les sourds en neuroimagerie lors de l'observation de divers gestes et mouvements biologiques.

Les études réalisées en IRMf ou en Topographie par Émission de Positons (TEP) souhaitaient initialement déterminer si le réseau cérébral impliqué dans le traitement de l'action humaine recouvrait celui de la langue des signes, entre les individus sourds signeurs et les individus neurotypiques. Alors que certaines études utilisent des pantomimes (Corina et al., 2007; Emmorey, Xu, Gannon, Goldin-meadow, & Braun, 2010; Fang, Chen, Lingnau, Han, &

Bi, 2016), deux autres études se concentrent sur un unique emblème (soit le pouce en haut/en bas) (Husain, Patkin, Kim, Braun, & Horwitz, 2012; Husain, Patkin, Thai-Van, Braun, & Horwitz, 2009) et d'autres encore, sur des séquences de gestes sans aucune signification tant pour les individus sourds signeurs que pour leurs pairs neurotypiques (Emmorey et al., 2010; Husain et al., 2012; MacSweeney et al., 2004, 2008; Newman et al., 2015; Petitto et al., 2000).

À ce jour, l'ensemble des études échouent à converger tant au niveau de la neuroimagerie que des résultats comportementaux. Ainsi, d'après deux études se basant sur la perception de pantomimes, les individus sourds signeurs présentaient une sous-activation des régions classiquement dédiées à la perception de l'action chez l'homme en comparaison à des pairs neurotypiques, et tout particulièrement au sein des régions impliquées dans le réseau des neurones miroirs (Corina et al., 2007; Emmorey et al., 2010). L'utilisation extensive de la langue des signes était explicitée comme étant à l'origine de cette sous-activation, par un phénomène de déshabituation. A contrario, des études portant sur le jugement de pantomimes (Fang et al., 2016), sur un unique emblème (Husain et al., 2012, 2009) ou sur une séquence de gestes sans signification (MacSweeney et al., 2004, 2008) sont en faveur d'un réseau du traitement de l'action comparable entre les personnes sourdes et neurotypiques. En outre, plusieurs études rapportent une activation cérébrale bilatérale dans le gyrus temporal supérieur et notamment le planum temporale chez les individus sourds en réponse à l'ensemble des stimuli (emblème, pantomimes et gestes sans signification) (Fang et al., 2016; Husain et al., 2012; Petitto et al., 2000) en l'absence de différence comportementale entre les groupes sur le plan des réponses correctes ou des temps de réponse (Fang et al., 2016; Husain et al., 2012).

Ainsi, les disparités soulevées restreignent la généralisation des résultats à plusieurs niveaux et supportent l'intérêt d'une nouvelle étude. Soulevons tout d'abord, les divergences quant aux régions cérébrales impliquées qui trouvent leur origine au niveau théorique, c'est-à-dire le modèle de réseau utilisé pour décrire les activations (réseau des neurones miroirs versus réseau de l'action humaine). En outre, la diversité des tâches, le faible nombre de stimuli (notamment pour ce qui concerne les emblèmes) et la nature du traitement demandé (perception versus jugement) entraînent également des divergences (Cardin, Smittenaar, et al., 2016; Fang et al., 2016). L'ensemble des études précédemment décrites concernent uniquement des individus sourds signeurs et majoritairement natifs de la langue des signes, restreignant la généralisation des résultats aux autres sourds, dits oralistes. Finalement, aucune différence comportementale entre les groupes n'est rapportée alors que des activations cérébrales bilatérales au sein des régions auditives sont décrites. Ces résultats sont inconsistants avec les études précédemment rapportées sur la sensibilité accrue des individus sourds à détecter les cibles visuelles en mouvement en lien avec un phénomène de plasticité intermodale. Nous tâcherons de répondre à l'ensemble de ces limites dans le premier article de cette thèse.

1.3 Le cas spécifique de la perte auditive : réorganisation cérébrale structurelle

Dans les précédentes sections, il a été démontré un lien étroit entre une réorganisation cérébrale de type structurel et les capacités sensorielles améliorées chez les individus privés d'un sens (Shiell et al., 2016; Shiell & Zatorre, 2016; Voss et al., 2014). Des auteurs émettent ainsi l'hypothèse que la réorganisation cérébrale à la suite d'une privation sensorielle serait à

mettre en lien simultanément avec des répercussions comportementales, mais également avec l'intégrité des structures cérébrales (Karns, Stevens, Dow, Schorr, & Neville, 2017). En effet, pour mieux comprendre et prédire l'impact de l'expérience, soit l'absence d'afférence auditive dès la naissance, sur la réorganisation cérébrale, il apparaît nécessaire d'évaluer l'intégrité des structures cérébrales chez l'individu sourd puis, l'intégrité des connexions anatomiques existantes qui permettent le transfert des informations entre les différentes modalités sensorielles. Cette causalité supporte l'importance d'étudier la matière blanche et la matière grise qui représentent des entités différentes du cerveau. Premièrement, la matière grise réfère à l'accumulation de corps cellulaires et de neuropiles qui constitue le cortex. La matière blanche, quant à elle, se compose principalement des axones myélinisés des neurones et constitue les grands faisceaux et les commissures (Purves, 2012). La maturation de la matière grise dans le cerveau évolue par le biais d'une augmentation massive jusqu'à l'âge de deux ans puis, elle connaît une diminution progressive à partir de quatre ans jusqu'à la fin de la vie (Silk & Wood, 2011). En ce qui a trait à la matière blanche, elle se développe, à nouveau, de façon exponentielle jusqu'à l'âge de deux ans, puis va continuer à augmenter tout en se spécialisant jusqu'à l'âge de 22 ans environ, pour décroître progressivement jusqu'à la fin de la vie (Silk & Wood, 2011). La maturation de la matière blanche et de la matière grise est influencée par des facteurs génétiques, l'environnement, mais également l'expérience (développement, vieillissement et maladies) (Lerch et al., 2017). Ainsi, puisque l'expérience agit sur le développement structural du cerveau, l'intégrité de la matière blanche et de la matière grise ont fait l'objet de nombreuses études chez les personnes privées de l'audition dès la naissance. Pour étudier ces différences, l'imagerie par résonnance magnétique s'avère l'outil de premier choix notamment en raison des diverses techniques d'analyse anatomique

dont elle dispose (par exemple : épaisseur corticale, morphométrie au niveau du voxel, imagerie par diffusion). Alors que les premières études de neuroimagerie anatomique datent d'une vingtaine d'années (Emmorey, Allen, Bruss, Schenker, & Damasio, 2003; Penhune, Cismaru, Dorsaint-Pierre, Petitto, & Zatorre, 2003), des études subséquentes et très récentes sont encore réalisées (par ex. Amaral et al., 2016; Zheng, Wu, Huang, & Wu, 2017) et une très grande variabilité persiste dans les résultats et contraint la compréhension du processus de réorganisation cérébrale structurelle chez l'individu sourd. À l'instar de la réorganisation cérébrale fonctionnelle, les modifications structurelles observées dans le cerveau des personnes sourdes sont parfois associées à des performances comportementales améliorées, notamment au niveau de la vision (par ex. Pénicaud et al., 2013). Toutefois, des altérations de la matière blanche sont fréquemment rapportées au sein des régions impliquées dans l'audition et sont ultérieurement associées à des contraintes en termes de restauration auditive. Ainsi, en raison des disparités obtenues entre les différentes études et les diverses interprétations fournies à ce jour, il nous est apparu nécessaire de réaliser une revue systématique de la littérature afin d'offrir un état des lieux des données structurelles, en termes de matière blanche et de matière grise, chez l'individu sourd congénital. Ce travail constitue le second article de cette thèse.

Chapitre II : Objectifs et hypothèses de recherche

2. Objectif général de la thèse

Comme en témoigne le premier chapitre, la réorganisation cérébrale ayant lieu chez l'individu privé de l'audition demeure complexe et les interprétations proposées ne font actuellement pas consensus. Dans ce contexte, l'objectif général de cette thèse est d'arriver à une meilleure compréhension de la réorganisation cérébrale chez l'individu sourd.

2.1 Objectif et hypothèses de la première étude

Par conséquent, le premier volet de cette thèse concerne la plasticité cérébrale de type fonctionnel, via l'IRMf. Nous souhaitons investiguer le lien entre les activations cérébrales et les performances comportementales des individus sourds lors d'une tâche portant sur les mouvements biologiques (emblèmes et pantomimes) en comparaison à des pairs neurotypiques.

Les hypothèses de cette étude sont que la perte précoce de l'audition entraîne un avantage comportemental lors de la détection de mouvements biologiques. Nous savons que les individus sourds sont plus rapides que les individus neurotypiques lors de la détection de cibles visuelles simples en mouvement (par ex. Shiell et al., 2014a). Ainsi, nous prédisons que les individus sourds vont être plus rapides que les individus neurotypiques pour reconnaître les mouvements biologiques. Sur le plan de la neuroimagerie, nous nous attendons à ce que les deux populations testées présentent un réseau cérébral équivalent en termes de traitement de l'action humaine (Fang et al., 2016; Husain et al., 2009). De plus, il est attendu que l'avantage sur le plan comportemental des individus sourds devrait être accompagné d'activations

cérébrales bilatérales au sein des régions normalement impliquées dans le traitement de l'audition (par ex. Shiell et al., 2016).

2.2 Objectifs de la seconde étude

Le second volet de cette thèse concerne la réorganisation cérébrale de type structurel. En raison de l'impact de l'expérience sur le développement de la matière grise et de la matière blanche, l'absence d'afférence sonore entraîne de multiples changements structurels dans le cerveau des individus sourds. L'objectif principal de cette seconde étude consiste à répertorier l'ensemble des changements structurels retrouvés auprès des individus sourds profonds par le biais de diverses techniques d'analyse en imagerie par résonance magnétique.

Une revue systématique de la littérature est ainsi proposée pour résumer les résultats hautement hétérogènes obtenus précédemment, offrir des liens avec les facteurs qui sont connus pour influencer l'étendue de la réorganisation corticale chez les individus sourds et proposer de nouvelles avenues de recherche. De plus, nous souhaitons discuter de ces altérations structurelles sous un angle plus clinique, et notamment de la façon dont les changements en termes de matière blanche et de matière grise peuvent avoir une influence sur l'acquisition du langage, le développement neurocognitif, mais également, sur les stratégies de restauration de l'audition.

Chapitre III

Article 1. Enhancement of Visual Biological Motion Recognition in Early-Deaf Adults: Functional and Behavioral Correlates

Enhancement of Visual Biological Motion Recognition in Early-Deaf Adults: Functional and Behavioral Correlates

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Abstract

Deafness leads to brain modifications that are generally associated with a cross-modal activity of the auditory cortex, particularly for visual stimulation. In the present study, we explore the cortical processing of biological motion that conveyed either non-communicative (pantomimes) or communicative (emblems) information, in early-deaf and hearing individuals, using fMRI analyses. Behaviorally, deaf individuals showed an advantage in detecting communicative gestures relative to hearing individuals. Deaf individuals also showed significantly greater activation in the superior temporal cortex (including the planum temporale and primary auditory cortex) than hearing individuals. The activation levels in this region were correlated with deaf individuals' response times. This study provides neural and behavioral evidence that cross-modal plasticity leads to functional advantages in the processing of biological motion following lifelong auditory deprivation.

Introduction

An increasing number of studies suggest that early sensory loss leads to the enhancement of the other intact sensory modalities (1). Several behavioral studies have shown that early-deaf people possess enhanced abilities for visual localization and visual motion detection (2). According to functional neuroimaging studies, the visual enhancements in early-deaf individuals are generally attributed to the recruitment of the deafferented auditory cortex (3–6). Therefore, the visual crossmodal activity of the auditory cortex is typically defined as compensatory, meaning that deaf people rely more on their intact visual system to encode their environment in comparison to hearing individuals (7). Some tactile (8–11) and language abilities (i.e., sign language and/or lip-reading) (12–17) are also associated with the recruitment of the auditory cortex in deaf people (1) and support the compensatory reorganization of the brain after early auditory deprivation.

This study's aim is to tackle the relevant topic of visual crossmodal plasticity following early auditory deprivation with the visual ability to perceive biological motion i.e. gesture sequences that characterize all living things (18). The study of biological motion is an interesting issue since with only minimal pieces of visual information, such as point-lights at the main joints of the human body, people can efficiently recognize human actions (18,19). Human movement recognition is essential for social cognition and interaction. With this ability, people can understand the gestural intentions of others and respond adequately (20). For the deaf individuals using sign language, the adequate comprehension of human action is specifically critical to rapidly detect the presence of linguistic movements (21). More

generally, the ability to quickly recognize human motion also represents additional visual cues for deaf individuals to interpret their environment despite the auditory deprivation (22).

Originally, the cerebral network associated to the understanding of action (biological movement, human action) was referred to as the mirror neurons system (23). The human mirror neurons system is formed by the inferior parietal lobule (IPL), the ventral premotor cortex (PMv) as well as the inferior frontal gyrus (IFG, BA 44/45) in the homologous brain of the macaque (24). Henceforth, it is commonly accepted that the cerebral network of action understanding in humans is broader than the previously cited regions and also includes the posterior superior temporal sulcus (pSTS), the supplementary motor area (SMA, BA 6), the primary somatosensory cortex (S1, BA 1/2), the intraparietal cortex (IPS), the posterior middle temporal gyrus (pMTG) at the transition to visual area V5, and fusiform face area/fusiform body area (FFA/FBA**)** (25). It is interesting that the neural responses associated to point-light biological motion recognition involve the same characteristic set of regions implicated in human action recognition (26).

In prior studies, several stimuli have been used to disentangle cerebral networks involved in either or both sign language and human action recognition processes between deaf native signers and hearing individuals. Among the human actions, meaningless gestures, pantomimes, emblems, and signs are conceptualized as a continuum in terms of linguistic properties, conventionalization, and semiotics characteristics (27). Pantomimes are non-communicative gestures that are oriented towards an object, an action or an event (28) who can convey meaning on their own without speech (27). Emblems are conventional communicative gestures (27) that are culturally influenced (29) and defined as non-verbal

action used to convey information to others (30) (for illustration see Figure 1). These two types of gesture are not language *per se*. They differ from sign languages since the latter are natural human languages that have evolved spontaneously in Deaf communities, and possess all of the linguistic structural properties and complexity of spoken languages (31). Although sign languages use the visual-manual rather than aural-oral modalities, the networks of brain regions recruited for spoken and signed language processing are largely overlapping (32).

To date, all of the previous studies on deaf signers fail to converge neuroimaging with behavioral results. Using fMRI, two studies have investigated the cerebral network involved in the passive observation of pantomimes by deaf native signers. These studies report a hypoactivation of the human mirror neuron system in the IFG, and the IPL in deaf signers individuals (21,33). On the other hand, some neuroimaging studies with pantomimes (34), sequences of meaningless gestures (35,36) or a single emblem (37,38) support a similar human action network between the deaf signers and hearing individuals. In the current study, we attempt to replicate and extend these findings to multiple emblems. This way, the present study offers a robust comparison of the human action network between emblems and pantomimes. Indeed, these two stimuli differ according to whether they aim to transmit information or not, since emblems represent communicative gestures whereas pantomimes represent non-communicative gestures (19). Additionally, only the emblems show some linguistic properties, such as phonological and morphological components (27). Furthermore, activation of the superior temporal gyrus (STG), including the primary auditory cortex and the planum temporale, has been observed across tasks requiring to recognize emblems, pantomimes, and meaningless gestures (14,34,37) despite the absence of behavioral differences in terms of accuracy or reaction time between deaf signers and hearing individuals.

Together, these findings suggest that lifelong deafness and/or sign language use could lead to alterations in the neural networks recruited to interpret manual communication, even when it is not linguistically structured. Furthermore, increased recruitment of traditionally auditory and language processing areas during gesture recognition may reflect that lifelong reliance on visual communication (sign language and lip-reading) (39) leads to alternative neural strategies for the processing of this information. Moreover, none of the prior studies have included early deaf people who are not signers but used rather spoken language and explore the distinct effect of linguistic experience and auditory deprivation on visual crossmodal plasticity. The goal of the present study was to compare neural responses to both emblems and pantomimes between early-deaf and hearing individuals, and for the first time to relate these to behavioral performance. Given the lack of convergence in previous studies, we expected that combined behavioral and fMRI results might seize compensatory brain plasticity in early-deaf individuals, independently of their primary mean of communication. To test our hypothesis, we measured the fMRI bold response to emblems and pantomimes recognition in early-deaf individuals who used or did not use sign language in comparison to hearing peers.

Methods

1. Participants

Thirty-five French-speaking adults participated in the present study. All the participants provided written informed consent prior to testing and all experiments were performed in accordance with relevant guidelines and regulations. This study was approved by the ethics committee and scientific boards of the Centre de Recherche Interdisciplinaire en Réadaptation du Montréal métropolitain (CRIR) and the Quebec Bio-Imaging Network (QBIN). One deaf

and two hearing participants were excluded from the study due to technical problems during fMRI data acquisition. A total of 32 participants were therefore included in the study: 16 early severe-to-profound deaf subjects (11 women, *Mean age* ± *SD* = 30.25 ± 4.69 years) were compared to 16 hearing participants (12 women, *Mean age* ± *SD* = 30.31 ± 5.42 years) matched on age, sex, and number of years of education. All subjects had a normal or corrected-to-normal vision and no history of neurological pathology. According to the Edinburgh handedness inventory index (Oldfield, 1971), five deaf and three hearing participants were left-handed. All participants were administered the Matrix Reasoning subtest of the Weschler Abbreviated Scale of Intelligence (WASI-II) (Wechsler & Hsiao-pin, n.d.), which is a brief evaluation of non-verbal intelligence, namely of nonverbal fluid reasoning (Strauss, Sherman, & Spreen, 2006). The results showed that both groups performed in the average to the superior level of ability, as indicated by T scores (deaf participants: $M \pm SD$ = 57.44 ± 4.85; hearing participants: $M \pm SD$ = 62.46 ± 4.45).

Deaf participants had a severe-to-profound hearing loss greater than 77 dB HL ($M \pm SD$ = 94.11 ± 9.93) in both ears as determined by certified audiologists. Specifically, 13 participants had a hearing loss greater than 90 dB HL at 500, 1000, 2000, 4000, and 8000 Hz in both ears while two participants were able to detect 500 Hz pure tones presented at 80 dB HL and 77 dB HL in their better ear. Four participants reported having hereditary congenital deafness whereas, for twelve participants, congenital or early deafness was due to unknown etiologies. Eight of the sixteen deaf participants were proficient signers and four of them were native deaf signers in the *Langue des Signes Québécoise* (LSQ). Eight participants had been using hearing aids since childhood, used spoken language only for expression and relied on lip-reading for reception (see Table 1 for detailed information about the participants).

2. Stimuli and experimental protocol

The stimuli consisted of 126 point-light animated videos representing 42 emblems (e.g. "*calm down*"), 42 pantomimes (e.g. "*playing guitar*"), and 42 scrambled versions of these biological motions (Figure 1). We carefully controlled point-light stimuli, which allowed us to isolate the effects of biological motion from possible confounding effects such as face and body perception. Point-light also allows us to isolate biological motion processing from more general visual motion perception, by including a control condition in which the starting positions of the points are randomized, but their motion vectors remain the same (19). Previous studies that used videos and often compared gesture conditions to non-motion control conditions, were thus limited in the interpretation of their results (43).

An event-related fMRI protocol was split in two runs both presented in random order across participants. The stimulation task was implemented on Psychopy with Python 3.4. Each run of six-minutes comprised 63 different videos (21 stimuli of each category). Safety instructions and imaging sequences were explained to the participants to familiarize them with the fMRI environment. The participants all performed a training trial of the biological motion task before the fMRI session. The instructions were presented before each run and the participants had to press a button once they were done reading them. Each video lasted between two to four seconds and was followed by an inter-stimulus interval randomly varying from two to ten seconds. Biological motion stimuli were projected on a screen at the back of the scanner and were presented to the participants through a mirror attached to the MRI head coil – at approximately 12 cm away from the eyes. With an fMRI-safe button response pad, participants were asked to press as fast and as accurately as possible with the correct button (1:

whether the video was a human motion with no communicative content (pantomimes condition), 2: a human motion with communicative content (emblems condition) or 3: a non-human motion (scrambled condition)). Participants performs the task with their dominant hand. Accuracy (percentage of correct answers) and response time were measured.

3. Statistical analysis on behavioral data

Accuracy and response time measures of the biological motion task were analyzed using a 3 x 2 repeated-measures ANOVA with point-light conditions (emblems, pantomimes and scrambled) as within-subjects factor and group (deaf and hearing) as a between-subject factor. A Greenhouse-Geisser correction was applied to the degrees of freedom and to the significance level to prevent the disrespect of the sphericity assumption. Because the duration of the videos varied and ranged from two to four seconds in each point-light condition, a two-way ANOVA was conducted to examine the influence of run (1 and 2) and point-light conditions (emblems, pantomimes, and scrambled stimuli) on stimuli duration. On average, duration time of the videos was 3047.62 ms (*SD* = 740.013) for emblems, 2857.14 ms (*SD* = 792.82) for pantomimes, and 2380.95 ms (*SD* = 734.28) for scrambled stimuli. The main effect of the run was not significant ($F(2, 120) = .000$; $p = 1.00$), suggesting that the two runs were similar in stimulus duration. However, there was a significant main effect of point-light conditions ($F(2, 120) = 10.43$; $p < .001$; $\eta^2 = .148$) suggesting that the duration of the stimuli differed among to the conditions. The interaction was not significant ($F(2, 120) = .000$; $p > .05$). Bonferroni *post hoc* tests showed that stimulus duration was significantly higher for emblems than for pantomimes ($p < .001$) and scrambled stimuli ($p < .001$) whereas no significant differences were found between pantomimes and scrambled stimuli ($p > .05$).

Consequently, these results show that emblem stimuli were significantly longer than the other two conditions. To address this, participants' response time was transformed into a global mean response time for all point-light conditions across groups. Each response time was then weighted by the duration of the video and multiplied by the global mean.

4. fMRI acquisition parameters

Whole-brain anatomical and functional images were acquired using a 3-T Trio Tim system (Siemens Magnetom, Erlangen, Germany) equipped with a 32-channel head coil. Multislice T2*-weighted fMRI images were obtained with a gradient echo-planar sequence using axial slice orientation (TR = 2200 ms, TE = 30 ms, FA = 90°, 35 transverse slices, 3.2 mm slice thickness, FoV = 192 x 192 mm^2, matrix size = 64 x 64 x 35, voxel size = 3 x 3 x 3.2 mm^3). Head movements were restrained using foam pads. A structural T1-weighted MPRAGE sequence was also acquired for all participants (voxel size = 1×1×1 mm^3, matrix size = 240 x 256, TR = 2.300 ms, TE = 2.98 ms, TI = 900 ms, FoV 256, 192 slices).

5. Processing of functional images

The fMRI data were analyzed using SPM 12 in a Matlab environment (Statistical Parametric Mapping, Centre for Neuroimaging, London, UK, http://www.fil.ion.ucl.ac.uk/spm, Matlab 8.5 (Mathworks, Natick, MA, USA). Standard preprocessing was performed (realignment, co-registration of functional and anatomical data). At the step of normalization, two distinct anatomical templates were created using DARTEL (44) (Diffeomorphic Anatomical Registration Through Exponentiated Linear algebra), namely, a template designed for hearing participants and another designed for deaf participants. Both templates were created separately for each group and they have been respectively normalized to the MNI template. A groupwise

registration using DARTEL was chosen to reduce possible deformations of the structures that are more difficult to match to the average template based on neurotypical individuals (44). The DARTEL templates are especially relevant given that previous studies have shown significant structural alterations between deaf and hearing individuals (45). Finally, spatial smoothing was performed (8-mm FWHM) after which linear contrast images were calculated to test main effects in each participant for each condition ([Emblems], [Pantomimes], [Scrambled]). These linear contrasts generated statistical parametric maps [SPM(T)].

6. Statistical analyses of fMRI images

Within-group analyses: One-sample *t*-tests were performed (FDR-corrected for multiple comparisons, $p < .05$) to compare individually each group's performance in the different point-light conditions ([Emblems > Pantomimes], [Emblems > Scrambled], [Pantomimes > Emblems], [Pantomimes > Scrambled], [Scrambled > Emblems], [Scrambled > Pantomimes]). A conjunction contrast (conjunction null hypothesis) characterized brain areas jointly activated by the contrasts [Emblems + Pantomimes] in both groups.

Between-group analyses: Two-sample *t*-tests were carried out (FDR-corrected for multiple comparisons, $p < .05$) to examine group effects in each point-light condition separately ([Deaf > Hearing] x [Emblems], [Deaf > Hearing] x [Pantomimes], ([Hearing > Deaf] x [Emblems], [Hearing > Deaf] x [Pantomimes]). The comparison of the brain activation during biological motion processing [(Emblems + Pantomimes)-scrambled] between deaf and hearing participants allowed very strict control of low-level stimulus features (19,46).

Finally, the behavioral differences ([Emblems - Pantomimes]) for response times was entered as covariates in the general linear model with group and point-light condition as factors. The correlation between group and behavioral differences was calculated using an *F* test (FDR-corrected for multiple comparisons, $p < .05$).

Results

1. Behavioral Data

Deaf and hearing groups were equivalent with regards to age ($t(30) = .035$, $p = .682$), number of years of education ($t(30) = 1.965$, $p = .06$), or on their performance on the fluid reasoning subtest ($t(30) = 2.32$, $p = .43$). We performed separate repeated-measures 3 x 2 ANOVAs with both accuracy and response times as the dependent variable. The analysis of correct responses showed a significant main effect of point-light condition ($F(1.93, 57.81) = 95.57$; $p < .001$; $\eta^2 = .76$), no main effect of group ($F(1,30) = .04$; $p = .85$) and no significant interaction ($F(1.93, 57.81) = 3.08$; $p > .05$). On average, deaf participants recognized 73.38% (*SD* = 5.33) of emblems correctly, 81.94% (*SD* = 6.59) of pantomimes and 99.62% (*SD* = 0.40) of scrambled stimuli as compared to respectively 68.94% (*SD* = 0.24), 87.69% (SD = 5.11) and 99.56% (SD = 0.65) for hearing participants. Bonferroni *post hoc* tests demonstrated that all the participants were more accurate in the scrambled condition in comparison to the pantomimes and emblems conditions and more accurate in the pantomimes condition than they were in the emblems condition ($p < .001$ for all differences).

The analysis of response times showed a significant main effects of point-light condition ($F(1.66, 49.88) = 37.69$; $p < .001$; $\eta^2 = .56$), no significant main effect of group ($F(1, 30) =$

0.14; $p = .71$) and, a significant Group × Condition interaction ($F(1.66, 49.88) = 4.63$; $p < .05$; $\eta^2 = .13$) (see Fig. 1D). Bonferroni *post hoc* tests demonstrated that the deaf and hearing participants were fastest at identifying the scrambled condition in comparison to the pantomimes and emblems, respectively ($p < .001$ for all differences). Only hearing participants exhibited a significant difference between the pantomimes and the emblems conditions, with faster responses for pantomimes ($p < .001$).

2. fMRI Data

All results reported as significant in this section survived a threshold of whole-brain $p < .05$ voxel-wise threshold, FWE-corrected. Anatomical labels for active regions are the most probable based on the Harvard-Oxford Cortical Atlas.

Biological versus Scrambled Motion: We first examined the areas significantly activated by biological motion relative to the scrambled condition [(Emblems + Pantomimes)-scrambled] in each group. As expected, the analyses revealed an overlap in the regions involved in the human action recognition network between the deaf and hearing participants (see Fig. 2). Both groups showed extensive bilateral activations that included posterior temporal-occipital regions including V5, pSTS, EBA, and FBA, parietal regions including the right SMG and bilateral SPL, frontal lobe regions including bilateral IFG, frontal operculum/insula, precentral gyrus, middle frontal gyrus, and SMA, and the thalamus bilaterally (see Table 2 for locations of peak activations). Extensive cerebellar activity was observed as well.

Between-group analyses: Beyond these areas of overlap, some areas showed significant activation only for the deaf group for the biological motion conditions [(Emblems +

Pantomimes)-scrambled]. The deaf group showed a significantly stronger bilateral response than the hearing participants in the STG, including the planum temporale (BA 22) and the primary and secondary auditory cortex (BA 41, 42) (see Fig. 3.B and Table 3). Additionally, only deaf individuals showed activation in the basal ganglia (specifically globus pallidus and the head of the caudate nucleus), and greater extent of activation than hearing individuals in the cerebellum (see Fig.3). In the hearing group, no brain region was found to be more activated than the deaf group (see Table 3).

Brain responses to emblems and pantomimes individually were examined (Deaf > Hearing x [Emblems]; Deaf > Hearing x [Pantomimes]; Hearing > Deaf x [Emblems]; Hearing > Deaf x [Pantomimes]). Again, the deaf group showed a significantly stronger bilateral response to hearing participants in the STG, including the planum temporale (BA 22) and the primary and the secondary auditory cortex (BA 41, 42) (see Fig. 4. and Table 3). Notably, the deaf group showed a stronger bilateral response for the emblems condition than for the pantomimes condition, including voxels mostly in the planum temporale and in the primary auditory cortex.

Laterality differences in the deaf group: We further investigated whether there were laterality differences within the STG clusters activated uniquely in deaf people. Pairwise comparisons were carried out between the average activity (maximum global coordinate, *66.0 -28.0 7.0*) in the STG, in both hemispheres in all point-light conditions. The results showed a significant difference in signal strength between the right and the left STG, both in the combined biological motion condition (Emblems + Pantomimes), ($t(16) = -8.42$, $p < .0001$ (Right: $M \pm SD = 2.31 \pm 1.07$; Left: $M \pm SD = 1.00 \pm .91$)) as well as in the emblems condition

(t (16) = -5.31, p < .0001 (Right: $M \pm SD$ = 2.31 ± .99; Left: $M \pm SD$ =1.22 ± .85)). A rightward asymmetry was found during processing of scrambled motion and emblems but no difference was found between the hemispheres in the pantomime condition (t(16) = -1.41, p > .15 (Right: $M \pm SD$ = 2.15 ± 1.24; Left: $M \pm SD$ = 1.61 ± .96)). Of interest, an extensive activation of the STG was found in the emblems condition in contrast to the scrambled and pantomimes conditions. The peak activation was located in the primary auditory cortex.

Correlations with Behavioral Performance: As demonstrated earlier, the behavioral results suggest that there was a significant interaction between hearing status with participants' response times (Fig.1). Therefore, the way this behavioral difference [Emblems - Pantomimes] translated into neural activations in the deaf group was explored. Whole-brain analyses with behavior differences (Emblems-Pantomimes response times factored out) as covariates were performed. We observed a significant relationship between behavioral measures and brain responses in the bilateral STG and the left precentral gyrus (see Fig. 4 and Table 4). A correlation analysis was carried out to specify the relation between the behavioral measures (response times) and the cerebral activations triggered in the left and right STG. Results indicate that the activation of the STG could predict response times, in the right hemisphere (r = .36, p = .04, R^2 = .13) and marginally in the left hemisphere (r = .35, p = .05, R^2 = .12). This finding suggests that, for the deaf individuals, stronger activation of the STG during the biological motion task leads to faster response times. Correlation analyses were also conducted on the left precentral gyrus to determine if behavioral results could be predicted by the cortical activity in this region. No significant correlation was found. This was true for the relationship between the peak activity in the precentral gyrus and response times (r = -.37, p > .05) in deaf individuals.

Discussion

The main goal of the present study was to combine, for the first time, behavioral and neuroimaging measures of emblems and pantomimes gesture recognition, between early-deaf and hearing individuals. In previous studies, inconsistent imaging results were found. A hypoactivation was reported in some cerebral regions involved in the human action network, namely the IPL and the IFG, by two studies investigating the observation of pantomimes in native deaf signers (21,33). These findings were explained by the predominant use of the visual modality in deaf individuals, not only to support their daily life, but also because of their extensive use of sign language. The latter could be seen as a training in human gestures decoding. The authors argue that this training could make native deaf signers less sensitive to human gestures and thus result in a cortical hypoactivation (33). More recently, a study (34) looked at congenitally deaf individuals who were native signers. With a pantomime's judgment task, the authors concluded that there was a robust activation of the human action network in individuals who experienced auditory deprivation in addition to using sign language. However, in this study, no relationship was found between deafs' linguistic experience and the strength of the cortical activations within the human action recognition network (34). The present study confirms that there is an overlap in deaf and hearing individuals' cortical activation network in response to biological motion processing. Both groups showed similar activations in the expected regions (25), that is, occipital, parietal, temporal, and inferior frontal regions during emblems and pantomimes recognition.

More importantly, the present results provide behavioral and neural evidence in favor of compensatory visual cross-modal activity experienced by early deaf people. As some previous

studies (14,34,37), we found significant bilateral activations of the STG, including the primary auditory cortex in the deaf group. Our findings corroborate previous work in the literature. Indeed, there are well-established associations between animal and human data (47) showing that deafness can lead to enhanced visual abilities (6,48), thus implying a cross-modal reorganization process where the visual modality recruits the auditory cortex (4,49,50). However, the evidence is unclear as to whether deafness can lead to both enhanced behavioral performance and a cross-modal activation of the primary auditory cortex by other sensory modalities or higher cognitive functions (1). Moreover, the literature on the possible behavioral enhancements experienced by deaf individuals is characterized by results that are both heterogeneous and inconsistent. This can be attributed to a variety of factors, such as sample characteristics (48). Indeed, variables such as the amount of residual hearing, the onset of deafness or etiology of deafness are known to influence the extent of cerebral plasticity (13,51). Thus, a majority of studies have specifically examined deaf native signers (51), while these deaf individuals represent only a small percentage of the deaf population (52). Overall, previous results cannot be generalized, and it is therefore complex to have a clear understanding of deaf individuals' cross-modal reorganization. In our study, differences were found between the behavioral performance and the cortical activation of regions altered by auditory deprivation in deaf compared to hearing participants. The results suggest that early-deaf individuals showed greater sensitivity to the processing of human action than hearing individuals. Specifically, deaf individuals identified emblems as fast as pantomimes in comparison to their hearing peers. These behavioral differences were directly correlated with the bilateral activation of the STG. These results differ from those of previous studies reporting the recruitment of auditory areas in the processing of emblems (37) but not of

pantomimes (34), and those reporting no behavioral differences between deaf and hearing participants (34,37). Additionally, a significant correlation was found between STG activations and response times. This correlation could suggest that the extent of STG recruitment in deaf individuals depends on their capacity to detect emblems more rapidly than pantomimes. This result is consistent with the previous literature showing that enhanced visual performances in deaf individuals are usually related to shorter reaction times rather than to accuracy (5), but must be replicated for exhaustive interpretation.

Furthermore, emblems overall led to more extensive bilateral activations than pantomimes in deaf individuals, especially in the STG (including planum temporale and primary auditory cortex). The activation of the primary auditory cortex, followed by the posterior region of the STG, involved in the dorsal pathway of language processing (53–55), suggests that emblems are more prone to be processed as linguistic material by early-deaf individuals. The linguistic processing of emblems, supported by the activation of the left STG, was reported in a study on prelingual deaf adults who were native signers (37). According to the authors, the linguistic processing of emblems is sustained by a leftward hemispheric asymmetry found in deaf signers in comparison to hearing participants. However, several neuroimaging studies propose that language processing implies a collaboration of both left and right pathways, as well as a cortico-sub-cortical network (53). In addition, the language network in the right hemisphere is classically related to the visual abilities involved in language processing (56) and explains the STG rightward asymmetry during recognition of visually communicative emblems by the deaf group.

The fMRI analyses performed in the present study addressed the implications of auditory deprivation and linguistic experience on visual biological motion processing. All our deaf participants presented profound-to-severe congenital deafness, but while half of them were proficient in sign language (four were native deaf signers), the other half was using spoken language as a first language. While not formally tested, the robustness of the cortical activations in the human action network suggests an absence of any linguistic experience effect. A particularly interesting finding of the present study is that the differences in human action processing are better explained by an effect of auditory deprivation since all the deaf participants experienced a bilateral activation of the STG. In future studies, a larger sample size of deaf individuals would be needed since deafness related factors are known to influence brain plasticity (e.g. deafness duration, amount of residual hearing, prior use of hearing aids) and should be considered in the analyses (13,51).

Functional and behavioral correlates converge to a human action sensitivity following early-deafness deprivation. This sensitivity does not appear to be modulated by linguistic experience but rather by auditory deprivation. Thus, the present findings are of importance not only because they contribute to the understanding of the visual cross-modal plasticity phenomenon in the deaf population, but also because they offer new avenues of research for rehabilitation strategies that would be better adapted to the daily effects of deafness.

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Table 1. Demographic and clinical data for the 16 deaf participants

Subject	Sex	Etiology	Age	Hearing aid	Hearing loss: left ear/right ear (dBHL)	Primary language	WASI T-score	Handedness
1	M	Unknown	36	No	100/100	Sign	54	R
2	F	Genetic	22	No	105/110	Sign (native)	63	R
3	F	Genetic	25	No	90/90	Sign (native)	52	L
4	M	Unknown	36	No	90/90	Sign	58	L
5	F	Unknown	29	Yes	115/110	Spoken	68	R
6	F	Genetic	25	Yes	93/95	Spoken	58	L
7	F	Unknown	29	No	90/90	Sign (native)	62	R
8	M	Unknown	25	Yes	87/92	Spoken	50	R
9	F	Unknown	33	Yes	103/102	Spoken	58	L
10	F	Unknown	34	Yes	106/106	Spoken	60	R
11	F	Genetic	36	No	90/90	Sign	52	R
12	F	Unknown	28	Yes	93/92	Spoken	62	R
13	F	Unknown	37	Yes	78/77	Spoken	58	R
14	F	Unknown	28	No	97/95	Sign (native)	58	R
15	M	Unknown	31	No	90/90	Sign	60	L
16	M	Unknown	30	Yes	101/106	Spoken	49	R

Table 2. Brain regions showing significant activations for the conjunction of biological motion (emblems and pantomimes)-scrambled in each group.

Anatomical region	Hemi	Cluster size	T	x	y	z	*p*-FEW corr (*p*<.05)	Other areas including Distance (mm)
		Deaf						
Precentral	L	4412	13.63	-54	2	43	.000	Postcentral (4.58) Frontal mid (10.25)
Fusiform	L	5543	12.62	-39	-43	-20	.000	Temporal inf (2.45) Cerebelum 6 (7.35)
Parietal inf	R	65	5.52	30	-46	49	.001	Parietal inf (1.00) Postcentral (4.58)
Thalamus	L	82	5.34	-12	-16	7	.003	Pallidum (11.70) Caudate (12.04)
Thalamus	R	6	4.74	6	-22	-11	.026	Lingual (10.05) Parahippocampal (10.82)
Fusiform	R	3	4.66	36	-4	-41	.034	Temporal inf (2.24) Temporal mid (6.71)
		Hearing						
Fusiform	R	1766	12.12	39	-43	-20	.000	Temporal inf (5.10) Cerebelum 6 (5.83)
Temporal mid	L	3874	11.80	-51	-70	1	.000	Occipital mid (2.45) Occipital inf (5.10)
Insula	R	1356	10.97	30	26	1	.000	Frontal inf tri (4.58) Putamen (5.74)
Cerebelum 7b	L	473	7.58	-12	-73	-44	.000	Cerebelum 8 (2.24) Cerebelum crus2 (5.00)
Thalamus	L	167	6.14	-9	-16	4	.000	Thalamus R (11.00) Pallidum (13.45)
Parietal inf	R	136	6.10	27	-49	49	.000	Parietal sup (1.73) Postcentral (5.20)

MNI coordinates (x, y, z) of the most significant cluster are given, along with the corresponding brain region for this cluster and the other areas including in each cluster, with distance (mm).

Table 3. Brain regions showing significant activations for the contrast of Deaf > Hearing in each point-light condition.

Deaf>Hearing								
Anatomical region	**Hemi**	**Cluster size**	**T**	**x**	**y**	**z**	***p*-FEW corr (*p*<.05)**	**Other areas including Distance (mm)**
Biological motion								
Temporal sup	R	969	12.32	66	-28	7	.000	Temporal mid (6.71) Supramarginal (11.18)
Temporal sup	L	916	10.20	-54	-34	10	.000	Temporal mid (2.00) Supramarginal (8.94)
Precentral	L	69	6.46	-57	-1	43	.001	Postcentral (1.73) Frontal mid (13.96)
Caudate	R	26	5.27	18	17	4	.007	Putamen (3.00) Pallidum (9.00)
Occipital Mid	L	32	5.15	-33	-58	7	.005	Calcarine (7.07) Precuneus (7.35)
Cerebelum 8	L	5	4.75	-3	-61	-32	.005	Vermis 8 (1.41) Vermis 9 (3.16)
Precentral	R	5	4.75	57	8	37	.005	Frontal inf oper (6.16) Frontal mid (6.78)
Emblems								
Temporal sup	R	755	10.04	66	-25	4	.000	Temporal Mid (6.08) Rolandic oper (11.36)
Temporal sup	L	819	8.80	-60	-31	7	.000	Supramarginal (9.90)
Precentral	L	22	5.50	-54	2	43	.008	Postcentral (4.58) Frontal Mid (10.25)
Caudate	R	2	4.58	18	17	4	.035	Putamen (3.00) Pallidum (9.00)
Pantomimes								
Temporal sup	R	213	7.79	66	-28	7	.001	Temporal mid (6.71) Supramarginal (11.18)
Temporal sup	L	144	6.10	-51	-37	10	.002	Temporal mid (2.45) Rolandic oper (9.49)

MNI coordinates (x, y, z) of the most significant cluster are given, along with the corresponding brain region for this cluster and the other areas including in each cluster, with distance (mm).

Table 4. Brain regions showing significant activations for the main effect of group with reaction time.

Anatomical region	Hemi	Cluster size	F	x	y	z	*p*-FEW corr (*p*<.05)	Other areas including Distance (mm)
Temporal sup	R	108	82.32	66	-28	7	.000	Temporal mid (6.71) Supramarginal (11.18)
Temporal sup	L	140	71.98	-63	-31	7	.000	Supramarginal (9.95)
Precentral	L	14	51.40	-57	-1	46	.007	Postcentral (2.45) Frontal mid (12.88)

MNI coordinates (x, y, z) of the most significant cluster are given, along with the corresponding brain region for this cluster and the other areas including in each cluster, with distance (mm).

Figure 1. Stimuli and behavioural results. (A) example of a emblem "calm down" (B) example of pantomimes "playing guitar" (C) example of a scrambled version (D) Behavioural result illustrating the reaction times (RT) according to both groups.

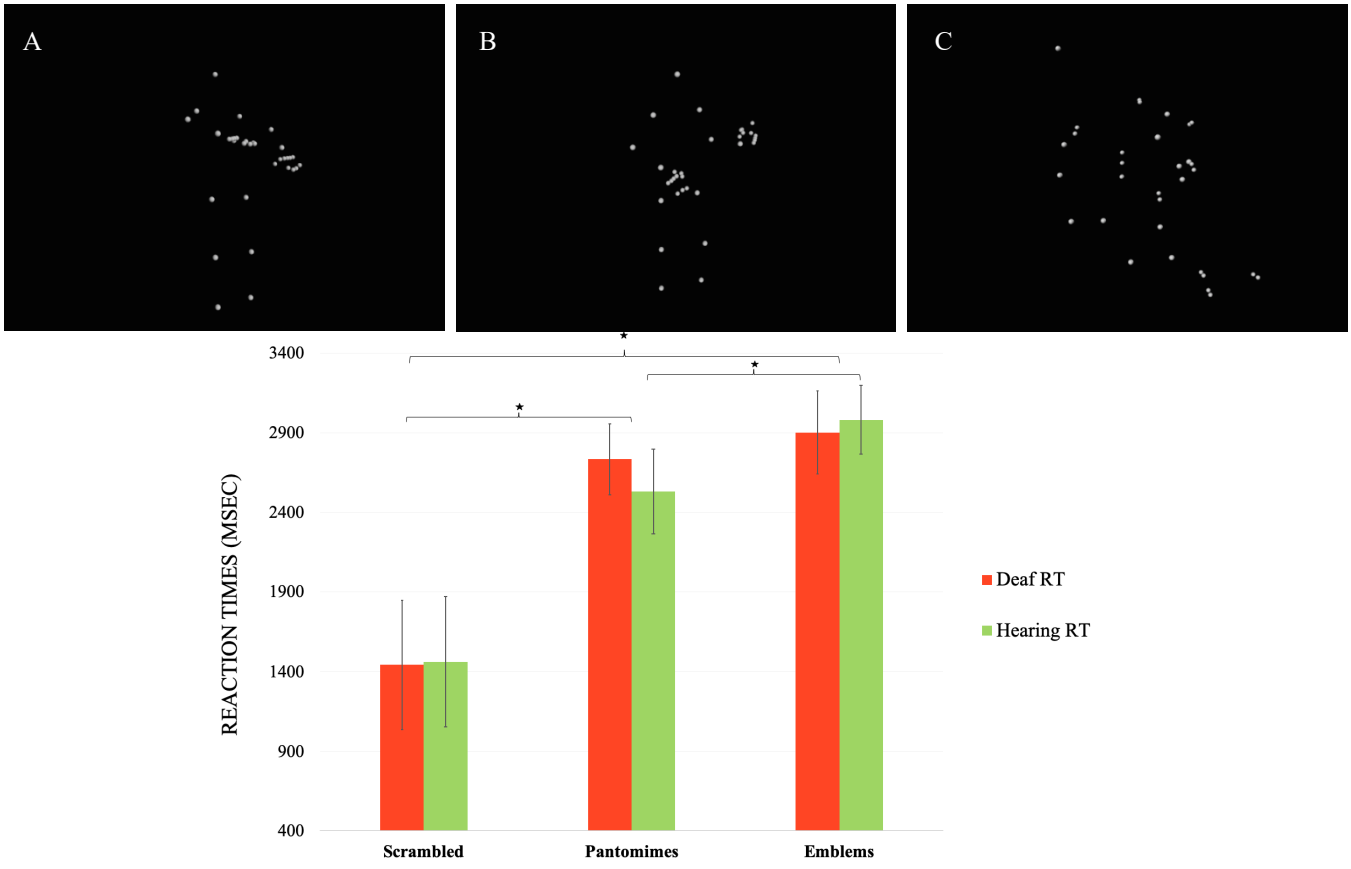


Figure 2. *fMRI data*. The conjunction of cortical activations implicated in biological motion processing [(Emblems + Pantomimes) - scrambled] by the group, deaf (Red) and hearing participants (Blue), Overlap (Purple).

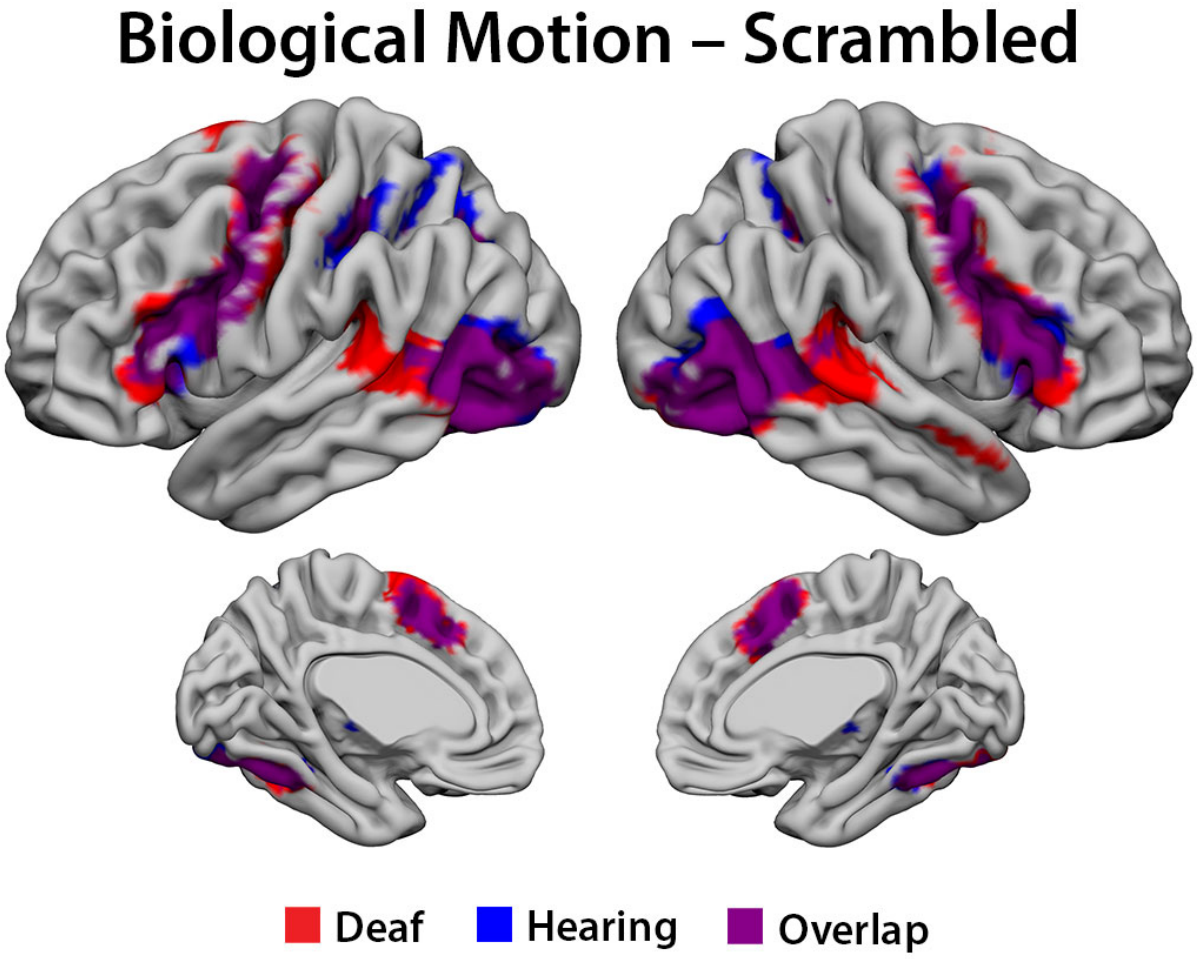


Figure 3. *fMRI data*. (A) The cortical activations implicated in Emblems only (Yellow), Pantomime only (Blue), and the Overlap (Green) by the group. (B) Significant difference between deaf and hearing participants in the biological motion condition, the image in the maximum global coordinate (66.0 -28.0 7.0). (C) Significant difference between deaf and hearing participants in the pantomime condition, the image in the maximum global coordinate (66.0 -28.0 7.0). (D) Significant difference between deaf and hearing participants in the emblem condition, the image in the maximum peak activation at coordinates (66.0 -28.0 7.0).

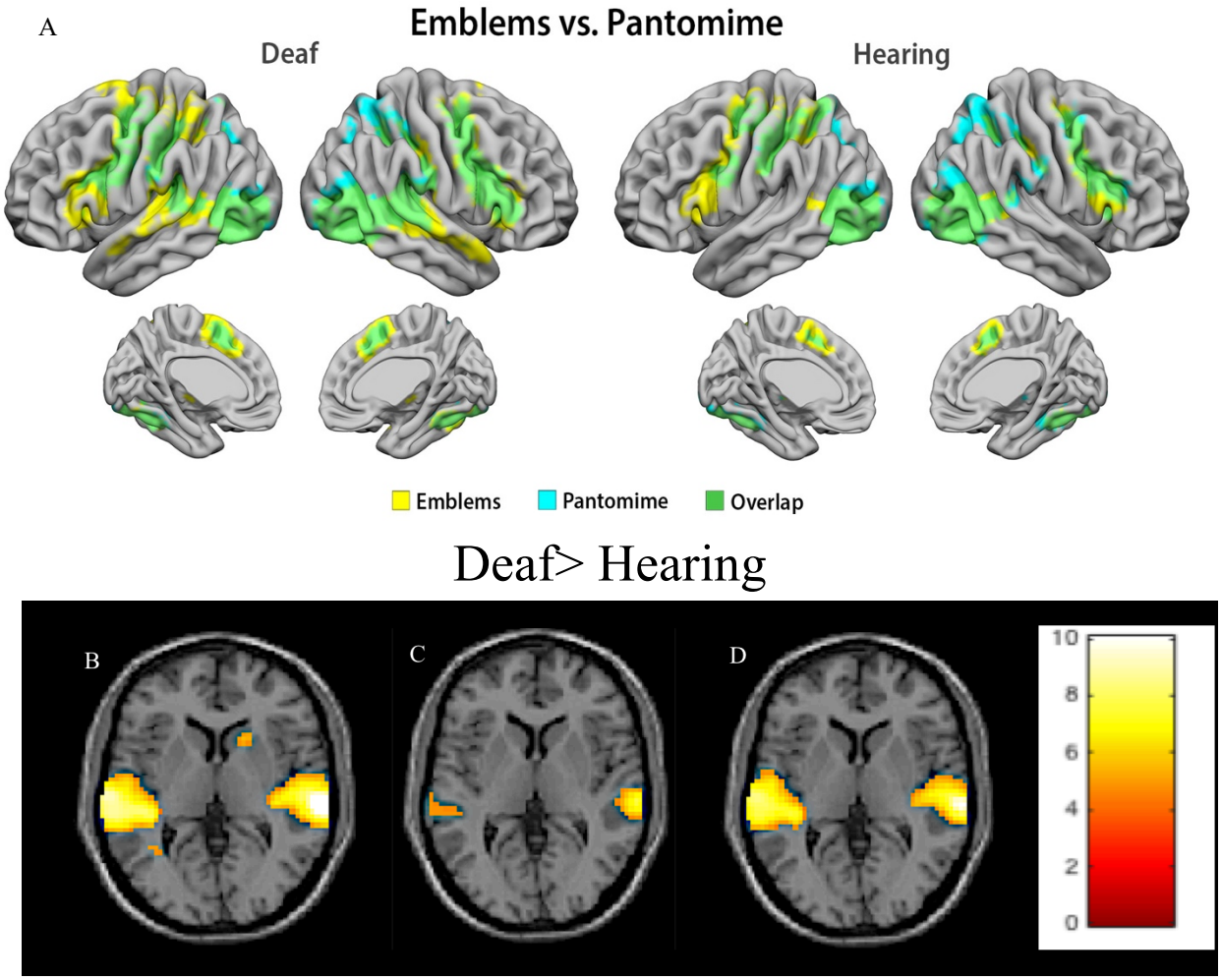
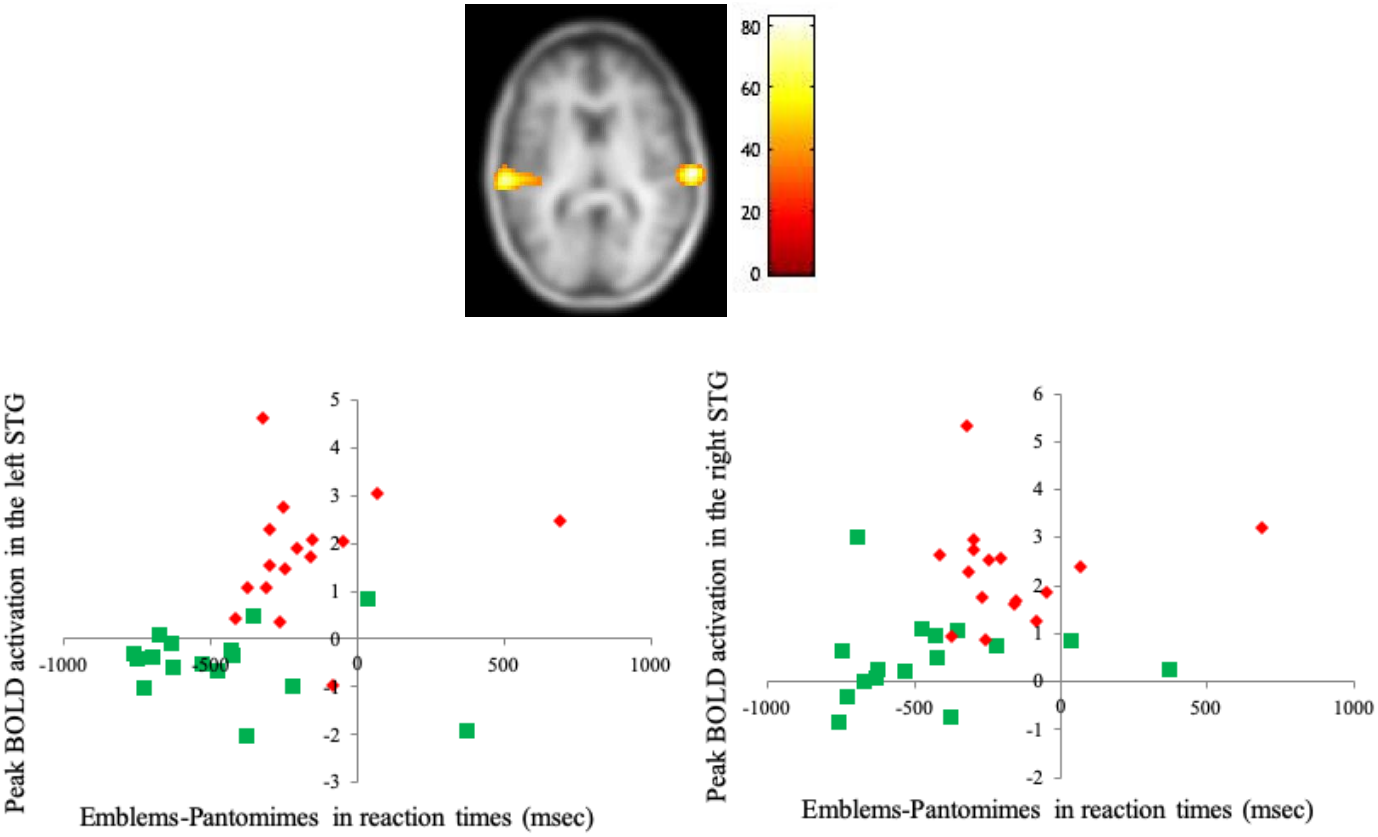


Figure 4. *fMRI data.* Regression analyses between cortical activity triggered by biological motion (Emblems - Pantomimes) and behavioral discrepancy (on reaction times) in the deaf group only. MNI coordinates for global maximum (66.0 -28.0 7.0). Graphs: Correlation plots of the blood oxygen level-dependent Emblems-Pantomimes responses in this region against reaction times (RT). Each data point represents a single subject, red for the deaf group and green for the hearing group.



Chapitre IV

Article 2. The impact of deafness on brain plasticity: a systematic review of the white and gray matter changes

The Impact of Early Deafness on Brain Plasticity:

A Systematic Review of the White and Grey Matter Changes

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ABSTRACT

Background: Auditory deprivation alters cortical and subcortical brain regions, primarily linked to auditory and language processing, resulting in behavioral consequences. Neuroimaging studies have reported various degrees of structural changes, yet multiple variables in deafness profiles need to be considered for proper interpretation of results. To date, many inconsistencies are reported in the grey and white matter alterations following early profound deafness. The purpose of this study was to provide the first systematic review synthesizing grey and white matter changes in deaf individuals.

Methods: We conducted a systematic review according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses statement (PRISMA) in 27 studies comprising 626 deaf individuals.

Results: Evidence shows that auditory deprivation significantly alters white matter across the primary and secondary auditory cortex. The most consistent alteration across studies was in the bilateral superior temporal gyri. Furthermore, reductions in the fractional anisotropy of white matter fibers comprising in the inferior-fronto-occipital fasciculus, the superior longitudinal fasciculus, as well as the subcortical auditory pathway are reported. The reviewed studies also suggest that grey and white matter integrity is sensitive to early sign language acquisition, attenuating the effect of auditory deprivation on neurocognitive development.

Conclusions: These findings suggest that understanding cortical reorganization through grey and white matter changes in auditory and non-auditory areas is an important factor in the development of auditory rehabilitation strategies in the deaf population.

Introduction

Neuroplasticity is an intrinsic property of the brain (Dennis et al., 2013) and refers to the brain's ability to reorganize itself in response to learning and the environmental interactions throughout life (Pascual-Leone, Amedi, Fregni, & Merabet, 2005). Early neuroplasticity increases the vulnerability of the immature brain, possibly leading to adverse development (Dennis et al., 2014). Thus, neuroplasticity can also be associated with a neurodevelopmental and behavioral pathology (Gilmore, Knickmeyer, & Gao, 2018), involving both functional and structural modifications and can lead to behavioral consequences (May, 2011). Therefore, given the absence of experience in the auditory cortex of congenitally deaf children, early deafness constitutes an excellent model to study neuroplasticity mechanisms in the human brain.

In neurotypical children, ontogenetic events support the development of the brain through neurogenesis, axonal and dendritic growth, synaptogenesis, synaptic pruning, and myelination (Anderson, 2001). These events are highly interdependent such that perturbations in one specific area of development can have long-term effects on the brain's structural and functional integrity (Grantham-McGregor et al., 2007). Indeed, intrauterine and early childhood development are critical to the proper maturation of cognitive abilities and behaviors, as brain development is characterized mainly by reorganization, "fine-tuning" or remodeling of primary circuits and networks after the age of two (Gilmore et al., 2018). Brain regions associated with primary functions such as perception (e.g., vision and audition) and gross motor abilities mature first and are followed by areas supporting spatial orientation and

language development; brain areas involved in executive function, attention as well as motor coordination appear to mature last (Gogtay et al., 2004; Grantham-McGregor et al., 2007).

Several studies have demonstrated a developmental decrease of synaptic plasticity in the auditory cortex after early deafness (for a review, see Kral and Sharma 2012). Consequently, neuroplastic changes occur at the youngest age in early deaf children and are generally related to a sensitive period (Sharma, Dorman, & Kral, 2005). In the particular context of early deprivation, this sensitive period corresponds to a window during which experience is critical for the development of sensory functions (Kral, 2013). In deaf children, this sensitive period principally occurs up to the third year of life and corresponds to a critical limit for auditory rehabilitation, especially as it relates to cochlear implantation (Kral, 2013). Based on these lines of evidence, the consequences of auditory deprivation on cortical maturation in congenitally or prelingually deaf children is of high importance for auditory rehabilitation, particularly for language acquisition and neurocognitive development.

With magnetic resonance imaging (MRI), numerous studies have acquired *in vivo* data to describe a plethora of cerebral structures in deaf individuals in comparison to hearing peers. *Morphometric analysis* was one of the first techniques used to describe anatomical reorganization in deaf individuals (Emmorey, Allen, Bruss, Schenker, & Damasio, 2003; Penhune, Cismaru, Dorsaint-Pierre, Petitto, & Zatorre, 2003). This technique allows the classification of cerebral tissues whereby grey matter, white matter and cerebrospinal fluid volumes can be calculated (Filipek, Richelme, Kennedy, & Caviness, 1994). Subsequently, *Voxel-Based Morphometry* (VBM) was developed to allow voxel-by-voxel assessment of tissue density in white and grey matter in typical and atypical brains (Wright et al., 1995).

Complementary to VBM is *Cortical Thickness* (CT), which measures the distance between white and grey matter and the distance between grey matter and the dura mater (He, Chen, & Evans, 2007) and *Tensor-Based Morphometry* (TBM), which enables measurement of volume differences in the brain (Ashburner & Friston, 2001). *Diffusion Magnetic Resonance Imaging* is used to analyze the integrity of white matter structures by estimating fiber structure through water molecule diffusion (e.g., Mori & Zhang, 2006; Mukherjee, Chung, Berman, Hess, & Henry, 2008). For example, *Diffusion Tensor Imaging* (DTI) determines whether or not water molecules diffuse in all directions and specifies the preferred diffusion direction within a given tract. The general index of the structural integrity and directionality of axonal fibers within a voxel (fractional anisotropy: FA) is the most frequently reported DTI measure. Finally, as an alternative to DTI, *Diffusion Kurtosis Imaging* (DKI) allows the measure of Gaussian and, more particularly, non-Gaussian properties of water diffusion (Lu, Jensen, Ramani, & Helpern, 2006).

In this systematic review, we first report the current state of knowledge regarding grey and white matter changes found in deaf individuals through various neuroimaging techniques (Volumetry, VBM, TBM, CT, DTI, and DKI). We then describe these structural changes as they relate to factors known to influence the extent of cortical plasticity. Finally, we interpret the reported findings in the context of recent advances and present our current understanding of these macroscopic cortical plasticity phenomena. We also discuss the predictive value of structural changes relating to language acquisition and neurocognitive development in deaf individuals, as well as how it can guide rehabilitation strategies.

Methods

This systematic review was conducted according to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses statement (PRISMA) (Moher, Liberati, Tetzlaff, & Altman, 2009).

1. Inclusion criteria

Studies were eligible if they included (1) a structural, anatomical or morphometric MRI brain analysis technique, (2) congenitally or prelingually deaf adults, adolescents or children, and (3) participants presenting severe to profound bilateral hearing loss. Studies involving unilateral or late-acquired deafness as well as animal data were excluded. Only articles published in a peer-reviewed journal in English or translated into English were considered.

2. Search strategy

Online searches on PubMed, including PubMed Central and Medline, PsycNET, including PsycINFO and PsycARTICLES, and Web of Science (Core Collection) were performed in April 2017, repeated in September 2017 and in April 2018 with relevant search terms. Search terms were: ("Deaf" OR "Hearing loss") AND ("Voxel-based morphometry" OR "VBM" OR "Diffusion tensor imaging" OR "DTI" OR "Cortical thickness" OR "White matter" OR "Grey matter" OR "Morphometric" OR "Neuroimaging"). All database literature coverage ranged from 1974 to present and no automatic filter was used for publication type (journal article, case reports, conference findings, reviews, etc.).

3. Study selection

The study selection procedure is presented in Figure 1. All studies were compiled to ensure the removal of duplicates; two distinct reviewers verified this procedure. Then, the first reviewer selected potential studies based on title, abstract and publication type. The second reviewer verified the previous selection and all articles that had been considered incompatible. After this screening, the two reviewers evaluated all the articles for full-text eligibility.

4. Data collection process

Extracted data for each study included (1) meta-study information (e.g., name of the authors, year of publication), (2) sample characteristics, including demographics (e.g., age) and hearing loss variables (e.g., onset and type of hearing loss, communication preference), (3) neuroimaging analysis (e.g., DTI, CT, VBM), measure used (e.g., region-of-interest [ROI] or whole-brain), MRI scanner strength, coordinates reference system, and (4) method and results of any significant (at least $p < 0.05$) group-based comparisons in the neuroimaging measures. For each brain region, reviewers took note of whether a significant alteration was found regarding volume, cortical thickness, fractional anisotropy (FA, the common index of structural integrity and directionality of axonal fibers within a voxel), axial diffusivity (AD, reflecting integrity of microtubules along the axon), radial diffusivity (RD, indexing levels of myelination), mean diffusivity (MD, reflecting mathematical combination of the RD and AD) or mean kurtosis (MK, reflecting structural changes in both anisotropic and isotropic tissue). Reviewers also specified if changes occurred in white or grey matter and in a specific hemisphere. Due to the lack of specific coordinates in most studies, MNI or *Talairach*

coordinates were not compiled and only brain regions were considered. Pertinent details from each study are presented in Table 1.

Results

1273 articles were identified from the databases using the selected keywords. Once the duplicates were removed, 764 articles were included in the selection process. After screening, 27 studies were eligible. Figure 1 shows the selection process according to the guidelines established by PRISMA (Moher et al., 2009).

1. Study selection and participants characteristics

The 27 studies were published between the years 2003 and 2017. Five studies reported data acquired using a combination of neuroimaging techniques. Nine studies used morphometric and volumetric analyses and four studies referenced cortical thickness. VBM findings were described in 10 studies while data acquired using DTI were reported in eight studies. Finally, a single study reported data obtained via TBM and one study via DKI. The MRI scanners had a magnetic field intensity of 1.5 Tesla for 12 studies and 3 Tesla for 15 studies. Analysis procedures differed from one study to another and also depended on neuroimaging technique. Thus, ROIs analyses were conducted in 15 studies while whole brain analyses were reported in 12 studies.

Compilation of study data showed acquisition of MRI data in 626 individuals presenting moderate to profound bilateral deafness, including 254 children and adolescents. Regarding deafness type, 14 studies focused on congenitally deaf individuals, 13 studies were conducted with pre-lingual deaf individuals and one study reported data from deaf individuals with post-

lingual deafness. With respect to the degree of hearing loss, 20 studies analyzed individuals presenting profound deafness. The degree of hearing loss was considered to be severe to profound in four studies and moderate to profound in three studies. Among the 27 studies, 10 included data from deaf individuals who were native signers, three studies focused on individuals who acquired sign language later in life and eight studies reported a preferential use of sign language without specifying the time of acquisition. Six studies did not report information regarding the means of communication of participants, although four of them dealt with deaf children that were candidates for cochlear implantation.

Among the 27 selected studies, the majority did not include information regarding the use of hearing aids. Five studies reported absence of hearing aids in the first years of life (Allen et al. 2013; Allen et al. 2008; Emmorey et al. 2003; Kim et al. 2009; Li et al. 2012) while two studies noted an absence of hearing aids at the time of testing (Amaral et al., 2016; Hribar, Šuput, Carvalho, Battelino, & Vovk, 2014). Additionally, five studies reported the use of hearing aids for all participants without indicating the duration of use (Kim et al., 2009; Li et al., 2013, 2015; Miao et al., 2013; Zheng, Wu, Huang, & Wu, 2017). Finally, four studies presented data of moderate to profound deaf children who were scanned while they were candidates to cochlear implant (Chang et al., 2012; Huang et al., 2015; Wu et al. 2016; Zheng et al., 2017).

Most participants in the control groups were hearing individuals. However, three studies presented findings from hearing participants whose primary language was sign language (hearing individuals born of deaf signer parents) (Allen et al., 2008, 2013; Olulade, Koo, LaSasso, & Eden, 2014). This comparison allows for the measurement of the potential effect

of sign language acquired as the first language since birth on brain plasticity by controlling for the impact of auditory deprivation. Two studies reported data of hearing adults that learned sign language (e.g., sign language interpreters).

2. Synthesized findings

To summarize data included in this systematic review, the majority of findings have been categorized according to neuroimaging technique and brain regions. In Figure 2, the data are presented in descending order according to the degree of consensus regarding the brain changes reported across the studies. In the next sections, we summarize brain changes according to major brain function and sensory modality.

2.1 Findings related to structures involved in auditory and language processing

The *superior temporal gyrus* is the brain area most commonly associated with structural modifications in deaf individuals. The superior temporal gyrus is mainly involved in auditory processing, but its left posterior part is specialized in language comprehension (Friederici & Gierhan, 2013). In the right hemisphere, the superior temporal gyrus is implicated in prosodic aspects of speech (Friederici, 2011). Reflected by several neuroimaging techniques, strong evidence supports the presence of white matter changes (reduced volume/density or a reduction in FA values) in the superior temporal gyrus of deaf individuals (Emmorey et al., 2003; Hribar et al., 2014; Huang et al., 2015; Karns, Stevens, Dow, Schorr, & Neville, 2017; Kim et al., 2009; Kim et al., 2014; Li et al., 2012 ; Miao et al., 2013; Olulade et al., 2014; Pénicaud et al., 2013; Shibata, 2007; Smith et al., 2011; Wu et al., 2016; Zheng et al., 2017). These changes are found in both hemispheres. Regarding DTI indexes, a reduction in FA appears to be related to an increase in RD in deaf individuals compared to hearing peers

(Karns et al. 2017; Li et al. 2012; Miao et al. 2013; Wu et al. 2016). One study reported equal RD and increased AD (Hribar et al., 2014). There is no agreement regarding grey matter.

The vast majority of reviewed studies report reduced volume, density and FA in fibers projecting to the primary auditory cortex (*Heschl's gyrus*) which is related to processing speech sounds. These changes are found in the left and right hemispheres (Emmorey et al. 2003; Hribar et al. 2014; Huang et al. 2015; Karns et al. 2017; Li et al. 2012; Miao et al. 2013; Smith et al. 2011; Wu et al. 2016; Zheng et al. 2017). Two studies reported similar white matter volumes between native deaf and hearing individuals (Leporé et al., 2010; Penhune et al., 2003). With regards to DTI, a reduction in FA was found to be related to an increase in RD (Karns et al. 2017; Li et al. 2012; Miao et al. 2013). One study reported no significant difference in RD and AD between deaf and hearing individuals (Hribar et al., 2014). Changes in grey matter in Heschl's gyrus were also found using morphometry and VBM techniques. One study reported increased in grey matter density (Smith et al., 2011), while another found a decrease (Olulade et al., 2014). Finally, four studies reported similar grey matter volumes of Heschl's gyrus in deaf and hearing individuals (Emmorey et al., 2003; Hribar et al., 2014; Kim et al., 2014; Li et al., 2015).

While there is an agreement between studies regarding the effects of deafness on the superior temporal gyrus and primary auditory cortex, only for white matter alterations, more heterogeneous findings have been reported for other structures. In particular, two types of structural changes have been reported in the inferior frontal gyrus, which is involved in speech production and semantic processing (Friederici & Gierhan, 2013). First, in the left hemisphere, a TBM study reported increased white matter volume in deaf adult native signers (Leporé et

al., 2010) whereas three studies observed the opposite effect (Kim et al. 2009; Olulade et al. 2014; Zheng et al., 2017). For grey matter, a morphometry study reported increased volume in native deaf signers (Allen et al., 2013) while other studies reported similar grey matter volume between adolescent signers and hearing individuals (Li et al., 2015).

The *planum temporale*, located above the superior temporal gyrus and partially coinciding with Wernicke's area, is considered to be part of the secondary auditory cortex. For white matter, two studies reported reduced density and FA in deaf adult signers (Hribar et al., 2014; Shibata, 2007). Alteration in FA were found to be related to a decrease in AD (Hribar et al., 2014). A morphometric study reported similar volumes between native deaf signers and hearing individuals in both white and grey matter (Penhune et al., 2003) while another found an increase in grey matter volume in native adult deaf signers (Emmorey et al., 2003).

Two studies regarding the *planum polare*, which is associated with auditory processing of voice and pitch attributes reported FA and AD reductions in adolescent and adult signers (Hribar et al., 2014) whereas increases in RD were found (Hribar et al., 2014; Miao et al., 2013).

The *middle temporal gyrus* is involved in linguistic processing, and more specifically in lexical-semantic processing (Friederici & Gierhan, 2013). It is also known to be a multimodal area that integrates auditory and visual information (Zatorre, 2002). In this region, two studies reported reduced white matter density in deaf children and native deaf signers (Smith et al., 2011; Olulade et al., 2014), whereas another found no volume differences between deaf and hearing participants (Zheng et al., 2017). One TBM study reported a bilateral increase in white matter volume in native deaf signers (Leporé et al., 2010).

The primary role of the *inferior fronto-occipital fasciculus* is language processing and, more specifically, semantic processing. It mainly connects areas such as the superior and middle frontal cortices, the inferior frontal and orbitofrontal cortices, but also the superior parietal, angular and fusiform gyri as well as the occipital lobe. Two studies reported reductions in FA in the right hemisphere of deaf signers (Hribar et al., 2014) and deaf adolescents (Miao et al., 2013), whereas one study found reduced FA in the left hemisphere of prelingually deaf adults (Kim et al., 2009). Hribar and collaborators (2014) also reported that the reduction in FA is related to a decrease in AD.

The *superior longitudinal fasciculus* connects the frontal and opercular areas with the superior parietal lobe, the angular, supramarginal and superior temporal gyri. Two studies reported changes in this fasciculus. A reduction in FA in fibers projecting to the right hemisphere was reported in prelingually deaf adults (Kim et al., 2009), whereas a decrease in white matter in the left hemisphere was shown in congenitally deaf signers (Meyer et al., 2007).

Four studies reported findings in the subcortical auditory pathway that connects the inferior colliculi and medial geniculate nucleus to the auditory cortex. In particular, some studies have reported a reduction of FA in fibers projecting to the auditory radiation of deaf children (Chang et al., 2012; Huang et al., 2015), including deaf children under three years of age (Zheng et al., 2017). Reduced integrity of the FA was reported in the inferior colliculi of deaf children (Huang et al., 2015) and a rightward asymmetry was found in native deaf signers (Amaral et al., 2016). In the superior olivary nucleus, a reduction in FA was reported in one study (Huang et al., 2015). In deaf individuals, no asymmetry or difference in terms of FA

compared to hearing individuals were found in the medial geniculate nucleus (Amaral et al., 2016; Zheng et al., 2017), whereas one study found a reduction in FA (Huang et al., 2015).

2.2 Findings related to structures involved in visual processing

The *primary visual cortex* is involved in visual processing, categorization and various changes have been reported in this region. Thus, three VBM studies have reported reduced white matter volume in children, deaf adolescents (Li et al. 2013; Li et al. 2012). One TBM study also reported increased white matter volume in native deaf signers (Leporé et al., 2010). Grey matter differences between deaf and hearing individuals have been consistently described in morphometric, CT and VBM studies. Five reported increased grey matter in this area (Allen et al. 2013; Allen et al. 2008; Li et al. 2013; Li et al. 2012; Pénicaud et al. 2013). These findings are observed in the left hemisphere in two studies with native deaf signers (Allen et al., 2013; Pénicaud et al., 2013) and in the right hemisphere in deaf adolescents using sign language (Li et al., 2013). The increase in grey matter volume seems to be related to the use of sign language as one study reports a reduction in grey matter volume in adult deaf signers who acquired sign language later in life (Pénicaud et al. 2013).

The *fusiform gyrus* is known to play a role in facial recognition and also in recognition of written words. In this area, a single VBM study reported white matter density changes in the right hemisphere of native deaf signers (Olulade et al., 2014). With regards to grey matter, the findings have been inconsistent. Whereas one study reported similar grey matter volume between adolescent signers and hearing individuals (Li et al., 2015), CT (Li et al., 2012; Li et al., 2013) and VBM data suggest a reduction in grey matter density in the left hemisphere of deaf adolescent and adult signers (Olulade et al., 2014).

In the left *lingual gyrus*, Olulade et al., (2014) reported a grey matter density reduction in deaf and hearing adult signers. A subsequent study reported no difference in terms of grey matter density in deaf adolescent signers. A decrease in grey matter volume in the left supramarginal gyrus was also observed in adult deaf signers, whereas Li et al. (2015) found no difference. Finally, one study using DKI reported similar FA in this area between prelingually deaf and hearing children.

2.3 Findings related to structures involved in multisensory processing

The *corpus callosum* is the largest white matter pathway in the brain. Its primary function is to coordinate and allows the interhemispheric transfer of sensory and motor information (Schulte & Muller-Oehring, 2010). In deaf individuals, studies suggest FA reductions of the fibers projecting through the splenium. This reduction was observed in native deaf signers (Karns et al., 2017), in both adolescents and adults sign language users (Miao et al. 2013; Li et al. 2012). Two studies reported similar white matter FA in the corpus callosum of deaf individuals and hearing participants, but also reported a decrease in AD (Miao et al. 2013; Li et al. 2012). One study reported increased FA in deaf individuals, located bilaterally in the major forceps of the corpus callosum, which is involved in the interhemispheric transfer of visual information (Kim et al., 2009). Finally, two studies reported reduced FA and increased RD (Karns et al., 2017; Miao et al., 2013), whereas another found reduced AD (Hribar et al., 2014).

The *cerebellum* is known for its role in motor function, as well as posture and balance. It is also involved in cognitive functions such as working memory, long-term memory, implicit and explicit learning as well as language (for a review, see Desmond & Fiez, 1998) and has been suggested to be involved in auditory processing (e.g., Petacchi et al., 2005). Three VBM

studies have reported a decrease in cerebellum white matter in deaf children and in adults using sign language (Smith et al. 2011; Olulade et al., 2014; Shibata 2007). One TBM study reported an increase in white matter volume in native deaf signers (Leporé et al., 2010). For grey matter, two VBM studies reported reduced density in deaf signers and native deaf signers compared to native hearing signers (Hribar et al., 2014; Olulade et al., 2014, although an opposite effect was shown in a study conducted with adolescent signers (Li et al., 2013).

The *thalamus* plays a significant role in the relay and integration of sensory afferences and motor efferences. Three DTI studies reported a reduction of FA in fibers projecting to the right internal capsule next to the thalamus in deaf adults, deaf adolescent and adult signers. One study also reported reduced AD (Hribar et al., 2014) while another reported increases in MD and RD in frontal and occipital thalamic radiation in late deaf signers (Lyness, Alvarez, Sereno, & MacSweeney, 2014). A single study reported a rightward volume asymmetry in the thalamus of native deaf signers (Amaral et al., 2016).

The *insula* contributes to several cognitive processes, as well as in multisensory integration (e.g., Naghavi et al. 2007). One study reported increased grey matter in deaf native signers. Olulade (2014) contradicted this finding by reporting the opposite pattern. As for white matter, one study reported reduced FA and AD in deaf signers (Hribar et al., 2014). Finally, a leftward asymmetry in grey matter was reported in native deaf signers by compared to native hearing signers in the posterior lobule (Allen et al., 2008).

2.4 Findings related to structures involved in motor processing

Neuroimaging data regarding the *precentral gyrus*, or primary motor cortex, are inconsistent. Whereas Lepore et al. (2010) reported a bilateral increase in primary motor cortex volume in

native deaf signers, another study found increased white matter density in the left hemisphere of native signers (hearing and deaf) (Olulade et al., 2014). A leftward asymmetry was also been reported in the hand region in deaf signers, while it is typically observed in the right hemisphere of hearing individuals (Allen et al., 2013).

The *basal ganglia* play a role in involuntary motor activity and muscle tone. In these nuclei, two studies (VBM and DTI) have reported reduced white matter density in the right hemisphere of deaf adults exclusively using sign language (Hribar et al., 2014; Meyer et al., 2007). A single study reported a grey matter increase in the caudate nucleus of native deaf signers (Olulade et al., 2014).

2.5 Findings related to structures involved in higher cognitive functions

The *middle frontal gyrus* is associated with higher cognitive functions such as executive functions, memory and language. Two studies using VBM and CT reported altered white matter density and thickness in the left hemisphere of adolescent signers (Li et al., 2012) and deaf adults (Kim et al., 2009). One TBM reported an increase of white matter volume in the right hemisphere in native deaf signers (Leporé et al., 2010). One VBM study also found increased grey matter density in the right hemisphere of native deaf signers and native hearing signers, suggesting an effect of sign language in the prefrontal cortex (Olulade et al., 2014).

The *superior frontal gyrus* is primarily involved in higher cognitive functions and, more specifically, in working memory (Du Boisgueheneuc et al., 2006). Regarding white matter, findings are inconsistent. One VBM study reported a leftward decrease of white matter density in deaf adults (Kim et al., 2009). One TBM study also found a bilateral increase in white matter volume in the superior frontal gyrus in native deaf signers (Leporé et al., 2010). With

regards to grey matter, two VBM studies and one CT study reported grey matter increases in the right hemisphere of deaf signer's (Li et al., 2012; Li et al., 2013; Olulade et al., 2014; Shibata, 2007). This effect seems to be associated with sign language as is also observed in native hearing signers. Finally, three morphometric studies reported opposite results, with grey matter reductions in the superior frontal gyrus of deaf signers (Hribar et al., 2014; Li et al., 2012; Li et al., 2013).

2.6 Findings from other structures without consistent observations

In the *postcentral gyrus* or primary somatosensory cortex, two studies reported reduced white matter in deaf children (Smith et al., 2011) and adults (Kim et al., 2009) whereas one TBM study reported a bilateral increase in white matter volume in native deaf signers (Leporé et al., 2010). For grey matter, a single study found reduced cortical thickness in deaf adolescent signers (Li et al., 2012).

In the *cingulate gyrus*, inconsistent findings have been reported. One study found increased grey matter volume in the left hemisphere of deaf adolescent signers and a decrease of grey matter density in the right hemisphere of native deaf and hearing signers (Olulade et al., 2014). One DTI study found similar FA and RD and a bilateral reduction in AD in the anterior region of the cingulate gyrus (Hribar et al., 2014).

In the *precuneus*, an increase in grey matter volume was reported in the right hemisphere of deaf signers (Olulade et al., 2014). Another study reported similar grey matter volume between deaf adolescent signers and hearing individuals (Li et al., 2015).

Discussion

The aim of the present systematic review was to identify key features of structural plasticity in deaf individuals by examining cerebral changes in grey and white matter. We provide an up-to-date synthesis that focused on structural changes identified with the following neuroimaging techniques: volumetry, VBM, TBM, CT, DTI, and DKI. Using the PRISMA method (Moher et al., 2009), 27 papers were selected that describe the structural changes reported in 626 individuals with a moderate to profound bilateral deafness, including 254 children and adolescents. This review provides converging evidence from several studies to determine specific or consistent changes in grey and white matter in congenital and prelingual deaf individuals. As the plasticity of grey and white matter are experience-dependent, the ontogenetic events occurring throughout development must be considered in the context of sensory loss. We thus emphasize the effect of auditory deprivation and more specifically the consequences of the early absence of aural experiences on the long-term development of brain anatomy.

Summary of main findings

1. Cerebral changes induced by auditory deprivation

Nearly all studies included in this review focused on cortical regions implicated in auditory processing: the primary auditory cortex (Helsch's gyrus) and secondary auditory cortex (Planum polare and Planum temporale). Evidence from these studies show white matter changes across all these areas. Specifically, reduced white matter volume and density, as well as reduced FA, was observed in deaf children, adolescents and adults. For the superior

temporal gyrus, which is involved in language processing, the majority of studies reported bilateral white matter changes in volume, density and FA. A large body of work suggests that the early absence of auditory stimulation leads to reduced myelinization in these areas (e.g., Hribar et al. 2014; Karns et al. 2017). Additionally, findings suggest that these changes are not sensitive to the means of communication used by deaf individuals (i.e., spoken or sign language). However, they are negatively correlated with auditory and speech perception in children candidates to cochlear implantation. Indeed, those with the poorest perceptive abilities after implantation show greater and broader changes in primary and secondary auditory areas.

Additional cortical and subcortical structures, rarely discussed in the context of neuroplasticity in deaf individuals, also contribute to auditory processing and are modified by auditory deprivation. Hence, white matter changes have been reported in the posterior part of the corpus callosum (or splenium), which allows interhemispheric connections between auditory areas (Zatorre, 2002). Anatomical differences have also been described in the anterior portion of the corpus callosum (or genu), which connects the left and right prefrontal and orbitofrontal regions (Chang, Lee, Paik, Lee, & Lee, 2012). These changes are negatively correlated with the auditory perceptive abilities of children candidates to cochlear implantation. One study showed a bilateral increase in white matter in the splenium, a portion of the corpus callosum involved in interhemispheric visual association (Kim et al., 2009). Changes in subcortical structures implicated in the auditory functions were also found. Reduced FA was observed in fibers projecting to the auditory radiation, the superior olivary nucleus, as well as the inferior colliculi. All of these changes are correlated with the speech perception outcomes of children fitted with a cochlear implant. With regards to grey matter, a

rightward volume asymmetry in native deaf signers was reported in subcortical structures (inferior colliculi and thalamus) (Amaral et al., 2016). These asymmetries are interpreted as constituting a mechanism for the transmission of visual information toward the auditory regions in deaf individuals (Amaral et al., 2016).

In sum, the evidence demonstrates significant changes in the main cortical and subcortical structures implicated in auditory processing, which appear to be present from an early age and have long-lasting effects. However, Li et al. (2012) reported a significant correlation between FA in fibers projecting to the superior temporal gyrus and the age of deafness onset. This result is consistent with the presence of a critical developmental period that is sensitive to auditory deprivation during postnatal life and critical for rehabilitation strategies (Kral, 2013). As it relates to experience-dependent-plasticity, one open question is whether the use of hearing aids modifies the extent of the reported structural changes. This question deserves to be thoroughly investigated since a relationship between the duration of hearing aid use and the extent of functional reorganization in the auditory cortex has only been shown in a functional connectivity study (Shiell, Champoux, & Zatorre, 2014b).

2. Cerebral changes related to language

Numerous brain areas and circuits are involved in language processing and production in the human brain (Friederici, 2011). Among these, the inferior frontal cortex, the superior temporal gyrus, and the middle temporal gyrus are believed to be the most important (Friederici, 2011). Typically, language production also requires the contribution of premotor and motor regions while language perception implicates the auditory and visual systems. The present review confirms the presence of brain changes in language-related areas in deaf individuals. There

was strong evidence for bilateral white matter changes in volume, density and FA in the superior temporal gyrus. Reductions in white matter volume and FA in the inferior frontal gyrus of the left hemisphere were also reported. However, the data do not provide robust evidence of middle temporal gyrus structural changes.

Four major fasciculi are involved in language processing. The dorsal pathway, connecting the frontal and temporal regions, includes the arcuate fasciculus and parts of the superior longitudinal fasciculus. These two fasciculi are involved in syntactic and speech repetition, respectively (Dick, Bernal, & Tremblay, 2014). The ventral pathway includes the uncinate fasciculus, also implicated in the primary syntactic process, and the inferio-fronto-occipital fasciculus, involved in semantic and comprehension processing (Dick et al., 2014). In deaf individuals, only some studies reported reduced density of FA in the superior longitudinal fasciculus, the uncinate fasciculus and the inferior-fronto-occipital fasciculus and none has presented findings regarding fibers projecting in the arcuate fasciculus. This demonstrates the need for future studies to evaluate the role of the ventral and dorsal language pathways in deaf individuals.

Moreover, three additional structures involved in language processing could be impacted by sensory deprivation and should be the focus of further research. Indeed, the supramarginal gyrus, which contributes to the processing of prosody, could present white and grey matter abnormalities. Alteration of the angular gyrus, which is involved in semantic processing, word reading, and comprehension, have been shown to be altered in a single study with children candidates to cochlear implantation (Zheng et al., 2017). Beyond these language functions, the angular gyrus is an essential structure in the context of sensory deprivation as it is a cross-

modal hub where sensory information (auditory, visual and tactile) converges and is integrated (Seghier, 2013). The insula is an important structure involved in auditory processing and the motor aspects of speech. More importantly, this structure also plays a role in multisensory integration at the level of audio-visual and visuo-tactile integration (e.g., Naghavi et al. 2007). Inconsistent findings are reported across the studies included in this review for both grey and white matter, although auditory deprivation seems to increase grey matter in the posterior insula. According to Allen et al. (2008), this change could be related to increased use of visual speech reading or stronger articulatory-based phonological representations of speech.

The reviewed data suggests that it may be necessary to differentiate structural changes according to means of communication (spoken or sign language). At the cerebral level, sign and spoken language share common neural bases, although some specificities have been reported. For example, higher activation of the posterior middle temporal gyri is observed in sign language when compared to spoken language (MacSweeney, Capek, Campbell, & Woll, 2008). The majority of studies detailed in this review involved deaf individuals who preferentially use sign language. However, in 10 studies, deaf participants were born of deaf parents and used sign language exclusively as a mean of communication. These deaf signers only represent 5% of the total deaf population and their linguistic abilities cannot be related to the majority of deaf individuals born in hearing families (Bavelier, Dye, & Hauser, 2006). Here, sign language appears to be a confounding factor when extrapolating functional information from anatomical changes. Nevertheless, comparing deaf individuals to a group of hearing native signers can isolate the effect of sign language that interacts with the effect's auditory deprivation. The present review identified three studies (Allen et al., 2008, 2013; Olulade et al., 2014) that directly compared the brain anatomy of deaf native signers to that of

hearing native signers. Early acquisition of sign language is associated with increased volume or density of grey and white matter in regions implicated in language processing (inferior frontal gyrus), executive functions (middle frontal gyrus), visuospatial and motor processing (precuneus, precentral gyrus) and multimodal sensory integration (insula). Deaf native signers also present specific brain differences in regions involved in auditory and language processing (superior temporal gyrus, inferior frontal gyrus, middle temporal gyrus) and executive functions (middle frontal gyrus), but also in visual (fusiform gyrus, calcarine sulcus), motor/sensorimotor (precentral gyrus, cerebellum, caudate) and multisensory integrative areas (insula).

These findings suggest that early auditory deprivation leads to specific brain changes according to the mean of communication (spoken or sign language). In young deaf children that are candidates to cochlear implantation, lower auditory perception scores are correlated with a decrease in FA in regions involved in linguistic processing (superior temporal gyrus, Heschl's gyrus, angular gyrus, genu of corpus callosum, inferior frontal gyrus). These changes support the auditory deprivation hypothesis which suggests that the absence or deterioration of auditory experience impacts the development of speech and spoken language as well as other cognitive functions such as executive functions (Beer, Kronenberger, & Pisoni, 2011). A second hypothesis suggests that this neurodevelopmental cascade can be explained by early language deprivation. This situation is often seen in deaf individuals for whom the auditory deficiency is only detected only once the acquisition of spoken language abilities is visibly delayed and is also often been to altered executive functioning (e.g., Figueras et al. 2008; Kral et al. 2016). A recent study has shown that native deaf signer children have similar executive functioning as hearing children matched by age and gender (Hall, Eigsti, Bortfeld, & Lillo-

Martin, 2017). Therefore, learning sign language appears to be associated with specific structural plasticity in multiple brain areas. These could act as a protection factor, minimizing the effect of auditory deprivation on neurocognitive development.

3. Cerebral changes induced by compensatory mechanisms

When comparing deaf to hearing individuals, numerous studies have reported enhanced abilities in deaf individuals in various sensory tasks, such as visual ones (e.g., Dye et al., 2007; Levänen & Hamdorf, 2001; Shiell et al., 2014a), higher cognitive functions, such as attention orientation (Colmenero, Catena, Fuentes, & Ramos, 2004) as well as recognition of emotional expressions and facial features (Arnold & Murray, 1998; Bettger, Emmorey, McCullough, & Bellugi, 1997). The principal explanation is that these behavioral enhancements are supported by cross-modal activation of auditory regions (Merabet & Pascual-Leone, 2009). Cross-modal plasticity refers to the recruitment of affected cortical areas by another sensory modality (Kral, Dorman, & Wilson, 2019). The review of previously published observations explains certain sensory compensatory mechanisms with structural plasticity in individuals with early auditory deprivation. Studies reviewed here suggest that grey matter changes may be associated with visual experience in deaf individuals. In fact, several functional neuroimaging and behavioral studies suggest that congenitally or early deaf individuals possess enhanced abilities for visual localization (for a review, see Pavani & Bottari, 2011) and visual motion detection (Shiell et al., 2014a). The present review supports the general agreement in VBM and volumetric studies in terms of grey matter increases in the visual areas of native deaf signers, leading to enhanced visual abilities (Allen et al., 2013; Pénicaud et al., 2013). It has also been suggested that an increase in grey matter is not only an effect of auditory deprivation but also of early sign language exposure, as demonstrated by contrasting native deaf signers with late deaf signers.

Indeed, one study reported grey matter reductions in the primary visual cortex of late deaf signers (Pénicaud et al., 2013). By apparent contrast, one study reported increased visual performance in the peripheral visual field, which was associated with thickness reduction in the primary visual cortex of deaf individuals (Smittenaar et al., 2016).

Atypical somatosensory change has also been observed when comparing deaf to hearing individuals, where deafness-induced cross-modal plasticity seems to support enhanced performance (e.g., Heimler & Pavani, 2014; Levänen & Hamdorf, 2001). Regarding motor development, a single study reported a delay in fine motor skills development in prelingually deaf children (Horn, Pisoni, & Miyamoto, 2006). However, in regions involved in motor and somatosensory processing, there is currently no consensus as to grey or white matter changes. Also, when looking at the postcentral gyrus or primary somatosensory cortex, a single study identified reduced grey matter density in deaf adolescents (Li et al., 2012).

To explore the relationship between functional and structural reorganization induced by auditory deprivation, it appears necessary to develop protocols that include specific and sensitive behavioral tasks associated with their anatomical neural substrates. For example, one recent study reported that an increase in cortical thickness in the right posterior superior temporal cortex was associated with visual motion detection abilities in early and profoundly deaf individuals (Shiell & Zatorre, 2016). For their part, Smittenaar and collaborators (2016) reported enhanced peripheral vision in congenitally deaf adults associated with reduced cortical thickness in primary visual cortex.

4. Issues regarding the interpretation of brain plasticity data

When discussing cortical reorganization, certain general aspects of cerebral plasticity could help the interpretation of the reported results. Indeed, recently, several studies have identified cortical changes as a result of experience-dependent plasticity. For example, when considering musicians, grey matter volume changes occur in auditory, motor, and visuospatial areas as soon as an individual engages in learning music. Then, extensive musical practice affects regions involved in higher cognitive processes such as executive functions, memory or emotions (e.g., Groussard et al. 2014). Accordingly, an increase in grey matter in somatosensory and auditory areas is usually interpreted as an adaptive plasticity phenomenon leading to enhanced performances, as demonstrated in opera singers (Kleber et al. 2016). However, a study shows contradictory results with a rapid increase in grey matter density in sensorimotor-related brain areas followed by a decrease after a few training sessions with a complex whole-body balancing task (Taubert et al. 2010). A careful interpretation is thus necessary regarding brain-behavior relationships when looking at grey matter differences since contradictory findings lead to the hypothesis that plasticity is functionally selective (Heimler, Weisz, and Collignon 2014).

Concerning white matter, the DTI technique is currently a powerful instrument for the study of anatomical correlates and changes at the levels of fibers (diameter, density) or myelinization. However, DTI is a relatively complex neuroimaging technique given the intricate nature of white matter and the extensive available choices of analyses. This complexity leads to many misconceptions regarding the interpretation of results (for an extensive review, see Jones, Knösche, and Turner 2013). A vast number of studies addressing clinical populations show white matter alteration, e.g.: Alzheimer's disease (Damoiseaux et al.

2009); schizophrenia (Qiu et al. 2010) and Tourette's syndrome (Neuner et al. 2010). In a neurotypical population, extensive piano practice is associated with an increased myelinization in children and is maintained with age (Bengtsson et al. 2005). Most studies in this review only report changes to the FA. The radial diffusivity (RD) which measures index levels of myelinization and the axial diffusivity (AD) which reflects the integrity of microtubules along the axon seem necessary for an exhaustive understanding of white matter plasticity. For example, in deaf individuals, three studies indicate that superior temporal gyrus reductions in FA following deafness can be better attributed to changes in RD than in AD (Karns et al., 2017; Li et al., 2012; Miao et al., 2013). A single study reports differences in the AD (Hribar et al., 2014). On the other hand, a reduction of FA in regions implicated in auditory and lingual processing appears to be consistent across the studies reported in this systematic review. However, heterogeneity in complementary measures (AD, RD, MD) suggests the importance of follow-up DTI studies.

Finally, an increase in cortical thickness seems to be associated with groups of neurons missing their migrating targets in the cerebral cortex leading to the formation of a neuronal nodule (Guerrini and Marini 2006). These authors propose a second hypothesis to explain structural abnormalities with the presence of polymicrogyria, an excessive number of convolutions distanced by enlarged sulci (Guerrini and Marini 2006). An increase in CT is therefore associated to a maladaptive plasticity process and is identified among several neurodevelopmental disorders such as reading impairment (Chang et al. 2005) and congenital amusia (Hyde et al. 2007).

Limitations

The diversity of developmental deafness profiles, observed in the 27 reviewed studies, considerably restricts generalization of the reported effects to the entire deaf population. Factors such as deafness onset, deafness duration, age of language acquisition, degree of hearing loss, amount of residual hearing, and use of hearing aids should ideally be considered in future analyses. Some of the studies in this review assessed cerebral changes relative to age of language acquisition (sign language) (Lyness et al., 2014; Miao et al., 2013) or age of onset versus duration of deafness (Li et al., 2012). Evidently, a larger sample size is necessary to adequately consider these multiple variables. Sample size is an important challenge in this area of research, as some of the reviewed studies reported findings from the same group of deaf individuals using different neuroimaging techniques. Also, all of the reviewed studies used a cross-sectional design. Longitudinal studies are needed to better understand the time course of deafness-related structural changes and to reduce the heterogeneity of deafness profiles. Long-term follow-ups would also allow for identification of structural changes as a function of means of communication or rehabilitation strategy.

Multiple constraints also concern the neuroimaging techniques themselves, their limits and the various types of analyses. While some studies used a whole-brain approach, others focused on regions of interest, based on previous research. In the context of this review, whole-brain interpretations of differences between deaf and hearing individuals were based solely on published findings that often omit to report null findings. Moreover, both methods require different corrections approaches (Genovese, Lazar, & Nichols, 2002) and a substantial number of reviewed studies did not apply correction methods to their findings.

Conclusion

The present systematic review aimed at regrouping the current scientific literature on brain changes following early auditory deprivation from 27 studies on 372 deaf adults and 254 deaf children. Auditory deprivation primarily alters brain structures of the primary and secondary auditory cortex and language areas. These structural changes appear to be modulated by individual variables (deafness onset, deafness duration and, mean of communication) and to influence behavioral performance during sensory and cognitive tasks. Many of these changes in cortical and subcortical auditory and language areas are negatively correlated with auditory and speech perception ability in deaf children with cochlear implants. Therefore, further neuroimaging studies are required to distinguish the heterogeneity in auditory and language outcomes in deaf children with a cochlear implant, and moreover, to optimize clinical prognosis and rehabilitation. Furthermore, early acquisition of sign language appeared to increase grey and white matter in both deaf and hearing individual. Consequently, the learning of sign language could be used as a protective factor in the neurocognitive development of deaf children. Nevertheless, the effect of sign language on neurodevelopmental outcomes of deaf children is still open for discussion.

Finally, we argue that some of the inconsistent findings may be related to deafness variables and methodological limitations of the reported neuroimaging studies. Therefore, future studies are needed to establish “best practice” guidelines for the analysis of structural brain changes in deaf individuals. To counter the issue of the generalization, we suggest well-powered studies and the addition of a hearing native signers’ group to isolate the confounded effects of sign language and auditory deprivation. We also propose longitudinal studies

comprising behavioral tasks that could help develop better rehabilitation strategies in deaf individuals.

Conflict of Interest

The authors declare that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

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Figure 1. Procedure for systematic review inspired by the PRISMA protocol (Moher et al., 2009)

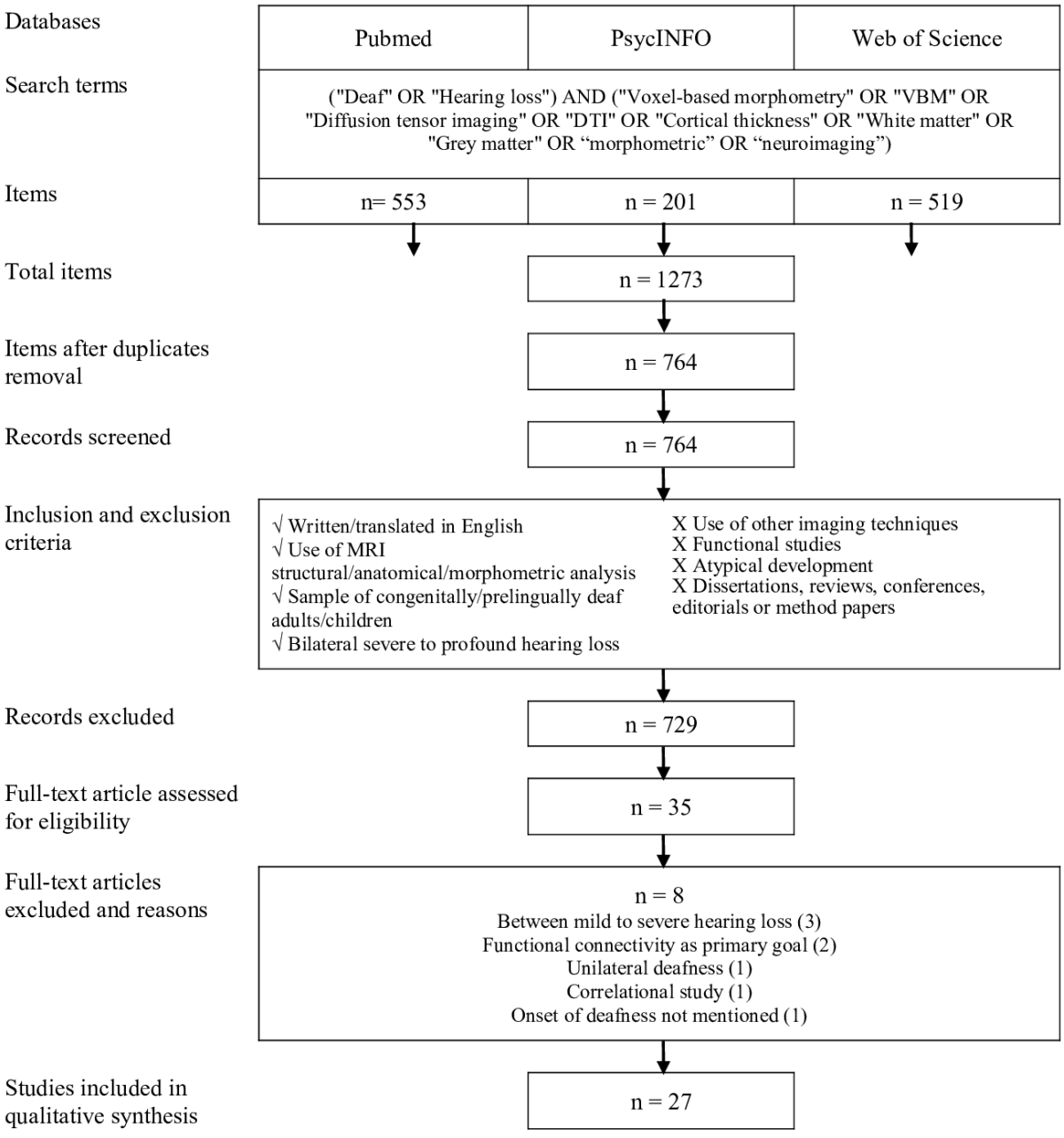

Figure 2. Overview of brain changes in 27 studies on deaf individuals

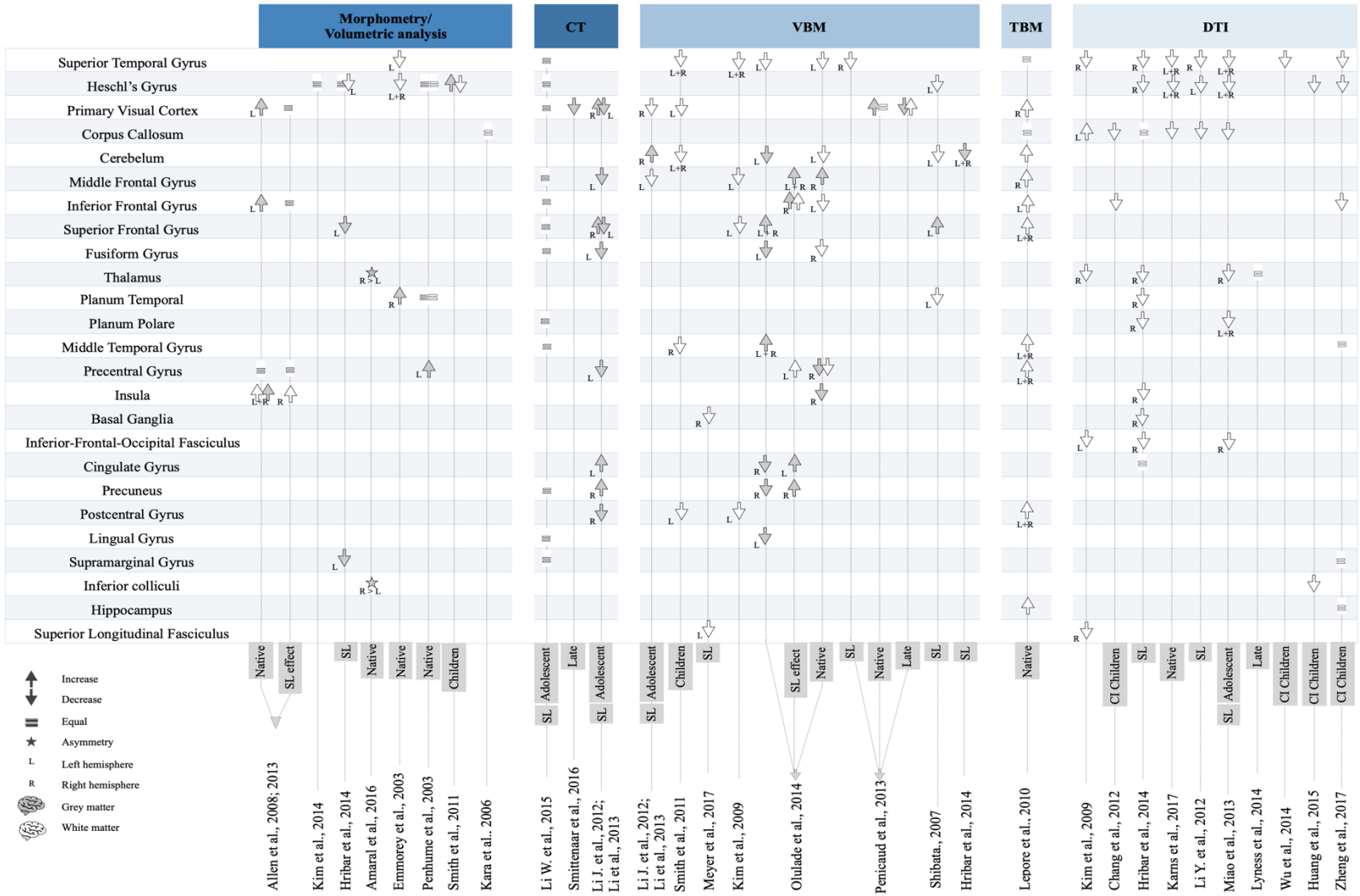

Tableau I. Main characteristic of selected articles for systematic review

Author(s)	Participants							MNI scanner strength (Testla)	Brain imaging technics	Analysis	Reference	Statistical correction
	Deaf (N)	NH (N)	Age (SD) (years)	Hearing loss	Onset	Hearing Aids (HA)	Communication preference					
Allen et al., 2008	25	25 16 CODA	28.3 (4.5)	Profound	Congenital	HA>2	NSL	1.5	MVA	ROI	NM	Bonferroni
Allen et al., 2013	25	25 16 CODA	28.3 (4.5)	Profound	Congenital	HA >2	NSL	1,5	MVA	ROI	NM	Bonferroni
Amaral et al., 2016	15	16	20.4 (NM)	Profound	Congenital	No HA	NSL	3	VA	ROI	NM	Greenhouse-Geisser & Bonferroni
Chang et al., 2012	18 CI candidates	0	5.9 (NM)	Profound	Prelingual	NM	NM	3	DTI	ROI	MNI	Uncorrected
Emmorey et al., 2003	25	25	28.3 (4.5)	Moderate to severe	Congenital	HA>2	NSL	1.5	VA	ROI	NM	Uncorrected
Hribar et al., 2014	14	14	35.4 (6)	Profound	Prelingual	No HA	SL	3	DTI, VBM, SBA, Manual volumetry	Whole brain	Talairach	Uncorrected
Huang et al., 2015	24 CI candidates	20	4.7 (1.0)	Profound	Prelingual	NM	NM	1.5	DTI	ROI	NM	Uncorrected
Kara et al., 2006	18	18	41.2 (7.5)	Profound	Prelingual	NM	NM	1.5	CT	ROI	NM	Uncorrected
Karns et al., 2017	23	26	28 (1.4)	Profound	Congenital	NM	NSL	3	DTI	ROI	NM	Uncorrected

Kim et al., 2009	13	29	29.3 (6.8)	Profound	Prelingual	HA	NM	3	DTI, VBM	Whole brain	MNI	Corrected (DTI) & uncorrected (VBM)
Kim et al., 2014 (1)	8	11	50.4 (6.1)	Severe to profound	Prelingual	No HA	SL	3	VBM	Whole brain & ROI	MNI	Bonferroni & FDR
Kim et al., 2014 (2)	11	11	50.9 (12.2)	Severe to profound	Postlingual	HA	SpL	3	VBM	Whole brain & ROI	MNI	Bonferroni & FDR
Lepore et al., 2010	14	1	29.5 (NM)	Profound	Prelingual	NM	NSL	1.5	TBM	Whole brain & ROI	Talairach	Corrected
Li J et al., 2012	16	16	14.56 (2.10)	Profound	Prelingual	NM	SL	3	CT, VBM	Whole brain	MNI	FDR
Li W et al., 2013	16	16	14.56 (2.10)	Profound	Prelingual	HA	SL	3	CT, VBM	Whole brain	MNI	FDR
Li W et al., 2015	16	16	14.56 (2.10)	Profound	Prelingual	HA	SL	3	VA	ROI	MNI	Bonferroni
Li Y et al., 2012 (1)	60	38	21.1 (2.26)	Profound	Congenital	No HA	NSL	3	DTI	Whole brain	MNI	FDR
Li Y et al., 2012 (2)	36	38	21.5 (1.54)	Profound	Prelingual	No HA	SL	3	DTI	Whole brain	MNI	FDR
Lyness et al., 2014	13	13 NHSL	39.08 (11.08)	Severe to profound	Congenital	NM	SpL & LSL	1.5	DTI	ROI	NM	FDR
Meyer et al., 2007	6	6	23.5 (NM)	Profound	Congenital	NM	SL	3	VBM	Whole brain	MNI	Uncorrected
Miao et al., 2013	16	16	14.56 (2.10)	Profound	Prelingual	HA	SL	3	DTI	Whole brain	MNI	Corrected

Olulade et al., 2014 (1)	15	15	23.4 (3.3)	Profound	Congenital	NM	NSL	3	VBM	Whole brain	MNI	Uncorrected
Olulade et al., 2014 (2)	15	15	28.2 (3.8)	Profound	Congenital	NM	SpL	3	VBM	Whole brain	MNI	Uncorrected
Penhune et al., 2003	12	10	29 (NM)	Profound	Congenital	NM	NSL	1.5	VBM, VA	Whole brain & ROI	Talairach	Uncorrected
Pénicaud et al., 2013	23	43	39.2 (12.2)	Severe to profound	Congenital	NM	NSL (9) SL (8) LSL (6)	1.5	VBM	Whole brain & ROI	MNI	Corrected
Shibata, 2007	53	51	21 (NM)	Profound	Prelingual	NM	SL	1.5	VBM	Whole brain	MNI	Bonferroni
Smith et al., 2010	16 CI candidates	26	1,167 (0.25)	Moderate to severe	Congenital	NM	NM	3	VBM, MVA	Whole brain & ROI	MNI	FDR
Smithenaar et al., 2016	14	15 NHSL	39 (10.2)	Severe to profound	Congenital	NM	LSL	1.5	CT	Whole brain & ROI	NM	Greenhouse-Geisser
Wu et al., 2014	92 CI candidates	0	4.9 (NM)	Profound	Prelingual	NM	NM	1.5	DTI	ROI	NM	Uncorrected
Zheng et al., 2017	72/20 CI candidates	38 NH	4.7 (1.0)	Severe to profound	Prelingual	HA	NM	3	DKI	ROI	NM	Uncorrected

Notes. CI Cochlear implant candidate; CODA, Children of deaf adults; NH, Normo-hearing; NHSL, Normo-hearing sign language users; NSL, Native sign language; SL, Sign language; LSL, Late acquired sign language; SpL, Spoken Language; VBM, Voxel Based Morphometry; TBM, Tensor Based Morphometry; CT, Cortical Thickness; DTI, Diffusion tensor imaging; DKI, Diffusion kurtosis imaging; SBA, Surface based analysis; VA, Volumetric analysis; MVA, Morphometric/Volumetric analysis; ROI, Region of interest; MNI, Montreal Neurological Institute; FDR, False discovery rate; NM, Not mentioned.

Chapitre V : Discussion Générale

5. De la privation à la restauration auditive

L'objectif général de cette thèse était d'enrichir notre compréhension actuelle de la réorganisation cérébrale à la suite d'une absence précoce d'afférence auditive. Dans les précédents chapitres, deux études ont été présentées portant sur la plasticité fonctionnelle puis, sur la plasticité structurelle chez l'individu sourd. À présent, nous souhaitons discuter de ces résultats à la lumière des données existantes dans la littérature. Par la suite, nous discuterons des limites de nos études en lien avec les enjeux méthodologiques qui entourent la population sourde dans le but d'offrir des suggestions pour les recherches futures.

5.1 Réorganisation cérébrale fonctionnelle

Dans la première étude, nous avons investigué la sensibilité des individus sourds à reconnaître des actions humaines, à savoir des emblèmes et des pantomimes. Le signal BOLD de participants sourds et neurotypiques a été enregistré par le biais de l'imagerie par résonnance magnétique fonctionnelle (IRMf) et mis en relation avec les données comportementales d'exactitude et de temps de réponse obtenues lors d'une tâche de reconnaissance d'actions humaines, sur la base d'animations de points lumineux. En premier lieu, nous discuterons des résultats comportementaux obtenus dans le premier article de cette thèse, en les comparant avec les connaissances scientifiques portant sur le lien entre cerveau-comportement au sein de la population des sourds.

5.1.1 Interprétation du versant comportemental

Le résultat comportemental principal de notre première étude démontre que les individus sourds présentent une sensibilité à la reconnaissance de l'action humaine en comparaison à des

individus neurotypiques, dans la mesure où ils reconnaissent aussi rapidement les emblèmes que les pantomimes. Chez les individus dont l'audition est intacte, on observe des temps de réponse plus courts pour la reconnaissance des pantomimes que des emblèmes, suggérant une faiblesse relative pour déterminer rapidement le caractère communicatif des emblèmes. Aucune différence entre les groupes n'est observée en ce qui concerne l'exactitude. Notre étude est donc la première à identifier une différence comportementale entre les emblèmes et les pantomimes chez les individus sourds et neurotypiques. Cette différence comportementale est cohérente avec les études antérieures sur de nombreux aspects et notamment, avec l'hypothèse d'une amélioration de la réactivité visuelle chez les individus sourds (Pavani & Bottari, 2012). Basée sur de multiples études de détection visuelle au cours desquelles les participants devaient détecter le plus rapidement possible des cibles présentées aléatoirement sur un écran (Bottari, Caclin, Giard, & Pavani, 2011; Bottari, Nava, Ley, & Pavani, 2010; Codina, Buckley, Port, & Pascalis, 2011; Loke & Song, 1991; Neville & Lawson, 1987), cette hypothèse suggère que les différences comportementales entre les individus sourds et neurotypiques sont observées au niveau des temps de réaction et non de l'exactitude des réponses (Pavani & Bottari, 2011a). Ainsi, plusieurs études convergent vers une diminution du seuil de détection du mouvement chez les individus sourds pour des stimuli visuels de points en mouvement (Bosworth & Dobkins, 2002; Hauthal et al., 2013; Neville & Lawson, 1987; Shiell et al., 2014a). Plus récemment, une étude portant sur les aspects phonologiques et morphosyntaxiques de la langue des signes démontre une différence comportementale en termes de temps de réponse entre des individus neurotypiques et sourds (Cardin, Orfanidou, et al., 2016). Les individus sourds se montraient plus rapides pour reconnaître des signes que des non-signes (séquences gestuelles présentant toutes les caractéristiques de la langue des signes).

Il est d'autant plus intéressant de noter ici que cette différence est observée indépendamment de l'expérience linguistique de ces individus, puisque ce résultat était obtenu suivant que les individus soient oralistes ou natifs de la langue des signes. Ainsi, notre première étude confirme que cette sensibilité visuelle se généralise pour des stimuli en mouvement de niveau plus complexe, peu importe l'expérience linguistique. Une meilleure détection visuelle du mouvement chez les sourds pourrait s'étendre, non pas uniquement à un niveau perceptif, mais bien à des processus cognitifs de haut niveau, permettant alors un traitement plus rapide. Ces données sont la résultante d'un mécanisme de compensation à la perte auditive qui permet l'amélioration de certaines compétences visuelles chez les personnes privées de l'audition, leur offrant des indices supplémentaires pour faire face à leur handicap auditif.

Alors que nous voyons se confirmer l'hypothèse d'une réactivité visuelle accrue chez les individus sourds, de nombreuses études antérieures ne démontraient aucune différence comportementale. Plusieurs mécanismes explicatifs étaient proposés. Dans une récente revue de la littérature, Alencar et collaborateurs (2019) regroupent l'ensemble des tâches dans lesquelles les individus sourds obtiennent des performances comportementales similaires à des pairs neurotypiques, telles que la direction du mouvement (Bosworth & Dobkins, 2002; Hauthal et al., 2013), la vélocité (Brozinsky & Bavelier, 2004), la discrimination d'orientation et la discrimination faciale (Parasnis, Samar, Bettger, & Sathe, 1996), ainsi que la sensibilité aux contrastes (Finney & Dobkins, 2001b). Cette absence de différence a été initialement interprétée comme pouvant subvenir chez les individus sourds en raison de la faible charge attentionnelle requise pour réaliser ses tâches de seuil de détection (Bavelier, Dye, & Hauser, 2006; Dye, Baril, & Bavelier, 2007). Toutefois, de nombreuses études subséquentes ont ultérieurement démontré des améliorations comportementales au sein d'habiletés visuelles tout

en prenant soin de contrôler pour le degré de charge attentionnelle requis au cours des tâches (Alencar et al., 2019).

Alternativement, une hypothèse proposée afin d'expliquer les résultats comportementaux observés chez les individus sourds consistait à dire que la privation auditive avait un impact majoré sur les fonctions de la voie visuelle dorsale (Bavelier & Neville, 2002). Classiquement, le système visuel de l'homme comprend deux voies visuelles principales : la voie ventrale et la voie dorsale (Ungerleider, Mishkin, Ingle, Goodale, & Mansfield, 1982). La voie ventrale dont les projections s'étendent du cortex occipital au cortex temporal est impliquée principalement dans le traitement des visages, des objets, des formes et de la couleur. La voie dorsale, quant à elle, possède des projections allant du cortex occipital et temporal vers le cortex pariétal. Elle est principalement impliquée dans les aspects visuels liés à l'information spatiale. Plusieurs auteurs suggéraient ainsi qu'en raison de l'utilisation quotidienne de la langue des signes par les individus sourds, ceux-ci démontraient d'une tendance à utiliser préférentiellement les indices visuo-spatiaux disponibles dans l'environnement, expliquant l'amélioration comportementale de certains processus visuels appartenant au traitement de la voie dorsale (par ex. Bosworth & Dobkins, 2002; Emmorey, Klima, & Hickok, 1998). Toutefois, cette hypothèse échoue à expliquer certaines absences de différences comportementales mentionnées ci-haut, telles que pour la vélocité (Brozinsky & Bavelier, 2004), ou encore la direction du mouvement (Bosworth & Dobkins, 2002; Hauthal et al., 2013), habiletés visuelles qui devraient être traitées par la voie dorsale. De plus, comme pour l'étude de Cardin et collaborateurs (2016), notre premier article démontre que les améliorations visuelles semblent être mieux expliquées par la privation auditive précoce et non par l'utilisation exclusive de la langue des signes.

La variabilité des résultats comportementaux obtenus lors de tâches visuelles auprès des individus sourds n'en reste pas moins un défi pour l'interprétation. Ce constat n'est pas spécifique à la privation auditive puisque le même phénomène d'incohérence comportementale est retrouvé également à la suite d'une privation visuelle précoce (Voss, 2018). L'hétérogénéité dans les profils d'individus sourds, section qui sera détaillée dans une section ultérieure, explique probablement en partie cette variabilité. Toutefois, nous pouvons également remettre en question le type de méthodologie expérimentale utilisé et tout particulièrement les tâches privilégiées. À ce sujet, une récente étude explicite la pertinence et la validité d'utiliser la réalité virtuelle dans le cadre de l'évaluation des fonctions visuelles (Bennett, Bex, Bauer, & Merabet, 2019). Nous pensons que l'utilisation de ce type de procédé méthodologique, dit écologique, permettra, sans nul doute, une meilleure estimation de l'impact au quotidien de la perte auditive, mais également de la compensation par les habiletés visuelles de l'individu sourd.

5.1.2 Interprétation du versant fonctionnel

Parmi les résultats majeurs qu'apporte notre première étude, citons en premier lieu la confirmation que les individus sourds et neurotypiques présentent un réseau de la reconnaissance de l'action humaine similaire. Ainsi, les activations cérébrales obtenues auprès de nos deux groupes concordent avec les récentes études démontrant que le réseau de l'action humaine implique le recrutement du gyrus inférieur frontal, du cortex prémoteur dorso-latéral, l'aire motrice supplémentaire, le lobule pariétal inférieur, le cortex somatosensoriel primaire, le sillon temporal supérieur, le cortex intrapariétal, le gyrus temporal médian associé à l'aire visuelle supplémentaire V5 ainsi que les aires du gyrus fusiforme spécialisées pour le traitement du visage et du corps (Caspers et al., 2010). Cette confirmation est importante, car

par le passé, des divergences étaient rapportées dans la littérature, du fait notamment que le traitement de l'action humaine par l'homme n'était interprété qu'au sein de la théorie du réseau des neurones miroirs (Fabbri-Destro & Rizzolatti, 2008). Ainsi, deux études étaient en faveur d'une hypoactivation du réseau des neurones miroirs chez les individus sourds signeurs lors de tâches impliquant des pantomimes (Corina et al., 2007; Emmorey et al., 2010), les résultats rapportés n'étaient toutefois pas corrigés pour comparaisons multiples. Ces auteurs suggéraient alors que la pratique extensive de la langue des signes par les individus sourds entraînait une extinction des activations typiquement attendues dans le réseau des neurones miroirs. À contrario, d'autres études obtiennent des activations cérébrales similaires entre les individus sourds natifs de la langue des signes et les individus neurotypiques lors de tâches impliquant la reconnaissance de pantomimes, de gestes sans signification ou d'un unique emblème (thumbs-up) (Fang et al., 2016; Husain et al., 2012, 2009; MacSweeney et al., 2004, 2008). Toutefois, la première étude de cette thèse se démarque des précédentes recherches dans la mesure où il s'agit de la seule à considérer simultanément les pantomimes et plusieurs emblèmes au sein des actions humaines. De plus, en incluant une condition contrôle dont les activations sont soustraites aux activations conjointes de la reconnaissance des pantomimes et des emblèmes (Zaini et al., 2013), nous isolons les activations liées plus généralement au traitement visuel de bas niveau afin de circonscrire nos résultats au traitement de l'action humaine.

En outre, notre première étude démontre qu'au-delà de ce recouvrement cortical entre les deux populations testées, les individus sourds démontrent d'un surplus d'activations cérébrales observées bilatéralement au sein du gyrus temporal supérieur. Par le passé, une étude observait une activation du gyrus temporal supérieur gauche, et notamment du planum

temporale chez des individus sourds natifs de la langue des signes lors d'une tâche comparant un unique emblème (thumbs up/thumbs down) avec des gestes sans signification (Husain et al., 2012). Pour ces auteurs, cette activation s'expliquait par un traitement en unité linguistique de cet emblème par les individus sourds, en lien avec leur pratique de la langue des signes, les amenant ainsi à traiter linguistiquement des actions humaines signifiantes. En ce qui a trait aux pantomimes, de très larges clusters bilatéraux autour du cortex auditif des individus sourds étaient activés dans la précédente étude, ne résistant toutefois pas à la correction pour comparaisons multiples (Fang et al., 2016). Rappelons qu'aucune différence comportementale entre les individus sourds et neurotypiques n'avait été obtenue au sein de ces deux dernières recherches. Nos résultats sont d'importance, ils démontrent que la reconnaissance des actions humaines par les individus sourds implique une série d'activations cérébrales, dont la principale est retrouvée bilatéralement au sein du gyrus temporal supérieur, incluant le planum temporale, mais également les cortex auditifs primaire et secondaire. De plus, nous démontrons d'une différence comportementale entre les deux populations testées. Cette différence comportementale en termes de temps de réponse pour les emblèmes chez les sourds est corrélée significativement avec l'activation du gyrus temporal supérieur droit et marginalement à gauche, chez les individus sourds. Dans un récent article, Singh et collaborateurs (2018) offrent une modélisation des deux explications à l'origine de la réorganisation cérébrale fonctionnelle chez les individus privés de la vision. Dans les prochaines lignes, nous tenterons d'appliquer ce modèle au cas spécifique des individus sourds, à la lumière de nos résultats.

La *première explication* énonce le rôle compensatoire de la réorganisation cérébrale chez les personnes privées d'un sens précocement (Singh et al., 2018). Ainsi, les activations

du cortex auditif par des stimuli visuels desservent un but compensatoire à la perte sensorielle. Nos résultats s'inscrivent au sein de cette explication et sont cohérents avec les multiples études préalables ayant démontré un avantage comportemental pour la détection du mouvement visuel chez les sourds en lien avec des activations dans les régions cérébrales impliquées principalement dans le traitement auditif (Bosworth & Dobkins, 2002; Fine et al., 2005; Finney et al., 2003; Hauthal et al., 2013; Neville & Lawson, 1987; Sadato et al., 2005; Shiell et al., 2014a; Vachon et al., 2013). Pour compenser leur handicap auditif, les individus sourds témoignent de plus amples ressources visuelles que les individus neurotypiques qui nécessitent la réallocation des régions auditives. Toutefois, cette première explication, à elle seule, ne peut expliquer l'ensemble des données obtenues auprès des individus sourds. Quelques études démontrent que l'amélioration comportementale visuelle des individus sourds est associée à une réorganisation intramodale, soit une activation accrue du complexe MT+ (V5) et des régions visuelles primaires (Bavelier et al., 2001; Bottari et al., 2011; Hauthal et al., 2013). Bien que nos résultats n'aillent pas en ce sens, nous soutenons cette hypothèse dans la mesure où le choix de soustraire notre condition contrôle dans nos analyses de contraste en IRMf ne nous permet pas d'obtenir cette information. En outre, Singh et collaborateurs (2018) explicitent que les améliorations comportementales chez les individus aveugles souffrent généralement d'une faible magnitude qui est, selon eux, comparable à celle observée par l'effet d'un entraînement chez des individus typiques. Ces données sont très intéressantes et mériteraient d'être opérationnalisées auprès des individus sourds. À notre connaissance, aucun chercheur n'a mesuré l'impact d'un entraînement visuel chez des individus sourds en comparaison à des individus neurotypiques. Il pourrait s'agir d'une belle perspective en termes de restauration auditive puisque la réorganisation cérébrale dans le cas d'une perte sensorielle

n'est pas bénéfique de façon univoque et s'avère aussi inadaptée à la restauration de la fonction. Dans le cas de la déficience auditive, il est largement admis que le recrutement du cortex auditif par les autres modalités sensorielles nuit aux performances auditives obtenues post-implantation (par ex. : Doucet, Bergeron, Lassonde, Ferron, & Lepore, 2006). Finalement, cette première explication questionne également le rôle de l'étendue de la réorganisation, puisque de nombreuses études (Cardin et al., 2013; Fine et al., 2005; Finney & Dobkins, 2001a; Sadato et al., 2005; Shiell, Champoux, & Zatorre, 2014b; Vachon et al., 2013) et notamment la nôtre, obtiennent une large activation du cortex auditif primaire et secondaire par diverses stimulations visuelles chez les individus sourds. Pourtant, l'impact comportemental de cette activation reste mineur. À raison, ce modèle questionne alors de l'unique rôle de la réorganisation corticale à des fins de compensation alors que nous observons une utilisation sous-optimale des ressources corticales et il nous renvoie à la précédente section de cette discussion portant sur un choix plus optimal des tâches comportementales à utiliser.

La *seconde explication* consiste à dire que la plasticité cérébrale à la suite d'une privation sensorielle est inévitable dans la mesure où elle résulte de connexions cérébrales présentes dès la naissance, dont la spécialisation s'oriente vers les fonctions sensorielles préservées (Singh et al., 2018). Alors que les régions sensorielles ont largement étaient considérées comme unimodales par le passé, de récentes évidences démontrent la nature multimodale des régions cérébrales sensorielles primaires (par ex. Driver & Noesselt, 2008) et dans le cas spécifique de cette thèse, du cortex auditif primaire et secondaire (Bizley et al., 2006; Lakatos, Chen, O'Connell, Mills, & Schroeder, 2007). En l'absence d'afférence auditive soit de la modalité dominante, ces connexions multimodales préexistantes dans le cerveau

typique se renforcent par l'inexpérience auditive, ce qui offre plus de place aux autres modalités sensorielles qui deviennent alors dominantes. Nos résultats et les activations cérébrales observés au sein du cortex auditif primaire et du cortex auditif secondaire lors de la reconnaissance d'actions humaines chez les individus sourds de notre étude peuvent également s'inscrire conjointement au sein de cette seconde explication. Il ne s'agirait alors plus d'une réorganisation cérébrale intermodale, mais multimodale. Cette hypothèse corrobore l'activation du cortex auditif primaire, du cortex auditif secondaire et associatif (planum temporale), lors du traitement fonctionnel de stimulations visuelles de bas niveau (Cardin et al., 2013; Fine et al., 2005; Finney & Dobkins, 2001a; Vachon et al., 2013) ainsi que pour les propriétés visuelles de la langue des signes (Cardin et al., 2013; MacSweeney et al., 2002; Petitto et al., 2000; Sadato et al., 2005). Les avantages comportementaux observés chez les individus sourds pourraient alors s'expliquer par des connexions top-down requises dans des tâches cognitives de plus haut niveau, nécessitant notamment une prise de décision (Singh et al., 2018). Par conséquent, ces connexions top-down pourraient également justifier l'absence de différences comportementales lors de tâches simples de détection visuelle (Alencar et al., 2019).

Finalement, une étude récente de connectivité fonctionnelle au repos réalisée auprès de chats, dont une surdité transitoire avait été induite, corrobore la présence concomitante et non exclusive de ces deux explications théoriques (Stolzberg, Butler, & Lomber, 2018). En effet, ces auteurs rapportent la présence simultanée d'altérations et de connexions accrues en termes de connectivité fonctionnelle entre le réseau auditif et certaines régions comprises dans les autres modalités sensorielles (visuelles, motrices, somatosensorielles). La reproduction de

cette étude auprès d'individus sourds nous semble une avenue très pertinente pour l'interprétation des données chez l'homme.

À la lumière des dernières sections, les évidences collectées à ce jour chez les individus présentant une déficience auditive précoce ne permettent pas d'entériner un modèle de la réorganisation cérébrale de la surdité uniquement sur la base des données fonctionnelles et comportementales. De nombreuses recherches également se sont intéressées à l'étude des structures cérébrales sensibles à la privation auditive précoce et constitueront le cœur de la prochaine partie.

5.2 Réorganisation cérébrale structurelle

5.2.1 Interprétation du versant structurel

À ce stade des avancées scientifiques et technologiques, l'élaboration d'une revue systématique de la littérature paraît nécessaire afin de résumer et d'intégrer les résultats des multiples études existantes et d'offrir un état des lieux des données actuelles en vue de proposer de nouvelles avenues de recherche. Par conséquent, notre second article répond à ce besoin et suggère de nouvelles pistes de compréhension des altérations cérébrales de type structurel chez les individus sourds. Nous désirons à présent mettre en relation les principales altérations structurelles observées dans notre second article avec la qualité du développement neurocognitif et de la restauration auditive.

Comme nous l'avons vu précédemment en introduction de cette thèse, la restauration auditive des personnes dont la perte auditive est apparue dès la naissance ou avant l'acquisition du langage constitue un défi permanent pour les professionnels (médecins,

orthophonistes, professeurs), pour les parents des enfants sourds et les sourds eux-mêmes. Dans les prochaines lignes, nous souhaitons modéliser la façon dont ces altérations cérébrales identifiées par notre revue systématique, en raison notamment de la proximité anatomique des régions cérébrales dédiées à l'audition et au langage, entraînent des répercussions sur les habiletés langagières des individus sourds. De plus, notre second article met en lumière toute une série d'altérations structurelles dans des régions cérébrales impliquées dans le traitement perceptif visuel, mais également des fonctions cognitives de plus haut niveau. Ces fonctions cognitives ont fait l'objet de multiples études chez les individus sourds et tout particulièrement auprès d'enfants sourds implantés, porteurs de prothèses auditives ou signeurs, une revue de la littérature à ce sujet est annexée à cette thèse et les données qui y sont recueillies seront intégrées à la présente discussion.

La qualité de la restauration auditive est classiquement évaluée par l'efficacité de l'enfant ou de l'adulte sourd, une fois appareillé ou implanté, à percevoir les sons et la parole dans des conditions de silence ou de bruit (Nikolopoulos, Archbold, & Gregory, 2005; Turgeon, Lazzouni, Lepore, & Ellemberg, 2014). En outre, la qualité de la restauration auditive a une influence importante sur le développement des habiletés langagières et des processus neurocognitifs subséquents (Houston et al., 2012; Kral, Dorman, & Wilson, 2019; Pisoni, Kronenberger, Harris, & Moberly, 2017) et bien évidemment sur leur qualité de vie (Castellanos, Kronenberger, & Pisoni, 2018). De nombreux facteurs associés au handicap sensoriel, tels que la durée de la surdité, la sévérité de la surdité, l'étiologie, le degré d'audition résiduelle, ou encore des facteurs associés à la qualité de la prise en charge, comme le diagnostic précoce, l'utilisation préalable d'une prothèse auditive et l'âge de l'implantation sont connus pour influencer le devenir perceptif des individus sourds et notamment l'efficacité

de la restauration auditive par l'implant cochléaire (par ex. Blamey et al., 2012; Giraud & Lee, 2007; Lazard, Giraud, Gnansia, Meyer, & Sterkers, 2012; Moberly, Bates, Harris, & Pisoni, 2016). L'implant cochléaire permet la conversion du signal acoustique en impulsions électriques qui viennent stimuler les fibres nerveuses auditives (Yawn et al., 2015). Bien que cette technologie existe depuis plus de trente ans, une importante variabilité interindividuelle persiste dans les performances de perception auditive et de perception de la parole obtenues post-implantation (par ex. van Wieringen & Wouters, 2014). L'origine de cette variabilité demeure encore inconnue, mais constitue l'un des thèmes de recherche les plus discutés de la littérature portant sur la déficience auditive.

Parmi les principales généralisations que nous obtenons à travers cet exercice de synthèse, nous pouvons en premier lieu conclure que la privation auditive précoce entraîne des altérations structurelles des fibres de la matière blanche au sein des régions corticales et sous-corticales impliquées dans le traitement de l'audition. Ces résultats sont rapportés sur tout le spectre des âges, soit autant chez de jeunes enfants sourds que chez des adultes sourds. De plus, ces altérations ont pour origine l'absence d'afférence auditive qui nuit au processus naturel de myélinisation au sein de ces structures cérébrales (Lebel & Beaulieu, 2011) alors même que les structures liées à la perception auditive font partie des régions qui arrivent les premières à maturation lors du développement cérébral typique (Gogtay et al., 2004). La récurrence de ces altérations structurelles tant chez les individus sourds oralistes que signeurs suggère une absence d'effet du mode de communication sur ces changements, mais bien un impact précoce de l'inexpérience auditive. Ce constat est confirmé par les quelques études ayant démontré une corrélation négative entre les altérations structurelles des régions auditives primaires et secondaires observées préimplantation avec les habiletés de perception de la

parole chez de jeunes enfants sourds ayant reçu un implant cochléaire (Huang et al., 2015; Wu et al., 2016; Zheng et al., 2017). À ce stade, il est peu probable que le mode de communication puisse avoir un impact sur le développement cortical des régions auditives, toutefois, l'absence de langage fonctionnel pourrait est contributif de ces altérations (Robinson, 1998), et nous y reviendrons dans les prochaines lignes. De plus, ces changements sont également observés auprès de jeunes enfants sourds candidats à l'implant cochléaire âgés de moins deux ans (Smith et al., 2011). Ces résultats concordent avec la période dite critique, soit la fenêtre temporelle au cours de laquelle le cerveau se montre particulièrement plastique pour répondre à la restauration de la fonction touchée (Kral & Eggermont, 2007). Au-delà de cette période, la réorganisation cérébrale au niveau fonctionnelle, soit le recrutement des régions auditives par les autres modalités sensorielles préservées (par ex. Shiell, Champoux, & Zatorre, 2014b; Vachon et al., 2013), ainsi que la réorganisation cérébrale structurelle des régions auditives (Huang et al., 2015; Wu et al., 2016; Zheng et al., 2017) sont définies comme des changements inadaptés à la restauration auditive par l'implant cochléaire (Heimler et al., 2014). Chez l'enfant sourd, l'implantation cochléaire est actuellement encouragée avant l'âge de trois ans (Sharma & Campbell, 2011). Ainsi, de meilleures capacités de perception de la parole sont retrouvées chez les enfants sourds implantés avant l'âge de deux ans (par ex. Svirsky, Teoh, & Neuburger, 2004).

Au-delà des altérations dans les régions cérébrales impliquées dans l'audition, l'inexpérience auditive précoce entraîne une cascade neurodéveloppementale sur les autres structures cérébrales du cerveau et par conséquent sur les fonctions cognitives qui y sont associées (Grantham-McGregor et al., 2007). Comme nous l'avons mentionné ci-haut, la proximité anatomique entre les régions auditives et celles impliquées dans le langage entraîne

des altérations structurelles chez les individus sourds. Ainsi, une altération de l'intégrité, du volume ou de la densité de la matière blanche au sein du gyrus temporal supérieur fait consensus dans notre travail de synthèse. À nouveau, une corrélation négative est obtenue entre les performances de perception de la parole obtenues post-implantation et l'altération structurelle observée initialement chez les enfants sourds candidats à l'implant cochléaire (Smith et al., 2011; Zheng et al., 2017). D'autres régions impliquées dans le traitement du langage connaissent des changements bien moins consensuels, telles que le planum temporale, le gyrus temporal médian, le gyrus supramarginal, le gyrus angulaire ainsi que l'insula (Allen, Emmorey, Bruss, & Damasio, 2008; Hribar, Šuput, Carvalho, Battelino, & Vovk, 2014; Olulade, Koo, LaSasso, & Eden, 2014). Alors que les données de neuroimagerie chez les enfants sourds sont assez rares, elles sont abondantes du point de vue comportemental et tout particulièrement en ce qui a trait aux habiletés langagières obtenues à la suite d'un implant cochléaire. Après trente années de recherche, les études portant sur les compétences langagières témoignent toujours d'une déviation sous la norme d'environ un écart-type chez les enfants sourds implantés (Cupples et al., 2018). Néanmoins, l'âge de l'implantation constitue un facteur déterminant puisque les enfants sourds implantés précocement présentent de meilleures habiletés langagières autant sur le versant expressif (Tomblin, Barker, Spencer, Zhang, & Gantz, 2005) que sur le versant réceptif (Niparko et al., 2010). À l'instar de la maturation sensorielle qui connaît une période critique, l'acquisition du langage a également été associée à une période critique (Chomsky & Lenneberg, 1967), soit qu'une absence d'exposition langagière de qualité avant l'âge d'un an entraîne un déclin de la maturation corticale et des retards au long court tout particulièrement sur le plan syntaxique. Ces observations ont été rapportées par le biais d'études portant sur le bilinguisme ou encore

auprès d'enfants sauvages (Friedmann & Rusou, 2015). Ainsi, les habiletés langagières des enfants sourds implantés plus tardivement, soit avant quatre ans, tendent à se rapprocher de celles des enfants implantés précocement, tout particulièrement en termes de richesse lexicale (Dunn et al., 2014), néanmoins, des fragilités sur le plan grammatical et syntaxique persistent (Friedmann & Rusou, 2015).

Si l'on poursuit la cascade neurodéveloppementale, des délais dans l'acquisition des habiletés langagières chez les enfants sourds entraînent des répercussions sur l'émergence des fonctions exécutives (Kuhn, Willoughby, Wilbourn, Vernon-Feagans, & Blair, 2014), c'est-à-dire les capacités à faire preuve de flexibilité, d'inhibition, de mise à jour, de catégorisation, de planification ainsi que sur la mémoire de travail (Miyake et al., 2000). D'après notre second article, des altérations structurelles sont observées chez les individus sourds privés de l'audition précocement au niveau de certaines régions impliquées dans les fonctions exécutives, à savoir, le gyrus frontal médian et le gyrus frontal supérieur. Ces altérations sont toutefois observées à partir de l'adolescence (Li et al., 2012; Li et al., 2013), se maintiennent une fois adultes (Olulade, Koo, LaSasso, & Eden, 2014; Shibata, 2007) et ce, uniquement chez des individus signeurs mais non natifs de la langue des signes. Sur le plan neurocognitif, de récentes études ont démontré que les enfants sourds natifs de la langue des signes présentaient des capacités en termes de fonctionnement exécutif se situant dans la norme en comparaison à des enfants typiques (Hall et al. 2017; Marshall et al. 2015). Toutefois, chez les enfants sourds bénéficiant d'un implant cochléaire, un rendement sous-optimal de plusieurs fonctions exécutives est rapporté (pour une revue exhaustive voir la Table 1 située en annexe), notamment en ce qui a trait à la mémoire de travail. À la lumière de ces observations, mais en l'absence d'études corrélationnelles entre altérations structurelles et fonctionnement exécutif

chez l'individu sourd, nous pouvons néanmoins spéculer que ces altérations dans les régions frontales sont la conséquence d'une sous-stimulation langagière en jeune âge. L'apprentissage tardif de la langue des signes ne leur permettant alors pas un développement neurocognitif optimal.

Dès le préambule de cette thèse, nous décrivions le lien étroit et complexe entre la privation auditive et le langage dans le contexte de la surdité. Nous comprenons à présent mieux la relation entre ces deux entités en termes de réorganisation cérébrale structurelle et notamment des répercussions qu'elle peut avoir sur le développement neurocognitif et la qualité de la restauration auditive. Il ne s'agit pas seulement de tenir compte de la seule période critique pour la restauration de la fonction atteinte, mais bien, dès le dépistage effectué, d'offrir un niveau de stimulation langagière suffisant en dépit de quoi, des déficits linguistiques tendent à perdurer et à se répercuter sur le bon développement cognitif, notamment des fonctions exécutives.

5.3 Limites et perspectives

5.3.1 Contraintes pour l'interprétation des données structurelles

À présent, certains aspects généraux de la plasticité cérébrale structurelle nous semblent importants à discuter et pourraient aider à interpréter les observations rapportées au sein de notre seconde étude. Comme il l'a été dit à plusieurs reprises, l'expérience entraîne des modifications corticales. Citons par exemple le cas de l'entraînement musical, pour lesquels des changements de volume de matière grise se produisent dans les régions auditives, motrices et visuospatiales dès qu'un individu entreprend un apprentissage musical (Groussard et al.,

2014). En outre, l'expertise musicale induit des modifications structurelles au sein des régions impliquées dans des processus cognitifs supérieurs tels que les fonctions exécutives, la mémoire ou le traitement des émotions (par exemple, Groussard et al., 2014). Par conséquent, une augmentation du volume de la matière grise dans les régions cérébrales impliquées dans les modalités somatosensorielle et auditives a été démontrée chez des chanteurs d'opéra (Kleber et al., 2016) et interprétée comme un mécanisme de compensation menant à des capacités améliorées. Toutefois, de multiples études démontrent de résultats contradictoires, à savoir une augmentation rapide du volume de la matière grise dans les zones cérébrales sensorimotrices suivie d'une diminution après l'entraînement (Calmels, 2019). Une interprétation prudente est nécessaire en ce qui concerne les relations cerveau-comportement lorsque l'on examine les différentes métriques d'analyses de la substance grise (densité/volume). En outre, une augmentation de l'épaisseur corticale semble être associée à des groupes de neurones manquant leurs cibles dans le cortex cérébral, ce qui conduit à la formation d'un nodule neuronal (Guerrini & Marini, 2006). Ces auteurs proposent une seconde hypothèse pour expliquer les anomalies structurelles avec la présence de polymicrogyries, un nombre excessif de convolutions distancées par des sillons élargis (Guerrini & Marini, 2006). Une augmentation de l'épaisseur corticale est donc associée à un processus de plasticité inadaptée et est identifiée parmi plusieurs troubles du développement neurologique tels que les troubles de la lecture (Chang et al., 2005) et l'amusie congénitale (Hyde et al., 2007). Actuellement, les altérations de la matière grise demeurent probablement la cause de multiples mécanismes cellulaires et moléculaires qui sont très peu connus (Zatorre, Fields, & Johansen-Berg, 2013).

En ce qui concerne la substance blanche, l'imagerie par résonance magnétique de diffusion est un puissant instrument pour étudier les corrélats anatomiques et les modifications en termes de fibres (diamètre, densité) ou de myélinisation. De multiples études portant sur des populations cliniques ont démontré une altération de la substance blanche, comme pour la maladie d'Alzheimer (Damoiseaux et al., 2009), la schizophrénie (Qiu et al., 2010) ou encore le syndrome de Tourette (Neuner et al., 2010). En lien avec l'expérience, une pratique extensive du piano est associée à une myélinisation accrue chez les enfants qui montre une tendance à se maintenir avec l'âge (Bengtsson et al., 2005). Toutefois, l'imagerie par diffusion est une technique complexe en raison de la nature de la substance blanche et du vaste choix d'analyses disponibles. Cette complexité conduit à de nombreuses erreurs d'interprétations dans les résultats préalablement obtenus telles que la prise en considération des croisements entre fibres à l'intérieur des voxels (Jones, Knösche, & Turner, 2013). Ainsi, la grande majorité des recherches inclues dans notre second article de thèse ne rapportent que des changements au niveau de la fraction d'anisotropie (FA) représentant l'index général de l'intégrité et de la direction des fibres à l'intérieur d'un voxel. Pourtant, la diffusion radiale (RD) qui mesure les niveaux d'indice de myélinisation et la diffusion axiale (AD) qui reflète l'intégrité des microtubules le long de l'axone, ou encore la diffusion moyenne (MD) qui combine les deux précédents indices, semblent indispensables à la compréhension exhaustive de la plasticité au sein de la matière blanche (Jones et al., 2013). Au sein de notre revue systématique, trois études mentionnent que la réduction de FA au sein du gyrus temporal supérieur à la suite d'une privation auditive précoce s'explique probablement mieux par les changements de diffusion axiale et radiale (Karns et al., 2017; Li et al., 2012; Miao et al., 2013). De plus, une réduction de la FA dans les principales régions impliquées dans le

traitement auditif et langagier semble cohérente à travers les études rapportées dans cette revue systématique. Cependant, l'hétérogénéité des mesures complémentaires (AD, RD, MD) suggère l'importance de poursuivre les travaux à la lumière des grandes avancées de cette technique.

La vaste majorité des études en imagerie rapportées dans le cadre de cette thèse comporte des analyses transversales chez des adultes sourds, et très peu chez les enfants ou les adolescents. Comme nous l'avons vu dans les précédents chapitres, ces recherches apportent des informations hétérogènes sur l'impact de l'expérience sur la plasticité. Dans une récente revue de la littérature, Alencar et collaborateurs (2019) proposent une représentation visuelle du décours temporel des performances visuelles chez les enfants sourds. Ce graphique permet d'illustrer la tendance chez les enfants sourds à obtenir de plus faibles performances lors de tâches de perception visuelles en comparaison à des enfants neurotypiques jusqu'à l'âge de 10 ans environ, puis après cet âge, les performances tendent à se normaliser pour finalement être meilleures que celles des adultes neurotypiques (Codina et al., 2011; Megreya & Bindemann, 2017; Parasnis et al., 1996). Il est à noter ici que les résultats comportementaux sont obtenus chez des profils très différents d'enfants sourds, à savoir tout particulièrement en termes de mode de communication (oralistes et signeurs). Parallèlement, les données référencées au sein de notre revue systématique permettent d'identifier des altérations structurelles de la matière blanche au sein du gyrus temporal supérieur, du gyrus de Heschl, du cervelet, du cortex visuel primaire ainsi que du gyrus temporal médian avant l'âge de deux ans (Smith et al., 2011). Ces altérations semblent évoluer progressivement jusqu'à l'âge de cinq ans (Huang et al., 2015; Zheng et al., 2017). Ces résultats sont obtenus auprès d'enfants sourds profonds qui sont candidats à l'implant cochléaire et pour lesquels l'oralisation constitue leur moyen de

communication préférentiel. Par la suite, nous observons un pic des altérations structurelles qui portent cette fois-ci sur la matière blanche et la matière grise, mais qui ne sont obtenues qu'auprès d'adolescents sourds signeurs (Li et al., 2012; Miao et al., 2013) et auprès d'adultes sourds dont le profil est hautement hétérogène en termes de mode de communication (natifs, signeurs, oralistes) (voir Table 1, Chapitre IV). La figure 1 permet ainsi d'illustrer le décours temporel de la réorganisation cérébrale en termes de plasticité fonctionnelle et structurelle.

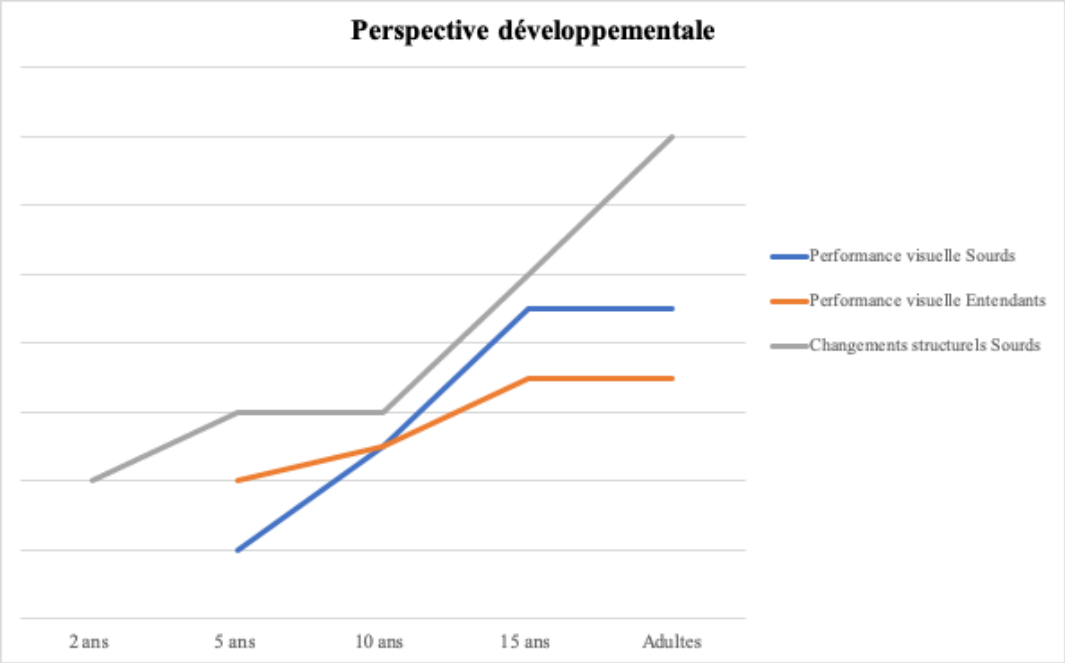

Figure 1. Graphique représentant les changements structurels et fonctionnels chez les enfants et adolescents sourds. Les données sur les performances visuelles des enfants sourds et entendants sont extraites de Alencar & al., 2019.

Parmi les hypothèses qui pourraient expliquer l'inconsistance des changements structurels dépendants de l'expérience, ainsi que les données développementales comportementales observées dans les diverses études auprès des individus sourds, le modèle d'expansion-normalisation s'avère très intéressant (Wenger, Brozzoli, Lindenberger, & Lövdén, 2017). Ce modèle décrit que durant l'acquisition d'une compétence, les structures

cérébrales du cerveau vont suivre une séquence dite d'expansion, de sélection et de normalisation (Calmels, 2019). Dans les premières étapes de l'apprentissage, le volume de la matière blanche et de la matière grise de certaines régions augmente en réponse à la spécificité de la tâche. Puis, avec le temps et la spécialisation, nous observons le phénomène classique de sélection et d'élagage de la matière superflue. Finalement, lorsque cette sélection est achevée, le volume de la matière cérébrale revient progressivement à sa taille initiale alors que les compétences comportementales se sont améliorées et peuvent continuer à se perfectionner. De toute évidence, ce modèle échoue à ce jour à s'appliquer à la population des individus sourds, mais vient expliquer les disparités dans les altérations structurelles retrouvées, notamment en termes de temps et d'interprétations offertes. En effet, selon ce modèle une altération à un moment donné pourrait faire partie d'un processus naturel de sélection de la matière cérébrale, ne pouvant plus être interprétée comme étant induit par la privation auditive. Il permet de soulever d'importantes questions : est-ce que les individus sourds présentent la même séquence alors que les altérations en lien avec la perte auditive sont présentes dès le plus jeune âge ? En raison de quelques améliorations comportementales dans les modalités sensorielles préservées, nous pouvons nous demander si les individus sourds ne présenteraient pas une version alternative de cette séquence, comprenant simultanément un processus de développement cérébral typique et alternativement, des altérations structurelles irrémédiables dans les régions auditives et langagières qui ont tendance à se montrer encore plus importantes une fois adulte ? Du point de vue de la restauration, ce modèle mériterait de questionner l'impact de l'implantation cochléaire sur ce processus, ou encore de la stimulation du nerf auditif par le port d'une prothèse auditive. Singh et collaborateurs (2018) offre une première réponse à ces questions. Selon ces auteurs, la présence concomitante des deux processus de

réorganisation cérébrale fonctionnelle ayant été décrits précédemment n'a pas pour unique rôle la compensation à la fonction perdue. En effet, la réorganisation fonctionnelle jouerait un rôle protecteur sur l'étendue de la réorganisation cérébrale afin d'éviter que les changements structurels observés impactent de façon inadaptée et irrémédiable le comportement de ces individus. Toutefois, notre graphique démontre que ces changements comportementaux ne semblent pas suffire à protéger le cerveau des adultes sourds, puisque les changements structurels augmentent à l'âge adulte, ne se restreignant plus aux régions dédiées à l'audition et au langage, mais pouvant altérer l'intégrité structurelle des régions telles que le corps calleux, le thalamus et le cervelet. À la lumière de ces questions, l'établissement de protocoles expérimentaux longitudinaux apparaît nécessaire. Ils permettraient de mieux comprendre le décours temporel et spatial de la réorganisation cérébrale, de déterminer quels aspects des expériences sensorielle et linguistique influencent le développement des autres modalités sensorielles (Dye & Bavelier, 2013) et pourraient offrir des informations cruciales à l'optimisation de la restauration auditive.

5.3.2 Limites en lien avec les spécificités de la population sourde

Le constat est permanent et se répète au fil des études dans le domaine de la déficience auditive. L'étude de la réorganisation cérébrale chez les individus sourds se complique par toute une série de contraintes inhérentes à cette population, qui sont désormais bien référencées, mais peu ou prou contrôlées. Parmi ces contraintes, citons l'importante variabilité en termes d'expérience auditive (degré de la perte auditive, audition résiduelle), de restauration auditive (prothèses auditives, implant cochléaire), du développement du langage

(exposition précoce à un mode de communication orale ou signée), ainsi que les comorbidités (syndromes génétiques, troubles neurodéveloppementaux).

Ainsi, mentionnons le fait que seul 5 % des individus présentant une surdité sévère à profonde congénitale sont exposés à la langue des signes précocement par le biais de leur famille sourde (Bavelier et al., 2006). En dépit de cette sous-représentation, les individus sourds natifs de la langue des signes restent la population ciblée prioritairement par les études en privation auditive (par exemple, 10 études sur un total de 27 dans le cadre de notre revue systématique de la littérature). Cette décision est motivée par le fait que les chercheurs s'attendent à observer plus de différences comportementales et cérébrales chez ces individus en raison de la concomitance de la privation auditive et de l'exposition précoce à la langue des signes. Pourtant, en les comparant à des individus neurotypiques, il est alors complexe de départager la part de ces deux facteurs sur les changements observés. Quelques études font le choix d'intégrer un second groupe contrôle d'individus neurotypiques natifs de la langue des signes, c'est alors sans compter que le développement du langage chez ces individus entendants implique l'acquisition simultanée de la langue des signes et de la langue orale, ce qui entraîne un développement cortical spécifique en raison de la double modalité langagière (Cardin et al., 2013).

D'autres chercheurs ont souhaité contrôler l'impact de l'utilisation de la langue des signes en incluant des individus sourds signeurs et oralistes (par ex. Cardin, Orfanidou, et al., 2016; Cattani, Clibbens, & Perfect, 2007). Ce choix nous apparaît particulièrement pertinent dans la mesure où comme il l'a été mentionné dans le préambule de cette thèse, de nombreux individus sourds naissent dans des familles entendantes, présentent des surdités sévères à

profondes, qui surviennent avant l'acquisition du langage, et pour différentes raisons, n'ont pas pu recevoir un implant cochléaire. Cet ajout d'un groupe expérimental constitué d'individus sourds oralistes peut paraître infondé dans la mesure où ils sont considérés comme plus proches des individus neurotypiques, ayant reçu des afférences auditives par le biais de prothèses auditives. Pourtant, ces individus sourds bien qu'oralistes continuent à souffrir de la déficience auditive sur bien des aspects. Ainsi, la compréhension de la parole dans le bruit et notamment la capacité à suivre une conversation sont les situations auditives qui présentent le plus bas niveau de satisfaction chez les individus sourds oralistes porteurs de prothèses auditives (Lopez-Poveda et al., 2017). Rappelons également que 51 % des utilisateurs sourds porteurs de prothèses auditives considèrent observer peu ou occasionnellement une amélioration de leur qualité de vie, impliquant les habiletés de communication au quotidien (Abrams & Kihm, 2015). Dans ce contexte, la déficience auditive représente un handicap persistant en dépit de l'amplification sonore ne leur permettant pas un développement typique du langage et un fonctionnement optimal au quotidien. En effet, une déviation de moins un écart-type en comparaison à la norme est ainsi retrouvée chez des enfants sourds porteurs de prothèses auditives précocement lors de tests évaluant les capacités de langage expressif et réceptif (Cupples et al., 2018). À l'instar de l'implant cochléaire, les prothèses auditives offrent insuffisamment d'informations spectrales pour une représentation phonétique de qualité suffisante à un développement typique du langage oral et du langage écrit, requérant alors l'apprentissage d'un soutien visuel d'aide au décodage phonologique tel que la lecture labiale et/ou le langage parlé complété (Bayard et al., 2019; Simon, Fromont, Le Normand, & Leybaert, 2019). Pour de plus amples informations, nous vous invitons à lire la première annexe de cette présente thèse qui porte sur le développement du langage oral et du langage

écrit chez les enfants sourds signeurs versus oralistes. De notre opinion, la nécessité d'utiliser des indices visuels en suppléance du décodage au langage oral chez les sourds porteurs de prothèses auditives peut s'apparenter, ou tout du moins se mettre à un niveau comparable, de l'impact de l'utilisation de la langue des signes sur la réorganisation cérébrale de l'individu sourd.

En raison des diverses limitations citées ci-haut, la meilleure pratique pour l'étude de la réorganisation cérébrale serait de tester quatre groupes incluant : des individus sourds oralistes et natifs de la langue des signes en comparaison d'individus neurotypiques oralistes et natifs de la langue des signes. De plus, selon Dye & Bavelier (2013), l'âge au moment du testing, une mesure de l'intelligence, l'étiologie, le degré de la perte auditive, l'âge de début de la surdité, l'âge d'exposition au langage, la maîtrise du langage (oral ou signé) sont des variables à rapporter de façon systématique dans la recherche en surdité. Nous ajoutons à cela le degré d'audition résiduelle (Shiell et al., 2014b), le degré de restauration auditive avec les prothèses auditives, l'utilisation du langage parlé complété et de la lecture labiale.

5.3.3 Limites spécifiques de la présente thèse et nouvelles avenues

L'une des limites de notre étude 1 provient du construit expérimental de la tâche destinée à l'imagerie par résonance fonctionnelle, qui subit alors un certain nombre de contraintes inhérentes à la technique et aux coûts qu'elle engendre. De façon optimale, la tâche aurait dû être préalablement testée en condition comportementale uniquement afin de déterminer si le nombre de stimuli, soit 126 stimuli (42 emblèmes, 42 pantomimes et 42 stimuli contrôle), représentait un nombre suffisant pour identifier des différences plus robustes dans le comportement des individus sourds, en termes de temps de réponse, mais également au niveau

de l'exactitude. À titre d'exemple, l'une des plus récentes études chez les individus sourds ayant démontré des différences comportementales en termes de détection du mouvement lors de stimuli visuels de bas niveau, comprenait une moyenne de 136,7 essais par stimuli (Shiell et al., 2014a). D'autre part, il aurait été pertinent pour mieux distinguer la part des processus linguistiques impliqués dans le traitement des emblèmes, d'intégrer des stimuli en langue des signes. Par cet ajout, nous aurions été en mesure d'identifier le potentiel recouvrement des régions cérébrales impliquées entre le traitement des emblèmes qui visent à transmettre de l'information et ceux impliqués dans la langue des signes. Finalement, un plus grand nombre de participants sourds aurait permis la réalisation d'analyses statistiques portant sur l'impact du mode de communication sur la réorganisation cérébrale des régions auditives dans le cadre de la reconnaissance du mouvement biologique. Les précédentes contraintes liées à la population sourde et détaillées ci-haut orientent également vers la pertinence de tester quatre groupes : les individus sourds natifs de la langue des signes, les individus sourds oralistes, les individus neurotypiques oralistes et un dernier groupe d'individus neurotypiques natifs de la langue des signes.

Concernant notre seconde étude, il est bien courant dans la littérature d'observer simultanément la réalisation d'une revue systématique et d'une méta-analyse, il s'agissait bien évidemment de notre première intention. Dans le cas spécifique des études en imagerie par résonnance magnétique, des logiciels sont désormais disponibles pour permettre ce type d'analyses statistiques sur des données d'imagerie structurelle (Vanasse et al., 2018). Toutefois, cet outil n'inclut que les analyses obtenues par le biais de la morphométrie au niveau du voxel, réduisant la possibilité de généraliser dans notre cas, les multiples altérations

rapportées à ce jour chez les individus sourds. Cette absence d'analyses statistiques restreint ainsi la portée des données obtenues et confine l'interprétation actuelle. Avec des analyses statistiques, mais également des analyses de régression, il serait alors possible de faire ressortir les régions cérébrales principalement altérées chez les individus sourds, ou encore de réaliser des cartographies cérébrales selon le décours temporel des changements structuraux dans le cerveau des personnes souffrant d'une déficience auditive précoce, de distinguer spécifiquement l'impact du mode de communication sur ces altérations cérébrales ou encore de faire ressortir statistiquement les régions cérébrales les plus à risque de changements en vue d'une implantation cochléaire.

5.4 Conclusion générale

Le présent projet de thèse visait à offrir une contribution unique à la compréhension de la réorganisation cérébrale chez les individus privés de l'audition. Par conséquent, nous sommes les premiers à démontrer que les individus sourds présentent une sensibilité accrue pour la reconnaissance du mouvement biologique. Cette différence comportementale corrélée à une activation cérébrale au sein des régions auditives primaire et secondaire supporte la présence de mécanisme de réorganisation cérébrale de type compensatoire à la perte auditive. La surdité entraîne ainsi des habiletés visuelles supérieures à celles des individus neurotypiques dans le but de compenser quotidiennement leur handicap auditif. De plus, la réalisation d'une revue systématique de la littérature portant sur les altérations structurelles observées chez ces individus sourds, contribuent à une meilleure appréciation des changements de réorganisation structurelle qui caractérisent la déficience auditive précoce, ainsi que leurs impacts sur le développement langagier et neurocognitif. Mis ensemble, ces résultats nous permettent d'offrir

une compréhension exhaustive des changements ayant cours dans le cerveau des individus sourds. Par le biais d'une perspective développementale, nous établissons les liens étroits entre ces deux types de réorganisation cérébrale, tout en offrant de nouvelles pistes pour la recherche dans le domaine de la surdité qui serviront ultimement au développement d'interventions plus optimales à la réalité de ces individus.

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Annexe 1

Cross-modal plasticity and central deficiencies: the case of deafness and the use of cochlear implants

Cross-modal plasticity and central deficiencies: the case of deafness and the use of cochlear implants

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ABSTRACT

The primary objective of this chapter is to describe the consequences of central deficiencies on the neurodevelopment of children. We are approaching this topic from the standpoint of congenital deafness – deaf from birth. Thus, we first present the current state of knowledge on cortical reorganization following congenital deafness. The allocation of auditory cortices to other sensory systems can enhance sensory processing and therefore the cognitive functions related to them. Second, we explore the linguistic development of deaf children. The English written system is speech-based. Consequently, its acquisition for deaf children is complex and atypical, usually leading to poorer achievements. Next, we explore the impact of neural prostheses such as cochlear implants on the neurocognitive and linguistic development of deaf children. In some cases, it allows the individuals to, at least partially, regain access to the lost sense. We also comment on the specific needs of the deaf population when it comes to neuropsychological assessment. Finally, we touch on the specific context of deaf children born of deaf parents, and therefore naturally exposed to sign language as their only means of communication.

Introduction

The brain has the remarkable ability to reorganize itself throughout life in response to experience. Thus, sensory deprivation is an excellent avenue to explore the phenomenon of cortical reorganization. The consequences of central deficiencies (e.g., deafness or blindness) on the brain have been the focus of numerous studies in the last decade. Importantly, understanding the cortical changes induced by sensory deprivation is a major challenge in the improvement of rehabilitation strategies.

In individuals deprived of a sense, the literature mostly concerns early or congenital sensory deprivation (e.g., born or from early years). The study of these profiles is preferred since, in the first years of life, the brain shows extraordinary development leading to changes in behaviors and cognitive processing. After those few years, its growth is rather characterized by "fine-tuning" and remodeling of the primary circuits and networks (Gilmore, Knickmeyer, & Gao, 2018). In the context of sensory deprivation, it is even more crucial since the brain shows critical periods during which it is more sensitive to environmental sensory input (Purves, 2012). Looking at the auditory system, this critical period is until approximately the 3rd year of life (Kral, 2013). Hence, the sensory deprivation that occurs before this sensitive period leads to far more important cortical changes.

Sensory-deprived individuals must then depend on the remaining senses to perform daily tasks. Consequently, cortical reorganization induced by early sensory deprivation leads to compensatory mechanisms, leading in turn to enhanced behavioral performances in preserved sensory modalities. These behavioral enhancements are ensured by a phenomenon of cross-modal cortical activation i.e. the activation of regions initially developed for the impaired

sense by sensory information intended for other sensory systems (Merabet & Pascual-Leone, 2009). For an extensive review of brain reorganization in the specific case of blindness, see Voss, 2018.

In congenital deaf individuals, such compensatory mechanisms are observed, although not as straightforward as in blind individuals. Still, deafness turns out to be a more exhaustive theoretical model than blindness in regard to the exploration of brain reorganization and rehabilitation in children and adults. Numerous studies have identified enhanced performances in specific visual tasks such as peripheral localization (for a review, see Pavani & Bottari, 2011), motion detection (Shiell, Champoux, & Zatorre, 2014), as well as facial discrimination and facial memory (Arnold & Murray, 1998; McCullough & Emmorey, 1997) in congenitally deaf adults in comparison to hearing individuals. A few studies also revealed similar performances in deaf individuals to hearing individuals in visual tasks such as color processing (Armstrong, Neville, Hillyard, & Mitchell, 2002) and divided attention (Dye & Hauser, 2014). More evidence suggests that, in congenitally or prelingually deaf adults, tactile (Auer & Bernstein, 2007; Karns et al. 2012; Levänen et al. 1998; Levänen & Hamdorf, 2001), biological motion (Simon et al. *unpublished work*) and language processes (i.e. sign language and/or lip-reading) (Fine, Finney, Boynton, & Dobkins, 2005; Lambertz, Gizewski, de Greiff, & Forsting, 2005; Petitto et al., 2000; Sadato et al., 2005) are supported by the cross-modal recruitment of the auditory cortex. Studies with deaf children show that visual stimulation activates the auditory cortex and prove that the cross-modal phenomenon is already present in childhood (e.g., Campbell & Sharma, 2016; Liang et al., 2017). To date, only one study demonstrated a correlation between structural reorganization concerning the cortical thickness in the auditory cortex with enhanced visual motion detection abilities in early and profoundly

deaf individuals (Shiell & Zatorre, 2016). No study established a link between brain reorganization and behavioral enhancement in deaf children or adolescents.

Regarding auditory rehabilitation, major advances were achieved through the cochlear implant (CI). The CI is a device that transforms the acoustic sounds in the environment in signals that can be transmitted to the auditory nerve via a set of electrodes that stimulate different areas of the cochlea. By stimulating the auditory nerve (e.g., Yawn et al. 2015), the CI is the most efficient way to restore audition in children or adults with severe or profound deafness (Wilson & Dorman, 2008). Considering the critical period for auditory development, there is a strong body a research that supports early implantation. For illustration, in 2012, approximately 38,000 children were fitted with a CI in the USA (NIDCD, 2017). However, in Canada, only 50% of the children fitted with a CI between 2000 and 2005 were younger than three years old (Fitzpatrick, Ham, & Whittingham, 2015). In Europe, trends indicate younger ages at implantation considering that 70% of children in need of a CI were implanted before the age of two (Bruijnzeel et al., 2017). This leads to considerable variability in CI profiles in children and consequently to variability in its effect on cortical plasticity and development.

With respect to cortical reorganization in line with auditory restoration, the prevailing view is that the reallocation of auditory regions to non-auditory stimuli is deleterious to rehabilitation and diminishes the auditory outcomes obtained post-CI (e.g., Champoux et al. 2009; Doucet et al. 2006; Giraud & Lee, 2007; Lee et al., 2007; Strelnikov et al., 2009). Anatomical connectivity studies show that prior to implantation, the structural integrity of the auditory cortex and neighbouring regions is strongly correlated to the speech perception outcomes of deaf children after they have received their CI (e.g., Huang et al., 2015; Wu et al.,

2016; Zheng et al. 2017). These studies have had a considerable influence on rehabilitation strategies for deaf children with CIs. Thus, rehabilitation methods based on the auditory modality are prioritized to those based on vision (e.g., lip-reading or sign language) (Hall, 2017). However, researchers have recently shown contradictory results in both human (Anderson, Wiggins, Kitterick, & Hartley, 2017) and animal data (Land et al., 2016). According to these last studies, enhanced cross-modal activity in auditory regions by visual stimulation (e.g., lip-reading for the case of human study) could ameliorate multisensory integration and shouldn't reduce CI efficiency. In turn, enhanced access to auditory experience is primordial for optimal neurocognitive development (Kronenberger et al. 2014).

Taken together, the available knowledge attests of the importance of pursuing research efforts on the compensatory mechanisms linked to cerebral plasticity in deaf individuals, and primarily in deaf children. Future data will allow us to adjust our rehabilitation strategies to the challenges brought by congenital deafness. For example, literacy and learning outcomes of deaf children are well documented but remain a significant challenge for the deaf community and the professionals accompanying them. The following sections will focus on the neurocognitive and linguistic profiles and barriers of deaf children with or without CI. In the last section of this chapter, we shall suggest good practices for the neuropsychological assessment of deaf children. Finally, we shall conclude with future research avenues.

Neurocognitive development of deaf children

Worldwide, 34 million children have a disabling hearing loss (World Health Organization, 2018) and the estimated incidence of congenital deafness is one to three newborns per thousand births (Nelson, Bougatsos, & Nygren, 2008). Despite technological and scientific

advances, such as newborn hearing screening (Pimperton et al., 2016), CI or digital hearing aids, the present literature regularly reports delays in language acquisition and neurocognitive development. However, a large variability in linguistic and cognitive profiles exists and is still left to be explained. According to Huber and Kipman (2012), auditory rehabilitation success depends on hearing variables such as deafness duration, age at cochlear implantation, but also social and education characteristics and the presence of comorbidities. Nonetheless, researchers stated that only 25% of the inter-individual variability observed in deaf individuals is explained by the hearing variables themselves (Moberly, Bates, Harris, & Pisoni, 2016).

1. Language development in deaf children

Language comes in different shapes and forms, but all share common importance. Whether it is read, written, spoken or signed, language is essential for cognition, educational and social development. As deafness leads to an absence of, or to incomplete auditory experience, deaf children will exhibit atypical pathways to language acquisition. Importantly, 96% of all deaf newborns have hearing parents (Mitchell, Young, Bachleda, & Karchmer, 2006). Thus, CI implantation followed by auditory rehabilitation remains the preferred avenue for the acquisition of spoken language. In this context, those who did not receive a CI show typically delays in language acquisition (Marschark & Spencer, 2010).

Regarding literacy, deaf children without CI tend to have poorer reading skills than hearing children (e.g., Kyle & Harris, 2010; Perfetti & Sandak, 2000; Qi & Mitchell, 2011; Trezek et al. 2010). Indeed, most deaf 18-year-old students' reading skills are equivalent to the fourth grade (Cheri Williams & Mayer, 2015). Research suggests that the reading performances of deaf children without a CI are positively influenced by the use of spoken

language (Kyle & Harris, 2010). Other factors such as lip-reading abilities, cognitive functions (i.e. working memory) and linguistic skills (i.e. vocabulary and phonological awareness) also participate in reading skills and could influence the reading efficiency of deaf children without a CI (Pisoni & Cleary 2003; Lyxell et al. 2008). Finally, it appears that while single word reading performances of deaf children without CI increase with time and education, reading comprehension ameliorates at a much slower rate (Harris, Terlektsi, & Kyle, 2017). As understanding what is read is the primary goal of reading, this can lead to various impairments throughout academic and professional achievement.

When it comes to spelling, deaf children without CI again show delays when compared to hearing children (Mayer, 2012; Williams & Mayer, 2015). Studies indicate that text compositions of deaf children are shorter, less fluent and comprise significantly more lexical and grammar errors than that of their hearing counterparts (Alamargot et al. 2007; Arfé et al. 2014). While factors such as working memory (e.g., Arfé et al. 2014) might play their part in spelling accuracy in deaf children, the inherent issue is the disparity between their linguistic exposure (spoken or sign language) and the demands of written speech-based systems (Mayer & Trezek, 2017).

2. The specific case of deaf children with a CI

2.1 Spoken language development in children with a CI

Taking into account the latter conclusion, one can expect better achievement when looking at deaf children with a CI. As typical reading and spelling acquisitions rely on speech-based learning, i.e. phonological awareness (Lundberg, 2002), that of CI users will depend on CI

efficiency i.e. speech perception outcomes (Simon, Fromont, Le Normand, & Leybaert, 2019). However, speech perception outcomes in deaf children with a CI are very heterogenous even when they are implanted early (e.g., Kang et al., 2004). Thus, deaf children with a CI remain at risk for poorer literacy outcomes compared to hearing children (Harris & Terlektsi, 2011; Harris et al. 2017; Moeller et al. 2007). However, while deaf children with a CI might still experience delays compared to their hearing peers, they obtain better scores in linguistic tasks than deaf children without a CI.

Post lingual deaf adults with a CI typically show impressive speech perception improvement during the first-year post-implantation (Ruffin et al., 2007), but it is not as straightforward for early deaf children. Literature suggests that implantation before the age of two leads to better speech perception outcomes (Svirsky et al. 2000; Svirsky et al. 2004; Peterson et al., 2010). However, many other factors, in addition to hearing variables mentioned above, need to be taken into account when attempting to predict CI outcomes, such as social and educational factors and communication mode (for a review, see Peterson et al. 2010). Nonetheless, the CI offers the possibility for deaf children to acquire speech perception abilities that are unreachable without it.

Another factor identified as a significant predictor for CI outcome is the development of expressive skills (Davidson, Geers, Blamey, Tobey, & Brenner, 2011). A longitudinal study examined the spoken language development of almost 200 deaf children with a CI (Niparko et al., 2010). Deaf children implanted before 18 months showed a developmental curve practically parallel to that of hearing children whereas those implanted after 18 months still showed improvement but with a lot more variability. Despite the age at implantation, deaf

children with a CI present persistent delay in expressive skill after three years following implantation (Niparko et al., 2010). With respect to intelligibility, Habib et al. (2010) showed that it improved with years of CI experience. They also observed that, when tested in the same period, children implanted before the second year of life were more intelligible than those implanted after. However, due to the poor auditory signal available, children fitted with a CI continue to show lower intelligibility than hearing peers (e.g., Poursoroush et al., 2015).

An essential skill for literacy development is phonological awareness. It refers to one's understanding that speech is made up of different units, leading to the ability to manipulate such units. In deaf children with a CI below the age of five, speech production and perception outcome measures correlate significantly with phonological awareness abilities (Ambrose, Fey, & Eisenberg, 2012). In other words, being already delayed in expressive capacities accentuates the risk of being delayed in phonological awareness. Again, reports show that children implanted early have better phonological awareness than those implanted later (James, Rajput, Brinton, & Goswami, 2007; Johnson & Goswami, 2010). Phonological awareness, as wells as speech perception and production, are important predictors of written language abilities (Geers & Hayes, 2011).

2.2 Written language development in deaf children with a CI

As mentioned above, the reading abilities of deaf children have received a lot of attention as it is an essential skill for academic progress. It is generally accepted that deaf children with a CI are better readers than those without (Arfe, Dockrell, & Berninger, 2014; Hayes, Kessler, & Treiman, 2011; Marc Marschark, Rhoten, & Fabich, 2007; Mayer & Trezek, 2017). Consequently, over half of the children with a CI perform as well as their hearing peers on

reading comprehension tasks (Geers, 2004). Some studies demonstrate that the age of implantation influences positively reading comprehension (Connor & Zwolan, 2004; Mayer, Watson, Archbold, Ng, & Mulla, 2016). Conversely, other studies suggest that better reading skills in deaf children with a CI are not associated with early implantation (Geers, 2003, 2004; Harris & Terlektsi, 2011). Geers (2003, 2004) instead suggests that it is related to exposure to spoken language prior and/or after receiving the CI. In a longitudinal follow-up, deaf teenagers with a CI showed delays representing approximately two years in reading comprehension compared to their hearing peers (Geers et al. 2008). While the CI offers an opportunity for better literacy development, deaf children with a CI require adapted interventions throughout their schooling especially during high school education.

The abilities to write and read are interrelated. Thus, the delay observed in reading achievement affects spelling skills in deaf children with a CI. However, spelling abilities in deaf children with a CI have not received as much attention as reading skills (see Mayer & Trezek, 2017 for a review). Some studies show that spelling skills in deaf children with a CI are below the expected level for their age on single word spelling or written expression tasks (Apel & Masterson, 2015; Mayer et al. 2016; Geers & Hayes, 2011). More precisely, it seems that deaf children with a CI produce the same proportion of correct single-word spelling, but in counterparts, they commit significantly less plausible phonological errors that hearing peers (Hayes et al., 2011; Simon et al., 2019). Deaf children with a CI write more accurately using a phonological strategy which is unavailable to deaf children without a CI (Mayer et al., 2016; Straley, Werfel, & Hendricks, 2016).

2.3 Academic achievement of deaf children with a CI

The variability in both reading and writing abilities calls for prudence when it comes to literacy expectations after implantation. A review summed up that while over two-thirds of parents and one-third of teachers reported that deaf children with a CI could easily take part in regular class activities, over 70% of the children fell below the class median (Punch and Hyde, 2011). A challenge raised by parents and teachers is the lack of specialized support for deaf children and adolescents with a CI. Especially when spoken language is functional, the deafness and its various impacts are often falsely seen as fixed (Punch & Hyde, 2011). While language skills are correlated to academic achievement, long-term teenage users of CI appeared to be better protected from schooling failure when parents were better educated and the CI was fitted at an early age (Diaz, Labrell, Le Normand, Guinchat, & Dellatolas, 2019). All in all, very few students with a CI seem to follow the same pattern of literacy development. This suggests the need for long-term interventions to support the children throughout their schooling.

2.4 Neuropsychological development of deaf children with a CI

For many decades, the relation between auditory privation and language acquisition of deaf children has been at the center of scientific concern. Since the brain is an integrated functional system, the neurocognitive development depends on the integrity of the sensory systems. Consequently, auditory privation has an impact on the cerebral and cognitive development of deaf individuals (Conway et al. 2009; Kronenberger et al. 2014). Pertinent studies on cognitive functions of deaf children with a CI are resumed in Table 1. Among neurocognitive measures, the Intelligence Quotient (IQ) composite score is the most used marker to assess the

intellectual ability of children and is a good predictor of academic achievements (Lezak, Howieson, Loring, & Fischer, 2004). In versions of the Weschler Intelligence Scale for Children (Wechsler, 1949), indexes of verbal IQ (e.g., verbal reasoning, vocabulary and general knowledge) and nonverbal IQ (fluid reasoning, processing speed, visuospatial and visuoconstructive abilities) are obtained via multiple sub-tasks.

Regarding verbal comprehension, while most deaf children with a CI perform within the normal range, all studies that assessed this skill show an inferior performance of deaf children with a CI compared to their hearing peers (Hashemi & Monshizadeh, 2012; Huber & Kipman, 2012; Park et al., 2015; Wu et al. 2008). However, a poor performance cannot always be simply explained by poor intellectual abilities but rather by sensory or verbal limitations and must be interpreted with caution (for specific instructions, see Wechsler, 2014).

Considering the nonverbal reasoning index (typically measured by the Leiter, Raven's Standard Progressive Matrices and the nonverbal IQ index of the WISC), the majority of studies demonstrate that deaf children with a CI perform similarly to their hearing peers (Cejas, Mitchell, Hoffman, & Quittner, 2018; Edwards, Khan, Broxholme, & Langdon, 2006; Geers et al., 2008; Park et al., 2015; Sarant, Hughes, & Blamey, 2010; Schlumberger, Narbona, Manrique, & Navarra, 2004; Shin, Kim, Kim, Park, & Kim, 2007). When looking at CI outcome, inconsistent results are reported regarding the prediction of expressive and receptive language abilities by nonverbal IQ (Cejas et al., 2018; Ann Geers et al., 2008; Nicholas & Geers, 2004; Sarant et al., 2010). Indeed, the causal relationship between nonverbal IQ and language is complex and appears like a dynamic developmental pattern with reciprocity (Botting, 2004). Thus, a recent longitudinal study observed that early IQ measure

(i.e., before three years old) is not predictive of cognitive functioning in deaf children with and without a CI. These outcomes appear to be highly influenced by the language background and degree of stimulation at home (Cejas et al., 2018). Therefore, the authors propose that before the school-age range, measures of nonverbal IQ are only useful to target deaf children needing of specific interventions.

Moreover, many other neurocognitive functions have been studied in deaf children with a CI. One of these is executive functions (EF) since typical language development underlies their emergence (Kuhn, Willoughby, Wilbourn, Vernon-Feagans, & Blair, 2014). EF are commonly defined as high-level functions such as mental flexibility, inhibition, updating, categorization, working memory and planning. For optimal functioning, good working memory capacities, comprehension and reasoning abilities are also required.

A behavioral measure of EF in daily life is a questionnaire filled out by parents (Behaviour Rating Inventory of Executive Function, BRIEF) (Gioia, Guy, Isquith, & Kenworthy, 1996). In deaf children with a CI, the results to this questionnaire show difficulty or delays in inhibition and working memory and a significant correlation between these processes and language outcomes post-CI (Beer et al. 2011; Kronenberger et al. 2014). With a formal neuropsychological evaluation, many EF were delayed in deaf children with a CI in comparison to hearing peers (Botting, 2004; Botting et al., 2017; Figueras, Edwards, & Langdon, 2008; Surowiecki et al., 2002) even when the tasks were controlled for the verbal content of instructions (Botting et al., 2017). In accordance with the BRIEF questionnaire, a positive correlation was found between EF and language abilities (e.g., vocabulary and receptive grammar) (Botting et al., 2017; Figueras et al., 2008). Working memory in deaf

children with a CI has been the focus of numerous and specific studies (for a review, see Pisoni et al. 2017) as working memory is a crucial prerequisite for the decoding of reading. The main hypothesis is that the auditory deprivation is the principal cause of EF delay in deaf children with a CI. However, two recent studies support that the language deprivation has more deleterious effect on EF than the auditory deprivation (Hall et al. 2017; Marshall et al. 2015). EF in sign language native deaf children were in the typical range in comparison to hearing children.

Since estimation indicates that 5.4% of deaf children with a CI are diagnosed with comorbid attention deficit and hyperactivity disorder (ADHD) (Cejas, Hoffman, & Quittner, 2015), some authors have focused on their attentional capacities. Classically evaluated with the Continuous Performance Task (CPT, Conners & Sitarenios, 2011) the results for sustained attention are heterogeneous. For example, the same attentional efficiency is found in deaf children with a CI compared to hearing and deaf children with hearing aids, closely matched on non-verbal IQ and age (Tharpe, Ashmead, Sladen, Ryan, & Rothpletz, 2008). Another study found lower sustained attention abilities in deaf children with a CI when compared to the normal range of the task (Horn, Davisa, Pisoni, & Miyamoto, 2004). Finally, the use of a CI accentuates auditory interference by the surrounding environment as deaf children with a CI underperformed six months post-implantation (Shin et al., 2007). It is well established that processing speed is negatively affected in children with ADHD (e.g., Cook, Braaten and Surman, 2018). With a coding task, deaf children with a CI show similar processing speed as hearing children (Maria Huber & Kipman, 2012). On the contrary, Kronenberger and collaborators (2014) found that school-aged deaf children with a CI are slower than their hearing peers. Additionally, it was demonstrated that verbal working memory is predicted by

verbal processing speed in deaf children with a CI. The authors posit that such measures should be taken into account when assessing the benefits of CIs as they may help understand inter-individual variability (AuBuchon, Pisoni, & Kronenberger, 2015).

Importantly, some neurocognitive components are underestimated in the field of rehabilitation with deaf children with a CI. Motor development is one of those components since gross motor skills and vestibular function are critical factors of the academic achievement and learning (e.g., Westendorp et al. 2011). Hearing children with vestibular dysfunction show delay in gross motor milestones (Verbecque et al., 2017). Considering the anatomical proximity between the cochlea and the vestibular system, deaf children are more at risk of a natural vestibular deficit (about 20 to 85%) (Maes, De Kegel, Van Waelvelde, & Dhooge, 2014). Moreover, the CI surgery can also lead to direct trauma to the vestibular structures (Verbecque et al., 2017). Studies have assessed motor skills in deaf children with a CI. Some of these children learned to walk later than their hearing peers and showed difficulty in balance (e.g., Horn et al. 2006; Schlumberger et al. 2004). The integrity of vestibular function is often not controlled in most studies concerning deaf children. Consequently, the link between motor and language profiles is often missing when deaf children with poor CI outcomes are discussed.

Discussion

The CI is well accepted and even encouraged for those with hearing loss (Fitzpatrick et al., 2015). However, its efficiency, in terms of speech perception and expression, still greatly varies from one individual to the next. This chapter shows that other neurocognitive abilities such as reading, writing, and cognition are heterogeneous in deaf children with a CI. Many

factors are important when trying to explain this variability. Some are linked to auditory experience: the age at deafness, deafness duration, residual hearing, use of hearing aids and age at implantation (e.g. Blamey et al. 2013; Lazard et al. 2012; Holden et al. 2013). Other factors significantly influence the development of deaf children but are frequently omitted in results interpretation. For example, comorbidity between deafness and other neuropsychological disorders is frequent such as learning disorders (7.20%) and ADHD (5.4%) (Castellanos et al. 2018; Cejas et al. 2015). Also, psychosocial factors should not be neglected when looking at CI outcome. Parental abilities have the potential to impact rehabilitation and family interventions are essential for optimal CI outcomes (Spencer, 2004; Cejas et al. 2018).

1. Essential considerations for neuropsychological assessment of deaf children

As mentioned above, many variables can impact the neurocognitive development of deaf children. These factors must systematically be considered when assessing their neuropsychological profiles. Clinicians must keep these variables in mind for a nuanced interpretation of performances.

Additionally, the tests typically used in neuropsychology were standardized with verbal instructions and with hearing individuals. Therefore, they are not optimal for deaf children because of the highly expressive and receptive language processing demands inherent to these measures (Phillips, Wiley, Barnard, & Meinzen-Derr, 2014). While clinicians wait for their validation for deaf children with respect to the intellectual quotient, many other tests can be privileged. To date, the most exhaustive neuropsychological evaluation of a deaf child can be

achieved with the Leiter scale (Phillips et al., 2014). This exclusively non-verbal battery is exempt of verbal instructions. It allows for the assessment of fluid intelligence (reasoning, abstraction, and discrimination), of attentional abilities and memory. Thus, a specific profile of the strengths and weaknesses of deaf children can be obtained.

2. The specific case of sign language native deaf children

Since the neurocognitive profiles of deaf children with and without a CI are most discussed in the literature, they are also at the heart of this chapter. However, the discussion of another profile of deaf children appears necessary to offer a complete vision of the challenges surrounding deafness and its rehabilitation. It concerns deaf children who are born in deaf families and are exposed naturally to sign language from birth, just as hearing children are to spoken language. Even if they represent a minority (4%), this profile is essential to discriminate the effects of auditory deprivation and language deprivation on cerebral and neurocognitive development.

The acquisition of sign language is commonly associated with poor outcomes post-CI regarding speech intelligibility and future academic achievements (Geers et al. 2017). Authors often come to this conclusion while the impacts of auditory and/or linguistic deprivation are confused or not discriminated well. Indeed, without proper consideration, it is practically impossible to know if the effects observed on CI outcomes are due to the lack of auditory experience or to the lack of linguistic experience. Sign language is a complete language with grammatical rules and a complex syntax (Sandler & Lillo-martin, 1999). Like any other language, learning and mastering sign language is an arduous and lengthy process. Although in reality, sign language is often introduced as a backup strategy when the deaf child has

difficulties or has failed to acquire spoken language. Presently, more and more studies are in favor of early exposure (as soon as birth) to sign language in deaf children whether or not they are waiting for a CI. Indeed, the linguistic development of sign language native deaf children seems to be similar to that of hearing children regarding lexical size (Anderson & Reilly, 2002) and grammatical development (Morgenstern, Caët, Collombel-Leroy, Limousin, & Blondel, 2010). Moreover, the early acquisition of sign language by deaf children seems to improve neurocognitive and psychosocial development in terms of reading skills (Goldin-Meadow & Mylander, 1984), working memory (Marshall et al., 2015), sustained attention (Dye & Hauser, 2014), EF (Hall et al., 2017), theory of mind (e.g., Meristo et al. 2007) and quality of life (Kushalnagar et al., 2011). It was also recently demonstrated that early sign language experience leads to higher performances for the recognition of affective prosody post-CI (Fengler, Delfau, & Röder, 2018). Thus, the early learning of sign language stands as a protective factor to the impacts of spoken language deprivation on neurocognitive, academic and psycho-affective development in deaf children with and without a CI.

Other challenges accompany the generalization of sign language usage in deaf children. Learning a new and complex language is a major barrier for hearing parents discovering their child's deafness. Additionally, parents usually suspect a hearing deficiency when there's a lack of reactions or babbling on the child's part, leading to late screening and diagnosis. At this point, the impacts of language deprivation could already be in place. Finally, most hearing professionals (teachers, speech therapist, psychologists, educators) working with deaf children are non-bilingual (with sign language). This is an essential condition to offer adapted language teaching and support.

The debate about sign language in the daily lives of deaf children remains. Longitudinal studies are necessary to discriminate between the most efficient rehabilitation strategies in the long run. Still, taken together, this information confirms the importance of systematic auditory screening at birth. This is a crucial process to allow hearing parents and professionals to make the best decision for the deaf child and ultimately to adapt to their specific needs concerning linguistic and societal environment.

In conclusion, focusing on congenitally deaf children in research was thought to allow for more comparable results and conclusions on CI outcomes. However, the present literature reports variability in speech perception and expression, on written language abilities and neurocognitive development. When CI outcome is poor, each of these domains can have a significant impact on social marginalization, educational and long-term functional achievement. Pisoni et al., (2017) state that research on sensory and neuropsychological strengths and weaknesses in poor CI outcome profiles is essential and pressing for the development of intervention protocols. Such evidence- and patient-based rehabilitation strategies are much needed to ensure that all deaf children with a CI may reach their full potential.

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Table 5.

General findings of studies investigating the cognitive functions of deaf children with a CI

First author (year)	Deaf children with a CI	Controls	Intelligence quotient		Higher-order functions		Attentional functions				Executive functions		Memory functions		
	N	N	Verbal	Non-Verbal	Abstraction	Visuo-spatial reasoning	Vigilance	Sustained attention	Selective attention	Processing speed	Planning	Inhibition	Short term memory	Working memory	Long term memory
Beer (2014)	24	21						X			O	X	O	X	
Burkholder (2004)	37	37											X		
Cleary (2001)	45	45												X	
Conway (2011)	24	31				O						O	O	O	O
De Giacomo (2013)	20	20		O											
Engle-Yeger (2011)	20	20													X
Fagan (2007)	26					O					X				
Figureas (2008)	22	22			O						O	X		X	
Geers (2008)	85			O											
Harris (2011)	110												X	X	
Hashemi (2012)	30	30	X												
Huber (2012)	40	40	X												
Kronenberger (2013)	53	53						X		X		X	X	X	
Kronenberger (2014)	24	79			X	O						X		X	
Lyxell (2009)	34	120												X	
Park (2015)	13		X												
Pisoni (2000)	43														
Pisoni (2003)	176	45												X	
Pisoni (2011)	181													X	
Pisoni (2016)	31	31												X	
Sarant (2010)	21			O											
Schlumberger (2004)	25	40		O	O	O					O				
Tharpes (2002)	9	10		O			O	O	O						
Watson (2006)	15	19												X	
Wu (2008)	60		X	O											

O : The authors report a typical performance in this cognitive domain

X : The authors report a deficit or a delay in this cognitive domain

Annexe 2

Spelling, reading abilities and speech perception in deaf children with Cochlear Implant

Spelling, reading abilities and speech perception in deaf children with a cochlear implant

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ABSTRACT

This study aims to compare word spelling outcomes for French-speaking deaf children with a cochlear implant (CI) with hearing children who matched for age, level of education and gender. A picture written naming task controlling for word frequency, word length, and phoneme-to-grapheme predictability was designed to analyze spelling productions. A generalized linear mixed model on percentage of correct spelling revealed an effect of participant's reading abilities, but no effect of hearing status. Word frequency and word length, but not phoneme-to-grapheme predictability, contributed to explaining the spelling variance. Deaf children with a CI made significantly less phonologically plausible errors and more phonologically unacceptable errors when compared to their hearing peers. Age at implantation and speech perception scores were related to deaf children's errors. A good word spelling level can be achieved by deaf children with a CI, who nonetheless use less efficiently the phoneme-to-grapheme strategy than do hearing children.

Introduction

Acquiring good reading and spelling skills is a challenge for deaf children. Children with congenital and profound deafness are at risk of under developing these abilities which are crucial to achieving a complete education and social integration in our society. Indeed, sensorineural hearing loss, if not appropriately rehabilitated, may result in linguistic and neurocognitive impairments (e.g., Kral, Kronenberger, Pisoni, & O'Donoghue, 2016). Studies demonstrated that 18-year-old deaf students have a median reading level equivalent to that of the fourth grade (Qi & Mitchell, 2011; Trezek, Wang, & Paul, 2010; Perfetti & Sandak, 2000). A similar trend was observed for spelling, where 17 to 18-year-old deaf students had a level comparable to that of 8 to 10-year-old hearing children (Mayer, 2010; Williams & Mayer, 2015).

Cochlear implants (CIs) are neuroprosthetic devices that provide direct electrical stimulation to the auditory nerve, bypassing the damaged hair cells in the cochlea. To date, CIs are the most efficient way to restore audition in severe and profound deafness (e.g., Wilson & Dorman, 2008). However, the spectral and temporal frequency information provided by CIs is not as detailed as the information delivered by the inner ear. Therefore, a large variability in speech perception outcomes characterizes pediatric CIs (e.g., Hyo et al., 2005; Lee et al., 2007). Age at implantation is a good predictor of success: deaf children who received early CI are most likely to show substantial benefits in language comprehension and production (for a review, see Kral, 2013). Nevertheless, even when age at implantation is controlled for, considerable variability remains in speech perception abilities. Due to the impoverished signal available, the acquisition of phonological structures of words remains difficult for CI users (Nittrouer, Sansom, Low, Rice, & Caldwell-Tarr, 2014). Deaf children who are less proficient with their CI rely more on lip-reading to perceive speech than hearing children, while those who are more proficient rely

more on auditory information (Bayard, Colin, & Leybaert, 2014; Huyse, Berthommier, & Leybaert, 2013; Rouger, Fraysse, Deguine, & Barone, 2008). Therefore, mental speech representations, regarding phonological structure and phoneme identity, are likely less accurate for less proficient CI users than proficient users.

The lack of precision of phoneme coding might impact acquisition of orthographic representations. In alphabetic languages like English or French, the acquisition of accurate word spelling is a long process that requires the ability to (1) use correspondences between phonemes and letters (or letter clusters) representing them, (2) memorize a specific succession of letters in a word when they are not entirely predictable from the phoneme-to-grapheme correspondences, and (3) use morphological knowledge (Williams & Mayer, 2015). English and French are considered to be inconsistent languages. A first indicator of consistency is the ratio of the number of phonemes to the number of letters. This ratio is higher in English (1.7:1) than in French (1.5:1) (Caravolas, 2004). Critically, in the French written system, the phoneme-to-grapheme correspondences used to spell words are far less consistent than the grapheme-to-phoneme correspondences used to read words (Ziegler, Jacobs, & Stone, 1996). A second indicator is the ratio of sound-spelling consistency (e.g., [o] in French can be written "*o*", "*au*", "*eau*") in relation to the total number of words including this specific speech sound (e.g., "*bateau*", "*vélo*", "*auto*"). French and English systems share similar sound-spelling consistency for this latter indicator (Caravolas, 2004). The use of phoneme-to-grapheme rules makes it possible to spell only about 50% of French words (Véronis, 1986). However, in French as in English, the words are rarely fully predictable or unpredictable, meaning that the ability to use correspondences between phonemes and letters must be completed by the ability to memorize a specific succession of letters in inconsistent words. Not only deaf children with a CI must master the two skills defined

above, but also possess accurate representations of the spoken language. English and French share, to a certain extent, a comparable orthographic complexity with some specific respective difficulties. Thus, the complexity of these two languages is definitively relevant to consider spelling abilities of deaf children with a CI.

A large number of studies reveal an age-appropriate reading level for deaf children with a CI (e.g., Harris, 2016; Harris, Terlektsi, & Kyle, 2017; Leybaert & Dominguez, 2012; Mayer & Trezek, 2017). By contrast, the number of studies documenting spelling accuracy and errors of deaf children with CI in English and French is limited (Apel & Masterson, 2015; Bouton & Colé, 2014; Harris & Terlektsi, 2010; Hayes, Kessler, & Treiman, 2011; Quick, Harrison, & Erickson, 2018; Roy, Shergold, Kyle, & Herman, 2015; Williams & Mayer, 2015; for a review see Mayer & Trezek, 2017). Hayes, Kessler, and Treiman (2011) conducted an important study of spelling with English-speaking deaf children with a CI. A picture spelling task was used to compare 39 deaf children with a CI to a group of hearing children. The data were analyzed using a generalized linear mixed model. Results showed that both hearing and deaf children with a CI spelled more accurately words that were short, frequent, and predictable (see Hayes et al., 2011, p. 529 for details of how predictability was computed). Hearing status interacted with predictability: a greater difference between predictable and unpredictable words was observed in hearing than in deaf children with a CI, suggesting that deaf children were less sensitive to the frequency of phoneme-to-grapheme correspondence. Age and reading comprehension had a positive effect on accuracy in both groups. Age at implantation did not influence the spelling of deaf children with a CI. Analysis of spelling errors helped clarify the strategies used by hearing and deaf children with a CI. Misspellings were scored as Phonological Plausible Errors (PPE, e.g., BRAINE for brain) or Phonological Unacceptable Errors (PUE, e.g., BRIANE or BRAN for

brain). The proportion of PPE compared to the total number of errors was lower in deaf children with a CI than in hearing children (44% vs 75%, respectively), confirming that deaf children with a CI used a phonological spelling strategy to a lesser extent than hearing children. Unlike in previous spelling studies (Aaron, Keetay, Boyd, Palmatier, & Wacks, 1998; Leybaert, 2000; Leybaert & Alegria, 1995; Leybaert & Lechat, 2001; Roy et al., 2015), deaf children with a CI made the same proportion of transposition errors (e.g., SORPT for sport) as the hearing group, which indicates that they do not rely more on root spelling memorization. In sum, deaf children with a CI reached equivalent accuracy as age-matched hearing children, with equal sensitivity to word length and word frequency. Deaf children's lower sensitivity to predictability and lower proportion of PPE both point toward an inaccuracy of the phonological representations or in the use of phoneme-to-grapheme correspondences in a highly inconsistent orthography.

There are very few studies on spelling accuracy of deaf children with a CI in French. Leybaert, Bravard, Sudre et al. (2009) recorded spelling productions of 33 French-speaking deaf children with a CI, and 20 hearing children matched by grade level (from 2nd to 5t^{h} grade). Deaf children with a CI were divided into two subgroups depending on whether they had been exposed to French Cued Speech or not (CS+ and CS-, respectively). Children were asked to spell dictated words from the *Batterie d'Évaluation du Langage Écrit* (BELEC) (Mousty & Leybaert, 1999). Accuracy was similar in the hearing and the CS+ groups and better than the accuracy of the CS- group, suggesting that deaf children's orthographic representations are related to the precision of their phonological representations acquired through exposure to CS (see Rees and Bladel, 2013 for similar evidence about English Cued Speech). The relationship between spelling scores, age at implantation and speech perception scores was not examined in this study. Colin, Lina-Granade, Ecalle, Pénillard & al. (2010) did a transversal research of 60 prelingual profoundly

deaf children with a CI, aged 7 to 10 years old. All had been exposed to French Cued Speech for a mean time of 48 months. A picture written naming task was used to assess spelling abilities. Words contained predictable and unpredictable phoneme-to-grapheme correspondences. After controlling for chronological age, the duration of use of the CI contributed to 14% of the additional variance of word spelling. There was no effect of duration of CS. Colin and collaborators concluded that spelling abilities are related to the amount of auditory experience with CI. Finally, Bouton & Colé (2014) compared ten deaf children with a CI, aged from 9.5 to 12.3 years with ten hearing children, paired on word reading abilities. Children had to spell dictated words from the BELEC (Mousty & Leybaert, 1999). Out of ten deaf children with a CI, six had scores within the confidence interval of the hearing children for predictable spelling grapheme-to-phonemes, indicative of phonological procedure. Eight deaf children with a CI had scores within the confidence interval of the hearing children for unpredictable phoneme-to-grapheme correspondences, indicative of the orthographic procedure. Thus, the results point to a larger deficit in the acquisition of accurate correspondences between phonemes and graphemes than to the use of word orthographic representation. Better word orthographic representations could be related to the fact that deaf children with a CI were older than hearing children and had benefitted from more exposure to written words.

This brief literature review highlights the need for a more comprehensive and accurate investigation of spelling abilities in French-speaking deaf children with a CI. The present study uses a picture written naming task to compare deaf children with a CI to hearing children regarding the percent of correct answers and the proportion of PPE and PUE. As in Hayes et al's study, we used a generalized linear mixed model analysis, which is a robust analysis to examine potential sources of variance. We investigated the effect of word frequency, word length, and

phoneme-to-grapheme predictability, by using fine-grained continuous measures of these variables. We also measured word reading level, auditory and audio-visual speech perception abilities as well as age at implantation as potential factors explaining the variance of French-speaking deaf participant's word spelling.

Three research questions are addressed in the present study:

1. Are there differences in word spelling accuracy between deaf children with a CI and hearing children when the two groups are matched for chronological age, word reading level, and school grade level? Do word frequency, word length and phoneme-to-grapheme predictability have a similar effect on deaf children with a CI and hearing children? Do children's reading abilities impact their spelling scores?
2. Do deaf children with a CI and hearing children differ in the type of spelling errors, i.e., less PPE and more PUE in deaf children with a CI?
3. Are speech perception scores and age at implantation related to spelling accuracy and the amount of PPE in deaf children with a CI?

Method

1. Participants

Twenty-five profound congenital deaf children with a CI (nine girls), with a mean age of 9.6 years (range from 7.08 years to 12.07 years) and from grades 2 to 6 were selected through deaf children services with the collaboration of professionals, mostly teachers and speech therapists. The mean age at implantation was 2.9 years old (*SD* = 1.6, range: 6 months to 82 months). According to their medical records, none of the deaf children with a CI had any other known physical, neurological or intellectual disability. All deaf children were born to hearing parents

with French as a native language. The etiology of deafness was genetic (n = 12), viral infection (n = 3) or unknown (n = 10). Seven deaf children benefited from one CI (without a contralateral device), 12 had a CI in one ear and carried a contralateral hearing aid and six benefited from bilateral CIs. The communication method reported in Table 1 reflects the language(s) used at home: spoken language only (n =13), spoken language + Sign Language (SL) (n = 1), spoken language + CS (n = 4), spoken language + CS + SL (n = 6). Thus, the sample was heterogeneous regarding age of implantation and communication methods. The participants' individual characteristics are presented in Table 1.

The control group included 25 hearing children (eight girls) with a mean age of 9.4 years old (range: from 7.10 to 12.10 years), and from grade 2 to 5. They were selected from a larger sample of 100 children to be paired to the deaf children with a CI group with respect to age, gender, and level of education. They had no known disability and were native speakers of French. The present study was approved by the ethics committee of the *Université libre de Bruxelles* in accordance with the ethical principles of the World Medical Association Declaration of Helsinki. Informed consent was obtained from parents before data collection and children gave their verbal consent.

2. Tasks

Word reading tasks. Word decoding skills (speed and accuracy) were assessed using the *Lecture en une minute* test (1-min reading Test), which requires the child to read out loud as many written words from a list as possible in one minute (Khomsi, 1999). This test is frequently used as a screening tool for word decoding skills. The words varied in decoding difficulty, from one-syllable stimuli (e.g., "*un*") to longer and more complex ones (e.g., "*gymnastique*"). According to

Bertrand, Fluss, Billard, & Ziegler (2010), the standardized values of this test, with an optimal threshold of 26 words correctly in one minute, make it possible to obtain a sensitivity index close to 91% and a specificity index of 98%. Among children with a score greater than 26, only 2% could be mistaken as poor readers, and 9% could be mistaken as good readers. Thus, this test provides a relevant measure to discriminate between poor and good readers. In our study, correct answers varied between 30 and 101. On average, deaf children with a CI read 56.20 (*SD* = 15.98) words correctly in one minute, compared to 66.2 (*SD* = 20.82) for hearing children: the two groups did not differ (F (1.49) = 3.49; p = .07).

Orthographic representations of written words were assessed using the *Test d'identification du mot écrit* (written word identification Test) (Khomsi, 1999). It consists of 50 written words, each presented with a picture. Children were asked to read each word silently and to decide whether or not it matched the picture. The 50 words consisted of 10 pseudo-synonyms (e.g., BOL for *tasse; "*bowl" for *mug),* 10 pseudo-homophone spellings (e.g., MEZON for *maison* "house*"),* 10 graphic homophones (e.g., PEINT for *pain* "bread") and 20 correctly spelled words. Correct answers varied from 34 to 50. On average, deaf children with a CI identified 43.08 (*SD* = 4.34) words correctly as compared to 43.48 (*SD* = 4.22) for hearing children.

These two tests are complementary: while the 1-min reading Test taps on reading speed and accuracy, the written word identification Test evaluates word form recognition. In addition, they are frequently used by French-speaking speech therapists and have been normalized across a population of 526 children. To standardize the words' reading values obtained by our participants, the raw scores of correct answers on each task were transformed into z-scores. Thereby, we obtained two standardized measures of words' reading values: reading speed (the 1-min reading test) and reading precision (Written Word Identification Test).

*Speech perception test***.** Auditory and audiovisual speech perception of deaf children with a CI were assessed using the *Test d'évaluation de la réception du message oral* (oral message reception evaluation Test) (Busquet & Descourtieux, 2003). Children were asked to repeat two lists of 50 disyllabic words as accurately as possible. Each list was presented in two modalities: auditory only (AO) and audiovisual (AV: auditory + lip-reading). The number of accurately repeated words was measured. This test is commonly used to determine children with CI speech perception efficiency yet does not include standardized values. According to the criterion generally accepted in clinical practice, less than 65% of correct answers refers to a low post-CI performance (Turgeon, Lazzouni, Lepore, & Ellemberg, 2014). On average, deaf participants with CI correctly repeated 42.16 (*SD* = 9.71) words in the AO condition, and 44.28 (*SD* = 6.00) in the AV condition. Raw scores for correct answers in each modality were standardized into two z-scores.

*Picture spelling task***.** A picture spelling task was designed for the present research. Picture presentation was chosen instead of spoken words to avoid auditory perceptive errors which could affect the access to orthographic representations and the phoneme-to-grapheme conversion process (see Hayes et al., 2011; Leybaert, 2000; Leybaert & Alegria, 1995; Leybaert & Lechat, 2001).

The theoretical framework of this test is based on the *Batterie d'évaluation du langage écrit* (BELEC, written language assessment Battery) which has normalized data from 217 children from 2nd to 4th grade (Mousty & Leybaert, 1999). Our picture spelling task assesses word spelling taking into account word frequency, word length, and phoneme-to-grapheme predictability. We selected 73 words from Manulex, a lexical database that supplies a grade-level frequency list of

1.9 million words provided by 54 French elementary (1st to 5th) schoolbooks (Ortéga & Lété, 2010). All words were easy to picture and had the following characteristics:

- *Word frequency*: frequency varied between 0.15 and 514.04 (estimated frequency of use for one million word). This variable underwent a logarithmic transformation to normalize distribution (Tabachnick & Fidell, 2001).
- *Word length*: length was defined as the number of phonemes, varying between two and seven phonemes.
- *Phoneme-to-grapheme predictability*: we calculated a phoneme-to-grapheme predictability measure for each word by using the Manulex measure of frequency with which a phoneme-grapheme combination appears, divided by the total frequency of the phoneme in the word, multiplied by 100. For example, PR-I-S-ON ("jail") was very predictable (92.32%) and N-ŒU-D ("knot") was less predictable (29.03%). Our selected words' predictability varied between 23.96% and 97.04%.

A black and white drawing representing each word was downloaded from the internet without copyrights. All stimuli characteristics are summarized in Table 2.

3. Spelling error analysis

We defined two error categories: phonologically plausible errors (PPE) and phonologically unacceptable errors (PUE). Phonologically plausible errors were misspellings in which each phoneme in the word was spelled in a correct left-to-right sequence by a letter (or letter group) that aligns with that phoneme. Examples of PPE are BAINOIRE for *baignoire* ("bathtub"); PONPIER for *pompier* ("firemen"). PUE were misspellings in which the word's phonological structure was not respected. Phonologically unacceptable errors included consonant substitutions e.g. BAKE for *bague* ("ring"), SABIN for *sapin* ("christmas tree"); vowel substitutions, e.g.

LONPE for *lampe* (“lamp”), MOLIN for *moulin* (“mill”); letter transposition, e.g. SIRTON for *citron* (“lemon”), GIATRE for *guitare* (“guitar”); phoneme deletion, e.g. SIQUE for *cirque* (“circus”), PISON for *prison* (“jail”) and phoneme addition e.g. LAPRIN for *lapin* (“rabbit”); FROMAGRE for *fromage* (“cheese”).

Children were asked to name each picture out loud and spell the name it. If the picture was incorrectly named, the experimenter asked questions to encourage the child to figure out the correct name. For each child, a percentage of correct spellings out of 73 words was calculated, then a percentage of PPE and PUE were calculated from the percentage of errors.

Results

The data were analyzed using a generalized linear mixed model (GLMM) (Baayen, Davidson, & Bates, 2008). We used the *R* software (R Development Core Team, 2016) with the *lme4* package and the *glmer* function (Bates, Maechler, Bolker, & Walker, 2015). This model allows simultaneous consideration of the variability associated with random effect variables, such as individuals and stimuli (Barr, Levy, Scheepers, & Tily, 2013). The maximally converging (Barr & al., 2013) random structure included intercepts for words and subjects, thus accounting for inter-item and inter-individual variability. As some of the variance can be accounted for using this random structure, the GLMM provides a robust statistical method in comparison to ANOVA-based approaches (Quené & Van den Bergh, 2008).

A first GLMM analysis tested the predictors tied to words (i.e. frequency, length and predictability) and the predictors tied to all subjects (i.e. hearing status, age at testing, level of education, sex, word reading scores) on the percentage of correct spelling. A second GLMM analysis tested speech perception scores (AO and AV) and age at implantation as predictors of

the type of errors made by deaf children with a CI. All of these predictors were considered as fixed-effect factors. Following Barr et al. (2013), we included the maximally converging random effects structure justified by the experimental design, i.e. random intercepts for words and subjects. We used a backward elimination procedure by removing predictors and interactions that were not significant. To compare GLMM models, we used the Akaike Information Criteria (AIC) (Saefken, Kneib, van Waveren, & Greven, 2014). Thereby, the model with a lower AIC, meaning both simple and precise, was preferred. We followed the recommendation from Wieling (2015) to keep a more complex model only if the AIC decreases by at least two (indicating that the model is about 2.7 times more likely to be true). Before carrying out these analyses, potential multicollinearity problems, where two variables are too strongly correlated, needed to be checked (Tabachnick & Fidell, 2001). A too strong correlation suggests that variables are redundant, and that one variable can be perfectly predicted by the other (r>.90). Regarding redundancy between variables on our selected words, we obtained a significant correlation between frequency and length R = -.10, (p <.001), frequency and predictability R = .14, (p <.001) as well as predictability and length R = .64, (p <.001). Regarding redundancy between variables on all participants, we also obtained a significant correlation between the level of education and age R = .83, (p <.001) as well as a significant correlation between the two measures of word reading (i.e. speed and precision) R =.54, (p <.001). Finally, regarding redundancy between variables measured for deaf children with a CI only, we obtained a significant correlation between the two modalities of the speech perception test R = .46, (p <.001). Considering the significant positive correlation between some of our variables of interest, we calculated Variance Inflation Factors (VIFs) to check the inflation of a given coefficient to explain the variance, due to its dependence to other predicting variables. According to Rogerson (2001), a VIF must be less than five, while a variance equal to one represents a non-inflated variance. In our study, VIFs were between 1.11 and 2.80 for each

predictor. As the postulate of multicollinearity was respected regarding previous correlations and VIFs, we performed the GLMM.

1. Spelling accuracy

We first investigated whether deaf children with a CI and hearing children produced an equal number of correct spellings. On average, deaf children with a CI spelled 63.67% (*SD* = 23.79) of words correctly as compared to 66.08% (*SD* = 21.08) for hearing children. Our data set contained 1162 correct answers for deaf children with a CI and 1206 for hearing children. Our first GLMM included all children, used spelling accuracy as the binary response variable, with words and subjects as random intercepts, and obtained an AIC index of 3309.9. An additional model included hearing status as a fixed effect. This new model revealed that hearing status was not a significant predictor (p = .77) and did not improve the previous model (i.e. the AIC index remained the same: 3309.9). This suggests that spelling accuracy of deaf children with a CI did not differ from that of hearing children. The absence of interaction between correct answers and hearing status alone did not justify running separate analyses for each group.

We ran GLMM models with other fixed effects (i.e. gender, age at testing, level of education, and the two measures of words reading) on all subjects, with or without hearing status as a factor. Hearing status did not interact with any of the other predictors. AIC indexes for sex (3308.5), age at testing (3303.4) and level of education (3303.4) were higher compared to reading abilities (3231). This result indicates that reading abilities (both precision and speed measures comprised in the same analysis) predict spelling accuracy best.

Then, we ran the same analysis including reading abilities as a fixed effect with all predictors tied to words (i.e. frequency, predictability and length). The model that included fixed

effects of reading abilities and predictors tied to words accounted for significantly more variance than the model containing only the random effects of words and children. Table 3 lists the estimated coefficients, their standard errors, z-values and associated *p*-values for the predictors that emerged as significant in the best model. The AIC index was 3231 with a significant interaction between the random and fixed effects ($p < .001$).

Reading abilities predicted best for spelling accuracy in all children ($p < .0001$): better readers made significantly fewer spelling errors than poorer readers. The analysis revealed significant effects for word frequency ($p < .0001$) and word length ($p < .05$) on spelling accuracy but no significant effect for predictability ($p > .05$). Children spelled more accurately frequent words than rare words and short words than long words. Predictability failed to obtain a significant effect in this GLMM. Alone, predictability had a significant effect on spelling accuracy ($p < .05$) and a strong correlation with word length ($r = .64$). To sum up, accurate spelling performances were better explained by reading efficiency, word frequency and word length. Deaf children with a CI were as equally influenced as hearing children by psycholinguistics variables tied to spelling.

2. Spelling errors

Our results, acquired on 25 deaf children with a CI, with French as a native language, were slightly different from those of Hayes et al. (2011). We ran the GLMM on error data, as previously described for spelling accuracy. We ran the data set on 656 errors (PPE and PUE) of the picture spelling task for deaf children with a CI and 518 for hearing children. On average, the proportion of PPE errors was 54% for the deaf children with a CI and 75% for the hearing children. A first GLMM included all children, used PPE and PUE as the binary response variable,

with words and subjects as random effects. The AIC index was 1308.5 with a significant interaction between the random effects (p <.001). The same analysis was rerun including hearing status as a fixed effect. We obtained an equal AIC index (1308.5) including a significant interaction between random effects (p <.001) and a significant effect of hearing status (z = -3,239, SE = 0.312, p <.01), suggesting that deaf children with a CI made significantly less PPE ($M \pm SD$ = 59.21 ± 25.10) than hearing children ($M \pm SD$ = 65.04 ± 20.48). Consequently, deaf children with a CI made a significantly higher proportion of PUE ($M \pm SD$ = 39 ± 25.09) than hearing children ($M \pm SD$ = 22.14 ± 15.05).

Next, we tested the predictors tied to words (i.e. frequency, length and predictability) and the predictors related to all participants (i.e. age at testing, level of education, sex and the two reading measures). No interaction of hearing status with the participants or word characteristics was significant. Since PPE and PUE errors could not be explained by predictors shared by deaf and hearing children, we ran separate analyses, one for each predictor and only on deaf children with a CI. The results of these analyses are presented in Table 4.

First, the age of implantation (AIC index = 785.6) was a significant predictor of PPE and PUE (p = .001). Deaf children who were fitted later with a CI made significantly less PPE and more PUE than deaf children with early implantation. Speech perception scores (AO and AV) were also strong predictors of errors, whereas AIC indexes were respectively of 784.6 (p = .002) and 780.8 (p = .001). Thus, the ability of deaf children with a CI to repeat words presented auditorily and audio-visually was a significant predictor of PPE and PUE.

Discussion

The main focus of this study was to understand the factors influencing deaf children's spelling errors by comparing their French word spelling outcomes to those of hearing children matched for chronological age, level of education, gender and reading abilities. Overall, findings indicate that hearing status does not influence the percent of correct answers: French-speaking hearing and deaf children with a CI matched for word reading level achieved a similar spelling accuracy, consistent with data obtained in English by Hayes, Kessler & Treiman's (2011). A main effect of reading abilities indicates that children with good word reading skills performed better at a spelling task than children with poor word reading level. This was true for both hearing children and for deaf children with a CI. This suggests that children's ability to produce the correct spelling of a word is related to their ability to recognize the written form and to read the word aloud (Perfetti & Sandak, 2000). Spelling accuracy was influenced by word frequency and word length in both groups, confirming that orthographic representations are more easily retrieved for frequent than for rare words, and for short than for long words. These results support theories of the importance of phonological processes in reading and spelling (Hanson, Goodell, & Perfetti, 1991; Perfetti & Sandak, 2000). However, a lack of effect of phoneme-to-grapheme predictability in our data does not seem to be in agreement with the role of phonological processes in spelling. This result can be interpreted as follows: there was a high correlation between variables between predictability and length, for the selected words of our spelling task. After our model first took into account word length and frequency, the measure of predictability did not add a significant contribution to the explanation of variance in spelling accuracy. Taken together, our results on accuracy suggest that deaf children with a CI and hearing children process spelling with similar resources: a lexical orthographic and a phoneme-to-grapheme conversion procedure.

Differences were also found between the two groups. Deaf participants made significantly less PPE and more PUE than their hearing peers, suggesting that deaf children with a CI may have underspecified phonological representations. Hayes et al. (2011) also found a difference in PPE between deaf children with a CI (44%) and hearing children (75%). Previous studies demonstrated that deaf individuals have orthographic productions that are phonologically less plausible than matched hearing peers (Colombo, Arfé, & Bronte, 2012; Harris & Terlektsi, 2010; Kyle & Harris, 2006; Sutcliffe, Dowker, & Campbell, 1999), which was also true for deaf children with a CI (Mayer, Watson, Archbold, Ng, & Mulla, 2016; Quick et al., 2018). Our measurement of word reading was based on recognition tasks, where children had to match a printed word to an orthographic representation and to a word pronunciation (for the 1-minute test). In these recognition tasks, children may rely on incomplete word orthographic representations, which may be yet sufficient to recognize printed words. By contrast, spelling is a process that requires a written production by relying either on a well-formed orthographic representation or by using accurate phoneme-to-grapheme correspondences. The components of phonologically based spelling still seem to challenge deaf children with CI, possibly due to their current hearing limitations. Analyses of spelling errors made by the deaf children with a CI suggest a relation between age at implantation and speech perception scores (AO and AV) on the one hand, and percent of PPE and PUE errors on the other. These results support the hypothesis according to which implantation age and speech perception scores are determinant factors in production of phonologically based spelling.

Our results, with deaf children fitted with a CI, stand in stark contrast to the spelling achievement reported for profoundly deaf children without a CI (Aaron et al., 1998; Harris & Terlektsi, 2010; Leybaert & Alegria, 1995). The deaf children in our sample were heterogeneous

regarding proficiency with their implant. One question to be investigated in future research is the extent to which deaf children with good CI efficiency as opposed to poor CI efficiency differ in their spelling abilities (as they do in audio-visual speech perception, see Huyse et al., 2013). One way to enhance the spelling productions of deaf children with a CI would consist in improving the precision of their phonological representations. This goal can be achieved by presenting deaf children with words produced in Cued Speech. Cued Speech is a system aimed at disambiguating speechreading thanks to manual cues (Cornett, 1967). The manual cues have been designed to disambiguate phonemes and words that appear visually similar on the lips. Deaf children with a CI exposed early and intensively to Cued Speech within their home communication appeared to develop accurate phonological spelling to the same degree as hearing children (Leybaert, 2000; Leybaert & Lechat, 2001; see Trezek 2017 for a recent review).

Regarding the comparison between French and English, our results suggest a certain degree of similarity between children's abilities with CI in both languages. Likely, cross-linguistic differences also exist. We note that the difference between percentage of PPE and PUE is much more pronounced in the study by Hayes et al. (2011) when compared to ours. This suggests that French-speaking deaf children with a CI would be more comfortable using phoneme-grapheme correspondences than English-speaking deaf children with a CI. This may be due to the fact that French is a language in which grapheme-phoneme correspondences are more predictable than in English. In any case, a real cross-linguistic comparison study should be done before drawing firm conclusions on this topic.

Limitations and conclusions

This study has some limitations. First, phonemic awareness and working memory abilities should have been tested to acquire a more direct assessment of spelling skills. Deaf children with a CI may have less developed phonemic awareness skills, when compared to hearing children, and this could have an effect on their spelling. Short-term and working memory skills are also related to spelling accuracy and types of spelling errors, as suggested by Arfé, Ghiselli, & Montino (2016). The verbal digit span of deaf children with a CI is generally lower than those of hearing peers (e.g., Pisoni & Cleary, 2003). Deaf children with a CI are a heterogeneous group, and some of them demonstrate equivalent verbal memory skills to their hearing peers (Willems & Leybaert, 2009). This could be critical in explaining the inter-individual variability in language performances, including in spelling. Thus, the current results should be replicated in a study involving additional control measures (i.e. working memory and phonemic awareness) and a more detailed investigation of the phonological and orthographic rules that remain difficult for some deaf children with a CI (see Bowers, McCarthy, Schwarz, Dostal, & Wolbers 2014 for a detailed analysis of linguistic components in deaf children' spelling).

Second, a larger sample size of deaf children with a CI would be needed to measure the impact of the communication mode and more precisely, the effect of exposure to cued speech (Colin, Leybaert, Ecalle, & Magnan, 2013; Leybaert & LaSasso, 2010; Leybaert et al., 2009) and sign language. In our study, most deaf children with a CI who were exposed to cued speech also benefitted from sign language, which precludes a serious investigation of the effect of these methods of communication.

To conclude, this is the first study to provide strong evidence demonstrating how spelling abilities and spelling errors of French-speaking deaf children with a CI are related to psycholinguistic variables (i.e. word frequency and length) and to reading ability. As expected, there was no difference in spelling accuracy between deaf children with a CI and hearing children carefully matched for age, gender and level of education. In deaf children, the use of phoneme-to-grapheme correspondences is related to age at implantation and speech perception abilities. Our data support the idea that deaf children with a CI could follow a typical trajectory of written language acquisition, with reading abilities predicting spelling outcome. Even though an amount of variability remains, rehabilitation strategies based on the results of this study should be reinforced to improve the literacy outcomes of deaf children with a CI.

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Table 1.

Characteristics of participants with CI

Subject	Sex	Age at the testing	Age at CI	Etiology	Grade school	CI	Communication	AO	AV
1	F	8.10	23	Unknown	3rd	Bilateral	Oral only	46	48
2	M	12.07	26	Genetic	5th	Bilateral	Oral only	44	46
3	M	11.10	27	Infection	5th	Bilateral	Oral only	36	48
4	M	10.08	10	Infection	4th	Bilateral	Oral only	30	37
5	M	7.08	6	Genetic	2nd	Bilateral	Oral only	47	48
6	M	8.06	94	Unknown	2nd	With HA	Oral only	47	48
7	M	10.00	21	Unknown	3rd	Monolateral	Oral with SL and CS	42	39
8	M	11.03	17	Genetic	5th	With HA	Oral with SL and CS	45	45
9	M	9.05	12	Genetic	3rd	With HA	Oral with SL and CS	48	49
10	M	11.08	31	Genetic	4th	With HA	Oral with SL and CS	31	30
11	M	11.09	24	Genetic	5th	Monolateral	Oral only	46	46
12	F	11.05	35	Genetic	5th	Monolateral	Oral only	49	47
13	M	9.05	46	Genetic	4th	With HA	Oral only	47	49
14	M	8.08	32	Unknown	3rd	Bilateral	Oral with SL and CS	43	42
15	F	8.11	30	Genetic	3rd	Monolateral	Oral with SL and CS	22	33
16	M	8.01	42	Genetic	2nd	Monolateral	Oral with SL	43	45
17	F	9.03	36	Unknown	3rd	With HA	Oral with CS	43	45
18	F	9.07	53	Unknown	4th	With HA	Oral only	47	47
19	F	9.09	36	Unknown	4th	With HA	Oral with CS	46	46
20	F	8.05	31	Unknown	4th	With HA	Oral with CS	47	47
21	M	9.03	33	Genetic	3rd	With HA	Oral with CS	49	50
22	M	11.03	48	Infection	5th	Monolateral	Oral with SL and CS	49	45
23	F	11.07	82	Unknown	6th	With HA	Oral only	48	50
24	M	10.00	34	Unknown	4th	With HA	Oral only	9	29
25	F	11.04	48	Genetic	6th	Monolateral	Oral only	50	48

Notes. Hearing Aid (HA); Sign Language (SL); Cued Speech (CS), Auditory Only raw scores (AO), Auditory Lip-reading raw scores (AV), Communication (Home).

Table 2.

Stimuli for picture spelling task and psycholinguistic characteristics by lexical item

Number	Items	Frequency (ranks - range)	Length (number of phonemes - range)	Predictability (%)	Translation (english)
1	Fée	112.55 – frequent	2 – short	63.89	Fairy
2	Sel	154.47 – frequent	3 – short	63.17	Salt
3	Roi	359.81 – frequent	2 – short	97.04	King
4	Seau	95.24 – frequent	2 – short	33.51	Bucket
5	Prix	98.36 – frequent	3 – short	71.57	Price
6	Dent	128.70 – frequent	2 – short	54.03	Tooth
7	Pont	189.23 – frequent	2 – short	65.94	Bridge
8	Nœud	200.16 – frequent	2 – short	29.03	Knot
9	Cœur	236.09 – frequent	3 – short	58.17	Heart
10	Bras	242.57 – frequent	3 – short	79.82	Arm
11	Pain	331.26 – frequent	2 – short	52.16	Bread
12	Main	402.92 – frequent	2 – short	53.49	Hand
13	Singe	62.87 – frequent	3 – short	67.69	Monkey
14	Radis	131.21 – frequent	4 – short	83.06	Radish
15	Bague	131.23 – frequent	3 – short	78.6	Ring
16	Lampe	138.13 – frequent	3 – short	71.41	Lamp
17	Sapin	218.40 – frequent	4 – short	74.22	Pine
18	Tapis	221.51 – frequent	4 – short	79.84	Carpet
19	Train	356.20 – frequent	3 – short	65.46	Train
20	Lapin	386.80 – frequent	4 – short	84.65	Rabbit
21	Moulin	102.72 – frequent	4 – short	84.77	Mill
22	Réveil	103.00 – frequent	5 – short	63.46	Alarm clock
23	Prince	110.03 – frequent	4 – short	72.6	Prince
24	Fraise	113.24 – frequent	4 – short	78.57	Strawberry
25	Langue	113.89 – frequent	3 – short	63.1	Tongue
26	Citron	126.20 – frequent	5 – short	77.39	Lemon
27	Étoile	172.61 – frequent	4 – short	88.75	Star
28	Cadeau	189.54 – frequent	4 – short	73.25	Gift
29	Cirque	216.58 – frequent	4 – short	65.91	Circus
30	Cheval	292.77 – frequent	5 – short	87.81	Horse
31	Quatre	394.84 – frequent	4 – short	80.21	Four
32	Souris	514.05 – frequent	4 – short	72.55	Mouse
33	Guitare	189.79 – frequent	5 – long	82.48	Guitar
34	Bouquet	194.11 – frequent	4 – long	47.95	Bunch
35	Feuille	246.03 – frequent	3 – long	70.83	Leaf
36	Fromage	292.47 – frequent	6 – long	90.35	Cheese
37	Fauteuil	150.59 – frequent	5 – long	58.95	Chair
38	Cartable	155.29 – frequent	7 – long	79.94	Schoolbag
39	Champignon	111.07 – frequent	6 – long	79.63	Mushroom
40	Grenouille	129.11 – frequent	6 – long	75.51	Frog
41	Cent	13.62 – rare	2 – short	27.39	A hundred
42	Ogre	47.53 – rare	3 – short	94.37	Ogre
43	Nain	50.56 – rare	2 – short	47.77	Dwarf

44	Quai	22.11 – rare	2 – short	23.96	Platform
45	Quart	0.42 – rare	3 – short	58.3	Quarter
46	Rébus	0.61 – rare	4 – short	76.77	Rebus
47	Marin	31.14 – rare	4 – short	86.64	Sailor
48	Grain	54.15 – rare	3 – short	64.04	Seed
49	Gaufre	0.15 – rare	4 – short	78.9	Waffle
50	Compas	0.15 – rare	4 – short	60.8	Compass
51	Ciseau	0.61 – rare	4 – short	56	Scissors
52	Vernis	3.89 – rare	5 – short	74.67	Polish
53	Prison	13.28 – rare	5 – short	92.32	Jail
54	Sirène	34.39 – rare	5 – short	73,032	Mermaid
55	Cerise	35.48 – rare	5 – short	75.15	Cherry
56	Timbre	36.15 – rare	4 – short	78.23	Stamp
57	Requin	42.58 – rare	4 – short	54.78	Shark
58	Muguet	58.89 – rare	4 – short	54.97	Lily
59	Cochon	81.74 – rare	4 – short	90.06	Pig
60	Cadenas	0.30 – rare	5 – long	75.82	Padlock
61	Poulain	28.19 – rare	4 – long	73.24	Foal
62	Abricot	28.58 – rare	6 – long	83,69	Apricot
63	Sifflet	32.69 – rare	5 – long	53,35	Whistle
64	Pompier	49.35 – rare	5 – long	49,34	Fireman
65	Poussin	75.56 – rare	4 – long	66.98	Chick
66	Serpent	78.97 – rare	5 – long	58,21	Snake
67	Aiguille	31.67 – rare	5 – long	62,15	Needle
68	Quartier	48.49 – rare	6 – long	58,55	Segment
69	Araignée	62.87 – rare	5 – long	73,41	Spider
70	Guirlande	13.96 – rare	6 – long	77,83	Garland
71	Baignoire	46.21 – rare	5 – long	87,19	Bathtub
72	Trompette	78.85 – rare	6 – long	57.81	Trumpet
73	Citrouille	8.14 – rare	6 – long	73.79	Pumpkin

Table 3.

Mixed Model Analyses of Correct responses for Deaf and Hearing Children

Predictor	Estimate	*SE*	*z*	*p*
Reading (precision)	0.813	0.172	4,727	.0000***
Reading (speed)	0.703	0.170	4,119	.0000***
Frequency	0.476	0.083	5,678	.0000***
Length	0.146	0.070	2,076	.0379*
Predictability	0.015	0.010	1,536	.1245

Note. This table presents the results of one analysis with all deaf and NH children.
*p <.05. **p <.01. ***p <.001.

Table 4.

Mixed Model Analyses of Errors (PPE, PUE) for Deaf Children

	Estimate	*SE*	*z*	*p*
Age of cochlear implantation	-0.038	0.001	-19.4	.0000***
Audition only	-0.843	0.273	-3,081	.0020**
Audition + lip-reading	-0.776	0.218	-3,552	.0003***

Note. This table presents the results of separate analyses, one for each predictor and only on deaf children with CI. *p <.05. **p <.01. ***p <.001.