

Université de Montréal

**Caractérisation des approches de stimulation tactile suite à une
lésion nerveuse périphérique avec allodynie à la main :
une étude de cas et une revue systématique**

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Caractérisation des approches de stimulation tactile suite à une lésion nerveuse périphérique avec allodynie à la main : une étude de cas et une revue systématique

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Résumé

Problématique : Les lésions nerveuses périphériques peuvent entraîner une allodynie mécanique (AM) qui est une douleur neuropathique provoquée par le toucher. L'AM peut limiter les activités et les habitudes de vie des patients. Les approches de stimulation tactiles sont des interventions prometteuses pour traiter l'AM. Cependant, aucune étude n'a encore investigué l'intégration d'une telle approche dans un programme de réadaptation multimodal. De plus, il n'existe aucune synthèse des connaissances sur ces approches pour le traitement de l'AM.

Objectifs :

- 1- Décrire l'intégration d'une approche de stimulation tactile dans un programme de réadaptation multimodal;
- 2- Recenser les approches de stimulation tactiles et évaluer les évidences de ces approches pour traiter l'AM à la main suite à une lésion nerveuse périphérique.

Méthodologie : 1- Étude de cas. 2- Recension systématique sur les approches de stimulation tactile.

Résultats : L'étude de cas montre une diminution de l'AM et une amélioration des incapacités chez un patient présentant un syndrome de douleur régionale complexe qui a participé à un programme de réadaptation multimodal intégrant une approche de stimulation tactile. La recension systématique montre qu'il existe deux approches de stimulation tactiles (la désensibilisation et la rééducation sensitive de la douleur) pour traiter l'AM à la main. Ces approches ne se distinguent pas quant au niveau d'évidence de leur efficacité pour traiter l'AM suite à une lésion nerveuse périphérique.

Conclusion : Les deux approches peuvent être utilisées par les cliniciens pour traiter l'AM en fonction de leur raisonnement clinique et des caractéristiques des patients.

Mots-clés : rééducation sensitive, désensibilisation, allodynie, thérapie de la main, sensibilité, douleur, douleur neuropathique, lésion nerveuse.

Abstract

Context: Peripheral nerve lesions can lead to mechanical allodynia (MA), that is a neuropathic pain provoked by touch. MA can limit patients' activities and life habits. Tactile stimulations are promising approaches to treat MA. However, to our knowledge, there is no study that has investigated how such approaches can be integrated into a multimodal rehabilitation program. In addition, there is no synthesis of current knowledge on the tactile stimulation approaches for treating MA.

Objectives:

- 1- To describe the integration of a tactile stimulation approach in a multimodal rehabilitation program;
- 2- To identify existing tactile stimulation approaches and to assess evidences of the use of these approaches to MA in the hand following a peripheral nerve lesion.

Method: 1- Case report. 2- Systematic review on tactile stimulation approaches.

Results: The case report shows an abolition of MA and an improvement of incapacities in a patient with a complex regional pain syndrome who participated in a multimodal rehabilitation program including a tactile stimulation approach. The systematic review identified two tactile stimulation approaches (desensitization and somatosensory rehabilitation of pain). Those approaches do not differ in their level of evidence in the treatment of MA following a peripheral nerve lesion.

Conclusion: The two approaches can be used by clinicians to treat MA. The choice of these approaches should be based on clinical reasoning and patients' characteristics.

Keywords: somatosensory rehabilitation, desensitization, allodynia, hand therapy, sensibility, pain, neuropathic pain, nerve lesion.

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Liste des sigles

AM : Allodynies mécanique

AROM: *Active Range of Motion*

CINAHL: *Cumulative Index to Nursing and Allied Health Literature*

CRPS: *Complex Regional Pain Syndrome*

DASH: *Disabilities of the Arm, Shoulder and Hand Questionnaire*

DN : Douleur neuropathique

GMI: *Graded Motor Imagery*

LNP : Lésion nerveuse périphérique

MINORS: *Methodological Index for Non-Randomized Studies*

MRSD : Méthode de rééducation sensitive de la douleur

NP: *Neuropathic Pain*

NSPA: *Numeric Pain Scale Assessment*

NRS: *Numerical Rating Scale*

PNI: *Peripheral Nerve Injury*

PRISMA: *Preferred Reporting Items for Systematic Reviews and Meta-Analyses*

QDSA: Questionnaire de la douleur Saint-Antoine

RPS: *Rainbow Pain Scale*

SDRC : Syndrome douloureux régional complexe

SMA: *Static Mechanical Allodynia*

SRM: *Somatosensory Rehabilitation Method*

TENS: *Transcutaneous Electrical Nerve Stimulation*

VAS: *Visual Analog Scale*

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Avant-propos

En clinique, les douleurs neuropathiques, dont l'allodynie mécanique (hypersensibilité au toucher), sont fréquemment observées lors des blessures à la main. L'allodynie mécanique a un impact sur les capacités en interférant avec les activités qui demandent un contact cutané sur la main (ex. laver la vaisselle, utiliser un clavier, etc.). Elles sont également difficiles à traiter en réadaptation, mettant souvent en échec les modalités de traitement demandant un contact cutané (ex. massage, mobilisations, port de gant compressif, massage de cicatrice, etc.). C'est pourquoi cette problématique est importante à traiter. En réadaptation, l'approche conventionnelle par désensibilisation est utilisée depuis quelques dizaines d'années au Québec pour traiter l'allodynie mécanique. Tel qu'observé en clinique, cette approche parvient à diminuer les douleurs chez certains patients, mais il demeure qu'une bonne proportion de patients ne sera pas soulagée. C'est une des raisons pour lesquelles certains thérapeutes sont à l'affût de nouvelles approches et méthodes d'intervention innovantes pour le traitement de l'allodynie. Dans les dernières années, la méthode de rééducation sensitive de la douleur a été développée en Suisse. En raison de mon intérêt clinique pour le traitement de l'allodynie, j'ai été la première ergothérapeute du Québec à me former à cette méthode au Centre de rééducation sensitive du corps humain de Fribourg. Suite à cette formation, j'ai enseigné cette méthode à mes collègues dans le cadre de formations continues.

Les milieux cliniques reconnaissent que la méthode de rééducation sensitive de la douleur diminue la douleur de type allodynie en plus d'améliorer les capacités des patients qui en sont atteints. De plus, cette méthode est appliquée avec succès en combinaison avec d'autres modalités thérapeutiques. Malgré ces observations cliniques, cette combinaison de traitement qui semble prometteuse n'est pas illustrée dans la littérature. Cela a mené à ma première

publication de ce mémoire soit une étude de cas présentant le raisonnement clinique qui sous-tend un programme multimodal incluant la méthode de rééducation sensitive, ainsi que ses effets potentiels sur la douleur et la performance motrice.

Cette étude de cas m'a amené à me questionner plus à fond sur les approches de stimulation tactiles. Quel est l'apport de la méthode de rééducation sensitive de la douleur, et plus encore des approches de stimulation tactiles, dans le traitement des douleurs de type allodynie ? Plus précisément, quel éventail de modalités de traitement est inclus dans ces approches de stimulation tactile ? Quel est le niveau de preuve de ces approches pour traiter l'allodynie ? Ces réflexions m'ont amené à faire une recension systématique des écrits comme première étape afin de tenter de répondre à ces questionnements.

En résumé, dans le cadre de ce mémoire, la publication d'une étude de cas a permis d'illustrer mon cheminement clinique pour le traitement de l'allodynie tout en assurant un transfert de connaissances aux cliniciens. Ce premier article permet de fournir des pistes de réflexion pour soutenir le raisonnement clinique dans le cadre de l'utilisation des approches de stimulation tactiles incluses dans un programme multimodal pour traiter l'allodynie. Quant à la recension des écrits, elle a permis d'explorer l'éventail des interventions par stimulation tactiles disponibles ainsi que les effets de ces approches et leurs niveaux d'évidence. Il est espéré que l'ensemble de cette démarche dans le cadre de ma maîtrise de recherche saura être utile aux cliniciens qui travaillent quotidiennement à améliorer les capacités et la vie quotidienne de ces patients avec allodynie.

Chapitre 1 : Introduction

1.1 Neuropathies

Les neuropathies sont définies comme des atteintes du système nerveux périphérique qui incluent, entre autres, les polyneuropathies et les mononeuropathies (Merskey & Bogduk, 1994). Les polyneuropathies consistent en des neuropathies diffuses et bilatérales (Merskey & Bogduk, 1994), c'est-à-dire qui affectent plusieurs nerfs dans l'ensemble du corps. Elles incluent un ensemble de conditions diverses dont la polyneuropathie diabétique, la polyneuropathie des soins intensifs et la maladie de Charcot-Marie-Tooth. Quant aux mononeuropathies, elles consistent en des atteintes focales, c'est-à-dire des atteintes qui touchent une seule branche nerveuse (Merskey & Bogduk, 1994). Les neuropathies sont souvent désignées par l'appellation de « lésions nerveuses périphériques (LNP) ». Leur prévalence est importante. Par exemple, en considérant le syndrome du tunnel carpien, qui est une atteinte focale par compression du nerf médian, cette mononeuropathie affecterait à elle seule 6,4% de la population néerlandaise (Atroshi et al., 1999; de Krom et al., 1992). D'ailleurs, les mononeuropathies sont le plus souvent causées par des atteintes mécaniques ou traumatiques (i.e., compression, étirement, lacération) qui affectent le système musculo-squelettique, mais également un nerf (Sunderland, 1951). Ainsi, dans un centre de trauma à Toronto (Canada), 2,8% des admissions pour trauma présentent des LNP sur une ou plusieurs branches nerveuses, dont plus de la moitié touche le membre supérieur (Noble, Munro, Prasad, & Midha, 1998). De ce fait, les LNP qui affectent les nerfs médian, radial et/ou ulnaire figurent parmi les diagnostics les plus fréquents en thérapie de la main (Keller et al., 2016).

Les mononeuropathies entraînent des coûts importants pour les systèmes de santé et d'indemnisation à travers le monde. À titre d'exemple, une étude suédoise réalisée en 2005 a estimé que les coûts des traitements et de la réadaptation de travailleurs ayant subi des mononeuropathies traumatiques nécessitant une intervention chirurgicale, était d'une médiane de 31 186 EUR et de 51 238 EUR par travailleur pour des lésions des nerfs ulnaire et médian respectivement (Rosberg et al., 2005). Plus de 87% de ces coûts résultait du manque à gagner en raison des pertes de productivité chez les travailleurs (Rosberg et al., 2005). Ces données ne sont pas disponibles pour la population canadienne, mais il est raisonnable de croire que ces lésions entraînent des coûts tout aussi importants au Canada.

Par ailleurs, des études ont démontré que les LNP, incluant les mononeuropathies, engendrent des incapacités importantes qui peuvent limiter les habitudes de vie, le retour en emploi et affecter la qualité de vie des personnes affectées (Hundepool et al., 2015; Novak, Anastakis, Beaton, Mackinnon, & Katz, 2011a). En effet, une étude a démontré qu'un an après une lésion ayant entraîné une neuropathie focale au membre supérieur nécessitant une chirurgie, 40% des patients ne sont toujours pas encore retournés en emploi (Bruyns et al., 2003). Chez ces patients, le niveau de capacités et le retour en emploi seraient liés, entre autres, à la sévérité de la lésion, la sensibilité à la main, la force de préhension et l'intensité de la douleur (Bruyns et al., 2003; Hundepool et al., 2015; Novak, Anastakis, Beaton, Mackinnon, & Katz, 2011b).

1.2 Douleurs neuropathiques

Les LNP peuvent entraîner des douleurs neuropathiques (DN) (Haanpää et al., 2011; Treede et al., 2008). La DN est définie comme une « douleur causée par une lésion ou une maladie du système nerveux somatosensoriel » (Merskey et al., 1994). Plusieurs études suggèrent que la présence de DN aurait un impact important sur les capacités des personnes ayant une LNP au membre supérieur. Par exemple, on a démontré une corrélation positive et significative entre la sévérité de la douleur et la perception des incapacités en lien avec les déficiences au membre supérieur évaluée à l'aide du questionnaire DASH (*«Disabilities of the Arm, Shoulder and Hand Questionnaire»*) chez des patients présentant des douleurs suite à une LNP (Novak et al., 2011b). D'autres études ont également mis en évidence un lien entre la douleur et les incapacités chez des personnes présentant des douleurs lors des traumas avec LNP affectant la main (Bailey, Kaskutas, Fox, Baum, & Mackinnon, 2009; Clement, Duckworth, Jenkins, & McEachan, 2016; Lozano Calderon, Paiva, & Ring, 2008; Souer, Lozano-Calderon, & Ring, 2008).

La DN peut se présenter sous plusieurs formes, dont la douleur spontanée (névralgie) ou provoquée (allodynique). La névralgie est une douleur neuropathique qui peut être perçue de manière incessante ou intermittente et qui apparaît spontanément, c'est-à-dire sans stimulation provoquante. Ce mémoire ne porte pas sur la névralgie, mais plutôt sur la DN de type allodynique. L'allodynique est définie comme une douleur provoquée par une stimulation normalement non-douloureuse (Merskey & Bogduk, 1994). Elle est précisée selon la modalité du stimulus provoquant la douleur, par exemple l'allodynique thermique provoquée la température

chaude et/ou froide et l'allodynies mécanique (AM) statique ou dynamique provoquée par un toucher immobile (statique) ou mobile (dynamique). L'AM n'est pas exclusivement neuropathique, car elle peut survenir par exemple dans des conditions inflammatoires (Merskey & Bogduk, 1994). Pour être considérée de type neuropathique, elle doit être corrélée avec des signes cliniques témoignant de l'atteinte neuropathique (symptômes, signes). Les mécanismes pathophysiologiques en cause dans l'AM ne sont pas précisés, mais il est possible que différents changements dans le système nerveux central au niveau spinal et supraspinal y contribuent, dont une neuromodulation et une neuroplasticité maladaptative. (Cervero & Laird, 1996; Cruciani, Stacy, & Knotkova, 2011; Finnerup, Otto, McQuay, Jensen & Sindrup, 2005; Latremoliere & Woolf, 2009; Osborne, Anastakis, & Davis, 2018; Wall, Xu, & Wang, 2002; Woolf, 2011).

À notre connaissance, les prévalences des DN et de l'AM ne sont pas connues au Canada. Cependant, dans la population générale du comté d'Olmsted (Minnesota, États-Unis), il est rapporté que la prévalence des DN varie entre 3,0 et 12,4% selon l'instrument de dépistage utilisé (Yawn et al., 2009). Également, dans la population générale en France, une étude a démontré une prévalence de 6,9% de personnes qui présentent de la douleur chronique avec des caractéristiques neuropathiques (Bouhassira, Lantéri-Minet, Attal, Laurent, & Touboul, 2008).

Quoiqu'il n'y ait pas, à notre connaissance, d'étude qui en fasse état, il est raisonnable de croire que l'AM à la main puisse avoir un impact important sur les habitudes de vie (activités courantes et rôles sociaux) en interférant avec les capacités spécifiques à l'utilisation de la main qui impliquent un contact cutané, telles que la dextérité et la force de préhension. Dans la

perspective où l’AM peut affecter la réalisation des activités, les interventions qui permettent de la réduire pourraient améliorer les capacités, ainsi que l’autonomie dans les habitudes de vie chez les personnes avec LNP affectant la main.

1.3 Syndrome douloureux régional complexe

L’AM est présente dans plusieurs conditions de santé. Par exemple, on peut l’observer chez certaines personnes atteintes de polyneuropathie diabétique (Scholz et al., 2016), chez certains grands brûlés (Nedelec et al., 2016; Schneider et al., 2006) et chez certains patients atteints de syndrome douloureux régional complexe (SDRC) (Harden et al., 2010; Packham, Spicher, MacDermid, Michlovitz, & Buckley, 2018). Le SDRC, qui est le diagnostic du patient présenté dans la première étude de ce mémoire, est caractérisé par une douleur continue disproportionnée par rapport à l’évolution habituelle après un trauma ou une autre lésion (Harden et al., 2010). La douleur, spontanée ou évoquée par un stimulus externe, est située dans une région du corps et non pas dans un territoire nerveux spécifique ou un dermatome. La douleur montre habituellement une prédominance distale d’anomalies sensitives, motrices, sudomotrices, vasomotrices (œdème) et/ou trophiques » (Harden et al., 2010). Deux sous-types de SDRC ont été définis : le SDRC-I et le SDRC-II. Le SDRC-I est associé à tout type de trauma, spécifiquement les fractures ou les lésions des tissus mous (Harden et al., 2010). Le SDRC-II est défini comme étant associé à une évidence physique ou électrodiagnostique de lésion nerveuse majeure (Harden et al., 2010). Bien que le SDRC-I n’est pas associé à une lésion nerveuse majeure, il demeure qu’il peut inclure des signes et symptômes caractéristiques des DN (ex : AM). Il n’est donc pas surprenant qu’il ait été démontré que certains SDRC-I soient

associés à certains types de LNP (Birklein & Schmelz, 2008; Oaklander & Fields, 2009). Une récente étude a trouvé que 54,3% de leurs participants avec SDRC-I rapportaient des symptômes d'AM, et que les signes cliniques confirmaient objectivement l'AM chez 27,6% de ces participants (Dietz et al., 2019). Par exemple, dans la première étude de ce mémoire, le patient présentait un SDRC-I avec AM. Chez ce patient, il n'y avait pas diagnostic de LNP majeure, mais la présence de LNP était objectivée, entre autres, par la diminution de la sensibilité tactile dans le territoire d'un nerf périphérique, la présence de paresthésies et d'AM à la main. Les hypothèses des mécanismes pathophysiologiques responsables des douleurs évoquées dans les cas de SDRC sont, comme pour les douleurs neuropathiques, liées à des modifications inadaptées du système nerveux central (Juottonen et al., 2002; Maihöfner, Handwerker, Neundörfer, & Birklein, 2003).

1.4 Traitement des douleurs neuropathiques

Plusieurs interventions ont été développées pour traiter les DN, dont l'approche médicamenteuse dont les effets sont limités (Attal et al., 2006; Dworkin et al., 2007; Mason, Moore, Derry, Edwards, & McQuay, 2004; Silver, Blum, Grainger, Hammer, & Quessy, 2007; Vranken, 2009). Les interventions non médicales incluent, entre autres, la psychothérapie (Moura et al., 2012), l'électrothérapie (*TENS : Transcutaneous Electrical Nerve Stimulation*) (Gibson, Wand, & O'Connell, 2017), la désensibilisation (Yerxa, Barber, Diaz, Black, & Azen, 1983) ainsi que la méthode de rééducation sensitive de la douleur (MRSD) (Spicher & Quintal, 2013). Dans la pratique clinique, la psychothérapie (Evans, Fishman, Spielman, & Haley, 2003; Moura et al., 2012) est plutôt considérée comme un adjuvant et est combinée aux autres

modalités. Quant au *TENS* et à la désensibilisation, les protocoles d'utilisation ne font pas consensus compte tenu que les populations dans les études sont hétérogènes et que les évidences sur leur efficacité sont limitées (Gibson et al., 2017; Lewis, Coales, Hall, & McCabe, 2011; B. Pleger et al., 2005; Yerxa et al., 1983). La MRSD (Spicher, Quintal, & Vittaz, 2015) utilise un protocole d'utilisation qui est standardisé, mais une fois de plus, les populations étudiées sont hétérogènes (Nedelec et al., 2016; Packham et al., 2018; Spicher, Mathis, Degrange, Freund, & Rouiller, 2008). De plus, les devis des études qui ont porté sur la MRSD fournissent seulement des évidences limitées de son efficacité.

La littérature propose que les programmes multimodaux personnalisés sont à privilégier dans le traitement des DN (Deng, Luo, Hu, Fang, & Liu, 2016; Finnerup, Otto, McQuay, Jensen, & Sindrup, 2005) incluant celles associées au SDRC (Packham & Holly, 2018). Dans la perspective où les approches de stimulation tactiles pourraient induire une réorganisation du système nerveux central (Flor, 2002; Flor, Denke, Schaefer, & Grusser, 2001; Wand, Abbaszadeh, Smith, Catley, & Moseley, 2013) et qu'elles ne sont pas des approches adjuvantes puisqu'elles visent à traiter la condition, il y a donc lieu de s'intéresser à leur inclusion dans un programme multimodal. Il devient donc pertinent de vérifier leurs potentiels effets bénéfiques sur la DN.

1.4.1 Désensibilisation

Les approches de stimulation tactiles utilisées en clinique incluent la désensibilisation et la MRSD, qui sont deux approches différentes de traitement pour la DN évoquée par stimulation

mécanique, c'est-à-dire l'AM. Historiquement, la désensibilisation (Yerxa et al., 1983) a été la première méthode développée pour une population avec AM à la main et selon nos observations en clinique, elle semble encore à ce jour la plus utilisée. Cette approche de désensibilisation utilise l'application de textures directement sur la surface allodynique. Au fur et à mesure de l'amélioration de la condition du patient avec la diminution de l'intensité de l'AM, cette approche promeut d'adapter progressivement les textures plus douces vers des textures plus rugueuses dans le but de modifier à la hausse le seuil de perception de la douleur (Yerxa et al., 1983). Autrement dit, l'objectif est que ce soit des stimulations tactiles plus importantes qui soient nécessaires afin de déclencher la douleur. Jusqu'en 2003, la désensibilisation était la seule approche de stimulation tactile connue et utilisée en réadaptation pour traiter l'AM.

1.4.2 Méthode de rééducation sensitive de la douleur

Puis, les limites cliniques de la désensibilisation, on fait en sorte que des alternatives à cette approche ont été développées. Tel qu'observé en clinique et tel que discuté avec le premier auteur à avoir décrit la MRSD, une certaine proportion de patients avec AM ne répondent pas positivement à l'approche de désensibilisation, ou pire leur symptomatologie douloureuse augmente. C'est pourquoi la MRSD a été développée et son protocole d'utilisation a été publié dans un manuel en 2003 (Spicher, 2003). Cette méthode a été développée non seulement pour les atteintes sensitives et douloureuses à la main, mais également pour les autres territoires cutanés du corps humain (Spicher et al., 2015). La MRSD se différencie de la désensibilisation, entre autres en raison du fait qu'elle inclut des évaluations standardisées pour l'AM : l'allodynographie et l'arc-en-ciel des douleurs. La première évaluation est une cartographie de la surface allodynique évaluée avec un monofilament Semmes-Weinstein de 15.0g (#5.18), reportée sur papier millimétrée. La deuxième évaluation objective la sévérité de l'allodynies avec

une série de sept monofilaments (de 0.03g à 15.0g), correspondant à la plus petite force d'application causant de la douleur dans la surface allodynique. Elle se différencie aussi dans le protocole de traitement de l'AM qui comprend deux modalités, soit 1) l'enseignement thérapeutique et l'encadrement du patient pour diminuer voire éviter le toucher avec la surface allodynique et 2) des textures uniquement douces, et de la vibration mécanique de faible amplitude (fréquence de 100Hz, amplitude de 0.06mm) appliquées à distance, c'est-à-dire sur une zone anatomique proximale à la surface allodynique (Spicher et al., 2015). Elle vise à ce qu'une stimulation non-nociceptive soit perçue comme étant effectivement non-douloureuse (Spicher et al., 2015) et ainsi modifier à la hausse le seuil de perception de la douleur. Les deux approches (désensibilisation et MRSD) utilisent des textures afin de normaliser les perceptions sensitives. Cependant, elles se distinguent en ce qui a trait à la batterie d'évaluations, l'encadrement thérapeutique, le site d'application des stimulations tactiles et le type de textures employées.

1.5 Problématique

Plusieurs modalités d'interventions sont souvent combinées dans les programmes en réadaptation pour traiter l'AM. Cependant, force est de constater que les études portant sur les interventions pour traiter l'AM ne reflètent pas la pratique clinique réelle. En effet, ces études investiguent de manière isolée les effets d'une ou l'autre des modalités d'interventions sans toutefois considérer leur combinaison dans un programme d'interventions multimodal. Ceci limite l'applicabilité clinique des résultats des études portant sur des modalités isolées. À ce jour, aucune étude n'a encore décrit de quelle façon ces modalités d'interventions sont utilisées

en combinaison et en séquence dans le cadre d'un programme multimodal. Pourtant, plusieurs recommandations cliniques issues de la littérature, préconisent l'utilisation de programmes multimodaux (Deng et al., 2016; Finnerup et al., 2005), ce qui reflète la pratique clinique actuelle en réadaptation où la combinaison des interventions est préconisée.

Par ailleurs, il existe à l'heure actuelle des synthèses de connaissances de la littérature portant sur l'utilisation des approches de stimulation tactiles pour traiter l'hypoesthésie tactile à la main (Miller, Chester, & Jerosch-Herold, 2012; Oud, Beelen, Eijffinger, & Nollet, 2007). Cependant, il n'existe, jusqu'à présent, aucune synthèse des connaissances sur l'utilisation des approches de stimulation tactiles pour traiter l'AM. Cette lacune dans la littérature a des conséquences tant sur la pratique clinique que sur la recherche dans ce domaine. En effet, il n'est pas clair quelles sont toutes les interventions de stimulation tactiles existantes pour traiter l'AM qui ont effectivement fait l'objet d'études. De plus, on ne peut statuer parmi ces interventions lesquelles disposent des meilleures évidences concernant leurs effets bénéfiques sur l'AM. Par conséquent, il est difficile de comparer les interventions existantes et de sélectionner la ou les meilleures approches de traitement pour l'AM.

Les études qui sont présentées et discutées dans ce mémoire tentent de répondre à cette problématique. Elles portent plus précisément sur l'utilisation des approches de stimulation tactiles pour traiter l'AM à la main chez des patients présentant des mononeuropathies. Ce mémoire présente 1) de quelle manière une approche de stimulation tactile pour traiter l'AM peut être intégrée à un programme de réadaptation multimodal, et 2) une synthèse des

connaissances sur les différentes approches de stimulations tactiles pour traiter l'AM consécutive à une LNP qui ont fait l'objet d'études.

1.6 Objectifs

Le premier objectif de ce mémoire est de décrire l'intégration de la MRSD dans le cadre d'un programme de réadaptation multimodal chez un patient présentant un SDRC avec AM qui n'avait pas eu d'effets bénéfiques avec des interventions conventionnelles. Afin de répondre à cet objectif clinique, une étude de cas a été effectuée et publiée (Quintal, Poiré-Hamel, Bourbonnais, & Dyer, 2018). La MRSD a été utilisée dans un cadre de traitement multimodal puisque la désensibilisation classique n'avait pas eu les effets escomptés et que la MRSD possède un caractère innovant.

Comme le programme de réadaptation multimodal présenté dans l'étude de cas inclut une approche de stimulation tactile, cela a questionné l'impact relatif de ce type d'approche dans ce programme, et donc les niveaux d'évidences cliniques des approches de stimulation tactiles répertoriées dans la littérature. Ainsi, le deuxième objectif de ce mémoire est de recenser les approches de stimulation tactile étudiées dans la littérature et leurs niveaux d'évidences en lien avec le traitement de la douleur chez des personnes avec AM à la main. Afin de répondre à cet objectif de recherche, une recension systématique a été effectuée. Cette méthodologie a été utilisée pour ses capacités à identifier, évaluer et résumer les résultats d'études sur une problématique de santé précise (Gopalakrishnan & Ganeshkumar, 2013). De plus, ce type de méthodologie, qui résume les évidences scientifiques, améliore l'accessibilité aux données

probantes pour les utilisateurs de connaissances, ces derniers n'ayant pas toujours le temps de lire et d'analyser l'entièreté de la littérature disponible sur un sujet précis (Gopalakrishnan & Ganeshkumar, 2013).

1.7 Organisation générale du mémoire

Suite à l'introduction ([Chapitre 1](#) : Introduction) présentée précédemment, ce mémoire se compose de quatre autres chapitres. Ainsi, afin de répondre aux deux objectifs généraux de ce mémoire, deux articles rédigés en anglais sont présentés ([Chapitre 2](#) : Manuscrit, article #1 et [Chapitre 3](#) : manuscrit, Article #2). Suivent une discussion générale ([Chapitre 4](#) : Discussion) présentant des éléments non discutés dans les articles et une conclusion ([Chapitre 5](#) : Conclusion). Les tableaux et figures se retrouvent directement à la suite des articles correspondants. Toutes les [Références](#) bibliographiques associées aux cinq chapitres se trouvent à la fin de ce mémoire. À noter que les mots en bleu servent également de renvois à des parties spécifiques du mémoire (ctrl+clic pour suivre les liens).

Chapitre 2 : Manuscrit, article #1

Le chapitre 2 vise à répondre au premier objectif de ce mémoire : décrire l'intégration de la MRSD dans le cadre d'un programme de réadaptation multimodal chez un patient présentant un SDRC avec AM qui n'avait pas eu d'effets bénéfiques avec des interventions conventionnelles.

2.1 Management of long-term complex regional pain syndrome with allodynia: A case report.

Les résultats de cette étude de cas ont sous forme d'article a été soumis au *Journal of Hand Therapy* le 28 novembre 2017, a été révisé le 16 janvier 2018, puis accepté pour publication le 22 janvier 2018 (Quintal, Poire-Hamel, Bourbonnais, & Dyer, 2018). Le consentement du participant a été obtenu avant la soumission à l'éditeur et la publication de l'article.

2.1.1 Apport de l'étudiante et de chacun des co-auteurs

Isabelle Quintal a été la principale responsable de chacune des étapes menant à la publication de ce manuscrit, c'est-à-dire : l'élaboration du devis, la collecte de données, l'analyse de données, la rédaction du manuscrit, la soumission au journal ainsi que gérer les révisions demandées.

Laurent Poiré-Hamel ergothérapeute clinicien, a participé à la collecte de données, à l'analyse des données ainsi qu'à la rédaction du manuscrit.

Daniel Bourbonnais et Joseph-Omer Dyer ont dirigé la principale responsable de ce manuscrit, Mme Quintal, dans l'ensemble des étapes menant à la publication finale.

Management of long-term complex regional pain syndrome with allodynia: A case report

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[https://www.jhandtherapy.org/article/S0894-1130\(17\)30407-6/fulltext](https://www.jhandtherapy.org/article/S0894-1130(17)30407-6/fulltext)

2.2 Abstract Study design: Case report.

Introduction: Conventional rehabilitation alone may not be effective in reducing symptoms in some patients with complex regional pain syndrome.

Purpose of the study: This case report portrays the benefits of a new tailored rehabilitation program for a 39-year-old patient suffering from upper limb complex regional pain syndrome with severe touch- evoked pain (static mechanical allodynia).

Methods: This patient had previously received conventional rehabilitation for a year and a half including physical and nonsurgical medical interventions that did not improve symptoms or function. In the search for an alternative, this patient was referred to occupational therapy to try a tailored rehabilitation program, drawing on multiple strategies used sequentially according to the patient's tolerance and symptom evolution. During this 22-month program, the following methods were added (listed chronologically): somatosensory rehabilitation of pain method, graded motor imagery, pain management modalities, active mobilizations, strengthening exercises, and task simulation. The patient successively showed resolution of mechanical allodynia, decreased pain, reduction of tactile hypesthesia and improvement in active range of motion, strength, and function. These improvements allowed him to return to work.

Discussion: This suggests that a tailored rehabilitation program combining somatosensory rehabilitation of pain method, graded motor imagery and more conventional approaches could improve symptoms and functional status in patients with upper limb complex regional pain syndrome, even with persistent refractory symptoms.

Conclusion: The addition of the somatosensory rehabilitation of pain method and the graded motor imagery approach to conventional therapy could be considered in cases of complex regional pain syndrome that do not respond to conventional rehabilitation alone.

2.3 Introduction Complex regional pain syndrome (CRPS) incidence varies between 5 and 26 cases per 100,000 per year (Marinus et al., 2011), and is seen twice as often with the upper limb as with the lower limb (Sandroni, Benrud-Larson, McClelland, & Low, 2003). It is characterized by the presence of regional painful symptoms, seemingly disproportionate, associated with sensory, motor, sudomotor, vasomotor edema, and/or trophic signs (Harden et al., 2010). Those impairments can severely affect the function of the upper limb (Galer, Henderson, Perander, & Jensen, 2000). Evidence suggests that people with upper limb CRPS suffers longer than those with lower limb CRPS (de Mos et al., 2009). About two-thirds of patients with CRPS continue to show substantial limitations of their independence 1 year after the onset of symptoms (Bean, Johnson, & Kydd, 2014; Borchers & Gershwin, 2014; de Mos et al., 2009; Geertzen, de Brujin-Kofman, de Brujin, van de Wiel, & Dijkstra, 1998; Sharma, Agarwal, Broatch, & Raja, 2009). Patients with CRPS often have significant somatosensory symptoms (Gierthmuhlen et al., 2012; Rommel, Malin, Zenz, & Jänig, 2001). Among these symptoms, abnormal painful sensations such as hyperalgesia and allodynia, as well as skin sensibility disorders, are often the leading cause of complaints and decreased function (Huge et al., 2011; Savas, Baloglu, Ay, & Cerci, 2009). Hyperalgesia refers to increased pain due to a painful stimulus (Huge et al., 2011; Merskey & Bogduk, 1994), whereas allodynia is pain evoked by a normally painless stimulation (Lolignier, Eijkenkamp, & Wood, 2015). The term “allodynia” encompasses several forms, including thermal allodynia evoked by heat or cold and mechanical allodynia evoked by static or dynamic touching. Hyperalgesia and allodynia are seen twice as frequently in CRPS than in other pathological conditions affecting the upper limb, such as neuropathic conditions and discrete musculoskeletal

entities (e.g. osteoarthritis, rotator cuff disease, frozen shoulder, or healing fracture) (Mailis-Gagnon, Lakha, Allen, Deshpande, & Harden, 2014; Merskey et al., 1994). Allodynia has been reported in 74% of patients presenting with CRPS (Harden et al., 1999). The neurophysiological mechanisms responsible for painful symptoms in CRPS are not fully understood. Evidence suggests that in both types of CRPS, without (CRPS type I) and with nerve damage (CRPS type II), it is possible to observe neuropathic pain that is attributable to somatosensory impairments (Bruehl, 2010; Packham, MacDermid, Henry, & Bain, 2012; Treede et al., 2008). There is also evidence that these somatosensory impairments may contribute to the development of painful symptoms via peripheral and central sensitization mechanisms in CRPS (Goh, Chidambaram, & Ma, 2017; Smart, Wand, & O'Connell, 2016). For example, it is possible to observe reorganization in the primary somatosensory cortex (S1) that would be associated with sensitization mechanisms contributing to painful symptoms in these patients (Maihöfner et al., 2003).

Since peripheral and central sensitization mechanisms (Campero, Bostock, Baumann, & Ochoa, 2010) might contribute to pain chronicization in CRPS (Borchers & Gershwin, 2014), interventions that seek to regulate these mechanisms may be helpful in preventing such chronicization. The somatosensory rehabilitation of pain method (SRM) described by Spicher (2008) and graded motor imagery (GMI) (Moseley, 2004) are two innovative approaches that could potentially target these mechanisms. SRM uses peripheral somatosensory stimulation that can potentially act on peripheral sensitization mechanisms. Moreover, this method does not require active movement, which can be an interesting asset

in individuals in whom active mobilization can exacerbate symptoms. SRM consists of avoiding or reducing any cutaneous stimulation as much as possible in the skin area where touch evokes pain (i.e. allodynic area), while stimulating the somatosensory system at a distant site (with a soft fabric or light mechanical vibration in a comfortable territory proximal to the allodynic area). The SRM approach contrasts with that of the conventional desensitization approach, which promotes stimulation of the allodynic area with stimulations that are initially mild and then stronger as the person becomes accustomed to them and feels less pain (Yerxa et al., 1983). Evidence shows that SRM can reduce static mechanical allodynia (SMA) in patients with neuropathic pain (Nedelec et al., 2016; Packham et al., 2018; C. Spicher et al., 2008; Spicher et al., 2015). A retrospective case series on SRM showed a significant decrease in pain among burn survivors with SMA (Nedelec et al., 2016). Recently, a retrospective case series showed a reduction in the severity of SMA following SRM in patients with upper limb CRPS (Packham et al., 2018).

Conversely, GMI uses sensorimotor integration processes to reduce central sensitization and integrates the progression of active movements in its advanced stages. GMI is a hierarchical rehabilitation method in which patients must perform increasingly demanding tasks to create new neural connections targeted at normalizing the representation of the affected body part in the primary somatosensory cortex. GMI involves three stages of rehabilitation progression: (1) left/right discrimination, (2) explicit motor imagery, and (3) mirror therapy (G Lorimer Moseley, Butler, Beames, & Giles, 2012). Evidence shows that GMI alone can have beneficial effects in chronic pain conditions (Bowering et al., 2013) and CRPS (Daly & Bialocerkowski, 2009; Moseley, 2006; Walz,

Usichenko, Moseley, & Lotze, 2013). Therapies that preserve the integrity of the cortical somatosensory representation of body parts affected by CRPS may reduce pain symptoms in these patients (Moseley & Flor, 2012). The beneficial effects of GMI in CRPS could be explained by its ability to regulate cortical reorganization mechanisms involved in CRPS painful symptoms.

Although CRPS can be treated with medication and conventional physical therapy, these therapeutic approaches do not always reduce pain and improve function satisfactorily (Daly & Bialocerkowski, 2009). Currently, more evidence is needed for existing CRPS clinical guidelines (Daly & Bialocerkowski, 2009; Perez et al., 2010). There is still a need to develop new mechanism-based treatment approaches to achieve better results in the treatment of pain and somatosensory symptoms in CRPS (Bharwani et al., 2017). SRM and GMI are two different mechanism-based intervention approaches that may be potentially used in combination or as a complement to conventional rehabilitation (pain management modalities, active mobilizations, strengthening exercises, and task simulation) to treat CRPS. In the present case, this combination was used for a patient who did not respond to conventional rehabilitation alone.

2.4 Patient description Mr. B, a left-handed 39-year-old sub-Saharan African living in Quebec, Canada, was diagnosed in October 2012 with CRPS affecting his right upper limb. This condition resulted from a work-related accident that occurred in August 2012 while he was a plant production worker. During this accident, Mr. B sustained a right wrist injury involving ligament tears (triangular fibrocartilage complex and scapholunate ligaments). A few days after the injury and until December 2013, Mr. B received conventional treatments, including physical rehabilitation (conventional desensitization approach, contrast baths, passive mobilizations, active mobility, and strengthening exercises), prescribed medications (pregabalin and celecoxib), and pain management medical interventions (several stellate ganglion and venous blocks) without any subjective improvement. Due to the lack of improvement in his condition, he was referred by his plastic surgeon for occupational therapy at our private clinic to try a new rehabilitation approach.

2.4.1 Initial clinical examination

On his first visit to the occupational therapist in February 2014, Mr. B complained of intolerable pain in his entire right upper limb that was causing severe limitations and a fear of using his arm. He kept his hand held protectively against his trunk. On visual inspection, his affected hand looked waxy, swollen, and atrophied compared to his other hand. The pain was located on the dorsal side of his hand and thumb. It increased when he used his hand or when it was slightly touched (not able to tolerate any covering) or exposed to cold. He described his symptoms as follows: feeling of constant numbness, intermittent burning sensations, and shooting pain up to his right shoulder. With regard to hand function,

he reported being unable to move the entire arm from shoulder to fingers because of the pain. He could not use his right hand for any daily living activities. The only possible active limited motions were those of the index and thumb. He could use a pinch grasp with his index/thumb to hold light objects (eg, paper, fork, and so on) for no more than a few seconds because of the pain. He also complained of lack of strength and endurance in the affected hand. He was very emotional when he spoke about his accident or his condition. The mere act of talking about his arm was enough to trigger signs of emotional distress. Living with this pain was a great source of emotional burden to him. He felt unable to think of anything but this pain, unable to plan new projects. At the end of the first meeting with the occupational therapist, he reported being deeply discouraged, that he had no hope of healing to the point of wondering why he continued to seek treatment.

2.5 Methodology

2.5.1 General organization of patient's care

The patient was invited to participate in a tailored rehabilitation program which consisted of two components: (1) rehabilitation sessions supervised by an occupational therapist at the clinic (30 to 60 minutes in duration) and (2) home sessions managed by the patient. Rehabilitation sessions at the clinic and at home included the same interventions, that is, SRM and GMI combined with conventional treatments. Conventional treatments consisted of follow-up and advice on pain management modalities (i.e. medication, transcutaneous nerve stimulation (TENS), cryotherapy), active mobilizations, strengthening exercises, and task simulation. The patient had his own TENS and could

apply ice at home. The sessions at the clinic were held twice a week for the first 15 weeks and once a week thereafter for the rest of the program, which lasted 22 months. During the sessions at the clinic, all assessments and treatments were performed by the same two occupational therapists, alternately, depending on the availability of Mr. B and the therapists from one week to the next. One of the two therapists had SRM certification (56 hours of training in this method). Clinical evaluations included pain, SMA, tactile hypoesthesia, active range of motions (AROMs), hand muscle strength, upper limb function, and judgment of left-right discrimination assessments. Not all evaluations were performed at each session but periodically depending on condition evolution and the therapists' judgment. Only one or two assessments were done at each rehabilitation session to assess pain severity and prevent any flare-ups.

2.5.2 Clinical assessmentsPain intensity

Pain was assessed by the means of three evaluations: the Numeric Scale Pain Assessment (NSPA), the Visual Analog Scale (VAS), and the French version of the McGill Pain Questionnaire (*Questionnaire de la douleur de Saint-Antoine* [QDSA]). The NPSA measures pain intensity on a scale from 0 to 10, where 0 represents no pain at all, and 10 the worst possible pain (Farrar, Young, LaMoreaux, Werth, & Poole, 2001). The VAS evaluates pain intensity on a 10-cm scale, where 0 cm represents no pain at all, and 10 the worst possible pain (Scott & Huskisson, 1976). It is a valid tool for measuring pain at a specific point in time (Kersten, White, & Tennant, 2014).

The QDSA assesses pain by the patient's selection and rating of qualifiers that represent the sensory and emotional dimensions of pain (Boureau, Luu, & Doubrère, 1992). The QDSA has a maximum score of 64 points which includes 36 points for sensory and 28 points for affective qualifiers. When completing the QDSA, patients are asked to rate the minimum and maximum pain intensity they felt during the 24 hours preceding the assessment.

NSPA, VAS, and QDSA were used in this case because these assessments are complementary and allow for a thorough assessment and better understanding of the pain felt by the patient. The NSPA assessment is user-friendly for day-to-day assessment because it is short. The VAS was useful to help map the allodynic area by the allodynography technique which will be presented in the following section. As for the QDSA, it was used periodically to evaluate the evolution of CRPS pain phenomenon in its sensory and affective characteristics over several weeks.

Static mechanical allodynia Skin surface with SMA and severity of allodynia were assessed using the allodynography method and the Rainbow Pain Scale (RPS), respectively. The allodynography method consists of mapping the skin area that exhibits SMA. The borders of the allodynic cutaneous area are determined by the skin points where the static touch with a Semmes-Weinstein monofilament of 15 g (No. 5.18) causes a pain increase of 1 cm on the VAS scale from the baseline pain (i.e. pain level at rest assessed just before the evaluation) or where the monofilament causes a pain of at least

3 cm if the baseline pain was below 2 cm. For example, if the patient has a baseline pain level of 4 cm on the VAS, allodynography will look for the skin point where the 15 g monofilament causes pain at an intensity of 5 cm. However, if the patient has baseline pain of less than 2 cm (e.g. 0 cm), the allodynography will look for a skin point that produces pain that reaches at least 3 cm when the patient is touched with the 15 g monofilament. The mapping is made along the longitudinal line of the damaged cutaneous nerve branch, finding the most proximal and distal points (i.e. borders) of the allodynic area. The same procedure is applied in a transverse direction to find the most medial and lateral points of the allodynic surface. A polygon is then drawn by joining the 4 points with a line to give an approximate representation of the allodynic area (Spicher et al., 2015).

The RPS assesses the severity of SMA in the allodynic area mapped by allodynography at the skin point where the patient indicates feeling the worst touch-evoked pain. RPS uses 7 monofilaments of different sizes (see [Table 1](#), p.59) to determine the minimal application force required to increase the baseline pain by 1 cm on the VAS or to reach 3 cm if the baseline pain is below 2 cm. Each of these 7 monofilaments corresponds to a color, which is associated with an application force (g) and a number. The smaller the application force needed to evoke pain with the monofilament, the greater the SMA severity.

Tactile hypoesthesia Skin surface with tactile hypesthesia and severity of hypoesthesia were assessed by the esthesiography method and the static 2-point discrimination test, respectively. The esthesiography method consists of mapping the skin area with tactile hypoesthesia. On the dorsum of the hand, the borders of the hypoesthetic area are determined by the skin points where the patient does not detect the static touch with a Semmes-Weinstein monofilament of 0.4g (No. 3.61), which is the expected normal value for tactile sensibility on the dorsal surface of the hand (Bell-Krotoski, Fess, Figarola, & Hiltz, 1995; Spicher et al., 2015). The mapping is done in a longitudinal direction, finding the most proximal and distal points (i.e. borders) of the hypoesthetic area. The same procedure is applied in a transverse direction to find the most medial and lateral points of the hypoesthetic area. A polygon is then formed by joining the 4 points with a line to give an approximate representation of the hypoesthetic area (Spicher et al., 2015).

The static 2-point discrimination test uses a 2-point esthesiometer to vary the distance between 2 points of tactile stimulation. The test consists of evaluating the minimum distance between 2 points from which the person can distinguish between static touch with 1 or 2 points (Spicher, Hecker, Thommen, & Rouiller, 2005).

Active range of motion, strength, and function Hand and wrist active range of motion (AROM) were assessed by goniometry (Pendleton & Schultz-Krohn, 2013). Hand strength was assessed by grip strength, and by palmar and key pinch strength. Grip strength was

assessed with a standard, adjustable-handle JAMAR dynamometer (Mathiowetz et al., 1985). Palmar and key pinch strength was assessed with a B&L Engineering pinch gauge (Mathiowetz et al., 1985).

The patient's perception of his arm function was assessed by means of the Disability of the Arm, Shoulder and Hand Questionnaire (DASH), which assesses the patient's perception of his level of disability to use his upper limbs. The higher the score, the less functional the patient perceives himself (Angst, Schwyzer, Aeschlimann, Simmen, & Goldhahn, 2011; Dowrick, Gabbe, Williamson, & Cameron, 2005; Hudak, Amadio, & Bombardier, 1996). It has been demonstrated that the original DASH outcome measure has good construct validity, test-retest reliability, and responsiveness to change (Beaton et al., 2001). We used the Canadian French version of the DASH, which shows good acceptability and psychometric properties, comparable to those obtained with the original version (Durand, Vachon, Hong, & Loisel, 2005). The DASH score shows a high correlation with grip force ($r=0.47$), but the correlation is much weaker with range of motion ($r=0.24$) (Beaton et al., 2001; De Smet, 2007).

Finally, about every month and sometimes more often if needed, semi-structured interviews were used to assess the patient's perception of his level of functional independence.

All assessments, except RPS (i.e. severity of mechanical allodynia), were performed once a month, in order to avoid exacerbating the patient's painful symptoms. RPS assessment was carried out more often, that is, every week, to ensure adequate monitoring of allodynia, and to adjust SRM and GMI activities accordingly.

Tailored rehabilitation program The tailored rehabilitation program was designed to treat the patient by avoiding pain, respecting his tolerance and allowing gradual recovery of his upper limb active mobility and function. In order to achieve these objectives, the same rehabilitation program was performed at the clinic and at home: it combined conventional therapy with appropriate pain management, sensory and motor rehabilitation methods, as well as occupational therapy. As part of this program, the patient was required to go to therapy at the clinic once or twice a week for assessment of his condition, and also for the set-up, progression, and teaching of the daily exercise program (performed at the clinic and at home). Throughout the rehabilitation program, the patient continued to take the same medication as that used during the initial rehabilitation, before the tailored rehabilitation program (i.e. pregabalin and celecoxib). The program was reviewed weekly with the patient to optimize his functional activities and independence while minimizing evoked pain.

Table 2 (p.60) shows the sequence of assessment and treatment methods performed during the tailored rehabilitation program. The program started from the first visit on February 19, 2014 with the SRM described by Spicher et al. (2015), until the very

last session at the clinic in December 2015. In the presence of SMA, SRM involves an initial step of allodynia treatment and then a second step which is the treatment of the tactile hypoesthesia underlying allodynia (when SMA is resolved). SRM is initially aimed at reducing SMA in two ways: by encouraging the patient to adhere to the precaution of avoiding touching the allodynic area and by performing tactile stimulation away from the allodynic territory. More precisely, the tactile stimulation is performed at a distance on a proximal territory (working territory) with a comfortable light fabric or mechanical vibration, 8 times a day for 1 minute (or less long) without evoking pain. In this case, we used shaved beaver fur, with a progression in the location of the working territory, from proximal to distal. Since it was not possible to stimulate the right upper limb without pain, the right thoracic territory was first stimulated. The tactile stimulation at a distance from the allodynic area began on February 25, 2014, on the anterior branch of the 12th thoracic nerve until the following April 25, then progressed to the anterior branch of the third thoracic nerve. Once SMA was resolved, the underlying hypoesthesia was treated using the approach advocated in the second step of SRM. This step consists in using direct stimulation on the area with fabrics and light vibration in a progressive way. In the beginning of the approach, a tactile stimulation of 15-second duration is applied 12 times a day. This tactile stimulation is then gradually pursued by increasing its duration and decreasing its daily frequency in order to ultimately reach a stimulation of 5-minute duration applied 4 times a day.

The patient started the GMI method on February 25, 2014 ([Table 2](#), p.60). GMI involves three stages of rehabilitation progression: (1) left/right discrimination, (2) explicit

motor imagery, and (3) mirror therapy (Moseley et al., 2012). The protocol used was from the GMI Handbook (Moseley et al., 2012). This protocol describes the progression of the exercises according to the different stages of the methods and the normal outcomes expected at the end of each stage. The protocol does not give details as to exercises frequency or duration but rather recommends that these parameters be adjusted according to each patient's capability. Therefore, these parameters have been adjusted to minimize pain as much as possible, depending on the clinical reasoning and the information shared between the therapist and the patient. In order for him to do these activities at home, he received a package of 50 homemade pictures representing left and right hands in various positions. During the first stage of GMI, he was instructed to complete left/right discrimination sessions 4 to 6 times a day or more if he could tolerate it. During these sessions, he had to identify left/right laterality as fast as possible, without getting a conscious mental representation of the hand, as if he had to guess quickly. Each left/right discrimination session was timed with a digital stopwatch. The total duration of each session was divided by the number of pictures guessed to obtain the average time allocated to each picture. Average time by picture and percentage of correct answers were monitored until the patient reached the expected normal values for that activity, that is, a rate of correct answers of 80% or more, as well as an average time per image of 2.0 ± 0.5 seconds (Moseley et al., 2012). During the second stage of GMI, Mr. B was asked to close his eyes and visualize his right hand in a static position.

Promoting self-efficacy through patient education was an important goal that the therapists considered during the course of rehabilitation. Notably, upon initiation of SRM,

the therapists provided the patient with tips to avoid as much as possible touching the allodynic area. This teaching was intended to allow the patient to better understand the evolution of his condition and how to manage the challenge of respecting the precaution of not touching the allodynic area. Moreover, the therapists suggested solutions and adaptations that the patient could integrate daily, like pain control modalities or assistive devices such as the use of a nonslip surface to help him open jars. Thus, the patient was trained to use TENS, as well as cryotherapy (cold) and superficial heat as pain control modalities during the entire program. TENS and cryotherapy were performed on the ulnar nerve palmar branch because this nerve branch is in the vicinity of the allodynic area but not directly on the painful territory. The site of TENS stimulation on a neighboring nerve branch was chosen according to the literature (Bouhassira & Attal, 2012). Conventional TENS was applied at least once every day, or more if needed, for 20 minutes per session, as recommended (Bouhassira & Attal, 2012). Ice was applied many times a day, from 1 to 10 times if needed, for only 1-2 minutes, so it did not exacerbate the pain. Pain management education included instructions to take breaks regularly during his daily tasks to prevent pain exacerbation. Heated gloves were provided to the patient so that he could protect his hand from the cold during the winter period.

The second step of the SRM (treatment of the underlying hypoesthesia) also started on October 31, 2014 and ended in December 2015. Rehabilitation of tactile hypoesthesia was performed using direct stimulation of the area for reduced tactile sensation with soft fabrics and light mechanical vibration provided by a vibration generator. During the last few months of rehabilitation, the reduction of pain led to the introduction of strengthening

exercises and simulations of work-related tasks in preparation for his new job (**Table 2**, p.60). In addition, discussions were initiated with the patient regarding possible work station adaptations and adapted computer technology devices. Throughout the tailored rehabilitation program, the patient was instructed to stop exercise sessions momentarily when he experienced any increase of pain. The final assessment with Mr. B was performed on December 8, 2015 (**Table 2**, p.60).

2.6 Results

2.6.1 Pain outcomes

The NPSA score generally decreased during the tailored rehabilitation program. NPSA at rest decreased from 7 in February 2014 to between 0 and 3 in December 2015 (**Table 3**, p.61). From July 2014, Mr. B started reporting a consistent decrease in pain, except for certain weeks during which he experienced pain crises. The QDSA scores decreased steadily throughout the program, from 17 to 40 points in February 2014 to 8-14 points at the end of the program in December 2015 (**Table 4**, p.62). Fluctuations in pain severity associated with certain exacerbation crises were observed between June 2015 and December 2015. Because of these fluctuations, the level of pain at rest (NPSA at rest) and the pain sensory component of QDSA measured at the end of the follow-up in December 2015 were slightly higher than the measurements taken in June/August 2015 (**Table 3** and **Table 4**, pp.61 and 62).

With respect to symptoms of mechanical allodynia, the allodynography was initially impossible to draw because it spread throughout the right upper limb. For this reason, the

severity of allodynia was measured using the RPS, before allodynography could be performed in this patient. SMA severity at the beginning of the program was “red” on the RPS, which is the highest severity level on this scale (**Table 1**, p.59). In this patient, this means that a static application force of 0.03 g was enough to provoke an increase in baseline pain of 1 cm on the VAS scale (i.e. from 6 cm to 7 cm). SMA severity gradually decreased during the following months of rehabilitation. During the same period, the cutaneous area with SMA also decreased. Thus, it was possible to perform the first allodynography in this patient on April 1st, 2014 (**Figure 1**, p.63). This allodynography showed an allodynic skin surface of approximately 120 cm² and was associated with an SMA severity reaching the “violet” level on the RPS. The allodynic surface regressed in the first months of rehabilitation and disappeared on October 31, 2014. As of November 2014, mechanical allodynia disappeared in this patient (**Table 2**, p.60).

2.6.2 Hypoesthesia outcomes

Once allodynic symptoms were resolved, rehabilitation was able to target treatments to address tactile hypoesthesia. The resorption of the allodynic area made it possible to highlight the presence of an underlying hypoesthetic area. In December 2014, the esthesiography assessment demonstrated a surface of tactile hypoesthesia of 270 cm². This surface of tactile hypoesthesia decreased in the following months, completely disappearing in May 2015 (**Table 2**, p.60). In parallel, the patient also showed constant improvement in the static 2-point discrimination test. In January 2015, the first static 2-point discrimination assessment revealed a result of 21 mm (**Table 2**, p.60). This result was slightly higher than normal values. Two-point discrimination consistently improved in the following months to

reach 12 mm in September 2015, which corresponds to the expected normal value (Spicher et al., 2015; Von Prince & Butler, 1967).

2.6.3 Active range of motion, strength, and functional outcomes

It was initially impossible to measure AROM because of the pain felt by the patient and his reported fear of movement. During the first months of the rehabilitation program, the patient was unable to move his entire upper limb due to pain, except for light movement with his thumb and index finger. Because of these limitations in active mobility, it was considered appropriate not to introduce mobility or strengthening exercises at this stage. Goniometric measures commenced in November 2014, after the allodynia had resolved ([Table 2](#), p.60). The AROM at the wrist ([Figure 2A](#), p.64) and right thumb ([Figure 2B](#), p.64) showed a general pattern of improvement throughout the tailored rehabilitation program. Some wrist (ulnar deviation, supination) and thumb (metacarpophalangeal extension, interphalangeal extension, carpometacarpal abduction, metacarpocarpus extension) movements reached normal values during the program. For example, the wrist ulnar deviation improved from 15° to 40°, and the forearm supination improved from 35° to 90°. Other wrist (flexion, extension, radial deviation, pronation) and thumb (metacarpophalangeal flexion, interphalangeal flexion) movements remained below normal amplitudes. Those movements that failed to reach normal amplitudes were also those that showed fluctuations in amplitude losses corresponding to periods of symptom exacerbation during the course of rehabilitation.

Strength evaluations started in February 2015 because the improvement of the patient's condition made it possible to perform these assessments (**Table 2**, p.60). There was improvement in grip strength (**Figure 3A**, p.65) from 25 to 59 pounds and improvement in key pinch and palmar grip strength (**Figure 3B**, p.65), ranging from 5 to about 17 pounds for key pinch in the affected hand. These outcomes remained below the expected normal values for this patient at the end of the rehabilitation program. On October 31, 2014, when allodynia was completely resolved, Mr. B reported that he could move his upper right limb in a limited way, without evoking pain. He was instructed to perform light exercises and to start using his limb to perform light tasks at home (e.g. putting forks and knives on the table) (**Table 2**, p.60).

2.6.4 Motor imagery outcomes

The patient reached normal values for the first stage of GMI (i.e. 80% correct answers with an average time per image of 2.0 ± 0.5 seconds on the left/right discrimination test) on March 7, 2014. Therefore, on this date, the second stage of GMI (i.e. explicit imagery) could be started (**Table 2**, p.60). At the beginning of the second stage, he was unable to imagine his right hand without increasing pain in his right upper limb. Explicit imagery of the left hand showed that he was able to tolerate the visualization of the left hand in static positions without an increase in pain. When the patient was able to tolerate this level of visualization (left hand static positions), he was asked to imagine his left hand in progressively more complex situations. The explicit imagery of the right hand was painful throughout this stage. The third stage of GMI, that is, mirror therapy, was tried, but it resulted in pain exacerbation in the right limb as soon as he saw the reflection of his left

limb in the mirror. GMI was stopped on September 1, 2014 after Mr. B said that he did not want to pursue this method (**Table 2**, p.60).

2.6.5 Functional outcomes

DASH scores of 39% and 30.5% were obtained in July and August 2015, respectively. These results suggest a gradual improvement in function of the affected upper limb. Moreover, the semi-structured interviews provided a good idea of the functional evolution of this patient. DASH results were corroborated by the improvement in function described by the patient. At the initial assessment, he reported using his left hand most of the time and for the majority of his activities. Moreover, he needed to take frequent breaks and a lot of time to complete functional tasks. Using the nonaffected limb could increase the pain in the affected limb. He reported being independent in terms of personal care activities, and being partially independent for domestic activities, performing only light tasks (e.g. dusting). The most difficult task was doing the dishes. With respect to his hobbies, he had difficulty with reading and writing because of the pain and reported decreased attention and concentration due to medication. In October 2014, the patient started to be able to use his right arm more during personal care tasks. For example, he reported being able to hold his jacket with his right upper limb for a short period of time and being able to tolerate wearing his heated gloves. At that time, Mr. B told us he was satisfied with the improvements in his right hand. In November 2014, Mr. B attended a university course in which he had to use a computer keyboard but continued to type only with his left hand. Then, from March 2015, he reported that he was satisfied about starting using his right hand in domestic chores and could

perform physical activities (e.g. at the park) with his children. However, he confirmed that his current functional level was still below the capabilities he had before the accident. At the final assessment, Mr. B reported continuing increases in the use of his right hand, almost like before the accident, except for washing his back. He still needed help from his wife for this task and did not want any technical aid to compensate.

2.6.6 Participation outcomes

During the summer of 2015, the patient started returning to his leisure activities. He said he was proud of being able to get back to canoeing. He also reported that he did not want to return to his old job because of the bad memories related to the events that occurred there and also because his previous job did not allow him to optimally perform his duties with his new level of function. He participated in vocational retraining for an office job, using a computer most of the time. We offered him adapted devices, but he declined because he wanted to feel like everyone else.

2.7 Discussion

Even though desensitization and other conventional therapies can be effective for treating hyperalgesia and mechanical allodynia in CRPS patients (Pleger et al., , 2005; Rho, Brewer, Lamer, & Wilson, 2002), some patients with neuropathic pain and somatosensory impairments may feel an escalation of pain and those approaches may then become ineffective (Sebastin, 2011). In these cases, other alternative approaches must be considered in the hope of getting better results. For example, evidence suggests that if conventional

therapies do not work alone, one should consider combining physical rehabilitation with pain management modalities and psychological intervention for managing painful conditions (Stanton-Hicks et al., 2002).

In this case report, conventional rehabilitation did not improve persistent trophic symptoms, persistent pain, and SMA, which resulted in a fear of using the affected limb and continuing therapy. With the consent of the patient, new approaches were undertaken. A tailored rehabilitation program began with modalities aimed at reducing the area of pain and pain intensity. It was important to provide patient education and promote patient self-efficacy in order to facilitate his adherence to treatment. The clinical reasoning underlying these choices took many features of the clinical case into consideration: pain intensity, total skin area with touch-evoked pain, pain phenotype, and duration of symptoms since the initial accident. The pain phenotype involved mechanical allodynia, which affected the entire upper limb. From the beginning of the rehabilitation program, the priority was to treat the allodynia using the SRM method, a choice that may be explained by the fact that the severity of allodynia limited the patient's ability to participate in other types of intervention (e.g. passive mobilizations), as well as resume functional use of the limb.

The GMI method was quickly incorporated into the tailored rehabilitation program because its goals were compatible and complementary to those of SRM. Pain management through SRM targets the somatosensory system directly at a peripheral site, whereas GMI

targets the representation of the affected body part in the cortex. Application of the GMI method matches well with SRM, with the precautions being similar in both methods (to not increase pain during treatment). This is an important consideration, as decreased effectiveness of GMI has been reported when used in combination with traditional management, which could lead to conflicting messages regarding pain management during treatment (Johnson et al., 2012).

Another priority was to educate the patient about CRPS pathophysiological mechanisms and allodynia and pain management strategies. Once the pain intensity was reduced and allodynia completely resolved, conventional interventions could be progressively reintroduced, such as active mobility and strengthening exercises, while continuing SRM for managing hypoesthesia and pain to prevent any flare-ups. SRM was continued to improve sensorimotor function by normalizing hypoesthesia. Evidence suggests that under the area of mechanical allodynia lies a tactile hypoesthetic area (Spicher et al., 2008). This combination and sequence of treatments helped Mr. B reduce his pain and allodynia, normalize his tactile sensibility, and improve his AROM and strength. Finally, the tailored rehabilitation program allowed this patient to return to his daily living, productivity and leisure activities.

In this case, it is difficult to distinguish between the effects attributable to SRM and those of GMI since these two approaches were introduced at the same time. The SRM itself may have helped to improve this patient's painful symptoms and this reduction in pain may

in turn have promoted active movement. Such an effect could result from the influence of SRM peripheral stimulation on the sensitization mechanisms involved in CRPS. GMI could also contribute to the improvement of the patient's condition. It is possible that GMI influenced the central sensitization mechanisms involved in CRPS. It can be hypothesized that the fact that the patient was unable to perform the last stage of GMI suggests that the central sensitization mechanisms would still be effective and contribute to the residual symptoms that were still observed at the end of the program. This combination of conventional rehabilitation, SRM, and GMI showed a reduction in allodynia and pain intensity. Also, it must be emphasized that SRM and GMI are therapies that require strict patient adherence and activities performed outside of the sessions supervised by therapists. In this case, the patient was very attentive which must have contributed to the success of the therapy. He followed the instructions closely, especially for SRM, which made him an ideal candidate for this method. The therapist's involvement in motivating the patient to participate in the treatment and exercises is also important. The specific combination of SRM and traditional pain management modalities used concordantly with SRM principles (i.e. stimulations not applied directly to the area of allodynia) may be an appropriate option for persons with persistent long-standing CRPS with allodynia. This combination and the sequence of interventions could also be applied earlier in the rehabilitation process, if patients with mechanical allodynia that could benefit from this approach can be identified. Other important considerations may be the underlying causes for limitations of movement. If a pathology threatens to produce permanent loss of movement and sequelae on mobility, one should consider not beginning the tailored program with SRM but using this method later in the rehabilitation process (e.g. CRPS after flex or tendon repairs and/or nerve repairs

at the wrist). In addition, care should be taken to avoid touching the allodynic area during SRM so the pain does not result in a decrease in active patient mobilization, and possibly a decrease in function due to underuse. The instruction to avoid stimulating the painful area does not mean that the patient must not move or even limit his active mobility. In each CRPS case, it is important to carefully investigate all potential problems and prioritize them in order to select the most appropriate interventions in a tailored rehabilitation program. However, there is a significant proportion of persons with CRPS whose pain is the primary limitation for active movements, and these persons are potential candidates for SRM.

As for the limitations of this case report, it would have been interesting to have more objective measures of the level of autonomy and function/disability of the upper limb. DASH assessments could have been performed more often during rehabilitation, but due to time constraints, pain assessments were prioritized. Introducing DASH assessment earlier in rehabilitation would have reported functional improvements during the program more objectively. Another limitation of the study is the fact that the patient did not have a logbook in which to record all the interventions he used. Moreover, keeping a logbook would have provided better documentation of all the interventions performed at home and the patient's level of adherence to the therapy. The fact that this tailored rehabilitation program may not be readily implemented in all clinical settings could also be considered a limitation. The SRM as it was used in this case study is not accessible in all clinical settings since one of the therapists was specifically trained to apply this method. In fact, although any therapist can apply SRM, certification assures that the clinician has completed the training and can

apply the method in accordance with the published protocol. It is not clear that the method would be as effective if applied by untrained therapists.

2.8 ConclusionThis case report shows the successful use of a tailored rehabilitation program based on the combination of SRM and GMI methods for treating a patient with persistent CRPS with allodynia in the upper limb. Therapists have a choice among several interventions that can help patients with CRPS. However, the challenge is to be able to choose the most appropriate interventions, in the right order, based on the patient's condition and personal priorities. “There is critical lack of high-quality evidence for the effectiveness of most therapies for CRPS, and there is a need for larger trials in order to formulate an evidence-based approach for these patients” (O'Connell, Wand, McAuley, Marston, & Moseley, 2013). SRM appears to be a promising method for initiating the treatment of patients with CRPS who have responded poorly to conventional therapies. In addition, GMI appears to be a complementary method that can help patients with recalcitrant CRPS to reduce pain and progress in their active mobility so that they may be able to participate in strengthening exercises. A tailored rehabilitation program using a sequence of methods starting with SRM and followed by GMI appears to be a promising approach in persistent CRPS with significant functional limitations. Further studies will be needed to corroborate this type of approach and to better understand the neurophysiological mechanisms involved.

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Table 1 : Characteristics of the Semmes-Weinstein monofilaments used to assess static mechanical allodynia (SMA) severity using the Rainbow pain scale assessment

Nominal value = \log_{10} [force (mg) $\times 10$].	Force (in g)	Severity on the Rainbow pain scale (color)
2.44	0.03	Red
3.22	0.2	Orange
3.84	0.7	Yellow
4.17	1.5	Green
4.56	3.6	Blue
4.93	8.7	Indigo
5.18	15.0	Violet

Table 2 : Sequence of assessments and treatments performed during the tailored rehabilitation program.

Epochs														
2014 2015														
Assessments	Feb.	Mar.	Sep.	Oct.	Nov.	Dec.	Jan.	Feb.	May.	Jun.	Sep.	Nov.	Dec.	
	Allodynia severity (Rainbow scale)	Red (Start)	Red	Indigo	Violet	No more SMA (Stop)	N.A.	N.A.	N.A.	N.A.	N.A.	N.A.	N.A.	N.A.
Aesthesiography (surface in cm ²)	N.A.	N.A.	N.A.	N.A.	N.A.	270 (Start)	84	23	0 (Stop)	N.A.	N.A.	N.A.	N.A.	N.A.
Static two-point discrimination threshold (mm)	N.A.	N.A.	N.A.	N.A.	N.E.	N.E. (Start)	21	30	N.E. (Stop)	21	12	N.E.	N.E.	N.E.
AROM	N.E.	N.E.	N.E.	N.E.	At wrist (Start)	At thumb (Start)	E	E	E	E	E	At wrist/thumb (Stop)	N.E.	N.E.
Strength	N.E.	N.E.	N.E.	N.E.	N.E.	N.E. (Start)	N.E.	N.E.	Grip (Start)	Palmar/Key pinches (Start)	E	E	E	Grip/ Palmar/ Key pinches (Stop)
Treatments	SRM/ GMI stage1 (Start)	GMI stage2 (Start)	GMI (Stop)	T	AROM exs. (Start)	T	T	T	T	Strgth exs (Start)	T	T	T	Task sim. (Start)

Legend: AROM = Active range of motion; AROM exs. = Active range of motion exercises; Strgth exs. = Strength exercises; Task sim. = Task simulations; Start = Beginning of assessment or treatment; Stop= End of assessment or treatment; N.A. = Not applicable; N.E. = Not evaluated; E: Ongoing assessment; SRM = Somatosensory rehabilitation method; GMI = Graded motor imagery; T: Ongoing treatments;

Table 3 : Numeric pain scale scores on the right hand during the tailored rehabilitation program (minimal-maximal pain scores in the 24 hours preceding the assessment)

	Month (year)				
	February (2014)	November (2014)	February (2015)	June (2015)	December (2015)
At rest	6 - 7	5 - 6	4 - 5	0 - 0	0 - 3
Active	N.E.	N.E.	N.E.	4 - 4.5	2.5 - 3
Legend: N.E.= Not evaluated					

Table 4 : QDSA pain intensity scores on sensory, affective domains and total on the right hand during the tailored rehabilitation program (minimal-maximal pain scores in the 24 hours preceding the assessment)

	Date				
	February 21 st 2014	July 25 th 2014	February 06 th 2015	August 4 th 2015	December 8th 2015
Sensory (/36)	9 - 23	7 - 27	4 - 19	0 - 11	7 - 14
Affective (/28)	8 - 17	5 - 15	0 - 2	0 - 5	0 - 0
Total (/64)	17 - 40	12 - 42	4 - 21	0 - 16	8 - 14

Figure 1 : Allodynography on the right hand performed on April 1st, 2014, showing the skin area innervated by the superficial branch of radial nerve on which static mechanical allodynia (SMA) was found.

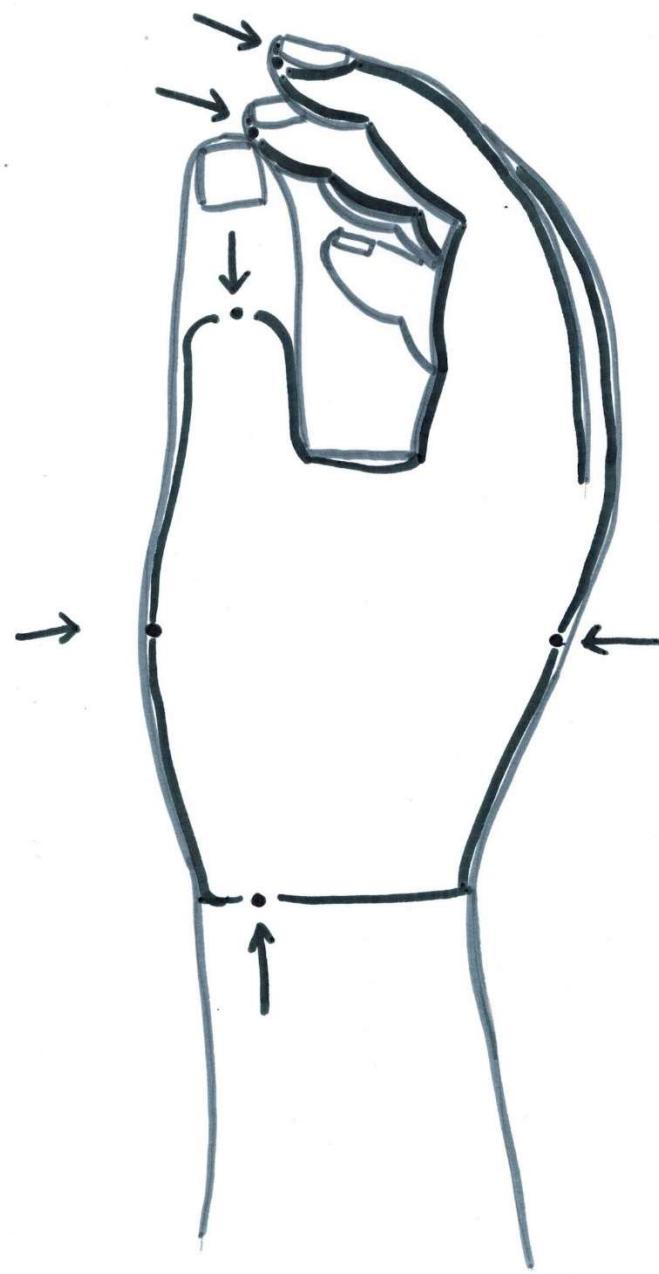


Figure 2 : Active range of motion (AROM) of the right forearm wrist (A) and thumb (B) of the subject were measured at four different epochs during the tailored rehabilitation program. AROMs at the thumb are presented for flexion and extension of metacarpophalangeal (MP) and interphalangeal (IP) joints (MP); abduction and extension of carpometacarpal (CMC) joint.

Normal active range of motions are indicated in parentheses.

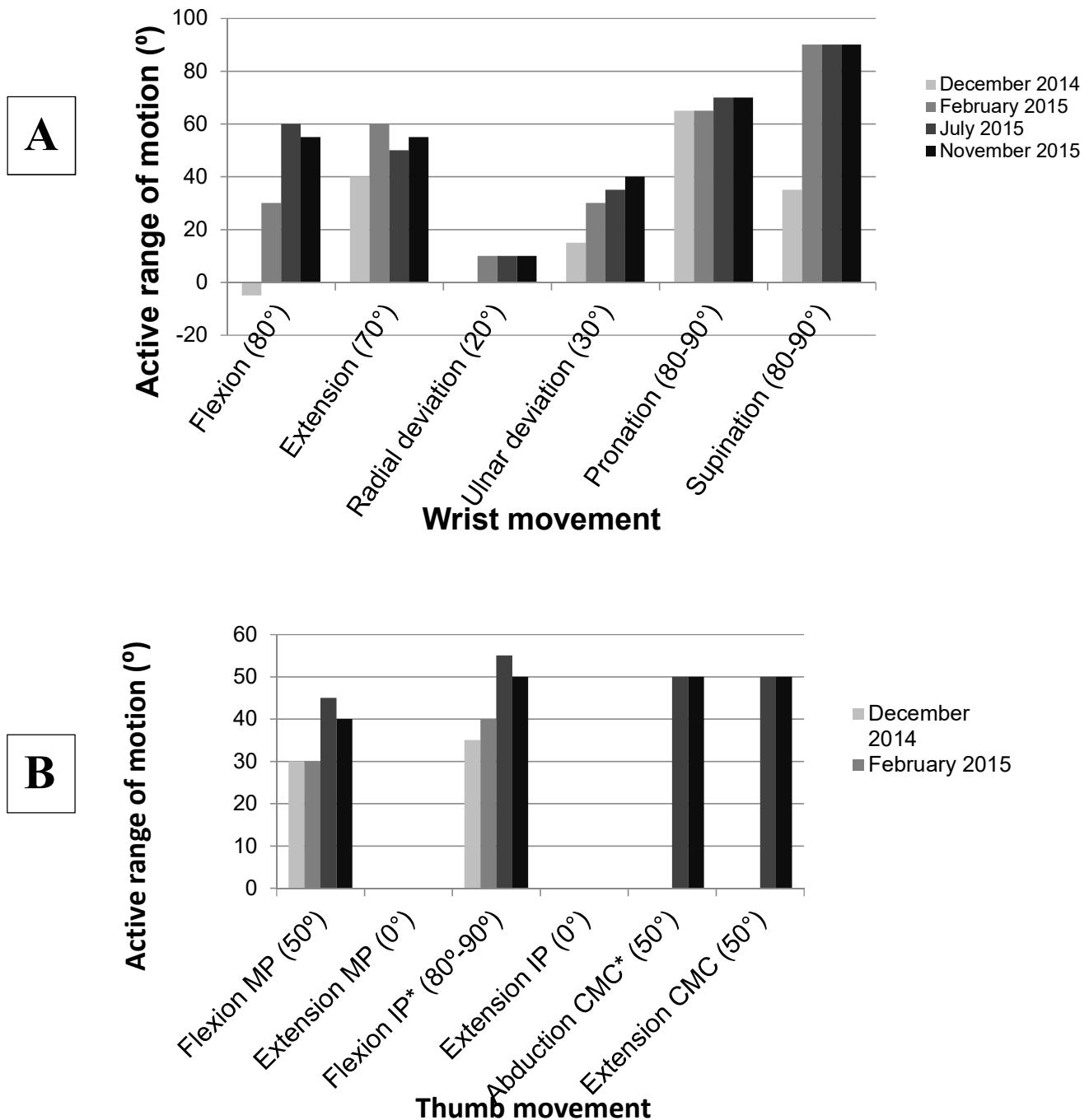
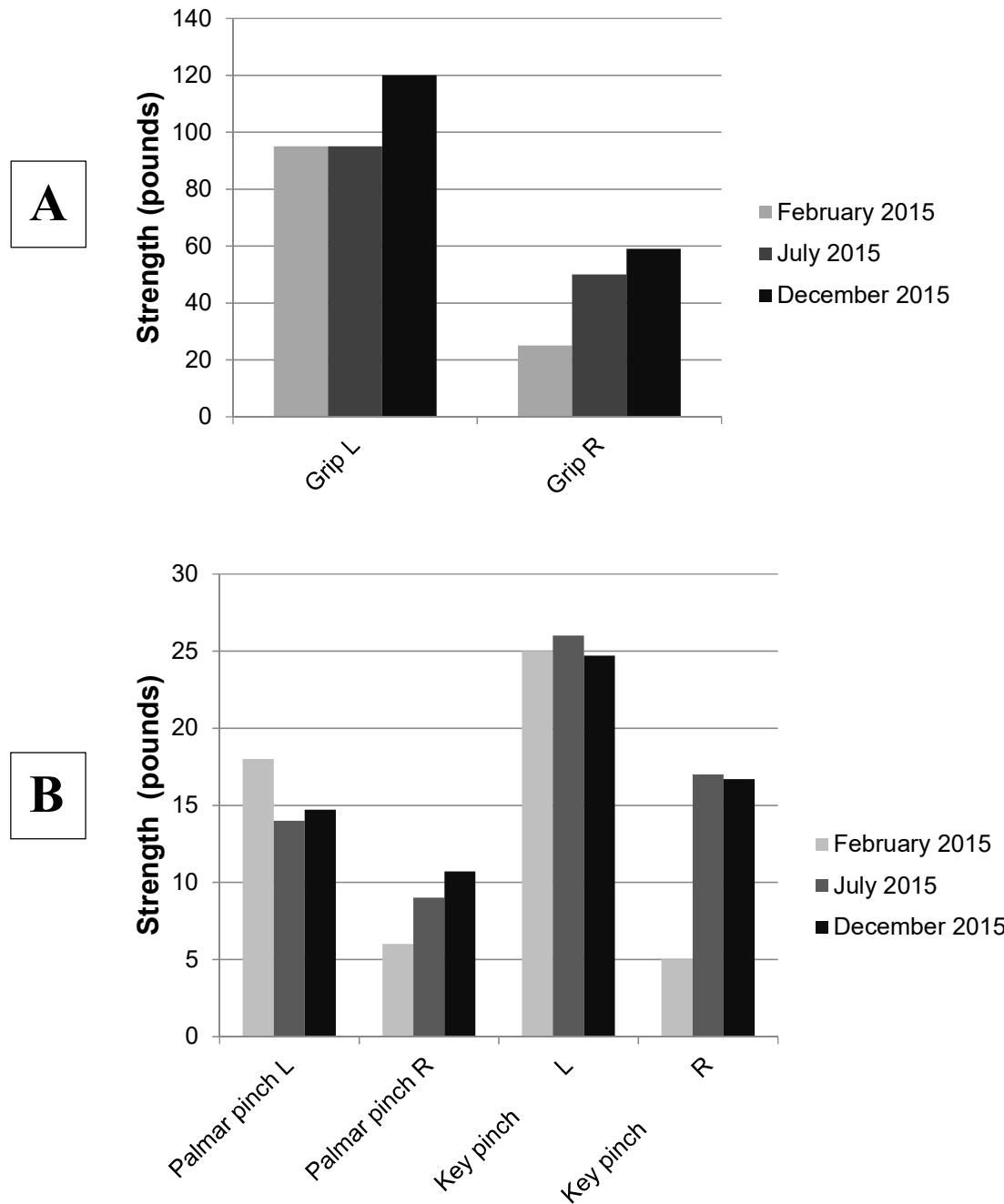


Figure 3 : Grip (A) and pinch (B) strengths (in pounds) on the left (L) and right (R) sides were measured at three different epochs during the treatment tailored rehabilitation program.

Normal values of grip strength for an individual of the same age and gender than the patient in this case report are approximately 115 pounds for grip and 26 pounds for palmar and key pinches, respectively.



Chapitre 3 : manuscrit, Article #2

Le chapitre 3 vise à répondre au deuxième objectif de ce mémoire : recenser les approches de stimulation tactile étudiées dans la littérature et leurs niveaux d'évidences en lien avec le traitement de la douleur chez des personnes avec AM à la main.

3.1. Tactile stimulation programs in patients with hand dysesthesia following a peripheral nerve injury: a systematic review

Les résultats de cette recension systématique sous forme d'article ont été soumis au *Journal of Hand Therapy* le 14 octobre 2019, a accepté avec révision le 31 décembre 2019, puis est actuellement en révision auprès de l'éditeur depuis le 20 mars 2020 (Quintal, Carrier, Packham, Bourbonnais & Dyer, en révision, 2020). La confirmation de soumission de cet article se trouve en « Annexe 1 » de ce mémoire.

3.1.1 Apport de l'étudiante et de chacun des co-auteurs

Isabelle Quintal a été la principale responsable de chacune des étapes menant à la publication de ce manuscrit, c'est-à-dire : l'élaboration du devis, la collecte de données, l'extraction des données, l'analyse de données, la rédaction du manuscrit et la soumission au journal.

Alexis Carrier a participé à la collecte de données, à l'extraction et l'analyse des données ainsi qu'à la rédaction du manuscrit.

Tara Packham a participé à l'analyse des données et à la rédaction du manuscrit.

Daniel Bourbonnais et Joseph-Omer Dyer ont dirigé la principale responsable de ce manuscrit, Mme Quintal, dans l'ensemble des étapes menant à la soumission.

Tactile stimulation programs in patients with hand dysesthesia following a peripheral nerve injury: a systematic review

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3.2 Abstract*Background:* Nerve injuries affecting the hand can lead to neuropathic pain including spontaneous and touch-evoked unpleasant abnormal sensations (dysesthesia). Diverse approaches based on tactile stimulation are often used to treat touch-evoked dysesthesias in hand rehabilitation. There is a lack of literature synthesis on the various approaches based on tactile stimulation that can be used for treating hand dysesthesia following nerve injury.

Purpose: The objective of the study was to summarize the current evidence on tactile stimulation programs for managing touch-evoked hand dysesthesia due to peripheral nerve injury.

Study Design: Systematic review performed according to PRISMA standards.

Methods: The search was carried out on Medline, Embase, CINAHL and the Cochrane Library databases. The selected studies had to present patients with touch-evoked dysesthesia following nerve injury who were treated with tactile stimulation approaches to reduce pain. Descriptive data including patient characteristics, interventions, and outcome measures extracted from the included studies. The MINORS scale assessed the methodological quality and the risks of bias for each included study.

Results and conclusions: Seven studies met the inclusion criteria. These studies present tactile stimulation interventions that are heterogeneous, among others, for populations and stimulation modalities. Painful symptoms appear to diminish in patients with touch-evoked hand dysesthesia whatever the tactile stimulation program is used. However, the included studies present significant risks of bias that limit the confidence in these results. Therefore, it is not possible to rule on the beneficial effects of tactile stimulation to treat touch-evoked hand

dysesthesia. Future studies with more rigorous methodological designs are required to verify the potential benefits of these approaches.

3.3 Introduction Nerve injuries, including neuropathies, are among the top ten diagnoses seen in the field of hand therapy (Keller et al., 2016). Focal neuropathies are impairments that affect a single peripheral nerve. They most often result from a mechanical injury such as compression or trauma (Sunderland, 1951). The incidence of focal neuropathies following trauma is estimated at 5% (Robinson, 2000), with 80% of these neuropathies affecting the upper limb (Noble et al., 1998). Given the higher incidence of upper limb focal neuropathies, these conditions are frequently encountered in hand rehabilitation.

The signs and symptoms of neuropathies often include sensory loss, motor weakness, autonomic dysregulation, and neuropathic pain (NP). This later is defined as “pain caused by a lesion or disease of the somatosensory nervous system” (IASP, 2012). Neuropathic pain from a peripheral nerve injury may affect functional performance and quality of life. Several studies have found a correlation between neuropathic pain intensity and decreased function in patients with peripheral nerve injuries (Novak, Anastakis, Beaton, Mackinnon, & Katz, 2010; Novak & Katz, 2010; Rosén & Lundborg, 2000). NP includes dysesthesia, which is defined as “an unpleasant abnormal sensation, whether spontaneous or evoked” (IASP, 2012). Allodynia can be a special case of dysesthesia where pain is evoked by normally painless stimuli (IASP, 2012). In the case of allodynia, pain may be evoked by different types of stimuli, such as cold and heat (thermal allodynia) or a light touch on the skin (mechanical allodynia). When mechanical allodynia is present in the hand, it can particularly affect function by causing pain during tasks

that require the sense of touch. This underscores the need to treat hand dysesthesia in patients with peripheral nerve injuries (PNI).

Although a systematic review has already been done on tactile stimulation programs for hand hypoesthesia following PNI (Miller et al., 2012), there is currently no literature synthesis describing such programs and their effects on hand dysesthesia. A variety of methods based on tactile stimulation are used to treat dysesthesia. These methods often include programs such as the desensitization method (Abrams, 2018; Jerosch-Herold, 2011; Lewis et al., 2011; Yerxa et al., 1983) and the Somatosensory Rehabilitation Method (SRM), which have been used to treat dysesthesia in different parts of the body (Spicher & Quintal, 2013). The use of these programs in hand dysesthesia may pose challenges. Program outcomes may differ from those obtained with other body parts because of the specific functional and somatosensory characteristics of the hand. The purpose of this systematic review is to summarize the current evidence on tactile stimulation programs for managing hand dysesthesia due to PNI. This review may prove useful to hand therapists in determining which tactile stimulation programs are most appropriate for treating patients with hand dysesthesia arising from PNI.

3.4 Method This systematic review was planned, performed, and reported based on the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) statement. The protocol for this systematic review is registered in the PROSPERO database under file number #78685765 (https://www.crd.york.ac.uk/PROSPERO/display_record.php?RecordID=122479).

3.4.1 Eligibility criteria All articles in French or English on studies that included PNI or NP conditions, hand dysesthesia, tactile stimulation programs and pain outcomes were included in the review. A tactile stimulation program was defined as a rehabilitation modality using mechanical stimulation (e.g., touch or vibration) on the skin, applied by a healthcare professional and/or taught to a client as part of a home program. Exclusion criteria were as follows: a) examination of only cold or heat dysesthesia, b) polyneuropathy or phantom limb pain as a primary diagnosis, c) dysesthesia secondary to chemotherapy, and d) studies on healthy subjects, newborns or animals. These other populations were excluded as their healing mechanisms are probably different from PNI. Commentaries, narrative reviews, clinical practical guidelines and conference abstracts were excluded. Foreign language studies were excluded due to the cost and time involved with translation.

3.4.2 Sources of information and search strategyA systematic review was conducted up to June 23, 2019 with consultation from an information specialist using the following databases: Medline, Embase, Cumulative Index to Nursing and Allied Health Literature (CINAHL) and the Cochrane Library. These databases were selected because of the volume and variety of healthcare articles indexed. The keywords used to conduct the search included: dysesthesia, allodynia, paresthesia, hyperesthesia, hyperalgesia, hyperesthetic, sensory, cutaneous, skin, tactile, superficial, somatosensory, touch, vibration, stimulation, reeducation, re-education and rehabilitation. Various combinations of these keywords were entered into each database in sequential order to achieve optimal results. For example, the Medline search was as follows: (exp Paresthesia/ OR exp Hyperesthesia OR exp Hyperalgesia OR (dysesthesi* OR hyperesthesi* OR allodyn* OR paresthesi* OR hyperestheti* OR hyperalg*).ab,kf,kw,ti.) AND (((cutane* OR skin OR tactile* OR superficial OR sensory OR somatosensory OR vibrat* OR touch*) ADJ2 (stimulat* OR reeducat* OR rehabilitat* OR re-educat*).ab,kf,kw,ti.) OR ("desensiti*".ab,kf,kw,ti.)). A manual database search was also done using reference lists of relevant scientific articles and reviews of additional articles.

3.4.3 Selection of studies The initial list of references was imported into Endnote, a bibliographic management software program. Once duplicate references were removed, two reviewers (IQ, AC) selected relevant references. All titles of references were screened and reviewed for eligibility. For references whose titles seemed relevant, the abstracts were checked to further validate the eligibility criteria. Copies of full-text articles whose abstracts seemed to meet the eligibility criteria were obtained and read in their entirety for initial selection, and then for an in-depth review, quality assessment, and final selection. Discrepancies were either discussed until a consensus was reached or resolved by a third independent reviewer (JOD).

Data extraction process Two independent reviewers (IQ, AC) used a data extraction form to summarize and interpret key aspects identified during the review of the selected studies. These included study design, patient characteristics (injury or health condition, duration of symptoms prior to intervention, PNI or NP), sample size, intervention characteristics, outcome measures, follow-up time, outcomes (quantitative and qualitative) and study quality. Outcomes related to pain intensity, allodynia surface, allodynia severity and cases where allodynia was resolved were extracted. This information was tabulated using an Excel spreadsheet. When articles addressed heterogeneous sites of dysesthesia, hand dysesthesia information was extracted separately whenever possible. Authors were contacted in order to retrieve additional information on the hand dysesthesia subgroup. Data extraction discrepancies between the two reviewers were either discussed until a consensus was reached or resolved by a third independent reviewer (JOD).

Summary measures of quantitative data including measures of central tendency and variability, as well as mean differences and effect size were extracted whenever possible. Measures were considered statistically significant if the 95% confidence interval did not include zero (0) and/or the p-value was ≤ 0.05 .

Quality and risk of bias assessment. Study quality was assessed using the Methodological Index for Non-Randomized Studies (MINORS). This index has good internal consistency ($\alpha=0.73$), moderate-to-good inter-reviewer agreement ($k=0.61-1.00$) and test-retest reliability ($k=0.59-1.00$) (Slim et al., 2003). MINORS is a 12-item standardized tool used to determine the methodological quality of non-randomized studies. Each item of this tool is given a score of 0 (“not reported”), 1 (“reported but inadequate”), or 2 (“reported and adequate”). The items examine the following: the aim of the study, inclusion of patients, prospective collection of data, appropriate and unbiased endpoints of the study, follow-up period, loss to follow-up, and prospective calculation of the sample size. The scores are added together, with maximum total scores reaching 16 or 24 for non-comparative and comparative studies respectively. Blind scoring was conducted independently by both reviewers (IQ, AC). Discrepancies were either discussed until a consensus was reached or resolved by a third independent reviewer (JOD).

Risk of bias was assessed by one reviewer (IQ) and re-evaluated by two reviewers (DB, JOD). This was done for each of the seven studies in accordance with the five principal types of bias (Shuster, 2011): selection, performance, attrition, detection and reporting. Those risks of bias were classified as low, moderate or high.

3.5. Results The search strategy identified 1388 studies, including 370 in Embase, 453 in Medline, 72 in CINAHL, and 0 studies in Cochrane. Once the duplicates were removed and the titles and abstracts were screened, 38 potential studies remained. **Figure 14**(p.89) shows the flow diagram for the search results. Seven studies (Bellugou, Allieu, de Godebout, Thaury, & Ster, 1991; Göransson & Cederlund, 2011; Love-Jones et al., 2009; Nedelec et al., 2016; Packham et al., 2018; Spicher, Mathis, Degrange, Freund, & Rouiller, 2008; Wider et al., 2006) were retained for this systematic review based on the inclusion and exclusion criteria. One of the selected studies was a prospective case-series study (Göransson & Cederlund, 2011). Three studies were retrospective case-series studies (Nedelec et al., 2016; Packham et al., 2018; Spicher et al., 2008), one study was a case report study (Wider et al., 2006), one was an experimental study (Love-Jones et al., 2009), and one was a proof-of-concept study (Bellugou et al., 1991). Therefore, the final seven studies included in this systematic review were all uncontrolled studies.

3.5.1 Study characteristics

Key study characteristics are summarized in **Table 15**(p.90). The studies were classified in chronological order by type of intervention and within the same intervention. Three studies examined the use of a tactile desensitization program (Bellugou et al., 1991; Göransson & Cederlund, 2011; Love-Jones et al., 2009), three assessed the use of the Somatosensory Rehabilitation Method (SRM) (Nedelec et al., 2016; Packham et al., 2018; Spicher et al., 2008), and one examined the use of a glove and tactile stimulation (Wider et al., 2006). Each study looked at different populations: hand injuries (Bellugou et al., 1991), hand and upper limb injuries (Göransson & Cederlund, 2011), NP patients with different aetiologies (n = 2) (Love-Jones et al., 2009; Spicher et al., 2008), burn survivors (Nedelec et al., 2016), upper-limb complex regional pain syndrome (Packham et al., 2018), and painful hand and moving fingers (Wider et al., 2006). Two studies (Bellugou et al., 1991; Wider et al., 2006) included only hand conditions. PNIs were diagnosed in three studies (Göransson & Cederlund, 2011; Love-Jones et al., 2009; Wider et al., 2006) and strongly inferred (rather than diagnosed) in four studies (Bellugou et al., 1991; Nedelec et al., 2016; Packham et al., 2018; Spicher et al., 2008). NP was mentioned (but not diagnosed) in four studies (Love-Jones et al., 2009; Nedelec et al., 2016; Packham et al., 2018; Spicher et al., 2008). The study characteristics, including type of population and intervention, were mostly heterogeneous across all studies selected.

3.5.2 Intervention characteristics The specific characteristics of the interventions used in each study are detailed in **Table 26**(p.93). All three desensitization studies (Bellugou et al., 1991; Göransson & Cederlund, 2011; Love-Jones et al., 2009) described the interventions and the adjustments made to the parameters in detail. The various parameters (material, frequency, duration, stimulation territory) of each of these three studies were well explained. Two studies (Bellugou et al., 1991; Göransson & Cederlund, 2011) recommended adjusting the parameters to the patients' symptoms (i.e., no pain increase or as tolerated) while the other study (Love-Jones et al., 2009) suggested that adjustments should match the maximum pain tolerated by patients. Clinical decision criteria for ending the treatment was not described in any study. Overall, the studies that addressed desensitization described heterogeneous interventions.

In terms of SRM studies, all three articles (Nedelec et al., 2016; Packham et al., 2018; Spicher et al., 2008) provided detailed descriptions of a standardized intervention procedure, with nearly the same parameters for material, frequency and duration of stimulation. However, the stimulation territory was determined based on the allodynic area being assessed. All three studies recommended not to increase the pain level during the program, because the stimulation territory should be comfortable to the touch and contact with the dysesthetic area should be avoided. Most patients reported hypoesthesia in the same cutaneous area where the allodynia was previously observed. This was the case in all three SRM studies (Nedelec et al., 2016; Packham et al., 2018; Spicher et al., 2008). These studies advocated to systematically continue the intervention, treating hypoesthesia with tactile stimulation directly on the area affected. For the study that examined the use of a glove and tactile stimulation (Wider et al., 2006), no details on the intervention were provided. Of the seven studies included, three of them (Bellugou et al.,

1991; Göransson & Cederlund, 2011; Nedelec et al., 2016) briefly mentioned whether or not other concurrent interventions were used. One study provided a thorough description of medication as a concurrent intervention (Wider et al., 2006).

3.5.3 Outcome measures Detailed information on outcomes measures are presented in **Table 15**(p.90). Two studies reported only on pain outcomes (Bellugou et al., 1991; Wider et al., 2006), one study focussed only on dysesthesia-specific outcomes (Spicher et al., 2008), and four studies reported on both pain and dysesthesia outcomes (Göransson & Cederlund, 2011; Love-Jones et al., 2009; Nedelec et al., 2016; Packham et al., 2018). Pain outcomes were measured with the Numerical Rating Scale (NRS) (Love-Jones et al., 2009), the Visual Analog Scale (VAS) (Göransson & Cederlund, 2011) and the McGill Pain Questionnaire or the French version of that tool, the *Questionnaire de la douleur Saint-Antoine* (Nedelec et al., 2016; Packham et al., 2018). Two studies used a detailed subjective assessment of pain (Bellugou et al., 1991; Wider et al., 2006). Of the studies assessing dysesthesia, four employed a variety of measuring techniques (**Table 15**, p.90). Two SRM studies assessed change in allodynia severity using Semmes Weinstein monofilaments (Nedelec et al., 2016; Spicher et al., 2008). The three SRM studies provided the number of cases where allodynia had resolved (Nedelec et al., 2016; Packham et al., 2018; Spicher et al., 2008). Only one study (Göransson & Cederlund, 2011) assessed a concept other than pain (i.e., occupational performance), yet that goes beyond the scope of this review. Although the studies assessed pain and/or dysesthesia, this was done using heterogeneous outcome measures.

Length of follow-up varied greatly, including after 60 minutes intervention (Love-Jones et al., 2009), six weeks (Göransson & Cederlund, 2011), two to three months (Nedelec et al.,

2016), or the amount of time needed to achieve rehabilitation goals (Packham et al., 2018; Spicher et al., 2008). Follow-up time was not specified in two of the studies (Bellugou et al., 1991; Wider et al., 2006).

3.5.4 Changes in pain/dysesthesia Four studies presented a statistical analysis of pain/dysesthesia outcomes (**Table 15**, p.90) and showed statistical differences between treatment initiation and the last day of follow-up. With respect to desensitization, Göransson and Cederlund (2011) found a statistically significant improvement in pain with use or on contact (i.e., touch) ($p<0.001$), reduced pain at rest ($p=0.001$), and reduced size of the hyperesthetic area ($p<0.001$). Love-Jones et al. (2009) found a statistically significant decrease in the size of the allodynic area ($p<0.001$) but did not find a statistical difference in pain scores. In terms of the SRM studies, Nedelec et al. (2016) found a statistically significant decrease in the size of the allodynic area ($p=0.002$). Packham et al. (2018) found a statistical difference in pain scores ($p<0.001$) with a strong effect size (Cohen's d : 1.64) for this variable following the entire tactile stimulation program used to treat allodynia and hypoesthesia. The other studies reported quantitative or qualitative improvements in pain scores and/or dysesthesia but did not perform a statistical analysis. Overall, all studies reported improvements in pain/dysesthesia, but some did not document these findings using inferential statistics. It was not possible to conduct a statistical synthesis (meta-analysis) of the results due to the heterogeneous characteristics (populations, interventions, outcome measures, follow-up) across studies.

3.5.5 Quality of selected studies

The MINORS scale scores for quality of research ranged from 2 to 10 out of a maximum score of 16 for non-comparative studies. Two studies (Bellugou et al., 1991; Wider et al., 2006) had a score of 2, one study (Nedelec et al., 2016) a score of 6, two studies (Packham et al., 2018; Spicher et al., 2008) a score of 9 and two studies a score of 10 (Göransson & Cederlund, 2011; Love-Jones et al., 2009). Ratings are presented in **Table 37**(p.97). Five studies clearly stated their research aim/objective (Göransson & Cederlund, 2011; Love-Jones et al., 2009; Nedelec et al., 2016; Packham et al., 2018; Spicher et al., 2008). Four studies (Göransson & Cederlund, 2011; Love-Jones et al., 2009; Packham et al., 2018; Spicher et al., 2008) mentioned the inclusion of consecutive patients. Two studies involved a prospective collection of data (Göransson & Cederlund, 2011; Love-Jones et al., 2009). One study (Packham et al., 2018) showed endpoints appropriate to the aim of the study. No study conducted an unbiased assessment of the study endpoint, except for the study by Love-Jones et al. (2009) who performed a single-blind assessment where patients were unaware of the aim of the study. Three studies (Nedelec et al., 2016; Packham et al., 2018; Spicher et al., 2008) had an appropriate follow-up period and two studies (Göransson & Cederlund, 2011; Spicher et al., 2008) reported a loss to follow-up rate less than 5%. Calculations for determining sample size and power were not performed in any of the studies. Comparator groups were also not used in any of the studies.

3.5.6 Risk of bias The five types of bias (selection, performance, attrition, detection, reporting) were present in most of the studies (**Table 48**, p.98). Only one study (Göransson & Cederlund, 2011) showed less risk of bias, which was considered high in one category, moderate in three categories and low in one category. Three of the studies demonstrated greater risk of bias (Packham et al., 2018; Spicher et al., 2008; Wider et al., 2006). Two types of bias (detection and reporting) were predominant for a high risk of bias across all seven studies. These risks of bias were mainly related to the unvalidated assessment tools chosen (*Questionnaire de la douleur Saint-Antoine*, severity of dysesthesia and size of dysesthetic area) and to the fact that the results were reported only in a descriptive manner.

3.6 Discussion This review summarizes the current evidence on tactile stimulation programs from seven articles (involving 218 participants in total), for managing hand dysesthesia following a peripheral nerve injury. It also assesses the methodological quality of these studies. Findings may be useful to hand therapists in determining which tactile stimulation program is most appropriate for their clients, and how to apply and adjust parameters for each of these programs.

3.6.1 Heterogeneity of populations

The significant heterogeneity of the populations examined in all the studies (which did not specifically address hand dysesthesia following a PNI) makes comparing their results a difficult task. Moreover, the population in five of the studies (Bellugou et al., 1991; Göransson & Cederlund, 2011; Love-Jones et al., 2009; Nedelec et al., 2016; Spicher et al., 2008) was heterogeneous. This makes it difficult to isolate the effects on the specific population studied in

this systematic review (i.e., hand dysesthesia following a PNI). Most of the studies failed to demonstrate reasonable evidence of a PNI or NP. Lastly, duration of symptoms prior to treatment was heterogeneous in three studies (Göransson & Cederlund, 2011; Nedelec et al., 2016; Packham et al., 2018), with acute (< 3 months) and chronic populations being combined. This heterogeneity suggests that those interventions can be used in a broad spectrum of clients, although acute patients can exhibit spontaneous recovery not related to treatment (i.e., neuropraxia) (Seddon, 1942; Siddiqui, Benjamin, & Schubert, 2000; Sunderland, 1951).

3.6.2 Heterogeneity of interventions

Another facet of heterogeneity in the studies reviewed is the essential construct of tactile desensitization, which implies a reduction in sensitivity through exposure. However, exposure was employed two different ways in the studies selected for our review: 1) direct flooding of tactile stimuli to the painful area with the goal of improving the pain threshold (Bellugou et al., 1991; Göransson & Cederlund, 2011; Love-Jones et al., 2009; Wider et al., 2006), which implies an effect at the level of the dorsal horn in the spinal cord (Moayedi & Davis, 2013); and 2) use of tactile stimuli on an adjacent territory where contact was normal to tolerable (Nedelec et al., 2016; Packham et al., 2018; Spicher et al., 2008), and intended to provide sensory re-education, implying an effect at the level of the somatosensory cortex (Pleger et al., 2005). This second element is also employed in the SRM studies, and thus reflected in the studies by Nedelec et al. (2016), Packham et al. (2018) and Spicher et al. (2008). However, there is a lack of consensus on any taxonomy related to sensory re-education (or re-learning) and desensitization. A 2011 Delphi process (Jerosch-Herold, 2011) reported 84% of respondents endorsed desensitization by immersing the hand in different textures (i.e., flooding) as an essential component of sensory

relearning programs; however, the research question did not discriminate between treatment of numbness or dysesthesia after nerve injury.

3.6.3 Description of the interventions

Most of the studies reported enough detail about the intervention for the study to be replicated. With respect to desensitization, the three studies (Bellugou et al., 1991; Göransson & Cederlund, 2011; Love-Jones et al., 2009) described a wide variety of intervention parameters. As for the SRM studies, the intervention parameters were standardized across the three studies (Nedelec et al., 2016; Packham et al., 2018; Spicher et al., 2008). There were no specific details for the use of glove or tactile stimulation (Wider et al., 2006). Overall, it seems that the interventions, including desensitization and SRM, were predominantly used in a home setting a few times a day (1-8 times) for a few minutes (1-10 minutes) each time. The exercises were generally reviewed with the clinician during appointments. Most of the studies suggested not to increase the level of pain and recommended that the stimulation be tolerable. All in all, it seems that a vast array of dysesthesia interventions can be easily applied in practice, as they require minimal equipment and are primarily carried out as a home program. However, none of the studies reported sufficient detail on co-interventions, with the exception of medication in one study (Wider et al., 2006). It is therefore difficult to attribute the reported results exclusively to tactile stimulation programs.

3.6.4 Outcomes

Many outcome measurement tools were used across the seven studies included in this review. Most of the instruments used, such as the NRS, VAS or McGill Pain Questionnaire,

were neither specific to dysesthesia, nor to neuropathic pain. On the other hand, five studies (Göransson & Cederlund, 2011; Love-Jones et al., 2009; Nedelec et al., 2016; Packham et al., 2018; Spicher et al., 2008) did use specific measurement tools to assess severity or the area affected by dysesthesia. However, none of these instruments had been assessed for validity and reliability at the time these studies were conducted, although reliability of allodynography has recently been reported (Packham, Spicher, MacDermid, & Buckley, 2019). It is therefore difficult to definitively conclude that the changes measured by these instruments reflect a change in dysesthesia. There was no other available validated instrument for specifically assessing hand dysesthesia when these studies were conducted. Nevertheless, a new tool is currently available to assess hand sensitivity, including dysesthesia and any sensitivity impairment (Packham, MacDermid, Michlovitz, Cup, & Van de Ven-Stevens, 2018). The statistical analysis performed on pain scores for some of the studies shows statistical significance for reduced pain with desensitization (Göransson & Cederlund, 2011; Love-Jones et al., 2009) and the SRM (Nedelec et al., 2016). The effectiveness of these interventions is unknown as no effect size was reported. Follow-up time varied greatly, which made it impossible to compare the results across studies. In the three studies on desensitization (Bellugou et al., 1991; Göransson & Cederlund, 2011; Love-Jones et al., 2009), the follow-up time was too short. Consequently, the long-term effects of these interventions could not be assessed or reported.

3.6.5 Methodological quality and risks of bias

The current literature on this topic is largely centred on small case-series studies, case studies, experimental studies, and proof-of-concept studies. Those non-comparative studies do not allow the effectiveness of interventions or the effect of tactile stimulation programs

themselves to be established. Moreover, the seven studies that comprised this review, including non-comparative studies, used mainly low-quality methodologies. The low scores on the MINORS scale were predominantly due to the lack of prospective collection of data, an unbiased endpoint and the prospective calculation of the study size.

With those results reflecting the low methodological quality of the studies, it is not surprising that risks of bias were found across all studies. Although the outcomes of the studies suggest promising benefits, it is difficult to comment on the effect of those interventions described in the literature due to poor methodological quality and significant risks of bias. One study (Göransson & Cederlund, 2011) showed improvements in clinical outcomes while being the least likely to be biased compared to the other studies included in this review. Overall, the studies included in this review suggest that interventions based on tactile stimulation would have beneficial effects on pain, as measured with VAS. However, it is not possible to confirm that the improvements noted in the studies reached clinical significance since there is as yet no study on the minimal clinically important difference for the VAS score in patients with touch-evoked neuropathic pain following nerve injury (Olsen et al., 2017). Based on the literature published to date, tactile stimulation programs show low evidence for decreasing hand dysesthesia following PNI.

3.6.6 Limitations

This review has its limitations. There is currently no consensus in the literature for terminology related to dysesthesia and tactile stimulation program. Dysesthesia and tactile stimulation program key words were chosen for this systematic review because they are used in

broad categories that employ a variety of terms. It is therefore possible that some articles were missed because of the keywords chosen. Studies on persons with type 1 complex regional pain syndrome (CRPS 1) were not included, such as those by Lewis et al. (2011) and Pleger et al. (2005), because these studies failed to meet the PNI inclusion criteria for this review. In fact, there is an unresolved debate about the role of nerve injury and small fiber neuropathy as a part of CRPS 1 (Oaklander & Fields, 2009), despite tactile desensitization being recommended as a core intervention in recent clinical practice guidelines (Goebel A, 2018). Even if the studies on CRPS 1 were included, that would not have changed any conclusions made because the methodologies and results of those CRPS1 studies are similar to the studies included in this present review. Other relevant studies may also have been missed as a result of the exclusion criteria relating to foreign languages.

3.7 Conclusion This systematic review sought to gather evidence on commonly recommended interventions in the treatment of hand dysesthesia in patients following a PNI. The studies reviewed suggest that tactile stimulation programs may play a role in decreasing hand dysesthesia. Nevertheless, this review suggests inconclusive evidence and inconsistent implementation of those tactile stimulation programs. All studies included have a low to very low level of evidence. We suggest that there are two main types of tactile stimulation programs: desensitization and the Somatosensory Rehabilitation Method (SRM). Of these two programs, only the SRM is a standardized intervention. Regardless of the technique chosen, tactile stimulation should be used 1-12 times daily for 1-10 minutes and should be increased based on the patient's response (tolerable symptoms or no increase in pain). Additional high-quality methodological studies are needed to establish best practices for tactile stimulation programs used to treat hand dysesthesia.

The identified gaps in the current evidence on tactile stimulation programs for hand dysesthesia following a PNI provide an opportunity for future research studies. There is a need for methodologically rigorous retrospective and prospective case series. These future studies should use internationally accepted terminology for hand sensitivity and pain (IASP, 2012). They should have precise inclusion and exclusion criteria to obtain a more homogenous population identified as having a PNI and/or NP. Studies should measure outcomes with validated assessment instruments related to the concepts being measured. It would also be interesting to assess the effects of those programs on other parameters, such as hand function and quality of life. Finally, randomized controlled trials would accurately determine the effectiveness of tactile stimulation programs for decreasing hand dysesthesia in comparison to

traditional treatment, and present opportunities for comparing the effectiveness of flooding and relearning approaches.

Conflicts of interests

Two authors (IQ, TP) have given paid presentations that included this topic, and three authors (IQ, TP, JOD) are either authors of some of the material included in this review or have other publications with authors of the included studies.

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Figure 1 : Prisma Flow Diagram for the search results

(Moher, Liberati, Tetzlaff, Altman, & Group, 2009)

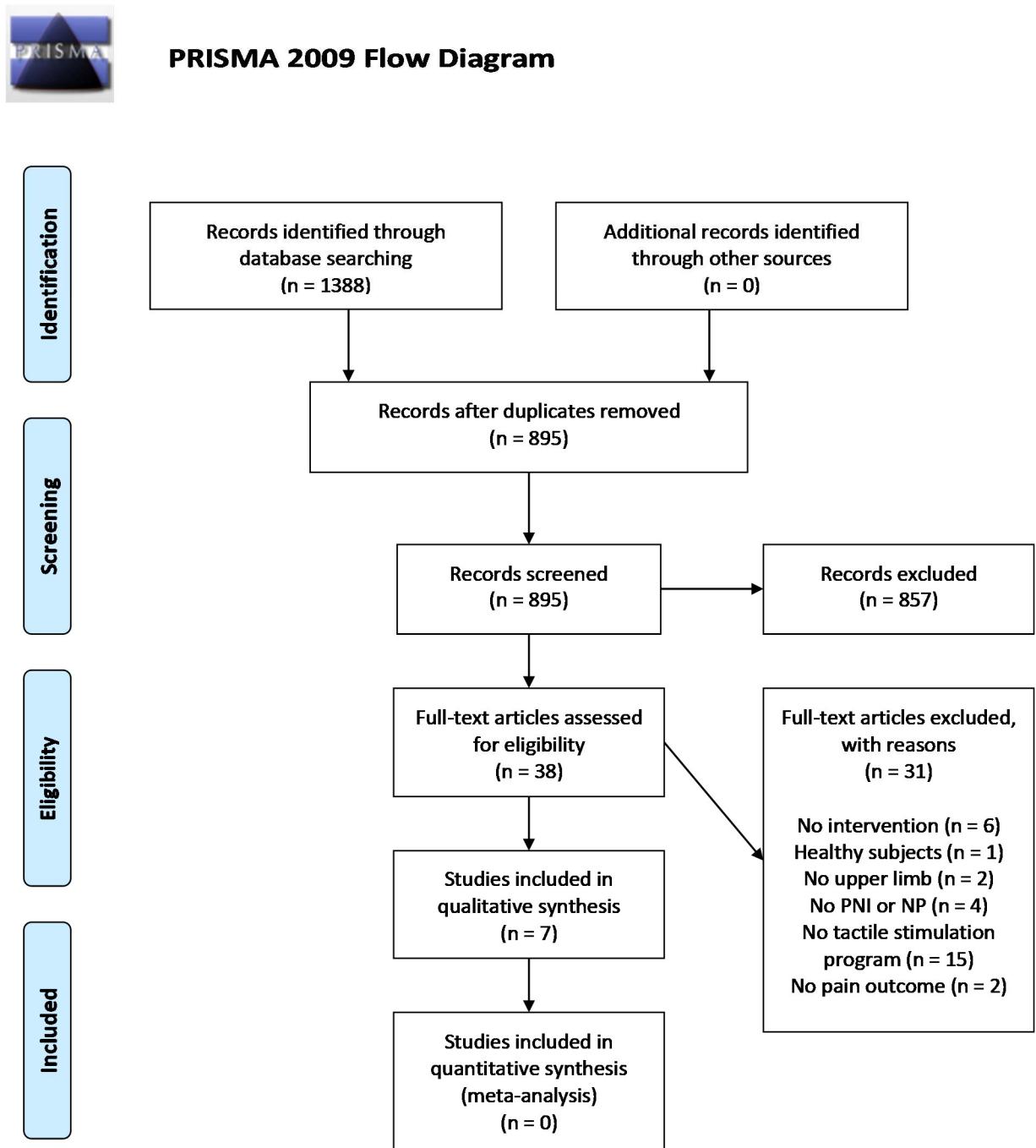


Table 1 : key characteristics of each study

Study article (date)	Study type	Population	Intervention	Outcomes measures	Follow-up time	Results	MINORS score
Bellugou et al. (1991)	PC	<p>Hand injury n = 60 participants</p> <p>Pain duration*: Unknown</p> <p>PNI: Authors suggest that all participants have at least lesions affecting sensory receptors</p> <p>NP: Authors report painful hypersensitivity in all patients</p>	D	<p>Pain: Descriptive</p>	6-7 weeks	<p>Pain 85% to 90% of participants showed reduction in pain or no pain (“complete healing”) 10% of participants showed no improvement in pain Reduction in pain can be observed within 2 to 3 weeks, with a maximum decrease within 6 to 7 weeks</p>	2
Love-Jones et al. (2009)	ES	<p>Neuropathic pain patients with different aetiologies and body parts affected n = 18 participants</p> <p>Pain duration*: 27 months (range: 3-336)</p> <p>PNI: n = 6 medically diagnosed (post herpetic neuralgia)</p> <p>NP: reported by all participants</p>	D	<p>Evoked pain: NRS</p> <p>Allodynic area: Marked out the area with a cotton swab then marked out a grid using perforated bubble wrap</p>	Reevaluated every 10 min post-intervention for up to 1 hour	<p>This study differentiated responders (R) from non-responders (NR). R is defined as having more than 30% reduction in the allodynic area</p> <p>Pain: no significant change in evoked pain intensity at any point post-treatment in both R and NR. R: NRS -0.1 mm ± 0.6 (p>0.05) (n = 9) NR: NRS -0.1 mm ± 0.8 (p>0.05) (n = 9)</p> <p>Allodynic area: maximum area shrinkage was seen 20 minutes post-treatment in R. R: Shrinkage of 48% ± 9% (p<0.001) of the area (n = 9)</p>	10
Göransson & Cederlund (2011)	PCS	<p>Hand and upper extremity injury n = 39 participants</p> <p>Pain duration*: 9 weeks (range: 3-104)</p> <p>PNI: medically diagnosed (n=10)</p> <p>NP: not able to differentiate nociceptive pain from NP</p>	D	<p>Pain: VAS</p> <p>Hypersensitive area: Drawn out patient then measured</p>	6 weeks	<p>Pain with use/touch: significant decrease VAS: -15 mm [95%CI: -30, -7] (p<0.001)</p> <p>Pain at rest: significant decrease VAS: -6 mm [95%CI: -18, 0] (p=0.001)</p> <p>Hypersensitive area: significant decrease Area: -850 mm² [95%CI: -1696, -300] (p<0.001)</p>	10

Spicher et al. (2008)	RCS	<p>Neuropathic pain patient s (different aetiologies and body parts affected) n = 43 participants</p> <p>Pain duration*: 35 months (SD 21, range: 7 - 523)</p> <p>PNI: Injured nerves identified, not medically diagnosed</p> <p>NP: reported by all participants</p>	SRM	<p>Allodynia surface: Allodynography (mapping) with a 15g S-W monofilament and VAS</p> <p>Allodynia severity: Rainbow pain scale (with S-W monofilament)</p>	Time required to eliminate the allodynia (and treat hypoesthesia)	<p>Mechanical allodynia: Disappearance of allodynia in all participants within an average of 70 ± 66 days (8 to 206) Note: To be included in this retrospective study, participants had to demonstrate disappearance of allodynia.</p> <p>Allodynia severity: Overall duration to move on to the next monofilament: 24 days From green (1.5 g) to blue (3.6 g): $49.9 \text{ days} \pm 32.9$ From blue (3.6 g) to indigo (8.7 g): $33.7 \text{ days} \pm 20.8$</p>	9
Nedelec et al. (2016)	RCS	<p>Burn survivors with different body parts affected n = 17 participants (15 with allodynia out of a total of 17, n=4 hands)</p> <p>Pain duration*: 486 days (SD 596, range: 45 - 2373)</p> <p>PNI: Assumed (receptors minimally injured as a result of burns)</p> <p>NP: All participants described symptoms compatible with neuropathic pain</p>	SRM	<p>Pain: QDSA</p> <p>Size of area affected by mechanical allodynia: allodynography with a 15g S-W monofilament and VAS</p> <p>Mechanical allodynia threshold: Rainbow pain scale (expressed in percent of improvement)</p>	2 to 3 months	<p>Pain: reduction in QDSA score $-3.1\% \pm 7.7$ at 1 month (n=8) $-8.9\% \pm 14.1$ at 2 months (n=8) $-22.7\% \pm 22.8$ at 3 months (n=6)</p> <p>Size of area affected by mechanical allodynia: significant reduction at 3 months ($p = 0.002$) (n=5)</p> <p>Mechanical allodynia threshold: improved $27 \pm 21\%$ at 2 months (n=14) $29 \pm 26\%$ at 3 months (n=12)</p> <p>Out of the 15 participants with allodynia, 11 responded well to treatment (reduction in size of area and threshold), including 8 upper-limb injured participants. All hand injured participants showed resolution of allodynia (n=4). 4 participants did not respond to this treatment (2 shoulders and 2 lower extremities).</p>	6

Packham et al. (2018)	RCS	Upper limb with CRPS type II n = 88 nerve lesions (51 of which presented with allodynia) Pain duration* : 31 months (range: 1 month - 25 years) PNI : mentioned, not medically diagnosed NP : All participants had neuropathic pain	SRM	Pain : QDSA Allodynic area : allodynography using a 15g S-W monofilament and VAS	Time required to eliminate the allodynia (and treat hypoesthesia)	Pain : reduction in QDSA score Initial score: 48.1 ± 17.7 (range 5 – 99) Final score: 20.1 ± 20.0 (range 0 – 75) (at the end of treatments) Significative decrease of pain ($p<0.001$) Allodynic area : 49 out of 51 lesions showed a complete resolution of their allodynia In two lesions, treatments were discontinued because the therapist felt it would not be beneficial Average length of treatment to resolve allodynia : 81 days (SD 76.4; range: 5-381)	9
Wider et al. (2006)	CR	Painful hand and moving fingers n = 1 participant Pain duration* : not specified, but at least 7 months and at most 14 months PNI : medically diagnosed (carpal tunnel syndrome) NP : Participant reported hyperalgesia	Use of a glove and tactile stimulation	Pain : Qualitative	Not specified	Pain : Immediate and complete disappearance of pain upon tactile stimulation or use of a glove.	2

* duration of symptoms before initiating the intervention

Legend:

PC: Proof of concept, ES: Experimental study, PCS: Prospective case series, RCS: Retrospective case series, CR: Case report

CRPS: Complex regional pain syndrome

PNI: Peripheral nerve injury, NP: Neuropathic pain

D: Desensitization, SRM: Somatosensory rehabilitation method, NRS: Numeric rating scale, VAS: Visual Analog Scale, QDSA: *Questionnaire de la douleur Saint-Antoine*

DVCS: distant vibrotactile counter stimulation

S-W monofilament: Semmes-Weinstein monofilament

Table 2 : Description of intervention according to study

Study Author (date)	Type	Intervention (setting)	Description	Parameters	Adjustments	Other interventions
Bellugou et al. (1991)	D	Particles Textures Vibration Heated sand Paraffin bath Hydrotherapy Other therapies: LFUS, massage (Not specified whether done at home or in clinic)	<p>Particles: With multiple very light stimuli. The patient places his/her hand in a box with diverse particles (ex: rice, chickpeas) and moves his/her fingers in it.</p> <p>Textures: With various textures (cotton, velvet, Velcro) barely sweeping the skin. Ideally, the patient stimulates the sensitive area himself/herself with the texture in his/her healthy hand. Otherwise, the patient can use the injured hand to touch the texture directly, or the texture is placed on a wood stick and the therapist rubs it on the patient's sensitive skin. A series of brushes ranging from soft to coarse (Garros clavier), can also be used.</p> <p>Vibration: With a vibration generator.</p> <p>Heated sand flow: With fine sand particles in a bin, bombarding the hand</p> <p>Paraffin bath: With paraffin wax and paraffin oil at a temperature of 38° Celsius in a bin. The patient quickly dips his/her hand into the bin, allows the paraffin to dry for 3-4 seconds, then repeats this procedure 5-6 times.</p> <p>Hydrotherapy: With heated sterile water. The patient manipulates a soft sponge with his/her hand.</p> <p>Other therapies (LFUS, massage): Not described.</p>	<p>Textures/Particles 3-4x / day for 10 min</p> <p>Vibration 2-3x / day for 10 min</p> <p>Heated sand flow Not described</p> <p>Paraffin bath 5 min</p> <p>Hydrotherapy Not described</p> <p>Other therapies (LFUS, massage) Not described</p>	<p>Particles: Finger movements and types of particles are adjusted as the intervention should not increase the pain felt: progression of particles to more coarse, bigger and/or heavier particles.</p> <p>Textures: Types of textures are adjusted as the intervention should not increase the pain felt.</p> <p>Vibration: Frequency and amplitude of vibration is adjusted in order to not provoke pain. Also, if the targeted area is painful, the vibration is applied to an adjacent area. The session is stopped whenever pain is felt.</p> <p>Heated sand flow: The patient places his/her hand and forearm relatively deep into the bin or removes it completely to not provoke pain.</p> <p>Paraffin bath: This technique is contraindicated if it is not tolerated.</p> <p>Hydrotherapy: Temperature of water is modulated to be tolerated.</p>	Massage, hand therapy, motor reeducation for some of patients

Love-Jones et al. (2009)	D	Repeated tactile stimulation (Clinic) Repeated heat pain stimulation (Clinic)	Repeated tactile stimulation: The clinician strokes a cotton swab within the sensitive area and in a control mirror territory. Repeated heat pain stimulation: A thermode is applied to the affected side (or mirror areas)	Repeated tactile stimulation: 10x, within 1 minute at a speed of 2-3 cm/s Repeated heat pain stimulation: 1x, 2 minutes 10 heat ramps (2 degrees C/s up to the HPT and sustained for a further 4s, as tolerated)	Repeated tactile stimulation: stimulation territory is adjusted as the patient feels maximum tolerable pain. Repeated heat pain stimulation: stimulation territory is adjusted as tolerated.	Not mentioned
Göransson & Cederlund (2011)	D	Textures (Home)	Textures: Ranging from soft cotton to Velcro hooks. Massage with textures in the same direction, within the hypersensitive area, using the same speed and pressure every time until numbness occurred.	Textures: 3x / day, 2-5 minutes	Type of textures is initially chosen so that it is barely tolerated. After 1–3 weeks, most patients could progress to a rougher texture, still tolerated.	OT treatment: home program updated, not otherwise described. 2-8x/month
Spicher et al. (2008)	SRM	DVCS with textures (Home) DVCS with vibration (Clinic) Precautionary advice (Clinic) and application (not mentioned) Rehabilitation of the hyposensitivity (and underlying hyposensitivity) (not mentioned)	DVCS with textures: OT shows the patient how to apply textures in the appropriate cutaneous territory, perceived as comfortable. DVCS with vibration: Not described. Precautionary advice and application: OT shows the patient how to avoid contact with the allodynic area. Rehabilitation of the hyposensitivity (and underlying hyposensitivity): not described	DVCS with textures: Rabbit skin 6x / day for 2 min DVCS with vibration: 1x / week Frequency: 100 Hz Amplitude: 0.06mm Duration not specified Precautionary advice: each therapy session 1x /week Rehabilitation of the hyposensitivity (and underlying hyposensitivity): not described	DVCS: Territory for vibration and textures is adjusted as it is perceived comfortable. “During the course of treatment, it will become possible for the patient to progressively invade the “old” allodynic territory with the same comfortable stimulus.” Precautionary advice: defined at each therapy session. Rehabilitation of the hyposensitivity (and underlying hyposensitivity): not described	Not mentioned

Nedelec et al. (2016)	SRM	DVCS with textures (Home) DVCS with vibration (Clinic) Precautionary advice (Clinic) and application (Home) Sensory reeducation for hypoesthesia (Home and Clinic)	<p>DVCS with textures: The patient applies fur or a soft, comfortable fleece to a territory proximal from the allodynic area.</p> <p>DVCS with vibration: The same territory is stimulated with a vibration generator.</p> <p>Precautionary advice and application: OT records the patient's daily activities and treatments (e.g., pressure garments, massage, etc.) where direct pressure might be applied to the allodynic area. Using problem-solving collaboration, they find "alternative approaches so that pressure stimulation could be avoided during activities/treatments while accomplishing the daily activities or alternatively, determine that the activities should be discontinued or delegated until the mechanical allodynia is eliminated."</p> <p>Sensory reeducation for hypoesthesia: Once the allodynia is resolved, sensory re-education is initiated. Touch discrimination is performed with the eraser end of a pencil and requires the patient to discriminate when he/she is being touched, whether it is static touch or moving in a straight/curved line, with their vision obscured.</p> <p>Texture perception is performed with three different textures that the patient perceives as comfortable directly on the hypoesthesia area and to a normal control anatomically similar territory.</p> <p>Vibratory stimulation is performed with a vibration generator (in the clinic) or with another device at home (e.g., hand-held vibrator) on the hypoesthesia area.</p>	<p>DVCS with textures: 8x / day for 1 min</p> <p>DVCS with vibration 1x / week for 10 min</p> <p>Precautionary advice: Revised during therapy session with the patient every 2 weeks</p> <p>Precautionary application: Should be applied at all times</p> <p>Sensory reeducation for hypoesthesia: Touch-discrimination: 12x / day for 15 sec, then gradually progressing to 3x / day for 10 min Textures: 12x / day for 15 sec, then gradually progressing to 4x / day for 5 min Vibration: 1x / week (clinic), 5 min maximum 1x / day (home), 5 min maximum</p>	<p>DVCS with textures: Types of textures and stimulated area adjusted as it is perceived as comfortable. The targeted territory is revised every 2 weeks.</p> <p>DVCS with vibration: Zone of application and intensity of the vibration are adjusted as it is perceived as comfortable. The targeted zone is revised every 2 weeks.</p> <p>Precautionary advice and application: Personalized for every patient, revised every 2 weeks.</p> <p>Sensory reeducation for hypoesthesia: Touch discrimination begins with a static touch or moving straight line, then when the client is able to discriminate those touches, a curved line is added to the exercises.</p> <p>Textures are applied if the client is able to perceive 5 g (between 4.56 and 4.74 monofilament).</p> <p>The vibration generator is set at 100 Hz and the amplitude adjusted to 0.1 mm above the level the participant could perceive. Excessive painful vibratory stimulation is avoided.</p>	Cortisone (n=3) Not mentioned	injection otherwise
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Packham et al. (2018)	SRM	DVCS with textures (Home) DVCS with vibration (Clinic) Precautionary advice (Clinic) and application (Home) Sensory reeducation (not mentioned)	DVCS with textures: A comfortable texture (e.g., rabbit fur, plush microfleece), is applied using a light stroking motion to a territory of skin with normal sensation, in the area of the identified injured nerve. DVCS with vibration: Vibration stimulation is applied to the same territory as DVCS with textures, using a vibration generator. Precautionary advice and application: OT reviews activities of daily living with the participant and together they identify sources of evoked pain (e.g., rubbing of clothing and use of tools). They find “strategies to avoid stimulation and/or delegate provocative tasks,” in order to “minim[ize] evocation of pain by temporarily limiting touch and consequently functional use of the painful zone.” Sensory reeducation: to address the residual hypoesthesia	DVCS with textures: 8x / day for 1 min maximum DVCS with vibration: 1x / week for 10 min Precautionary advice: At every appointment Precautionary application: Applied at all times Sensory reeducation: not mentioned	DVCS with textures: Area of skin stimulated, type of texture and duration of stimulation is adjusted so the participant perceives it as “the most comfortable”. DVCS with vibration: Territory of application and intensity are adjusted for normal sensation. Advices: Strategies are personalized for every participant and chosen to minimize pain. Sensory reeducation: after the allodynia has abated	Not mentioned
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Wider et al. (2006)	Other	Use of a glove Tactile stimulation (Not specified whether done at home or clinic)	Use of a glove: Not specified Tactile stimulation: In the painful area. Not otherwise specified.	Use of a glove: Not specified Tactile stimulation: Not specified	Use of a glove: Not specified Tactile stimulation: Not specified.	Treatment medication (gabapentin, amitriptyline and baclofen).
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Legend:

D: Desensitization, SRM: Somatosensory rehabilitation method, NRS: Numeric rating scale, VAS: Visual Analog Scale, QDSA: *Questionnaire de la douleur Saint-Antoine*

OT: occupational therapist

DVCS: distant vibrotactile counter stimulation

LFUS: low-frequency ultrasound

Table 3 : Distribution of the seven articles included in the review according to the number of articles having obtained one of the three scores (0, 1, 2) for each item of the MINORS scale

Items	Scores		
	2	1	0
1 - A clearly stated aim	5/7	2/7	0/7
2 - Inclusion of consecutive patients	4/7	0/7	3/7
3 - Prospective collection of data	2/7	0/7	5/7
4 - Endpoints appropriate to the aim of the study	1/7	5/7	1/7
5 - Unbiased assessment of the study endpoint	0/7	1/7	6/7
6 - Follow-up period appropriate to the aim of the study	3/7	3/7	1/7
7 - Loss to follow up less than 5%	2/7	3/7	2/7
8 - Prospective calculation of the study size	0/7	0/7	7/7

Table 4 : risks of bias for each study

Study	Selection	Performance	Attrition	Detection	Reporting
Bellugou et al. (1991)	moderate	moderate	moderate	high	high
Love-Jones et al. (2009)	moderate	moderate	moderate	high	high
Göransson and Cederlund (2011)	moderate	moderate	moderate	high	low
Spicher et al. (2008)	high	moderate	high	high	high
Nedelec et al. (2016)	moderate	moderate	moderate	high	moderate
Packham et al. (2018)	moderate	moderate	high	high	high
Wider et al. (2006)	high	high	high	high	high

Chapitre 4 : Discussion

Le présent mémoire inclut deux articles portant respectivement sur une étude de cas clinique et une recension systématique (Quintal, Carrier, Packham, Bourbonnais & Dyer, soumis 2019; Quintal, Poiré-Hamel, Bourbonnais & Dyer, 2018). Ces études ont permis de répondre à deux objectifs. Le premier objectif était de décrire l'intégration d'une approche innovante de stimulation tactile dans le cadre d'un programme d'intervention multimodal chez une personne présentant un SDRC avec AM. Avant ce programme d'intervention, cette personne avait participé à des interventions conventionnelles pendant 19 mois sans améliorations cliniques de sa symptomatologie douloureuse et de sa fonction du membre supérieur atteint. Cette étude de cas montre que l'utilisation de la MRSD dans le cadre d'un programme multimodal a amélioré les mesures cliniques de douleur, des amplitudes articulaires actives, de la force de préhension, des forces de pince digitales ainsi que la perception des incapacités aux membres supérieurs et d'autonomie dans tous les domaines du rendement occupationnel (soins personnels, activités productives, loisirs) (Townsend & Polatajko, 2013) chez cette personne.

Comme le programme multimodal présenté dans l'étude de cas incluait une approche de stimulation tactile, cela a questionné l'impact relatif de ce type d'approche dans ce programme, et donc les niveaux d'évidences cliniques des approches de stimulation tactiles répertoriées dans la littérature. Ceci a mené à la formulation du deuxième objectif qui consistait à recenser dans la littérature et évaluer les études portant sur les approches de stimulation tactile en lien avec le traitement de la douleur chez des personnes avec AM à la main découlant d'une LNP. La recension systématique a permis d'atteindre ce second objectif de répertorier et évaluer les

études en question, en plus de documenter les niveaux d'évidence des approches de stimulation tactile pour diminuer la douleur chez cette population.

Cette discussion permet d'établir des liens entre les connaissances acquises dans l'étude de cas et la recension systématique, et ce sans réitérer les éléments de discussion déjà présentés dans les articles. L'accent est mis sur les conclusions de ces études et les implications cliniques qui en découlent. Les contributions à l'avancement des connaissances en ce qui a trait à la pratique clinique ainsi qu'en recherche sont également présentées. De plus, les lacunes qui demeurent au niveau des connaissances dans le domaine des approches de stimulation tactile pour traiter l'AM sont discutées, ainsi que les recommandations de recherches futures qui en découlent.

4.1 Deux approches de stimulation tactile

Deux types d'approches de stimulation tactile ont été identifiés dans la littérature pour la prise en charge de l'AM à la main suivant une LNP: la désensibilisation et la MRSD. Les données issues de l'étude de cas et les données probantes extraites de la recension systématique ne permettent pas statuer sur l'efficacité clinique relative de ces deux approches, et encore moins sur laquelle des deux approches est la plus efficace pour traiter cette condition. Néanmoins, dans une perspective clinique, les études suggèrent que les approches de stimulation tactile jouent un rôle pour diminuer la douleur chez les personnes atteintes d'AM à la main suite à une LNP. Il est alors intéressant de se pencher sur les facteurs qui pourraient contribuer aux effets observés. En prenant en référence un modèle biopsychosocial (Townsend & Polatajko, 2013), ces facteurs peuvent se situer tant au niveau personnel (physique, affectif, cognitif et spirituel) qu'au niveau

des occupations (soins personnels, productivité et loisirs) ou de l'environnement (physique, institutionnel, culturel, social).

4.1.1 Mécanismes neurophysiologiques

Quoique ne faisant pas partie des objectifs de ce mémoire, il est intéressant de mentionner qu'au niveau du domaine personnel, plus précisément au niveau physique, des mécanismes neurophysiologiques pourraient contribuer aux effets bénéfiques des deux approches de stimulation tactile sur la douleur. Tout d'abord, il est possible que les mécanismes neurophysiologiques au niveau spinal et supraspinal soient mobilisés par les approches de stimulation tactile. Les stimulations répétées des afférences cutanées pourraient exercer un rétablissement des mécanismes dérégulés de l'inhibition pré-synaptique (Guo & Hu, 2014) qui contribuaient alors à la sensibilisation centrale (Godde, Ehrhardt, & Braun, 2003; Godde, Spengler, & Dinse, 1996; Pleger et al., 2005; Schaefer, Rothmund, Heinze, & Rotte, 2004) et ainsi à l'allodynie (Woolf, 2011).

En effet, lors des traitements avec les approches de désensibilisation et de MRS, la stimulation tactile est réalisée de telle sorte de ne pas exacerber de douleur dans la surface allodynique (sous le seuil de perception de la douleur) et permettre à la personne de réapprendre à ressentir un stimulus non douloureux. Ainsi, il est possible que cette stimulation répétée sur une longue durée normalise, par un mécanisme de neuroplasticité, la transmission des afférences cutanées et diminue ainsi l'AM.

Par ailleurs, au niveau du domaine personnel également (Townsend & Polatajko, 2013), on ne peut exclure que la relation thérapeutique ou l'objectivation de la douleur par le thérapeute et le patient lors du processus thérapeutique puisse contribuer à la diminution de l'allodynie et l'hypersensibilité. À ce moment, l'influence des régions antérieures du cerveau contrôlant les aspects affectifs, émotionnels et cognitifs de la perception de douleur pourrait, de par leurs voies descendantes, agir sur les régions de relais des afférences nociceptives au niveau segmentaire et modifier les seuils de sensibilité de transmission synaptique des neurones et ainsi diminuer l'AM (Almeida, Roizenblatt, & Tufik, 2004; Iannetti & Mouraux, 2010; Pelletier, Bourbonnais, & Higgins, 2018; Perini, Bergstrand, & Morrison, 2013; Tracey & Mantyh, 2007). Par exemple, il est possible que l'action des faisceaux descendants du système médial telles les régions bulbaires rostrale ventromédiane (« *rostral ventromedial medulla* (RVM) ») augmente les seuils à la pression et au toucher et ainsi diminue la douleur (Heinricher, Tavares, Leith, & Lumb, 2009; Van Griensven, Strong, & Unruh, 2013). Ces mécanismes ou d'autres mécanismes de neuroplasticité pourraient contribuer les résultats positifs de l'étude de cas et aux autres études incluses dans la revue systématique. Quoique tout ceci reste très spéculatif, il demeure qu'il existe des substrats neurophysiologiques qui peuvent moduler l'expérience de la douleur par des stimulations tactiles.

Bien que ces hypothèses quant aux mécanismes d'action soient plausibles, des études fondamentales en neurophysiologie humaine devraient être réalisées afin de mieux comprendre les mécanismes neurophysiologiques responsables de la diminution de l'AM chez les patients appliquant les approches de stimulation tactile. Par exemple, l'électroencéphalographie ou encore les examens d'imagerie par résonance magnétique fonctionnelle pourraient permettre

d'observer les changements corticaux avec la stimulation tactile (Bosma, Hemington, & Davis, 2017; Davis & Moayedi, 2013; Godde et al., 2003; Osborne et al., 2018; Schaefer et al., 2004).

Malgré que les deux approches ne se distinguent pas en ce qui a trait aux mécanismes neurophysiologiques impliqués, elles se distinguent dans leur protocole d'application et leurs modalités de stimulation tactile. D'une part, la désensibilisation utilise une gradation de textures douces à rugueuses, en fonction du confort du patient, et ces stimulations sont appliquées directement sur la surface allodynique. D'autre part, la MRSD utilise exclusivement des textures très douces, et de la stimulation vibratoire qui sont confortables pour le patient, et qui sont appliquées à distance, sur un segment anatomique proximal à la surface allodynique. Il est donc possible qu'en raison de leur entrée sensorielle différente (directe pour la désensibilisation, ou à distance par rapport au site allodynique pour la MRSD), les mécanismes neurophysiologiques sous-jacents à la neuroplasticité décrits précédemment soient les mêmes, mais qu'ils soient exploités différemment dans ces deux types d'approches.

Par ailleurs, il est possible que les deux approches de stimulations tactiles se distinguent de par leur influence sur le domaine personnel (Townsend & Polatajko, 2013), soit sur les aspects cognitifs, affectifs et spirituels de la douleur. En effet, la MRSD fournit un cadre d'évaluation et de traitement centré sur le patient qui permet entre autres au patient de visualiser la surface allodynique (allodynographie) et d'avoir plus de contrôle sur sa condition via l'enseignement par le clinicien de la gestion de ses symptômes en fonction des activités (Spicher et al., 2015). Ce cadre de prise en charge qui fait partie du protocole de la MRSD ne fait pas partie intégrante de l'approche de désensibilisation. Ainsi, l'approche par MRSD pourrait

favoriser l'alliance thérapeutique, et l'autonomisation du patient de manière plus importante que l'approche de désensibilisation. En considérant cet aspect, il est possible que la MRSD permette de mieux mobiliser certains mécanismes de neuromodulation de la douleur par rapport à la désensibilisation. Ainsi, il est possible qu'une approche plus centrée sur le patient tel que la MRSD favorise davantage la neuromodulation du système médial, associé aux changements cognitifs, affectifs et motivationnels, que la désensibilisation. Cela pourrait expliquer en partie, du moins, le succès thérapeutique dans l'étude de cas présentée précédemment.

En plus du domaine des facteurs personnels, incluant l'aspect physique (mécanismes neurophysiologiques) ainsi que les aspects cognitifs, affectifs et spirituels de la douleur, il est possible que d'autres facteurs biopsychosociaux jouent un rôle dans la récupération des personnes avec allodynie. Tout d'abord, au niveau du domaine des facteurs environnementaux, des facilitateurs pourraient jouer un rôle dans la récupération (Townsend & Polatajko, 2013). Par exemple, un patient dont le tissu social serait aidant, optimisera ses chances de récupération. Également, au niveau des occupations (Townsend & Polatajko, 2013), des situations favorisant la participation sociale peuvent favoriser la récupération. Par exemple, un rôle social de parent auquel un patient tient énormément, peut jouer un rôle de facilitateur en ce sens que ce rôle va favoriser la mobilisation du patient dans les activités de la vie quotidienne, et pourra subséquemment favoriser l'intégration du membre atteint dans ces activités. Ceci n'est qu'un bref aperçu des facteurs pouvant contribuer aux effets des programmes de stimulation tactiles. Davantage d'études mériteraient d'étudier plus en profondeur ces aspects.

En somme, pour tenter de bien comprendre l'influence potentielle des approches thérapeutiques de gestion de la douleur, il faut considérer l'interaction entre les différentes

structures corticales du système médial et somatosensoriel qui définit l'expérience douloureuse dans ses dimensions nociceptives, cognitives, psychologiques et affectives (Almeida et al., 2004; Benaroch, 2006; Iannetti & Mouraux, 2010; Legrain, Iannetti, Plaghki, & Mouraux, 2011; Pelletier et al., 2018; Tracey & Mantyh, 2007). En plus de ces facteurs, l'aspect environnemental peut influencer l'expérience douloureuse.

Enfin, comme le choix d'approche ne peut être basé sur le niveau d'évidence et que les mécanismes neurophysiologiques impliqués ne sont pas précisés, le raisonnement clinique s'impose pour guider les cliniciens dans le choix de l'utilisation d'une ou de l'autre de ces approches.

4.2 Sélection des approches de stimulation tactile

Bien que les deux études de ce mémoire ne permettent pas de statuer sur la supériorité d'une approche de stimulation tactile par rapport à l'autre, il est possible cependant de proposer des pistes de réflexions cliniques pour guider le choix entre ces approches selon le profil et l'évolution clinique du patient en suivi. Tout d'abord, l'étude de cas montre que parfois l'utilisation d'une approche ne donne pas les résultats escomptés, et qu'il est alors temps d'envisager l'utilisation de l'autre approche. Dans ce cas précis, le raisonnement clinique soutenant le changement d'approche a été guidé par le manque de résultats bénéfiques de la première approche utilisée (i.e. désensibilisation) et le fait que le patient ne croyait plus en cette approche. En effet, des évidences démontrent que la relation thérapeutique, incluant la confiance et la communication avec les professionnels de la santé, ainsi que la confiance dans les soins prodigués est primordiale afin d'optimiser les succès thérapeutiques (Cole & McLean, 2003;

Hall, Ferreira, Maher, Latimer, & Ferreira, 2010; Klaber & Richardson, 1997; Stewart, 1995).

On ne peut alors exclure la possibilité que la relation thérapeutique et le contexte de traitement aient pu contribués aux résultats observés dans l'étude de cas.

Par ailleurs, la recension systématique souligne que peu importe le type d'approche utilisé, les paramètres devraient être ajustés afin de ne pas exacerber la douleur du patient. Cette indication, propre aux procédures des deux approches, peut alors guider le clinicien dans son raisonnement. Ainsi, si malgré des efforts raisonnables de la part du clinicien et du patient pour ajuster l'approche, le patient continue de rapporter une augmentation des symptômes douloureux, il y a alors lieu de changer d'approche. L'attention portée au suivi des symptômes et des réactions du patient ainsi que le jugement clinique des cliniciens en fonction de ces indices sont donc primordiaux dans le choix et l'application de l'une ou l'autre des approches. Les cliniciens doivent également considérer le décours temporel de l'atteinte dans le choix de l'approche de traitement. En effet, tel qu'observé en clinique et dans les études par série de cas recensées dans la revue systématique (Bellugou et al., 1991; Göransson & Cederlund, 2011; Nedelec et al., 2016; Packham et al., 2018; Spicher et al., 2008), la récupération de l'AM prend habituellement plusieurs semaines de traitement. Il serait donc recommandé de changer d'approche si le premier choix d'approche ne donne pas de résultats escomptés après une certaine période de temps. Malheureusement, cette période de temps ne peut être précisée présentement et des études longitudinales sur le décours temporel de la récupération permettraient de préciser comment tenir compte de ce décours dans la sélection des approches après quelques semaines. Il demeure qu'il y aurait avantage à cibler dès que possible laquelle de ces approches serait la plus bénéfique pour le patient. En effet, si l'on peut introduire

l'approche la plus bénéfique le plus tôt possible dans la prise en charge, cela optimisera le temps investi dans la réadaptation. Néanmoins, notre étude de cas indique que la récupération de la sensibilité peut être observée même plusieurs mois après l'apparition des symptômes initiaux. Aussi, tel qu'observé en clinique, lorsqu'un bilan sensitif et de la douleur est refait dans les premières semaines suivant le début de l'application de l'approche de stimulation tactile, il est alors possible de détecter les candidats qui ne présentent pas d'évolution ou encore pour qui la douleur augmente malgré les ajustements de l'approche de stimulation tactile. Il est alors possible de changer d'approche en considérant ces informations.

Un autre moyen de cibler rapidement quel programme est à privilégier est de recueillir les informations pertinentes sur les objectifs, valeurs et activités significatives du patient. Ces informations aideront à déterminer lequel des programmes est le plus compatible afin de ne pas entraver la fonction et l'autonomie du patient. Par exemple, un patient ayant un emploi impliquant des tâches manuelles, ne pourrait pas respecter les précautions de diminuer le contact cutané avec sa surface allodynique à la main. Ainsi, dans ce cas précis, la MRSD ne serait probablement pas le premier choix de traitement. Il serait alors préférable de tenter d'abord l'approche par désensibilisation et d'observer les résultats subséquents. Encore une fois, dans le cas où les résultats ne sont pas bénéfiques, il serait possible de changer d'approche en cours de programme.

À la lumière du raisonnement exposé précédemment, dans une perspective clinique, la prise en charge doit tenir compte des différents cas de figure rencontrés. Il apparaît qu'une approche personnalisée au patient, sur la base du jugement clinique, soit à privilégier et donc

que les deux approches sont jugées utiles pour traiter les douleurs liées à l'allodynie à la main suivant une LNP. De plus, en considérant que ces approches sont non invasives, qu'elles présentent une bonne applicabilité clinique et qu'elles présentent peu de risque, les cliniciens ont intérêt à les connaître et à les utiliser avec leur clientèle. Dans les cas où le clinicien ne trouve aucun indice afin de guider son raisonnement clinique, et qu'il est ainsi incertain du choix d'approche, il pourrait même envisager d'y aller par essai-erreur. Il serait donc judicieux que les cliniciens connaissent ces deux approches, plutôt que d'en connaître et utiliser systématiquement une seule de ces approches pour tous les cas d'allodynie mécanique.

4.3 Approche multimodale

Un des défis cliniques de l'utilisation de ces approches de traitement par stimulation tactile réside en leur incorporation dans un programme multimodal. En effet, « le traitement des douleurs neuropathiques demeure difficile malgré les nouveaux traitements disponibles, et aucun traitement à lui seul donne des résultats pour chaque condition et ne vise tous les mécanismes sous-jacents à l'atteinte » (Finnerup et al., 2005). C'est pourquoi un programme multimodal personnalisé est à privilégier chez la population atteinte de DN (Deng et al., 2016; Finnerup et al., 2005). Par exemple, en clinique, il est souvent observé que l'approche d'imagerie motrice graduée (Moseley et al., 2012) est intégrée dans un programme multimodal qui inclut aussi une approche de stimulation tactile, quelle qu'elle soit. Plusieurs autres modalités visant la DN sont également souvent ajoutées, telles que le TENS (Gibson et al., 2017), l'enseignement thérapeutique sur la douleur (Clarke, Ryan, & Martin, 2011; Louw, Zimney, Puentedura, & Diener, 2016), les techniques d'auto-gestion de la douleur (Barlow, Wright, Sheasby, Turner, & Hainsworth, 2002) et les exercices physiques actifs (Geneen et al.,

2017). Cela est sans compter la médication comme modalité ajoutée pour favoriser la modulation de la douleur (Attal et al., 2010; Dworkin et al., 2007; Vranken, 2009).

Tel qu'observé en clinique, certains patients présentent des symptômes et incapacités mixtes. Dans ces cas, en plus de tenir compte du traitement des douleurs neuropathiques, les programmes multimodaux doivent être personnalisés en fonction de la variété de symptômes et incapacités de chaque patient. Le raisonnement devra aussi tenir compte des priorités de traitement. Un exemple pertinent concerne les patients avec AM à la main qui ont subi une chirurgie récente. Les cliniciens se doivent alors de respecter le protocole postopératoire pré-établi avec le corps médical. Il y a alors lieu de se questionner sur les priorités de traitement. Dans ce type de cas, habituellement le protocole post-opératoire est autant que possible priorisé, et il devient difficile d'appliquer les approches de stimulations tactiles pendant ce protocole post-opératoire. En effet, les précautions de la MRSD visant à diminuer le contact cutané peuvent aller à l'encontre des protocoles post-opératoires qui demandent le port d'une orthèse ou encore des mobilisations passives impliquant un contact avec la surface allodynique. D'une autre façon, la désensibilisation qui implique le toucher des surfaces hypersensibles peut aller à l'encontre de ces protocoles, dans le cas où l'allodynie est près d'une plaie qu'il est contre-indiqué de toucher pour ne pas nuire à sa guérison. Dans ces deux cas de figure, la séquence d'application des modalités devient primordiale. Par exemple, une fois les quelques semaines de protocole post-opératoire passées, le clinicien pourra de nouveau envisager d'appliquer l'une ou l'autre des approches de stimulation tactile en fonction de son raisonnement clinique.

En bref, il apparaît que les programmes multimodaux sont à privilégier, malgré qu'ils représentent un défi au niveau de la prise de décision en lien avec le choix des modalités, dont les approches de stimulation tactiles, ainsi que leur séquence d'application.

À la lumière des résultats de la recension systématique et des réflexions amenées dans la discussion de ce mémoire, il est possible de reconsidérer rétrospectivement les choix effectués pour constituer le programme multimodal utilisé dans l'étude de cas présentée. En sachant que dans ce cas précis, la désensibilisation n'avait pas donné de résultats aux dires du patient, le clinicien aurait pu questionner la façon dont avait été appliquée cette approche. La fréquence quotidienne, la durée des exercices, le nombre de semaines, l'ajustement des paramètres, l'intensité de la douleur perçue pendant l'exercice de stimulation tactile font partie des éléments qui auraient pu être considérés. Dans le cas où les paramètres de la désensibilisation appliquée précédemment ne respecteraient pas ceux recommandés dans la recension systématique, la désensibilisation aurait pu être tentée de nouveau avec des ajustements conséquents. Par exemple, si le patient avait été questionné et avait rapporté une augmentation de la douleur pendant l'application de textures rugueuses, la désensibilisation aurait pu être tentée de nouveau avec des textures plus douces respectant la gestion de la douleur. Cependant, tel que discuté précédemment, la MRSD, de par son encadrement supplémentaire par rapport à la désensibilisation, favorise davantage l'alliance thérapeutique. En considérant cet avantage de la MRSD par rapport à la désensibilisation, il était plus plausible que la MRSD favorise le succès en réadaptation du patient traité qui n'avait pas eu de succès avec la désensibilisation. En plus, sachant que la motivation est un élément important de la réussite d'une réadaptation et sachant que le patient ne croyait plus en la désensibilisation, il y avait tout intérêt à opter pour la MRSD.

En effet, l'alliance thérapeutique, les émotions et la motivation peuvent moduler la perception de la douleur par le système médial. En bref, en prenant en compte les éléments de raisonnement clinique énumérés précédemment, le choix devrait demeurer le même : opter pour l'approche par MRSD.

Par ailleurs, dans le programme multimodal appliqué pour ce patient, on pourrait être tenté de reconsidérer rétrospectivement le choix des autres modalités et la séquence d'application de l'ensemble des modalités. En effet, il est tout à fait possible que des choix différents auraient pu également produire des résultats bénéfiques chez ce patient. Par exemple, les mobilisations actives auraient pu être tentées plus tôt dans le processus pour tenter de récupérer plus rapidement de la fonction au membre supérieur atteint. Ou encore l'imagerie motrice graduée aurait pu être mise en place plus tard dans le programme, afin de tenter de différencier les effets de cette approche avec ceux de la MRSD.

Dans cette étude de cas où un programme multimodal a été utilisé, il n'est pas possible d'isoler quelle est la part des effets bénéfiques qui sont attribuables à la MRSD. Il est même possible qu'aucun des gains observés ne soient pas liés à l'application de la MRSD. Ainsi, on ne peut exclure que d'autres modalités appliquées, tel que l'imagerie motrice graduée, aient été les principales responsables des effets sur la douleur. De plus, plusieurs facteurs pouvant influencer les résultats n'ont pas été mesurés, tels que le degré de participation ou encore la motivation du patient, qui peuvent être en partie responsables de l'amélioration observée.

En résumé, bien que les modalités responsables de la récupération ne soient pas clairement identifiées, il demeure que pour le même patient différents programmes multimodaux pourraient avoir des effets bénéfiques. Ces programmes peuvent varier dans le choix des approches, leurs séquences, l'ajustement des paramètres de chaque approche appliquée (fréquence, durée, matériel, etc.).

4.4 Contributions à l'avancement des connaissances

Les principales contributions à l'avancement des connaissances concernent directement les cliniciens. L'étude de cas supporte les cliniciens dans leur raisonnement clinique afin d'élaborer un programme multimodal intégrant entre autres une approche de stimulation tactile. Quant à la recension de littérature, elle est la première à faire une synthèse des connaissances sur les approches de stimulation tactile pour traiter une population souffrant d'AM à la main suite à une LNP. Les résultats de cette recension guident les cliniciens dans l'application des approches de stimulation tactile. Cette étude oriente aussi les cliniciens dans l'ajustement des paramètres de ces approches en fonction de la symptomatologie rapportée par leurs patients. Par conséquent, ces nouvelles connaissances acquises par les cliniciens amélioreront l'accès aux soins pour les patients souffrant de DN en ce sens qu'ils devraient se voir offrir ces programmes de stimulation tactile lorsque leur condition le requiert.

La discussion de ce mémoire amène ensuite des éléments intéressants, particulièrement en ce qui concerne le raisonnement clinique. En effet, il est espéré que les réflexions exposées dans ce mémoire guideront les cliniciens dans le choix d'une ou l'autre des approches de stimulation tactile afin de diminuer les douleurs chez une population présentant de l'AM à la

main suivant une LNP. De plus, ce mémoire devrait susciter des réflexions chez les cliniciens, concernant l’élaboration d’un programme multimodal ainsi que les priorités de traitements chez une clientèle avec des problématiques mixtes.

Malgré les études et la discussion présentées dans ce mémoire, il demeure des lacunes dans les connaissances actuelles que ce projet n’a pas permis de combler. En effet, quoique des idées soient avancées concernant le choix d’une ou l’autre des approches, il demeure qu’il n’existe pas de critère objectif précis guidant ce choix. La situation est similaire pour l’élaboration d’un programme multimodal, dont les critères pour en sélectionner les modalités et la séquence dans laquelle les appliquer demeurent inconnus. C’est donc encore à ce jour le raisonnement clinique des cliniciens, c’est-à-dire leur jugement clinique basé sur leur savoir expérientiel, et les données cliniques qui les guide principalement dans leur choix d’approche. De plus, il demeure des incertitudes concernant les mécanismes neurophysiologiques impliqués dans la récupération et donc la diminution de douleur observée. À la lumière de ces informations, il est présentement difficile d’évaluer l’efficacité de ces différentes approches par un essai clinique rigoureux. Ceci est dû au fait que pour faire ce type d’étude, il faudrait que les interventions soient standardisées, surtout en situation d’application d’un programme multimodal. Cela justifie entre-temps l’étude d’un cas clinique unique.

Les deux articles présentés dans ce mémoire discutent de recherches futures en recommandant entre autres des méthodologies plus rigoureuses ainsi que l’évaluation de davantage de variables, telles que la fonction du membre supérieur et la qualité de vie. À la lumière des réflexions de la présente discussion, il est également suggéré que les recherches

futures évaluent davantage les facteurs pouvant influencer l'évolution: la motivation du patient, le degré de participation du patient, etc. Les futures études pourraient tenter de départager les deux approches, à savoir pour quels patients et dans quelles conditions elles sont le plus utiles. Également, l'intégration de ces approches dans un programme multimodal devrait être explorée, afin d'améliorer l'applicabilité clinique des approches de stimulation tactile. Enfin, sachant par expérience clinique, que ces approches sont utilisées avec d'autres populations, par exemple avec les gens présentant de l'AM à la main ou au pied suivant un accident vasculaire cérébral, il serait fort pertinent d'étudier ces approches dans ces contextes de pratique.

Chapitre 5 : Conclusion

Des approches de stimulation tactiles variées sont couramment utilisées en clinique pour traiter la douleur et les troubles sensitifs chez les personnes avec lésions nerveuses. Avec l'étude de cas, l'approche de stimulation tactile par la MRSD a été démontrée dans le cadre d'un programme multimodal avec un patient présentant de l'AM à la main. Puis, avec la recension systématique, l'éventail des approches de stimulations tactiles comme modalités de traitement chez une population présentant de l'AM à la main suite à une LNP ont été répertoriées dans littérature. Deux approches ont été répertoriées, soient la désensibilisation, approche de stimulation directe de la surface allodynique, ainsi que la méthode de rééducation sensitive de la douleur (MRSD), approche de stimulation à distance de la surface allodynique. Les évidences actuelles démontrent qu'il n'y a pas plus de preuve d'efficacité d'une approche de stimulation par rapport à l'autre chez cette population. Néanmoins, nous proposons que les deux approches de stimulation tactiles sont utiles cliniquement. Pour ce faire, ces approches doivent être intégrées dans un programme multimodal visant le traitement des DN, de façon complémentaire aux autres modalités (médicamenteuse, comportementale, exercices physiques, etc.). Le choix d'une approche ou d'une autre doit être guidé en fonction du raisonnement clinique. Ce raisonnement doit tenir compte des caractéristiques du patient (symptômes, signes, valeurs, motivation, etc.), des priorités de traitement, du succès ou de l'échec d'une tentative précédente avec une des approches ainsi que des particularités des protocoles d'application des approches de stimulation tactiles.

Enfin, ces projets réalisés dans le cadre de mon mémoire mettent en relief que les programmes de stimulations tactiles pour améliorer les dysesthésies de la main devraient inclure

des critères d'inclusion et d'exclusion plus stricts basés sur des standards internationaux afin d'obtenir des populations homogènes. Aussi, l'utilisation de mesures avec de bonnes qualités psychométriques incluant des mesures de la performance du membre supérieur et de la qualité de vie devraient être mis de l'avant. Finalement, des critères rigoureux définissant la DN devraient être utilisés. Tous ces éléments supporteront la réalisation d'essais cliniques randomisés de qualité permettant de préciser l'effet thérapeutique des approches de stimulations tactiles pour traiter l'allodynie.

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Annexe 1 : Confirmation de soumission du manuscrit

Quintal I, Carrier A, Packham T, Bourbonnais D, Dyer JO. (2020, en révision). *Tactile stimulation programs in patients with hand dysesthesia following a peripheral nerve injury: a systematic review.* J Hand Ther.

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