

FACTORS INFLUENCING DECISION MAKING REGARDING
INTERVENTION NEEDS FOR INFANTS WITH TORTICOLLIS

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ABSTRACT

Background: Physiotherapy is the standard primary intervention for infants with torticollis, which is a common reason for consultation in pediatric settings.

However, the wide variation of intervention strategies and intensities proposed reflects the lack of clear understanding of these infants' needs.

Objective: To identify factors influencing decision making regarding intervention needs for infants with postural and congenital muscular torticollis.

Methods: Pediatricians' perspectives were gained through open-ended questionnaires, and physiotherapists' perspectives through focus groups and a national survey. Factors were identified and mapped to the International Classification of Functioning, Disability, and Health-Children and Youth (ICF-CY).

Results: All infants presenting with torticollis are thought to require intervention. Factors influencing the determination of intervention needs encompass all ICF-CY domains. Environmental factors represent an important proportion, supporting the importance of family-centered services.

Conclusion: Recommendations for assessment are presented and could guide decision making of therapists in providing optimal care for infants with torticollis.

ABRÉGÉ

Contexte: La variabilité des paramètres d'intervention en physiothérapie pour les enfants ayant un torticolis proposés dans la littérature reflète le manque de compréhension des besoins de cette population.

Objectif: Identifier les facteurs influençant le raisonnement clinique permettant de déterminer les besoins d'intervention des enfants ayant un torticolis postural ou congénital.

Méthode: La perspective des pédiatres (questionnaires qualitatifs) et la perspective des physiothérapeutes (groupes de discussion, sondage) ont permis d'identifier les facteurs influençant le raisonnement et les codes correspondants de la Classification Internationale du Fonctionnement, du Handicap et de la Santé – Enfants et Jeunesse(CIF-EJ).

Résultats: Les facteurs influençant le raisonnement clinique rassemblent des éléments de tous les domaines de la CIF-EJ, avec une emphase importante sur les facteurs environnementaux, supportant l'importance des services centrés sur la famille.

Conclusion: Des recommandations pour l'évaluation de cette clientèle sont présentées afin de guider le raisonnement clinique des thérapeutes dans le choix d'une intervention optimale.

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*Wisdom is to have dreams big enough
not to lose sight when we pursue them*

-Oscar Wilde

*La personnalité créatrice doit penser et juger par elle-même,
car le progrès moral de la société dépend exclusivement de son indépendance*

-Albert Einstein

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FORMAT OF THE THESIS

This thesis is presented using a manuscript-based format, and was prepared in accordance to the McGill Graduate and Postdoctoral Studies guidelines.

Connecting texts were inserted between chapters to guide the reader.

The first chapter of the thesis provides a literature review on torticollis and clinical decision making which guided the rationale and objectives of this study.

The second chapter, a manuscript submitted for publication, presents a structured literature review on assessment tools specifically developed or validated for infants with torticollis.

The third chapter, and second manuscript, is the core of this thesis and reports the study of clinical decision making regarding intervention needs for infants with torticollis.

Finally, a discussion chapter in which the main findings of all precedent chapters are summarized is presented. In this chapter, we also present an assessment strategy for infants with torticollis.

STATEMENT OF AUTHORSHIP

I certify that I am the primary author of all manuscripts contained in this thesis. I claim full responsibility for the content and style of the text here included.

CONTRIBUTION OF AUTHORS

Manuscript 1: *Assessment of infants with postural and congenital muscular torticollis: a literature review*; Julie Fradette, Isabelle Gagnon, Eileen Kennedy, Laurie Snider, Annette Majnemer¹ (submitted to POTP in July 2010)

This manuscript has been submitted to Physical and Occupational Therapy in Pediatrics in July 2010 and is presented in chapter 2 of this thesis in its integral format, with minor editing. Julie Fradette performed the literature search and synthesis. Isabelle Gagnon, Annette Majnemer, Eileen Kennedy, Laurie Snider participated in the design of the search strategy and the interpretation of the findings. All co-authors participated in reviewing the content of the manuscript.

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This manuscript is presented in chapter 3. Appendices and figures that were not included in the submitted manuscript have been added in the present thesis, in order to provide additional information to the examiners and readers. Julie Fradette participated in the design of the study, in data collection for the focus groups, the questionnaires and the survey. She performed data analysis and synthesis. Isabelle Gagnon participated in the design of the study, in data collection for the focus groups, and to the recruitment of participants for the questionnaires and the survey. She also participated in data analysis for all phases. Annette Majnemer participated in the design of the study and in the recruitment of participants. Eileen Kennedy participated in the design of the study and in data collection of focus groups. Laurie Snider contributed to the design of the study. All co-authors participated in reviewing the content of this manuscript.

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CHAPTER 1 – LITERATURE REVIEW AND OBJECTIVES

Preface - This chapter aims at presenting a comprehensive but focused review of the literature, allowing for a better understanding of the rationale and objectives of this study. An overview of the methodology used to pursue the objectives is also presented.

1.0 Characteristics of torticollis

1.1 Definition and classification

Torticollis is primarily a clinical sign involving an abnormal posture of the head and neck. It usually presents as a head tilt in association with a head rotation in the opposite direction. It can be the clinical presentation of many different conditions.¹ Over 80 different pathologies have been reported in the literature to present as torticollis,² such as unilateral absence of sternocleidomastoid (SCM) muscle,³ ocular pathologies,⁴ osseous abnormalities,⁵ tumors,⁶ or neurological findings,⁷ among others. The differential diagnosis of the cause underlying the clinical presentation of torticollis is therefore primordial.

The most common type of torticollis is orthopedic⁸ and involves muscular abnormalities. Three main classifications of orthopedic torticollis have been proposed. Firstly, Cheng et al.⁹ described three clinical types of torticollis that are: sternomastoid tumour (infants with a palpable mass in the SCM muscle), muscular torticollis (infants with no mass but with a tight SCM muscle) and postural torticollis (infants with no mass and no tightness but with a head tilt or rotational preference). Second, some authors consider that orthopedic torticollis in infancy is a continuum ranging from little postural asymmetry ranging up to severe muscle tightness.^{10, 11} The third classification is twofold and consists of: (1) Congenital Muscular Torticollis (CMT) that presents at birth and is characterized by a unilateral shortening of the SCM with or without a fibrous mass, and (2) Postural Torticollis (PT) that is usually observed at later ages than CMT and is a limitation in neck Range of Motion (ROM) leading to the typical head tilt or

rotational preference observed in torticollis. This last classification has been retained for the present thesis because of its frequent use and of the variations reported in the literature in incidence, etiology and intervention for these two types of torticollis.¹²⁻¹⁵

1.2 Incidence

Torticollis is fairly common which translates into an important subset of patients seeking the advice of physicians and other health professionals. At the Montréal Children's Hospital, it is the second most common reason for referral to the physical therapy department with 384 new cases in 2008.

CMT is the third most common congenital musculoskeletal abnormality with an incidence ranging from 0.3% to 2%, which has been stable over time (Colonna 1918¹⁶ and Ballock 1996¹⁷). On the other hand, the incidence of PT is more controversial. While some authors report that the actual incidence for both PT and CMT is between 4% and 6% of live births,¹⁸ others believe that this represents an underestimation of the number of cases due to the lack of clear diagnostic criteria.¹⁹ The incidence could be as high as 16% of live births²⁰ when assessing head preference towards one side at birth; but some authors believe that this rotational preference must persist beyond birth to be considered as a postural torticollis.²¹

Many authors have noted an increased number of referrals for torticollis to pediatricians and physical therapists over the past decades.²²⁻²⁴ Even if this cannot clearly be linked to a higher incidence of torticollis, it is commonly believed that the increased number of cases is associated to the implementation of the Back to Sleep campaign instituted by the American Academy of Pediatrics in 1992.²⁵ This campaign recommended the placement of infants in the supine position during sleep in order to prevent Sudden Infant Death Syndrome (SIDS).²⁶ This led to a decrease in the time spent in a prone position during sleep, but also during awake time, which is probably due to parental misinterpretation of the recommendations

or to the lack of education by health professionals about the application of the recommendations. It has been hypothesized that this decreased time in a prone position was associated with decreased strength of neck muscles making muscle imbalances more apparent and problematic, leading to more frequent diagnosis of PT.²⁷ It is however important to consider that this increased number of reported cases could partly be due to an increased awareness of the problem by physicians.

1.3 Etiology and risk factors

The etiology of CMT and PT is the object of speculation by many authors, and even if no formal hypothesis has been verified, many have been proposed, which differ slightly according to the type of torticollis.

PT is sometimes considered to be caused by plagiocephaly, an oblique deformation of the skull that is highly associated with torticollis. There is a 64-84% co-diagnosis rate of these two conditions²⁸ and plagiocephaly is also reported as the most important risk factor for clinical manifestation of torticollis. Although there is an established relationship between these two pathologies,²⁹ there is some debate in the literature as to whether plagiocephaly is causing torticollis or the opposite. However, one likely contributes to perpetuating the other; a preferred head position leads to flattening of the ipsilateral occiput and unilateral skull flattening makes it more difficult to move out of a preferred head position.

Another hypothesis regarding the etiology of PT is the presence of disequilibrium between both SCM muscles that would lead to the head tilt observed. The limited active ROM could then lead to muscle tightness if the preferred position is maintained for long periods of time.^{10, 11}

The *in utero* malpositioning hypothesis, which applies to both CMT and PT, was based on knowledge of risk factors for these conditions. A constrained intra-uterine environment could lead to a restricted posture of the head, and may therefore predispose to torticollis. This hypothesis is supported by a high

incidence among this population of other orthopedic malformations,^{30, 31} breech presentation, male sex (usually of higher weight and length) and twin pregnancies. The risk factors for developing CMT, including the presence of plagiocephaly (OR 22.3; 95%CI 7.0-71.0), facial asymmetry (OR 21.8; 95% CI 6.6-71.7), primiparity which is associated with a tighter abdominal wall (OR 6.3; 95% CI 2.3-17.0), and greater birth body length (OR 2.0; 95%CI 1.5-2.4) also support this hypothesis.³² Although no study could be found specifically looking at risk factors for developing PT, many authors have identified risk factors for developing plagiocephaly. In a systematic review, Bialocerkowski et al. (2008)³³ summarized the 17 published studies on risk factors for deformational plagiocephaly (see Table 1.1). More recent studies of risk factors, such as the one by Van Vlimmeren et al.³⁴ (cohort of n=380) and the one by McKinney et al.³⁵ (n=2764 cases and 13817 controls) yielded similar results to the ones presented in the review with some variations in odds ratios. Those risk factors are also consistent with the *in utero* malpositioning hypothesis.

Table 1.1 Risk factors associated with deformational plagiocephaly (adapted from Bialocerkowski et al. 2008)

Risk factors	OR (95% CI)
Mother's education	2.5 (1.1-2.7)
Primiparity	2.9 (1.6-5.5)
Prematurity	3.3 (1.0-12.5)
Antenatal education	2.1 (1.4-3.2)
Neck problems (↓ROM)	2.7to 22.0 according to criteria used
Early established head preference	29.7 (8.7-101.0)
Gender (male)	1.9 to 5.4
Cerebrospinal fluid space (larger)	Not available
Inactive infant	2.8 to 3.3
Temperament of the infant	2.6 (1.1-6.3)
Snoring	5.6 (1.6-19.5)
Development	18.1(2.0-166.5)
Position of the infant (time spent in prone)	1.8 to 11.5 according to criteria used
Bottle vs Breast feeding	1.9 (1.2-3.1)
Firm vs soft mattress	2.0 (1.0-3.9)
Assisted delivery	2.5 (1.1-5.7)

Anecdotal reports of several cases of torticollis in some families, and the high incidence of twin pregnancies among infants presenting with torticollis suggested that the development of torticollis could relate to genetic factors.^{36, 37} However, the impact of environment that is similar within families and the constrained intra-uterine environment associated with twin pregnancies are also plausible explanations for the reported prevalence of torticollis within families.

More recently, it was supposed that CMT could be the consequence of a pre- or peri-natal compartment syndrome, a condition in which the blood supply to a muscle is restricted and constricted by connective tissue due to swelling of surrounding tissues.³⁸ The compartment syndrome hypothesis, associated with the higher risk of developing CMT following birth trauma (OR 4.2; 95%CI 1.3-14.5), support the birth trauma hypothesis.

Often called pseudotumor of infancy, or fibromatosis colli, the mass observed in the SCM of some infants with CMT is constituted of fibrotic tissue with undifferentiated myoblasts.³⁹ The pathophysiological process leading to this mass has not yet been elucidated. Histological studies of these masses led to suspicion of infectious, hemorrhagic and ischemic processes in the development of the pathology.⁶ Histological findings, often based on case reports or series, are sometimes contradictory regarding the presence or absence of signs of inflammation or past hemorrhage, making it difficult to appreciate the value of the different hypothesized etiologies.³⁹⁻⁴¹ Some studies have been done on animals to evaluate the possibility of muscle shortening in different conditions, such as infection, ischemia and hemorrhage, but once again results are contradictory.⁶

The etiology of torticollis in infancy remains a controversial subject. A more thorough knowledge of the risk factors and physiopathological processes could lead to a better understanding of the causes of torticollis and therefore enable the development of more appropriate intervention strategies for the prevention and treatment of this condition.

1.4 Clinical presentation

The main feature of CMT and PT is the limitation in ROM; however, problems in different spheres may coexist. In the context of our work, we use the International Classification of Functioning, Disability and Health – Children and Youth version (ICF-CY)⁴² to describe the different clinical features that may present in this population and the contextual factors that can influence the level of functioning of these infants. The ICF-CY is a framework published by the World Health Organization (WHO) that belongs to the WHO family of International Classifications. It describes an individual's functioning across domains of body structures, body functions, activities and participation. These domains of functioning are influenced by the health condition and by personal and environmental factors. The model underlying this classification is presented in Figure 1.1.

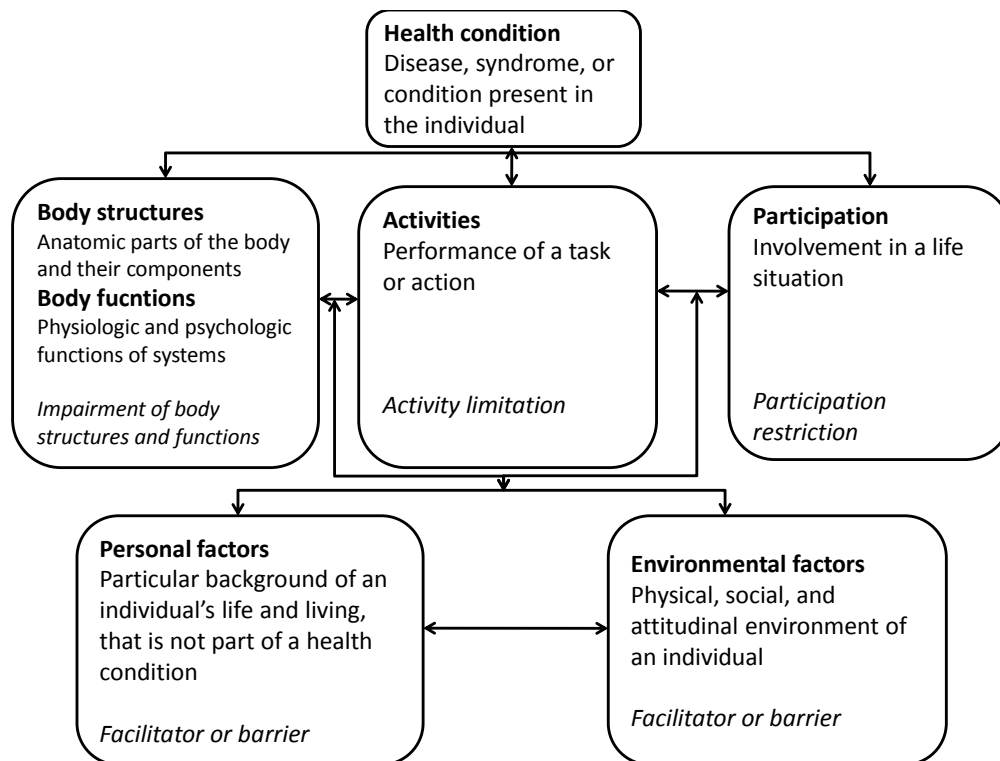


Figure 1.1 International Classification of Functioning, Disability and Health framework (adapted from ICF-CY 2007)

1.4.1 Body Structures

Neck muscles, mainly the SCM, but also peripheral muscles such as the upper trapezium and scalenes⁴³⁻⁴⁵ can show signs of atrophy and interstitial fibrosis.^{2, 39} They may present with a reduced length and flexibility. As a sign of thickening and fibrosis of the SCM, a laryngeal cough reflex can sometimes be induced by cervical rotation in infants with CMT as a result of the compression of the internal branch of the superior laryngeal nerve and of the internal jugular vein.⁴⁶ Skull bones can also be affected either by excessive pulling from the SCM muscle or by the association with plagiocephaly.¹⁰ Apart from plagiocephaly, other facial asymmetries⁴⁷⁻⁴⁹ can occur in infants with torticollis, as well as jaw tilt^{20, 50} and dental deformations.⁵¹

CMT is associated with a higher incidence of hip dysplasia occurring in 4 to 17% of infants with CMT,^{30, 52} while the general population risk is approximately 0.04%.⁵³ Other orthopedic malformations have also been correlated with the occurrence of CMT, such as clubfoot, infantile scoliosis, and pelvic obliquity.^{19, 31, 54, 55}

A few case reports of poor methodological quality also reported abnormalities of the skull base and cranial membranes and vertebral misalignment or subluxation.⁵⁶⁻⁵⁸

1.4.2 Body Functions

In terms of body functions, mobility of the cervical spine is limited⁵⁹ and can be associated with a contralateral hypermobility.⁶⁰ Reduced muscle power¹¹ and endurance⁶¹ of the neck muscles have also been noted in this population.

1.4.3 Activities and Participation

At the activity level, limitations of the ability to control their heads and to maintain various body positions have been reported in these infants. Also, one case-control study⁶² and one observational study¹³ reported that the postural

asymmetry observed in these infants may be associated with an early delay in the acquisition of gross motor abilities, such as rolling, sitting and crawling, when evaluated using norm-referenced tests. The results of Ohman et al. (2009) suggest that the developmental trajectory usually normalizes by one year of age, when conservative intervention is provided. The lack of evidence on participation with this clientele can be attributed to the lack of real expectation of participation at this young age, as the majority of cases present within the first year of life. However, one could postulate that the delays in gross motor abilities could limit participation and integration in daycare. Very few studies have looked at the long-term outcome of these children, providing little information on activities and participation at later ages in this population.

1.4.4 Environmental and Personal Factors

Personal factors affecting the functioning of infants with torticollis could include their age, the presence of associated medical conditions and the child's irritability and intrinsic motivation to move.⁶³ However, to our knowledge, no information is available concerning specific personal and environmental factors that may affect the course of development in infants with torticollis per se. Several studies in children with plagiocephaly have reported the important role of caregiving practices with respect to positioning for sleep and while awake,⁶⁴⁻⁶⁶ and the use of various positioning devices.⁶⁷ Therefore, considering the important relationship between these two pathologies,^{11, 68} it is likely that such environmental factors have also an effect on infants with torticollis. Also, other environmental factors such as the role played by parents in the stimulation of their child and intervention provided by health professionals could reduce the impact of torticollis on functioning of these infants.

1.5 Natural history

It is rare to find literature on the natural history of torticollis because intervention has been recommended as part of the standard care of this condition for many years. Later age at presentation was associated with a poorer outcome in two

intervention studies (n=101, n=821) suggesting that, with time, there is an increasing muscle tightness and less benefits from manual stretching.^{69, 70} Despite this fact, Coventry, in 1959,⁷¹ reported a series of 6 cases who demonstrated a resolution of their torticollis with observation only. However, the criteria for choosing observation without conservative or surgical management for this subset of infants were not described.

The fibrous mass sometimes found in infants with CMT, usually presents within the first weeks of life and resolves spontaneously within the first months.⁷² Although the exact pathophysiological process underlying the "disappearance" of the mass is still unclear, it is known that fibrous tissue is still present after the clinical resolution.⁷³ The fibrotic nature of the muscle leads to its tightness, and therefore limitations in ROM and asymmetrical postures don't necessarily resolve concomitantly with the disappearance of the mass. Infants presenting with a mass may therefore require monitoring beyond the disappearance of the mass.

The long-term impact of torticollis on motor and more global development without treatment is still unclear. Available studies which showed normalization of developmental trajectory by 12-18 months of age provided intervention^{13, 62}; therefore, it is difficult to speculate about the developmental outcome of these infants in the absence of intervention.

If left untreated, torticollis can lead to facial scoliosis, mandibular joint problems, and other cosmetic issues, as highlighted by the few case reports available.^{49, 74-76} While these are not life-threatening, they certainly can have a significant impact on quality of life, as they could persist through the infant's entire life.

2.0 Diagnosis

Although some authors have suggested that all infants be screened for torticollis to ensure early initiation of the intervention,¹⁹ usual pattern of identification and initiation of treatment is either through the report of parental concerns to their

pediatrician or physician, or during follow-up observations as part of well-baby care. In either case, the primary care physician has a pivotal role in the timely diagnosis of infants with torticollis. However, many health professionals can be accessed without referral from a physician and therefore these professionals can also play a role in diagnosis.

As mentioned above, torticollis can be the clinical presentation of a wide range of pathologies, and therefore one of the major roles of health professionals is to diagnose the condition underlying the clinical sign of torticollis. Peyrou et al. (2007) provided guidelines, based on available literature but not formally validated, regarding the diagnosis of torticollis and the tests that should be conducted according to the information available.¹⁴ They suggested that a medical history and a basic clinical examination should be performed to confirm PT or CMT. If these two methods leave the professional with doubts regarding the cause of torticollis, more advanced tests should be performed.

Many diagnostic tests can be prescribed by physicians' to guide differential diagnosis and rule out more serious medical and orthopedic issues. These tests include ultrasound (US), Magnetic Resonance Imaging (MRI), radiographs, computed tomography (CT), and fine-needle biopsy. US has been used by many investigators to describe the features of neck masses in infants using different variables from which two, echo texture and lesion-muscle ratio, are correlated with clinical outcome.⁷⁷⁻⁸³ MRI has been used in a few studies of infants with torticollis, but yet no predictive value or classification can be derived from MRI findings.^{80, 84, 85} Snyder et al.⁸⁶ revealed a positive predictive value of 40% when using radiographs in the diagnosis of torticollis. Given the low prevalence of clinically relevant bony abnormalities, this indicates that radiographs are not often helpful in this population. Van Vlimmeren et al. (2004) suggested that CT imaging be used when an infant presents with (1) an abnormal head shape associated with a normal posture or (2) an abnormal head shape and posture associated with asymmetries of the trunk or lower extremities.⁸⁷ Finally, the use

of invasive methods such as fine-needle biopsy is limited in clinical practice to infants with an unusual presentation of a neck mass.⁸⁸ The information from these tests can be used for prognostication and may guide, to a certain extent, the choice of intervention.

3.0 Intervention

Once a diagnosis of CMT or PT is established, standard management reported in the literature is to provide a conservative intervention,^{2, 15} which will be discussed in section 3.3. Although, conservative care leads to the resolution of torticollis in most cases,^{89, 90} some infants may require other types of intervention presented here under medical management and alternative therapies.

3.1 Medical management

When conservative management fails to resolve torticollis, physicians can use more aggressive strategies such as Botulinum toxin (Botox) injections and surgery. Botox is used to paralyze the SCM and other peripheral muscles. Four intervention studies reported gains in ROM and postural symmetry in children who failed to improve under conservative care and received these injections in conjunction with an intensive home program of manual stretching.⁹¹⁻⁹⁴

Surgery can be used to increase muscle length of the SCM allowing for increased passive ROM. Opinions vary regarding the optimal candidates for surgery.

Although the sole presence of a SCM mass was considered as an indication for surgery before the 1950s,^{48, 95} it is now generally accepted that surgery should be considered only after failure under conservative management. While many authors agree that infants who didn't respond to a course of 6 months of physical therapy should be considered for surgery,^{59, 96, 97} others argue that waiting until later ages may be beneficial because compliance to post-operative recommendations is usually better in older children.^{9, 98, 99} There is a paucity of evidence comparing outcomes following surgery based on different selection criteria. Also, different surgical techniques and post-operative management

strategies have been designed with the aim of optimizing functional and cosmetic status. But the optimal goal is to avoid surgery and currently, only a small subset of children with torticollis requires surgery.

3.2 Alternative therapies

Parents can also seek services from osteopaths and chiropractors for the treatment of their infant with torticollis. Although those alternative therapies are not mentioned in conventional referral guidelines, they are reported in the literature and are part of current treatment options. Chiropractic practitioners use manipulations of vertebrae to realign the spine and also apply soft tissue techniques. Three case reports (n=3) describe a successful resolution of torticollis using this approach.^{57, 100, 101} However, there are risks associated with manipulations of the cervical spine that parents should be aware of when deciding to opt for this therapy.^{102, 103} Osteopathy aims at treating the body as a whole functional unit and at promoting symmetry of the growing child.¹⁰⁴ It is frequently used as an adjunct therapy in the treatment of plagiocephaly, and could also be beneficial in cases of torticollis. One case series is available (n=6) on parasacro-coccygeal pressure but the study's methodology isn't described in enough detail to evaluate its quality.¹⁰⁵

3.3 Conservative management

As mentioned previously, conservative management is the standard care recommended for infants with torticollis. General objectives of conservative management include attaining full ROM, promoting symmetrical movements and postures, and preventing secondary flattening of the skull. Different health professionals can provide this type of intervention, but in current practice, it is physical therapists who will provide conservative management. Physical therapy is the most frequently reported conservative care for infants with CMT and PT in the literature.

Van Vlimmeren et al.⁸⁷ proposed guidelines for the management of torticollis, based on preliminary evidence but not on formal validation, suggesting that, if the infant presenting with PT is younger than 2 months or if a CMT is diagnosed, the infant should be referred to physical therapy. If the infant presents with PT at later ages, the parents should be given advice about positioning and be provided with an exercise program by their physician, and with reassessment three months later. If the asymmetry is not resolved at this time, the infant should then be referred to physical therapy.

Despite the recognition of physical therapy as the primary intervention for torticollis, the level of evidence regarding its effectiveness remains low, with no randomized controlled trial (RCT) available. The observational studies presented in the literature report a facilitated resolution of ROM limitation, head tilt and gross motor function delay. Many studies, however, focused on the need for surgery following conservative management providing little information on more specific outcomes for this population. Strategies currently used in the physical therapy management include manual stretching, specific handling and positioning strategies, active and passive ROM exercises, and a neuro-developmental approach to symmetry.⁹⁰ Literature also report the use of massage of the affected SCM, ultrasound and micro-current.^{46, 90, 106} A review of the literature on conservative intervention was performed. The following databases have been searched: PEDro (1929- May 2010), The Cochrane Library (1994-2010), CINAHL (1996-2010) and MEDLINE (1950- May2010). Keywords and subheadings (MeSh) used for the search of the databases were: (Torticollis MeSH, torticollis, Fibromatosis colli, positional preference). The titles and abstracts of English and French articles were screened and those mentioning the study of an intervention were retained. Then, retained articles were screened to retain solely those treating of conservative interventions. The studies that evaluated the effectiveness of conservative strategies are presented in Table 1.2 in a reversed chronological order (see next page).

Table 1.2. Observational studies and case reports on the effectiveness of physical therapy in infants with postural and congenital muscular torticollis

Abbreviations used in the present table: AIMS = Alberta Infant Motor Scale; AROM=Active Range of Motion; CMT= Congenital Muscular Torticollis; dev=development; DP= deformational plagiocephaly; HP=home program; min = minutes; LF=lateral flexion; MS>manual stretching; NDT=Neuro-developmental therapy; Phys ther = Physical therapy; PROM= Passive Range of Motion; PT= postural torticollis; ROM= Range of Motion; sec=seconds; TOT= Tubular Orthosis for Torticollis; tx=treatment; x=times.

Authors	Objective	Sample	Intervention	Comparison	Outcome measure	Results	Comments
Kim et al. (2009) ¹⁰⁶	Compared the effectiveness of micro-current to usual physical therapy	15 infants referred to phys ther for CMT (mean age at presentation 7.1 months)	30 minutes of microcurrent 100 μ amperes 8hz+2 minutes of MS 3x/week for 2 weeks Tx performed while infant asleep	30 minutes of phys ther ROM exercises, postural training and MS 3x/week for 2 weeks	Rotation PROM with protractor, head tilt in supine, muscle function scale and number of infants who cried	Head tilt decreased more in microcurrent group $p<0.01$ Rotation PROM improved more in microcurrent group $p<0.05$ Fewer infants cried	Method of attribution to groups not specified. Baseline comparison of groups was not available, except for age which was different between groups (infants in the experimental group significantly older)

Authors	Objective	Sample	Intervention	Comparison	Outcome measure	Results	Comments
Yim et al. (2009) ⁴⁶	Reported the outcome of infants presenting a cough reflex	24 infants with CMT presenting with a cough reflex sign (mean age at presentation 37.6 days)	5 min of ultrasound (no mention of parameters) + 20 min. of MS (1 sec+5-10 sec rest) 5x/week HP of MS by caregiver 1-2 x/day	No control group	Resolution defined as ROM in rotation 90° (independent of mass or other symptom resolution)	22/24 infants recovered full ROM within 19.2 days (mean) 2/24 underwent surgery; all 24/24 stopped showing cough reflex	Authors conclude that cough reflex could indicate that more vigorous intervention is needed although no comparison of intensity was made
Rubio et al. (2009) ¹⁹	Established the prevalence of moulded baby syndrome at birth and reported the effectiveness of early advice provided by a pediatrician	65 infants screened at birth for moulded baby syndrome with either DP, head rotational preference or head tilt	Advice on positioning and AROM exercises (no mention of parameters)	No control group	Follow-up at 2-3 months by phone, asking the parents about changes	Of those with head rotation preference 82% resolved, 3% stayed the same and 15% worsened Of those with plagiocephaly and head tilt 25% resolved, 33% improved, 17% stayed the same and 25% worsened	This suggests that a single session of advice and teaching of exercises may not be sufficient in an important proportion of infants. Outcome measure may be less reliable

Authors	Objective	Sample	Intervention	Comparison	Outcome measure	Results	Comments
Van Vlimmeren et al. (2008) ¹⁰⁷	Compared the outcome of physical therapy to general advice	65 infants with positional preference (no CMT) identified through screening at 7 weeks	8 sessions of Phys Ther between 7 weeks and 6 months Exercises and advice on positioning and handling Stimulation of global dev, play time in prone	Leaflet provided to controls with preventive measures to avoid positional preference. Randomisation of participants with sex stratification	Plagiocephalometry (standardized measure of DP), AIMS, Bayley Scales of Infant Dev II, PROM of the neck estimated visually	PROM and motor dev similar between groups. Caregivers in the treatment group demonstrated more symmetry in positioning and handling.	RCT with standardized outcome measures with primary focus on resolution of plagiocephaly
Schertz et al. (2008) ¹⁰⁸	Described the level of risk for developmental delays in infants with torticollis receiving physical therapy	101 infants with torticollis (PT and CMT), (mean age at presentation 2.9 months)	Phys ther once a week: MS (no mention of parameters) + developmental therapy with daily home program	No control group, used norms from the AIMS	AIMS, dev. quotient of cognitive function using the CAT-CLAMS, PROM of the neck, qualitative description of posture, hip and neck sonograms	Significant difference in number of infants with normal dev post-intervention. Cognitive scores within normal. Postural type associated with dev. delay	Use of controls rather than norms would have been better given that norms may be outdated because of changes related to sleep positioning. ^{62, 64}

Authors	Objective	Sample	Intervention	Comparison	Outcome measure	Results	Comments
Cottrill et al. (2007) ¹⁰⁹	Compared the outcome of infants treated with TOT collar and those receiving manual stretching	10 infants with CMT (no mention of the age of participants)	TOT collar and MS (no parameters specified)	MS alone	Head tilt measured using photographs and digitalization software	Mean head tilt improvement: TOT group 8.5°, control group 3.9° (No further statistical analysis performed)	Lack of methodological information provided (selection of subjects, method used to assign participants to groups, etc.)
Tatli et al. (2006) ¹¹⁰	Compared the outcome of physical therapy in infants with CMT to those with PT	311 infants with CMT or PT; 61% aged 0-6 weeks 39% aged 6-24 weeks	Positioning, MS (no mention of the duration of stretches) (5 series of 20 repetitions everyday)	Compared infants with CMT to those with PT	Recovery of symptoms (cosmetic, ROM)	95% with complete resolution (Defined as 5° or less of limitation in ROM)	Criteria used for resolution not necessarily representative of current practice
Rahlin (2005) ¹¹¹	Reported result of TAMO therapy in one infant	1 infant with CMT 4.5 months of age at presentation	TAMO therapy consisting of: NDT, active ROM, soft tissue mobilisation and HP	No control group	ROM measured using visual estimates, head posture at rest	"Completely resolved"	This is a case report, the positive results must be interpreted with caution.

Authors	Objective	Sample	Intervention	Comparison	Outcome measure	Results	Comments
Celayir et al. (2000) ¹¹²	Described outcome of early initiation of physical therapy	45 infants with CMT <4 months of age at presentation	HP of MS with 2 individuals 10x10 sec. each MS every 3 hours	No control group	Resolution (defined as no asymmetry and no limitation in rotation, ROM; estimated visually)	100% resolution of cases. Mean duration of treatment = 3.8 months	High rate of success but treatment has little external validity
Cheng et al. (2000) ⁴³	Reported the effect of clinical aspects on the outcome of physical therapy	1086 infants with PT and CMT aged 0-1 year at presentation	See Cheng et al. 1999 (next page) for treatment protocol; 24.5% received home program and 75.5% received MS	No control group. Various comparisons within the sample	Composite overall score, classification of ROM limitation (see Cheng et al. 1999) Clinical type of torticollis	Association between clinical type and overall outcome and between duration of treatment and ROM limitation	Prospective design Standardized assessment of ROM
Demirbilek et al. (1999) ¹¹³	Reported the effect of age at presentation on the outcome of physical therapy	57 infants with CMT and a SCM mass aged 0-7 years at presentation	HP of MS with 2 individuals Active exercises 40 repetitions 4-5x/day	No control group; compared different age groups at initiation of treatment	Need for surgery (persistence of symptoms > 3 to 6 months was considered as an indication)	0-3 months (0% needed surgery) 3-6 months (25%) 6-18 months (71%) 2-7 years old (100%)	Retrospective chart review design. No consideration of compliance with the intensive home program

Authors	Objective	Sample	Intervention	Comparison	Outcome measure	Results	Comments
Cheng et al. (1999) ¹¹⁴	Reported the effect of ROM limitation on the outcome of physical therapy	510 infants with CMT with a SCM mass, aged 0-1 year at presentation	HP of active positioning for infants with no limitation in ROM (10.8%) MS by therapist for infants with limited ROM (89.2%)	Compared different levels of ROM limitations (I- no limitation in passive, II- <15°, III- 16-30°, IV->30°)	Composite overall score: Excellent, Good, Fair or Poor, based on head tilt, side flexion and rotation deficit, residual band, parental assessment , and facial asymmetry	Overall score: Excellent 75.6% Good 15.1% Fair 2.6% Poor 6.7%. No correlation between ROM limitation group and overall outcome	Prospective design. Criteria for surgery provided
Ho et al. (1999) ¹¹⁵	Reported the result of manual stretching	91 infants with CMT aged 0-8 years (mean age, 6 months)	MS performed by therapist and parents (no mention of parameters)	No control group	Overall result of functional and cosmetic aspects: Good = both satisfactory Fair =either one unsatisfactory Poor = both unsatisfactory as judged by physician	Good 34.7%; Fair 31.9%; Poor 33.3%. 20 patients (21.9%) underwent corrective surgery	Retrospective chart review design. Lack of methodological information. 72/91 patients had follow-up data. Sample may not be representative of what is typically encountered in clinic

Authors	Objective	Sample	Intervention	Comparison	Outcome measure	Results	Comments
Taylor et al. (1997) ⁸⁹	Reported the result of an individualized home program	23 infants with CMT or PT Mean age of 3.8 months at presentation	Positioning and strengthening HP customized to each child (no standard parameters)	No control group	Composite index (head tilt, ROM, parental appreciation) Assessment procedures not provided	96% of infants had good to excellent results on the composite index	Lack of details on the treatment provided. Basis of individualization of intervention not provided
Cheng et al. (1994) ¹¹⁶	Reported the effect of initial ROM limitation on the outcome of phys ther	624 infants with CMT or PT (71.6% aged 0-3 months; 28.4% aged 4-12 months)	MS + active exercises, head righting daily Positioning at home	Compared different levels of ROM limitations (Cheng, 1999)	Treatment period (months)	Correlation between ROM limitation and treatment duration (Spearman $r=0.23$)	Retrospective chart review design. Standardized assessment
Emery (1994) ⁷⁰	Reported the effect of clinical factors on the outcome of phys ther	100 infants with CMT from 0-2 years of age	HP of MS requiring 2 individuals(LF+rot) 5x10 sec. 2x/day Positioning and handling suggestions. TOT collar if head tilt > 6° (30%)	No control group	Resolution of torticollis, ROM using goniometry and head tilt Duration of treatment	99% fully recovered Mean duration of tx 4.7 months (SD=5.1)	Author analysed factors influencing the duration of treatment. Those factors are discussed above in the text.

Authors	Objective	Sample	Intervention	Comparison	Outcome measure	Results	Comments
deChalain et Katz (1992) ¹¹⁷	Reported the effectiveness of physical therapy in preventing surgery	134 infants aged 0-9 months with CMT	HP of MS (LF+rot) parameters not mentioned Regular outpatient visits	No control group	Need for surgical intervention	60% resolved; 36% were lost to follow-up; 4% required surgery	Retrospective chart review design. Lack of methodological details. Criteria for surgery not described.
Binder et al. (1987) ¹¹⁸	Reported the effectiveness of conservative care in preventing surgery	85 infants with CMT from which 38.6% had a mass; 81.6% presented before 6 months of age	MS (neck and trunk) by parents Positioning and handling advices provided Soft collar if head tilt >45°	No control group	Resolution (defined as full passive ROM without resistance and no head tilt at rest)	70% of cases resolved if presented < 1 year of age 5% required surgery. No association found between recovery and fibrous mass	Outcome not clear in the remaining 25% of cases. No criteria provided for the need for surgery. Retrospective design.

Authors	Objective	Sample	Intervention	Comparison	Outcome measure	Results	Comments
Leung and Leung (1987) ¹¹⁹	Follow-up study that evaluated the long-term outcome of infants treated for torticollis	67 children who were treated for CMT as infants (mean follow-up of 6.5 years)	MS (LF+rot) performed by the therapist. Many repetitions per session. Treatment sessions at least 2x/week. Advice on positioning and massage	No control group	ROM using adapted goniometers Facial asymmetry rated subjectively. Overall assessment judged with Ling criteria (see below)	60% had full ROM and 12% had more than 10° of limitation when compared with contralateral side. Overall result in those with facial asymmetry at presentation: Good 15%, Fair 59%, Poor 26% Without facial asymmetry at presentation: Good 47.5% Fair 50% Poor 2.5%	No mention of criteria for discharge. Children followed may not be representative of the sample treated (67 out of 206 children were assessed)

Authors	Objective	Sample	Intervention	Comparison	Outcome measure	Results	Comments
Morrison and MacEwen (1982) ¹²⁰	Reported the outcome of conservative management	232 infants with CMT aged 0 to 6 years. 82% diagnosed before 3 months of age	HP of MS (LF+rot, flexion) at diaper change. Positioning advice. Heat +massage. Surgery if required	No control group	Overall result judged as: Excellent, Good or Poor (based on head tilt, mass, facial asymmetry)	Of those who didn't require surgery (84%) 71% excellent, 29% good	Retrospective chart review design. Age at presentation was associated with need for surgery
Ling et Low (1972) ¹²¹	Reported characteristics of infants presenting with CMT and the effect of the presence of a SCM mass on the outcome following conservative management	108 infants with data available on treatment (out of 150 infants reported in the study) – CMT with or without mass. 84.9% of the sample presented before 3 months of age	HP of MS (parameters not described)	Compared infants with and without a SCM mass	Overall result judged as: Satisfactory – normal or mild facial asymmetry with mild tightness of SCM; Poor – severe facial asymmetry with tightness of SCM	Mass group: 77% satisfactory; 23% poor 2% required surgery. No mass group: 50% satisfactory, 50% poor 42% required surgery, 8% declined surgery	Retrospective chart review design. 78% of the sample had a mass and infants without a mass were presenting at later ages which may have influenced the results

As observed in this table, the treatment program characteristics used in the different studies vary widely. For example, the treatment frequency ranges from therapy once every 3 weeks to stretching every 3 hours, with no rationale for these choices and none of these specific approaches has been proven to be more effective. Only one research team selected frequency of intervention based on the severity of the case. Cheng et al. (1994, 1999, 2000)¹¹⁶ divided their sample based on the ROM of the neck and assigned different treatment approaches and frequencies accordingly. They found positive results with both approaches.

Although ROM is an important indicator of the severity of torticollis, other factors influence the decision about need for intervention in clinical practice. Luxford et al. (2009) report in a recent survey of pediatric physical therapists that perceived parental competency, the family's daily routine, and the physiotherapist's preference for particular techniques influenced the selection of the parameters of stretching exercises for infants with torticollis.¹²²

Determinants of the duration and the outcome of physical therapy have been identified in the literature and include the extent of limitation in ROM, the presence of a mass in the SCM, age at presentation, male sex, side of involvement (right), and difficulties at birth.^{69, 70} Such determinants of outcome, and contextual factors, such as those identified by Luxford et al., may be important aspects to consider when deciding about the intensity of intervention to provide. However, unless those hypotheses are tested clinically, one can only speculate as to the extent to which these factors should influence the decision about intervention intensity and format.

The available literature on intervention for CMT and PT is confusing and of poor to moderate methodological quality. Physical therapists are therefore left with little sound evidence to guide their clinical decision making about the treatment that should be provided to infants with torticollis. Clinical decision making is a complex process that can be influenced by many factors. Little is known about

what is currently influencing the thought processes of therapists practicing with infants with torticollis but it has been studied in other populations and deserves further attention.

4.0 Clinical decision making

Clinical decision making is the thought process leading to the identification of need for intervention and to the choice of the specific intervention to provide to a patient within a given setting.¹²³ Many sources are available to inform those decisions such as the clinician's personal experience, leading expert opinions, and anxieties of the therapists.¹²⁴ Experience is one of the bases on which decisions can be made. Embrey et al. (1996) studied the differences in clinical decisions of experienced and novice pediatric physical therapists working with children with cerebral palsy using qualitative methodology.¹²⁵⁻¹²⁷ They found that experienced therapists showed more rapid adjustments during intervention, that their schematic representations of movement were more elaborated and that they showed more sensitivity to emotional and social needs than novices did. However, personal experience must not inappropriately outweigh higher levels of evidence. The use of experience may sometimes lead to erroneous belief of effectiveness based on previous positive results in a subset of patients while the majority of patients may not respond to the intervention. Alternatively, experience might confuse the effect of natural recovery with the effect of treatment.¹²⁴

Recently, an important emphasis has been placed on the benefits of using scientific evidence in decision making of health professionals. Evidence-based practice (EBP) is defined by Sackett et al. (1996 p.1) as "the conscientious, explicit, and judicious use of current best evidence in making decisions about the care of individual patients. The practice of evidence based medicine means integrating individual clinical expertise with the best available external clinical evidence from systematic research [...] with a thoughtful identification and compassionate use of individual patients' predicaments, rights, and preferences in making clinical decisions about their care. " ^{128, 129}

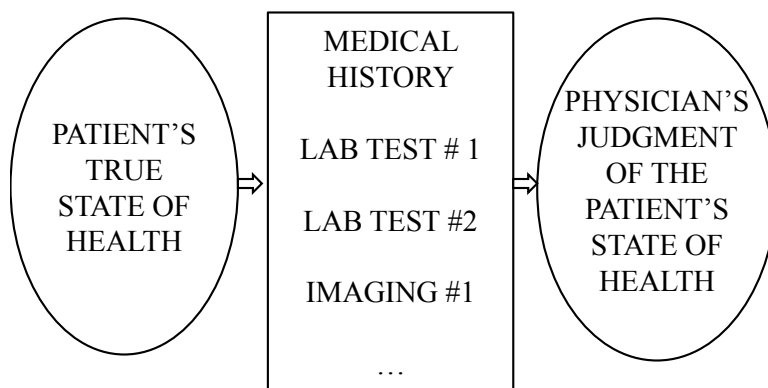
Research have shown that the use of algorithms in health care decision making are more reliable than clinical judgement alone.¹³⁰ Those algorithms use a statistical approach, such as Bayesian statistics, to evaluate the probabilities of different outcomes and weigh their respective impact on patients. In such algorithms, the patient can be involved to specify the importance that a given outcome has for them; this is also called utility.¹³¹ This can also translate into cost-benefit analyses. The clinical applicability of such methods has been questioned, as it is sometimes difficult to know all of the possible outcomes and to estimate accurately and reliably their probability of occurring. It is also considered by some to remove humanity and art from the profession. Schwartz reunified these two visions arguing that: "the art of medicine is the skilled application of medical science (p.23) ".¹²³

Different methods have been proposed to apply algorithms to medical problems. Decision analysis is one of these methods. It consists of delineating the problem into a decision tree and writing down the probabilities of all outcomes.¹³² Rothstein et al. proposed a systematic approach to guide physical therapists' decision making.¹³³ The Hypothesis-oriented Algorithm for clinicians II proposes to identify potential and existing problems, to formulate hypotheses as to their cause, to formulate intervention plans addressing the causes and to document the outcomes of the intervention. This methodology can be applied to many types of pathologies but doesn't necessarily facilitate the use of evidence.

One key element of decision making, whether or not using algorithms, is the cautious gathering and interpretation of data on patient problems. The American Physical Therapy Association, in its Guide to Physical Therapist Practice, proposes a model of patient management in which the steps leading to the determination of intervention are defined.¹³⁴ The first step consists of examination which is the process of obtaining a history, performing relevant systems reviews, and selecting and administering specific tests and measures to obtain data. Then

the therapist performs the evaluation that is a dynamic process in which the physical therapist makes clinical judgements based on data gathered during the examination. The evaluation is followed by the diagnosis which is the organisation of data to determine the prognosis and the most appropriate intervention strategies. After, the intervention, outcomes are used to measure the results, in different domains, of the patient management. This terminology will be used in the present thesis.

Schwartz applied the Lens Model, proposed by Egon Brunswick in *The conceptual framework of Psychology*,¹³⁵ to clinical decision making. This model illustrates that the doctor's judgment of a patient's health is made through the interpretation of medical data (e.g. medical history, physical exam, lab tests) which may be a more or less accurate representation of the patient's true state of health (see Figure 1.2). This highlights the importance of valid and reliable examination tools to accurately judge patient attributes. This is also consistent with the application of EBP that requires the use of psychometrically sound outcome measures to determine if the desired outcome is achieved.¹²⁴



**Figure 1.2 Lens model applied to clinical decision making by Schwartz
(Adapted from Schwartz 1986)**

Historically, physical therapists have focused their examination on physical aspects of functioning.¹³⁶ However, more recent conceptual frameworks have broadened the perspective. Mattingly studied the thought processes of therapists working with children and youth and found that five domains were taken into consideration: 1) understanding of the child's motivation, commitments and tolerances, 2) the environment in which the task is taking place, 3) knowledge of the child's physical and cognitive deficits and capacities, 4) perception of the therapeutic relationship, and 5) immediate and long-term goals.¹³⁷ These domains also reflect the increasing importance of family-centered care.¹³⁸ Family-centred care consists of values, attitudes, and approaches to health services recognizing that each family is unique; that the family is the constant in the child's life; and that they are the experts on the child's abilities and needs.¹³⁹ In this model of care, that was developed through advocacy of parents of children with disabilities and now supported by legislation in the United States,¹⁴⁰ health professionals should work collaboratively with families to determine the family's strengths and needs that will then guide the shared decision making regarding the services and supports required. Such care can easily be integrated within EBP, because EBP requires that patient preferences be taken into account. Moreover, there is evidence that patients who are involved in their own decision making are more likely to adhere to treatment regimens and experience better health outcomes.^{141,}¹⁴² For children, but more especially for infants, the parents are often the ones providing treatment to their child, and therefore should be integrated in the decision making process.

Research on clinical decision making demonstrates that a careful evaluation of probabilities and risks of given outcomes, taking into account patient preferences and evidence, is the best strategy to use when deciding about the type and frequency of intervention to provide. This evaluation of probable outcome should be based on the most valid and reliable tests to ensure the accuracy of the representation of the patient state of health and functioning. In pediatric settings, environmental context and family factors should be part of the decision making

equation. Currently, there is no available literature regarding the specific variables that should be considered in decision making for infants with torticollis.

Physical therapists working with infants with torticollis must make decisions about treatment that might affect the infant's outcome, and more specifically his or her level of functioning. These decisions include: determining which infants would benefit from physical therapy, expected outcomes of the intervention, the type of modalities and treatment parameters that should be applied to obtain the desired outcomes, and the degree to which parents will be expected to carry out the treatment of their infant.^{143, 144} Current clinical management of this population reveals that the perceived need for intervention vary widely among infants with CMT and PT. As part of preparatory work performed prior to the design of our study, the physical therapy charts of a consecutive sample of 293 infants treated for torticollis between April 2007 and November 2008 at the physical therapy department of the MCH were reviewed. The number of visits for physical therapy ranged from 2 to 19 per infant and the duration of therapy ranged from 20 days to 3 years. This wide level of variability suggests that not all infants are thought to require the same intensity of intervention. Greater understanding of the clinical decision making process used by physical therapists working with this population could help identify the specific variables considered in determining the intervention needs of this population.

5.0 Summary and objectives

Torticollis is a common problem encountered by pediatric physical therapists with many cases presenting in outpatient clinics. Indeed, it represented the second most common reason for consultation to the physical therapy department of the MCH in 2008. Physical therapy is considered as the standard primary intervention for infants with this condition. Although the literature on the effectiveness of this intervention is scarce and of poor to moderate quality, authors consistently report good outcomes. The optimal intervention strategies to resolve torticollis in a timely manner, however, remains unclear.

Clinical decision making literature suggests that standardized and reliable measures be used in the examination of the patients' condition to accurately describe their state, to guide the choice of intervention and to facilitate analysis of effectiveness of treatment on outcomes. Therefore, the best examination tools and outcome measures must be identified for our population of interest, to guide clinical decision making. In order to enlighten the design of the main study of this thesis, a literature review of examination tools and outcome measures that are specifically designed or validated for the torticollis population was undertaken and is presented in chapter 2. This review provides a summary of the type of attributes that are currently evaluated, as part of the decision making process, and illustrates the lack of consensus, and more importantly, the lack of adoption of a broad holistic approach to assessment.

The ultimate objective of this thesis is to identify the factors that should be considered in the decision making process of clinicians when determining the intervention needs of infants with postural and congenital muscular torticollis. In order to address this objective, we conducted a study to gain the perspective of pediatricians and physical therapists on intervention needs of infants with PT and CMT. A qualitative inquiry of the perspective of pediatricians was performed through open-ended questionnaires. Focus groups and a national survey were used to gain understanding of pediatric physical therapists' perspective.

The knowledge generated from this study could then guide the development of an examination strategy for this population of interest which would be particularly helpful to novice therapists for whom it may be more difficult to identify all aspects of the infant's condition and environment that should be considered in the determination of the level of need for therapy. Many physical therapists, as well as physical rehabilitation technicians, work alone in remote areas and may only occasionally assess infants with torticollis. With the increased incidence of torticollis, it is important to ensure that therapists in all treatment settings have the necessary information to make informed decisions about treatment needs.

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CHAPTER 2 - ASSESSMENT OF POSTURAL AND CONGENITAL MUSCULAR TORTICOLLIS: A LITERATURE REVIEW

Preface – The previous chapter highlighted the importance of using psychometrically sound assessment tools to guide decision making. The broad clinical presentation of torticollis was presented and suggests that multiple assessment tools or comprehensive measures are required to accurately depict the status of infants presenting with torticollis. This chapter presents a review of the assessment tools reported in the literature that have been specifically developed or validated for the torticollis population.

ABSTRACT

Introduction: Torticollis is a frequent reason for consultation in pediatric physical therapy. Physical therapists should consider different aspects in the choice of assessment tools to make evidence-based decisions about intervention for this population.

Purpose: To report assessment tools designed or validated for infants with torticollis and their psychometric properties.

Methods: A comprehensive literature review has been performed using specific keywords in PEDro, The Cochrane Library, CINAHL and MEDLINE.

Results: Out of the 2968 articles screened, 26 articles on 18 assessment scales or techniques were retained and analysed using a standardized method. ROM and muscle strength of the neck can be assessed using standardized and reliable methods for which norms are available. Techniques to assess asymmetry and the aspect of neck muscles have also been described. Four composite measures, which assess multiple aspects, are available, but none have been tested for their psychometric properties.

Discussion: A comprehensive assessment of torticollis is needed to reflect the complexity of the clinical presentation. Physical therapists may therefore have to resort to the use of tools designed for more generic populations to complement the information provided by the tools proposed in the literature to describe the infant's state.

1.0 Introduction

1.1 Definition and classification of torticollis

Torticollis is defined as an asymmetrical posture of the head and neck associated with a limitation in active or passive neck range of motion (ROM).¹ The two most common causes in infancy are Congenital Muscular Torticollis (CMT) and Postural Torticollis (PT). CMT is characterized by a unilateral shortening of the sternocleidomastoid (SCM) muscle with or without a fibrous mass² and is often associated with birth trauma or *in utero* malpositioning.³ PT is hypothetically related to an asymmetrical stimulation of the infant in its environment and to deformational plagiocephaly,^{4, 5} an oblique deformation of the skull without fusion of the cranial sutures.⁶ In PT, the infant's neck muscles can present with or without a unilateral shortening, but without any fibrous mass.

1.2 Incidence

CMT is the third most common congenital musculoskeletal abnormality with an incidence ranging from 0.3% to 2%, which has been stable over time (Colonna, 1918)⁷ and (Ballock & Song, 1996)⁸. On the other hand, the incidence of PT is more controversial. While some authors report that the actual incidence for both PT and CMT is between 4% and 6% of live births,⁹ others believe that this represents an underestimation of the number of cases due to the lack of clear diagnostic criteria.¹⁰ The incidence could be as high as 16% of live births¹¹ when assessing head preference towards one side at birth, but some authors believe that this rotational preference must be constant and present at later ages to be considered as a postural torticollis.¹² Many authors have noted an increased referral rate to pediatricians and physical therapists over the past decades.^{4, 13, 14} Even if this cannot clearly be linked to a higher incidence of torticollis, it is commonly believed that the increased number of cases is linked to the implementation of the Back to Sleep campaign instituted by the American Academy of Pediatrics in 1992.¹⁵ This campaign recommended the placement of infants in the supine position during sleep in order to prevent the Sudden Infant Death Syndrome (SIDS).¹⁶ Some authors hypothesized that the decreased time in

a prone position during sleep, but also during awake time, was associated with a decreased strength of neck muscles making muscle imbalances more apparent and problematic, leading to more frequent diagnosis of postural torticollis.¹⁷ It is however important to consider that this increased number of cases could partly be due to an increased awareness of physicians towards this problem.

1.3 Clinical presentation

Although limited ROM of the neck is the most striking clinical feature of both CMT and PT, torticollis can have an impact on different aspects. When looking at the available literature, we find that neck muscles, mainly the SCM, but also peripheral muscles such as the upper trapezium and scalenes,^{2, 18, 19} can show signs of atrophy and interstitial fibrosis^{3, 20} and have a reduced length and flexibility resulting in a limited ROM of the cervical spine²¹ which can be associated with a contralateral hypermobility.²² Reduced muscle power²³ and endurance²⁴ have also been noted in this population. As a sign of thickening and fibrosis of the SCM, a laryngeal cough reflex can be induced by cervical rotation in some infants with CMT as a result of the compression of the internal branch of the superior laryngeal nerve and of the internal jugular vein.²⁵ Skull bones can also be affected either by excessive pulling from the SCM muscle or by the association with plagiocephaly. Infantile torticollis is strongly associated with plagiocephaly with an estimated 64-84% co-diagnosis rate.²⁶ CMT is also associated with a higher incidence of hip dysplasia; in fact, from 4 to 17% of infants with CMT will develop this,^{27, 28} while the general population risk is approximately 0.04%.²⁹ A few case reports of poor methodological quality also reported abnormalities of the skull base and cranial membranes, and vertebral misalignment or subluxation.³⁰⁻³²

At a functional level, one case-control study³³ and one observational study,³⁴ reported that the postural asymmetry observed in these infants may also be associated with a early delay in the acquisition of gross motor abilities, such as rolling, sitting and crawling, when evaluated using norm-referenced tests. The

symmetry of the movements and positions is lacking and may perturb the normal course of the development. However, the developmental trajectory usually normalizes by one year of age when a conservative intervention is provided.^{33, 34} The lack of evidence on participation with this clientele may be attributed to the lack of assessment tools for participation at this young age, as the majority of cases present within the first year of life. However, one could imagine that the limited gross motor skills could limit participation and integration in daycare. Moreover, very few studies have looked at the long-term outcome of these children, providing little information on participation at later ages in this population.

Factors relating to the environment in which the infant evolves could also affect the outcome of torticollis but information regarding these factors is lacking in the torticollis population. However, in the plagiocephaly population, several studies have reported the important role of positioning for sleep, and more specifically for playtime while awake,³⁵⁻³⁷ as well as the use of various positioning devices.³⁸ Therefore, considering the important relationship between these two pathologies,^{23, 39} it is likely that such environmental factors could also have an effect on infants with torticollis. Also, the role that parents play in the stimulation of their child, prevention policies as well as interventions provided by physical therapists and other health professionals could reduce the impact of torticollis on functioning of these infants. In terms of personal factors, the child's irritability and intrinsic motivation to move⁴⁰ could influence the outcome of therapy.

1.4 Rationale and objective of the review

Physical therapists are faced with a significant number of referrals for torticollis.^{4, 14} It is one of the most common reasons for consultation to pediatric physical therapy outpatient clinics. Because the impact of a torticollis in infancy may be multi-dimensional and not limited to cervical ROM impairment, tools and scales that are comprehensive and specific to the torticollis population are essential for professionals involved in the care of those infants. Indeed, the use of standard,

valid and reliable tools is key for clinical decision making regarding optimal interventions as well as for assuring sound research methodology with this population. The primary purpose of this review was therefore to identify assessment tools, scales or checklists that have been developed or validated to measure domains and factors influencing functioning in infants with postural and congenital muscular torticollis. The second objective was to describe the psychometric properties of the identified assessment methods in order to facilitate physical therapists' and researchers' choice of appropriate examination tools for this population.

2.0 Methods

The following databases have been searched by the principal author (JF): PEDro (1929- May 2010), The Cochrane Library (1994-2010), CINAHL (1996-2010) and MEDLINE (1950- May2010). Keywords and subheadings (MeSH) used for the search of the databases were: (Torticollis MeSH, torticollis, Fibromatosis colli, positional preference, Plagiocephaly Non synostotic MeSH, plagiocephaly, and Craniosynostoses MeSH). Plagiocephaly has been included in the search strategy because of the important number of studies reporting data on infants having both plagiocephaly and torticollis. Articles relating to craniosynostosis have also been screened because the term plagiocephaly was not indexed prior to 1990. A screening of the general literature of torticollis revealed that the use of keywords such as outcome, measure, assessment or psychometric properties would be too restrictive, as many tools are only described as part of intervention or observational studies and are not the object of integral articles. Articles and references of the selected articles were screened using a standardized selection process. Selection process and criteria for articles that were retained in the review are presented in Figure 2.1.

Medical diagnostic tests used with this population have been described in the literature but are not reported here (see figure 2.1). Infants with torticollis may undergo these tests in order to ensure a proper diagnosis and to rule out more

serious medical and orthopaedic issues. Some information from these tests can be helpful to physical therapists for classification or prognostication. For example, a classification based on ultrasonographic images can help predict the need for surgery.^{2, 20, 41} Although the results of such tests may be available in medical charts, physical therapists cannot prescribe these diagnostic tests and can only make limited use of the information provided by those in the evaluation of infants with torticollis. Therefore, the literature regarding those tests will not be described in the present review.

Furthermore, multiple domains are routinely examined in infants with torticollis (e.g. craniofacial morphology, gross motor function). Research with this population report the use of various tools to measure these constructs (e.g. plagiocaphalometry for cranial morphology⁴² and the Alberta Infant Motor Scale⁴³ for gross motor function). These tools were, however, not developed and have not been formally validated for the torticollis population, and were therefore excluded from the present review. Reviews of tools for the examination of craniofacial morphology and gross motor function in other populations have been published and can be consulted for further information on these tools.⁴⁴⁻⁴⁶

To analyse the articles retained, the principal author (JF) used a standardised method to extract information from each article. This included year of publication, description of the sample, description of the assessment tool (number and type of items, scoring procedure, standardisation), and methods and statistics for the assessment of the psychometric properties. Results are presented in a descriptive manner. Guidelines provided by Streiner et al. were used to qualify the psychometric properties.⁴⁷

3.0 Results

Twenty-six articles were found that described 18 different assessment tools, techniques or criteria. From those, four are composite outcome measures and 14 are techniques or scales to examine body functions and structures. Their

description and summary of their psychometric properties are presented in Table 2.1; first composite outcome measures are presented in a reversed chronological order and then examination tools of body structures and functions are grouped according to the feature they are measuring.

As denoted in table 2.1, there is no tool available to measure factors from the environment that could affect the infant's outcome and needs. In a more research-oriented context, some efforts have been made to capture this dimension. Joyce et al. (2005)⁴⁸ used a 10 point Likert scale to assess parental satisfaction with outcome following Botox intervention. Littlefield et al. assessed the use of car seats, swings and other positioning devices to evaluate their impact on plagiocephaly, often associated with torticollis.³⁸ Kennedy et al. used a diary to measure parental habits of positioning and its effect on development in infants with plagiocephaly.³⁵

4.0 Discussion

The present review aimed at presenting the tools specifically developed or validated for the examination of infants with torticollis that could be used by physical therapists in the evaluation of this population. Eighteen different tools, techniques, criteria, or scales were identified.

Mobility of the neck is the body functions affected by torticollis the most frequently reported in the literature. Four authors provided techniques to assess ROM. Klackenberg et al. (2005)⁷² and Ohman & Beckung (2008)²² both tested the reliability of their techniques and found similar results. However, Ohman & Beckung are the only one to provide normative values for infants from birth to 10 months old based on empirical data. The report of ROM in clinical settings and in research should therefore incorporate the use such techniques, limit the use of over categorization (e.g. ROM limited or not) and favour the use of norms over the comparison with the unaffected side, considering that infants with CMT or PoT may present with hypermobility on the non-affected side.²² A survey of the

current assessment practices of pediatric physical therapist however revealed that although these techniques are available, about 86% are using visual estimates to measure ROM.⁴⁹

Literature on the clinical presentation of torticollis also report that muscle power and endurance can be affected. The Muscle Function Scale described by Ohman et al. (2009)⁷⁰ allows assessing reliably the muscle power of neck lateral flexors in infants with PoT or CMT. Norms are also available for infants from birth to 10 months old. Psychometric testing of the technique to measure rotators power (Rogers et al., 2008)⁷¹ and neck muscle endurance (Ohman et al., 2006)²⁴ could guide clinicians and researchers in the interpretation of the results obtained through the use of these techniques.

Measures of other clinical features of torticollis, including SCM characterization and asymmetry, were found and described. Two different methods have been reported to describe the characteristics of the SCM and surrounding neck muscles, but the description is limited to a subjective report of findings following palpation. More precise characterization of the size of the mass or of the thickening and tightness of the muscles could be beneficial, considering that the ratio of fibrosis in the muscle as been correlated to the outcome of intervention.²

The asymmetrical postures and movements found in infants with torticollis can be assessed using a tool designed by Philippi et al. (2004, 2006) (see Table 2.1). It aims at measuring asymmetry in infants aged 6 to 16 weeks and had undergone psychometric testing for face and content validity, internal consistency and inter-rater reliability. Internal consistency and inter-rater reliability were found to be good. But, although the authors claimed that their tool had face and content validity, the exact construct measured may be more related to muscle power or its integration in activities such as maintaining head and body positions or visual tracking, rather than actually measuring the construct of asymmetry. On a more technical note, the clinical applicability of this test in its original form may be

limited by the requirement of a video camera and by the restricted target population (ranging from 6 to 16 weeks). The limitation of the age range was justified by the authors by the fact that before 6 weeks, asymmetry can resolve on its own, and after 16 weeks the presence of active rotation can "mask" the presence of asymmetry, but it limits the potential of this test to assess improvement over time beyond 4 months of age.

The literature on the clinical presentation suggests that torticollis is a complex condition that has impacts that go beyond cervical ROM impairments or SCM muscle tightness. In an important proportion of the literature on torticollis, the focus is put on ROM and a more comprehensive understanding is lacking. The composite measures of outcome presented in this review attempted to assess the outcome of conservative and surgical interventions more globally. Variables included in these tools included: craniofacial asymmetry, head tilt, cosmetic appearance, and subjective parental assessment. These variables encompass many clinical features of torticollis, but do not include other aspects such as gross motor function and muscle power.

The comprehensiveness gained by the examination of these different variables has, however, often been gained to the detriment of accuracy. Many of the tools described included no standardized definitions to score the different items, and even though several measures have been used in clinical intervention studies, very few have been formally evaluated for their psychometric properties.

From these, the composite outcome measure designed by Cheng et al. (1999) that included six different variable each scored on a 4-point scale is probably the most responsive and reliable (see Table 2.1). This is because intervals between categorical variables are defined more clearly and their range is not too broad. Even if well described, the choice of the scoring options of this scale was not justified by authors. For example, classification of severe ROM limitation as being more than 15 degrees was not explained and may not be a responsive choice

of scoring option. Formal psychometric testing of this scale could be beneficial for clinicians and researchers.

With the increased number of infants presenting with torticollis in pediatric physical therapy departments, it becomes a priority to ensure effectiveness of the intervention provided to this population. The application of evidence-based practice principles is a key component in providing high quality care. However, the use of scientific evidence in clinical practice goes beyond the choice of effective interventions. Clinicians aiming to provide intervention based on evidence also need to use standardized, valid and reliable methods of examination to get an adequate representation of the patient's state and thereby judge accurately of the intervention that is needed.^{50, 51} The available tools designed or validated specifically for the torticollis population leave clinicians and researchers with few options to gain a comprehensive and accurate description of infants presenting with this condition.

One avenue of possible solution would be to use the International Classification of Functioning, Disability and Health – Child and Youth version (ICF-CY) as a framework for the examination of this population. The ICF-CY is a framework published by the World Health Organization that belongs to the family of international classifications.⁵² It describes an individual's functioning across domains of body structures, body functions, activities and participation. These domains of functioning are influenced by the health condition and by personal and environmental factors. It could be possible to link the different aspects found in the clinical presentation of infants with torticollis into the categories proposed in the ICF-CY and to identify examination tools designed to assess each domain in a more general pediatric population.

Barriers and facilitators to the use of valid and reliable examination and outcome measures are important to consider, considering that a significant proportion of therapists don't incorporate the available instruments in their routine assessment

with infants with torticollis and in other pediatric populations.^{49, 53} These barriers and facilitators include time, understanding of the meaning of scores, and availability of summarized information about measures among others, and should also be considered in the design of assessment strategies.⁵⁴ The summary provided by the present review could help clinicians in choosing appropriate examination strategies and thereby guide their decision making regarding the intervention to provide to infants presenting with torticollis.

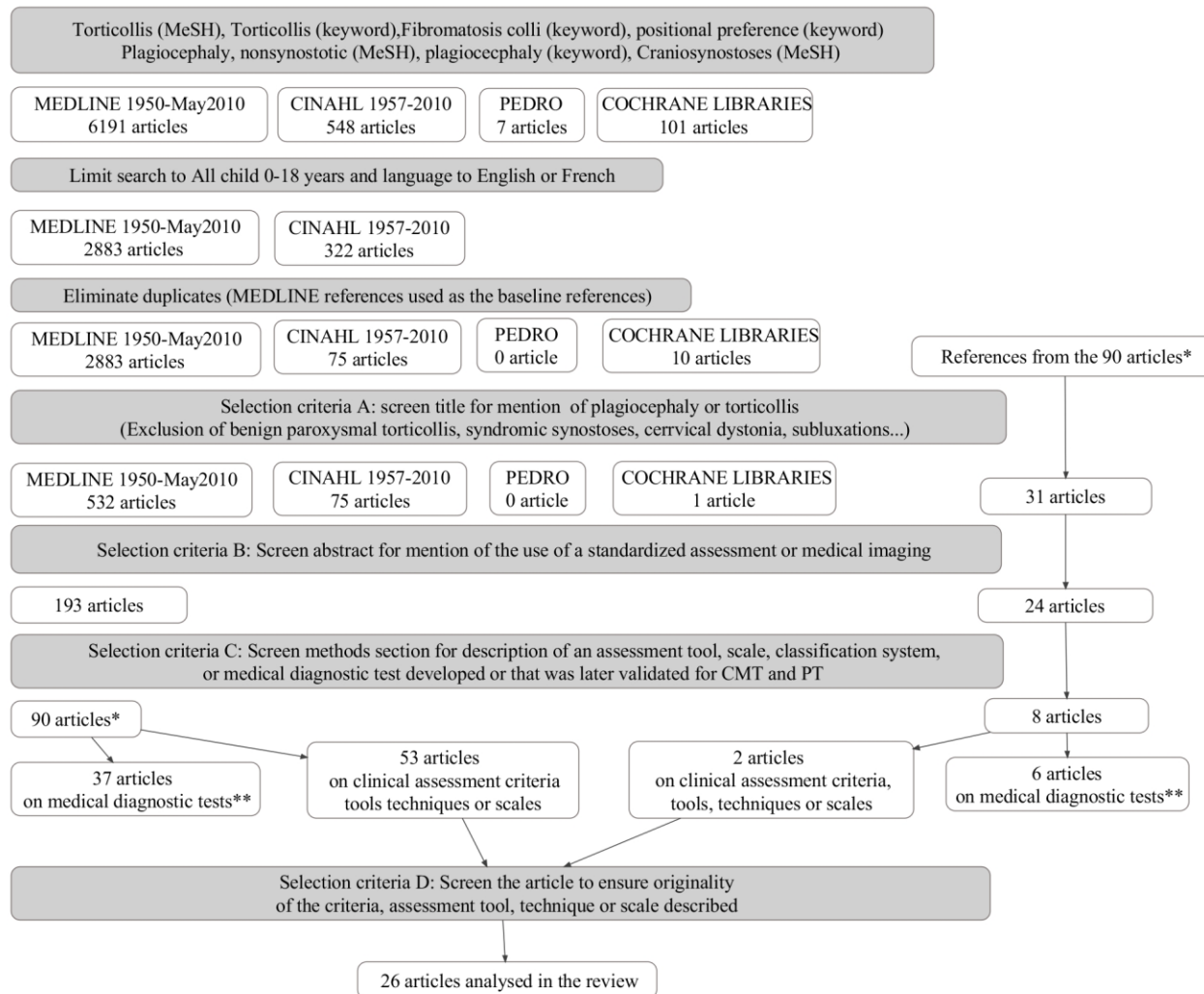


Figure 2.1 Search strategy and selection criteria for the inclusion of articles in the review

* References from the 90 articles meeting criteria C, were screened using the same process. See on the right side of the schema.

**Note that articles regarding medical diagnostic tests have been retrieved but will not be reported in this article. See text for justification.

Table 2.1 Composite measures of outcome and measures of clinical features of Postural and Congenital Muscular Torticollis

COMPOSITE MEASURES FOR EVALUATION OF THE OUTCOME OF INTERVENTION					
Authors	Construct	Population	Description	Scoring	Psychometric testing
Cheng, Tang, & Chen, 1999 ⁵⁵	Outcome measure of physical therapy intervention	Infants with CMT and PT	6 categorical variables: passive range of motion deficit, lateral flexion deficit, craniofacial asymmetry, head tilt, residual band in the SCM, and subjective assessment of the parents. The surgical version also includes scarring ⁵⁵	Each variable scored on a 0-3 point scale from which a total score out of 18 is calculated and then categorized as: Excellent (16-18), Good (12-15), Fair (6-11) or Poor(<6) The surgical version is scored out of 21	No psychometric testing and no norms available. Little information on psychometric properties can be drawn from the studies in which it has been used ⁵⁶⁻⁶⁰ since the results were reported as two categories leaving little room for interpretation of ranges of scores, validity of items, or their responsiveness
Taylor & Norton, 1997 ⁶²	Overall outcome of physical therapy	Infants with CMT (n=23)	Physical examination of the following characteristics: symmetrical head features, symmetrical facial features, passive cervical rotation $\geq 75^\circ$, passive cervical lateral flexion $\geq 40^\circ$, head righting complete, absence of head tilt at rest	Each variable scored 0 or 1, total score categorized as Excellent (6), Good (4-5), Fair (3), Poor (2-1)	No psychometric testing.
Lee, Kang, & Bose, 1986 ⁶³	Scoring system for the CMT assessment	Children aged 6-16 with CMT needing surgery	Facial asymmetry, neck movement, head tilt, scar, loss of column, and lateral band	Each item scored on a 0-3 point scale with descriptors of each score. Categorized as an Excellent, Good, Fair and Poor result	No psychometric testing, further used in clinical studies. ⁶⁴⁻⁶⁷

Coventry & Harris, 1959 ⁶¹	Outcome of treatment	Infants with CMT followed from 0-5 days up to 10 years of age	Visual and palpatory examination of tumour, apparent deformity of the neck, asymmetry of the skull and face, contracture of SCM, head tilt, tightness of neck muscles	Excellent= no remaining tumour, apparent deformity or asymmetry of the face or skull; Poor= contracture, head tilt, tight muscle, or asymmetry of the face or skull	No psychometric testing.		
MEASURES FOR EXAMINATION OF CLINICAL FEATURES							
Authors	Construct	Population	Description	Scoring	Validity	Reliability	Norms
Klackenberg et al., 2005 ⁷²	Mobility of the cervical spine-rotation	Infants with CMT	Infant in supine, head in midline and the head and body aligned along a sagittal axis marked with tape; shoulders stabilised by a 2 nd person	Standard goniometer (Medema) Movable arm of the goniometer in line with the infant' nose	Not tested The high face validity of goniometry is the main	Intra-rater ICC 0.77-0.99	Arbitrarily defined as 70-80°
Ohman & Beckung, 2008; ²²		Infants with CMT or PT	Infant in supine, stabilized at the shoulders	Arthrodial protractor, anatomical land-marks unspecified	argument for the absence of validation	Inter-rater r=0.71	110°(SD 6.2°) (5° less at 2 month)
Stellwagen Hubbard, Chambers, & Jones, 2008 ¹¹		Infants with CMT or PT	Infant in supine, no mention of stabilization	Eye-balling, result reported as chin moving past shoulder (100%), to shoulder (90%) or to mid-clavicle (70%)		Not tested	Not available
Leung & Leung, 1987 ⁷³		Children with CMT 6-7 years of age	A plumb-line attached to the centre of the child's chin while the head is rotated along a central vertical axis on a horizontal platform in front of the neck	Horizontal platform displaying a protractor. Measure is taken according to the plumb line's position		Not tested	Not available

Authors	Construct	Population	Description	Scoring	Validity	Reliability	Norms
Klackenberg et al., 2005 ⁷²	Mobility of the cervical spine-lateral flexion	Infants with CMT	Infant in supine Head was placed on the protractor in the neutral position (0°) Shoulders stabilised by a second person	Homemade protractor (75x60x9 cm), using the infant's nose as an anatomical landmark	Not tested The high face validity of goniometry is the main argument for the absence of validation	Intra-rater ICC 0.94-0.98	Arbitrarily defined as 60°
Ohman & Beckung, 2008 2009 ^{22, 70}		Infants with CMT or PT	Infant in supine stabilized at the shoulders	Using an arthrodial protractor, anatomical landmarks unspecified.		Intra-rater ICC 0.94-0.98	70° (SD 2.2)
Stellwagen Hubbard, Chambers, & Jones, 2008 ¹¹		Infants with CMT or PT	Infant in supine stabilized at the shoulders	Board with graph paper positioned under the head. Sagittal suture positions marked in neutral and maximal lateral flexion on the graph paper		Not tested	Not available
Leung & Leung, 1987 ⁷³		Children with CMT 6-7 years of age	A plumb-line attached to the occiput by a headband; the shoulders are steadied and kept horizontal	A goniometer is fixed to the headband and the measure is taken according to the plumb line's position.		Not tested	Not available
Karmel-Ross & Lepp, 1997 ⁶⁸	Aspect of SCM muscle	Infants presenting with CMT or PT	Palpation of SCM muscle from origin to insertion	Subjective record of the presence of fibrotic nodule, or diffuse fibrosis. Description of muscle tissue tension and elasticity	Not tested	Not tested	Not available

Authors	Construct	Population	Description	Scoring	Validity	Reliability	Norms
Freed & Coulter-O'Berry, 2004 ⁶⁹	Muscle tightness of neck muscles	Infants with CMT	Palpation of all superficial cervical muscles (no further description)	No description of particular aspects to evaluate	Not tested	Not tested	Not available
Philippi et al., 2004, 2006 ^{74, 75}	Asymmetry	Infants aged 6-16 weeks old with an asymmetry (including scoliosis, torticollis)	Infants with a video camera 2m above them: Head rotation in prone and supine starting from midline induced by auditory/visual stimulus (2 trials on each side) Cervical rotation deficit and trunk convexity for both positions assessed from video recordings	Each item scored on a 1-6 scale. A description for each score is given for every item. A total score ranging from 4 (symmetric) to 24 (very asymmetric) can be calculated. A difference of 3 points or more between the trunk convexity score and the cervical rotation deficit score determines the pattern of asymmetry	Mention of face and content validity by the authors, based on available literature (no use of expert panel) No other validation has been done	Inter-rater reliability ICC 91.5% (95% CI 86.0–97.0%) Assessed with 20 infants and 5 independent raters Internal consistency (Chronbach's α 0.84)	Not available
Rogers et al., 2008 ⁷¹	Muscle power of rotators of the neck	Infants with plagiocephaly, with or without PT	Infant in supine (most were younger than 4 months and had poor head control) Stimulated to rotate the head to the extreme in either direction by jingling a set of keys from side-to-side.	Estimated to the nearest 10° using the cervical spine as the axis. Several attempts made to ensure maximal effort.	Not tested	Not tested	Not available

Authors	Construct	Population	Description	Scoring	Validity	Reliability	Norms
Ohman et al., 2006 ²⁴	Muscle endurance of the neck	Children aged 2.5 months to 16 years old who had CMT or PT in infancy	Child in a sidelying position	Measure the time (in seconds) the child can hold the head upright and to compare sides	Not tested	Not tested	Not available
Ohman et al., 2009 ⁷⁰	Muscle power of lateral flexors of the neck	Infants in the general population and infants with CMT and PT	Infant held in a vertical position and then lowered to the horizontal position in front of a mirror Observe the head position and test both sides	Scale from 0 to 5 Score given according to the head position in relation to the horizontal line. The position has to be held for 5 seconds to obtain the score at a given level.	Construct validity was assessed using an expert panel (4 categories in original version and 5 in validated one) and to add description in degrees (was not used because of similar reliability without the description)	Inter-rater ICC all >0.90 Intra-rater weighted kappa (0.96 to 0.99) and ICC (0.93 to 0.98) Tested using photographs of 68 infants analyzed by 18 raters (physical therapists working in adult and pediatric settings and students)	With the 0-4 version (not the one validated) Age-mean (range) 2 months - 1.0 (0-2) 4 months - 2.6 (1-4) 6 months - 3.0 (2-4) 10 months- 3.4 (3-4)

ACTIVITIES, PARTICIPATION, PERSONNAL AND ENVIRONMENTAL FACTORS – NO TOOLS FOUND THAT ARE SPECIFIC TO THE TORTICOLLIS POPULATION

CMT: Congenital Muscular Torticollis; ICC: Intra-Class Correlation; PT: Postural Torticollis; SCM: Sternocleidomastoid muscle

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CHAPTER 3 – CLINICAL DECISION MAKING REGARDING INTERVENTION NEEDS OF INFANTS WITH TORTICOLLIS

Preface – The literature review presented in the two previous chapters illustrated that physical therapy is the standard intervention for infants with torticollis but that the optimal evaluation and treatment parameters to apply remain poorly defined. According to clinical decision making theories, health professionals rely on their examination to guide their choice of intervention parameters. However, the available tools for the torticollis population are either not psychometrically tested or do not provide a comprehensive representation of the complex clinical presentation of torticollis. There was therefore a need to better understand the intervention needs of this population in order to be able to design evaluation strategies and interventions that would be tailored to this population. This study aimed at gaining a better understanding of how health professionals working with infants with torticollis determine the intervention needs in this population.

ABSTRACT

Background: Physical therapy is the standard primary intervention for infants with torticollis, a common reason for consultation in pediatric settings. However, the wide variation in the frequency and type of intervention proposed in the literature reflects the lack of clear understanding of this population's needs.

Objective: To identify factors influencing clinical decision making regarding intervention needs for infants with postural or congenital muscular torticollis.

Methods: Focus groups and a survey of pediatric physical therapists, and questionnaires to pediatricians were used in order to generate a list of factors influencing decision making about intervention needs. Factors were mapped to the International Classification of Functioning, Disability and Health – Children and Youth version (ICF-CY).

Results: Pediatricians and physical therapists both report that intervention is needed in all infants presenting with PT and CMT. Factors across all domains of

the ICF-CY influence their decisions regarding the interventions to provide. An important subset of factors is related to the family and the environment.

Conclusion: Clinical decision making of physical therapists treating infants with CMT or PT should be based on a standardized family-centered evaluation encompassing all domains of the ICF-CY to ensure that the intervention needs of these infants are met.

1.0 Introduction

Congenital Muscular Torticollis (CMT) and Postural Torticollis (PT) are the most common causes of torticollis in infancy.¹ CMT is characterized by a unilateral shortening of the sternocleidomastoid (SCM) muscle with or without a fibrous mass while PT is a limitation in Range of Motion (ROM) leading to the typical head tilt of torticollis.²⁻⁵ The incidence of CMT varies between 0.3 and 2%⁶ while the incidence of PT is more controversial with reports ranging from 4 to 16%^{7, 8} of live births. This translates into an important number of families seeking health professionals' advice for this condition.

Physical therapy is the standard conservative care recommended for infants with CMT or PT and is usually associated with high rates of resolution.^{5, 9} Strategies frequently used in the physical therapy management include manual stretching, specific handling and positioning strategies, active and passive ROM exercises, and neuro-developmental approach to symmetry.¹⁰ Although a substantial number of studies on physical therapy intervention for this population have been published, the optimal treatment parameters that should be applied remain unclear. For example, the treatment frequency proposed in the different studies ranges from therapy once every 3 weeks¹¹ to stretching every 3 hours,¹² with no rationale for these choices and none of these specific approaches proven to be more effective.

Although a standardized intervention is generally applied to all participants of a study, Cheng et al. (1999, 2000) varied the frequency of intervention based on the severity of neck ROM limitation and found successful outcomes with both frequencies of intervention. ROM is an important indicator of the severity of torticollis, however other factors may influence the decision about treatment content and intensity in clinical practice. Luxford et al. (2009) report in a recent survey of pediatric physical therapists that perceived parental competency, the family's daily routine, and the physiotherapist's preference for techniques

influenced the selection of the parameters of stretching exercises for infants with torticollis.¹³

The breadth of treatment parameters and modalities proposed in the literature provide little understanding of the intervention needs of this population and leave therapists with little sound evidence to guide their clinical decision making. Little is known about what is currently influencing the thought processes of practicing therapists when treating infants with torticollis.

Research on clinical decision making show that a careful evaluation of probabilities and risks of given outcomes, taking into account patient preferences and scientific evidence is the best option to guide the choice of the type and frequency of intervention.^{14, 15} In pediatric settings, environmental context and family factors should undoubtedly be taken into consideration.¹⁶

The evaluation underlying decision making should be based on valid and reliable measures to ensure the accuracy of representation of the patient's state of health and functioning.^{17, 18} The literature on torticollis describes only a few valid and reliable measures, targeting single or limited aspects of the condition. Those fail to provide a comprehensive representation of the level of functioning and relevant contextual factors of infants with torticollis, providing little guidance to physical therapists in their decision making process. (see Chapter 2)

The absence of comprehensive and psychometrically sound assessment tools and the limited sound evidence on optimal intervention to provide reflect the limited understanding of the needs of infants presenting with torticollis. Such an understanding would allow the design of examination tools and outcome measures that represent accurately the burden of the condition and would help in the creation of more appropriate intervention strategies for these infants.

The objective of this paper is to identify the factors influencing the determination of intervention needs for infants with PT and CMT through an analysis of pediatric physical therapists' and pediatricians' clinical decision making.

2.0 Methods

2.1 Design overview and rationale

The present study included two phases to reflect different professionals' perspectives. First, the pediatricians' perspectives were sought in order to determine what aspects influenced their decisions regarding the management of infants with torticollis. Although van Vlimmeren et al. (2006)⁵ suggested that all infants with CMT and infants with PT presenting before the age of 2 months be referred directly to physical therapy, there is no evidence on the current and optimal management practices of pediatricians for this population. Therefore, there may be an important proportion of infants thought to have no need for specific intervention, especially in the subset of infants presenting with mild torticollis. A better understanding of the needs of this sub-population was sought through questionnaires sent to pediatricians.

In the second phase, a qualitative inquiry of clinical decision making of experienced pediatric physical therapists was conducted through two focus groups. The absence of literature on this topic, as well as the complexity of clinical decision making guided the choice of this method.¹⁹ Validation of the factors first identified in the focus groups through a national survey of pediatric physical therapists allowed estimation of the extent to which the factors identified were important in influencing decision making regarding intervention needs, using a larger sample. The survey also allowed exploration of additional factors as well as the current assessment practices for the evaluation of these factors.

Ethical approval for this research project was obtained from the McGill University Faculty of Medicine Institutional Review Board. All participants agreed to participate on a voluntary basis, after providing informed consent.

2.2 Pediatricians' perspectives

Design: Mailed open-ended questionnaires allowed tackling pediatricians' perspective with depth without requiring extensive amount of time.

Participants: Pediatricians registered to the Association des Pédiatres du Québec and working in the Montréal area (Québec, Canada) were contacted by mail (n=271). They were eligible if they were a) routinely assessing new infants for global development as part of well-baby care and b) referring infants to physical therapy for torticollis at least 10 times in the last year. Targeted email and phone reminders were made to pediatricians known to meet the inclusion criteria and to ensure representativeness of the sample (for example, to avoid over-representation of hospital-based versus community-based pediatricians). Eighteen pediatricians returned the completed questionnaire and the answers provided led to saturation of themes.

Procedure: A mailing list was obtained by the Association des Pédiatres du Québec for the Montréal area and all pediatricians (eligible or not) were mailed the description of the study, the consent form and the questionnaire that they were asked to complete and return by mail or fax. The questionnaire consisted of socio-demographic information and 6 open-ended questions which were targeting management provided by pediatricians and factors influencing the decision to provide different types of management (see Appendix 3A).

Analysis: The questionnaires were qualitatively analyzed using codes (see section 2.3.1 for detailed description of the coding procedure). Factors thought to be related to the intervention needs and quotes were extracted from the questionnaires. Descriptive statistics were used to describe the sample that participated and are presented in Table 3.1, but a lack of data about the whole population of pediatricians from the Montréal area meeting eligibility criteria restricted the ability to compare the respondents to the non-respondents and to estimate the response rate.

2.3 Physical Therapists' perspectives

2.3.1 Focus groups of experienced physical therapists

Participants: Purposeful sampling was used to select participants. Physical therapists were included if they were a) involved in the assessment and treatment of infants with torticollis for at least 2 years, b) seeing 3 cases of torticollis per week on average in the past year, and c) practicing in one of the two tertiary pediatric university teaching hospitals in Montréal. These criteria were chosen to ensure that the clinicians involved were aware of the current best practice and had seen many different cases of torticollis. Considering the limited number of potential participants as well as the relative focus of the themes, it was estimated that 2 groups with 4 to 10 participants would be sufficient;¹⁹ one group of 8 participants and another group of 4 participants were held, which represented a participation rate of 100% and indeed led to the saturation of themes. The physical therapists who participated all completed a BSc in physiotherapy leading to practice in a Canadian university, two of them had a master's degree in rehabilitation, two others had completed a BSc in another domain, and six of them reported to have completed extra-academic training (Neuro-Developmental Therapy, global postural re-education). Socio-demographic data of the participants are presented in Table 3.1.

Procedure: Invitation to participate to the focus groups was distributed to the therapists by the Chairs of the Physical Therapy Department of the two selected pediatric hospitals. The focus groups were held in the therapists' own work environment to facilitate open discussion and lasted approximately 90 minutes. The discussion was lead by the principal investigator (JF) who was assisted by a co-moderator (IG) who took notes on the discussion as well as comments on the process. The content of the discussion was audio-taped to allow for transcription and analysis of the content. Questions and probes were developed based on the available literature on torticollis intervention and clinical decision making and are presented in Appendix 3B. Questions referred mainly to factors considered as part of clinical reasoning that underlie the assignment of a given treatment intensity.

As an example, the therapists were asked to discuss the aspects that they would like to share with fellow students regarding the choice of treatment frequency for infants with torticollis.

Analysis: The focus group discussions were transcribed verbatim. A content analysis was applied. Codes were developed by the principal investigator and were used to generate a list of factors thought to be related to the need for more or less intensive course of physical therapy which was reviewed by the research team.²⁰ Since the aim of these groups was not to come to a consensus but rather to identify the full range of relevant factors, conflicting ideas for a given topic were not excluded.

In order to facilitate the organization of the results, the principal investigator mapped these factors with codes of the International Classification of Functioning, Disability and Health - Children and Youth (ICF-CY) (2007)²¹ using the methodology described by the ICF Research Branch.²² The ICF-CY is a framework published by the World Health Organization (WHO). It describes an individual's functioning across domains of body structures (BS), body functions (BF), and activities and participation (AP). These domains of functioning are influenced by the health condition and, by contextual factors divided into personal factors (PF) and environmental factors (EF).

2.3.2 Validation of factors identified through a national survey of pediatric physical therapists

Design: After the analysis of focus group data, a cross-sectional survey of pediatric physical therapists was conducted in order to estimate the level of agreement upon the factors previously identified as influencing decision making regarding intervention needs of infants with torticollis.

Participants: Different recruitment strategies were used to ensure broad participation and representation of pediatric physical therapists. All members of

the Paediatric Division of the Canadian Association of Physiotherapy (n=408) were invited to participate through the newsletter of the division. A reminder in the following newsletter was used as a strategy to increase the response rate. The members of Québec's Provincial Licensing body who had indicated being available for the treatment of infants with torticollis and plagiocephaly (n=410) were contacted by phone or email and recruited to participate. Finally, the Chairs of Physical Therapy Departments of 10 pediatric hospitals across Canada were contacted to solicit their physical therapists' participation. The participants were included if a) they were not working in the hospitals where the participants of focus groups were recruited, and b) had done at least 10 assessments of infants with torticollis in the last year. From the 70 physical therapists who agreed to participate, 25 (36%) did not meet inclusion criteria. The proportion of therapists who were not eligible from those who were asked to participate is unknown, but probably higher since those demonstrating an interest to participate may be more likely to work frequently with infants with torticollis and eligible to participate. Moreover, there is possible overlap between the members of the sampling sources. Therefore, the response rate cannot be estimated.

Forty-five physical therapists completed the survey and provided answers for 92.4% of the questions. Ninety-five percent of the participants graduated from a Canadian university, from which 62% received their professional training in the province of Québec. They were practicing in a variety of settings: 32% in private clinics, 22% in acute general hospitals, 18% in pediatric rehabilitation centers, 13% in pediatric hospitals, 10% in community-based services, and 4% in general rehabilitation centers. More details on their practice's characteristics are presented in Table 3.1.

Procedure: The web-based survey, using the Survey Monkey™ platform, was accessible for a period of approximately 2 months. Questions for the survey were developed following the analysis of the data from focus groups and addressed the validation of the identified factors and, secondarily, the use of assessment tools to

measure the different factors; the questions on the survey are presented in Appendix 3C. For each factor, the therapists were asked to identify the extent to which the given factor was influencing the identification of intervention needs on a scale ranging from 0 (to no extent at all) to 4 (to a very great extent). Therapists were also asked to report the five most influential factors in their clinical decision making without prior knowledge of the identified factors. The assessment practices of therapists were surveyed for each factor, except for some (e.g. age at presentation) as their clinical documentation usually consists of information provided by the parents or from the medical chart.

Analysis: Each participant first reported the five most influential factors in the determination of intervention needs. Their answers were related to the codes previously identified in the focus groups when possible and new codes were developed when necessary. The percentage of therapists who identified a factor from among the five most influential factors was calculated. Then, for each factor identified from the focus groups, the percentage of survey participants who rated the importance of the factor as 3 or 4 on the 5 point scale was calculated. This represented the percentage of therapists considering the factor as influencing their decision about treatment to a "great or very great extent". Factors that were not identified during the focus groups and that were mentioned during the survey were coded using the same method as described earlier. The assessment strategy of participants for each factor was described in terms of percentage using a given method. Descriptive statistics were used to describe the clinical practice of participants.

3.0 Results

3.1 Who should be referred to physical therapy? - *Pediatricians' perspectives on intervention needs*

Pediatricians reported high rates of referrals to physical therapy (median: 100%, range: 15-100%). Even if some reported lower rates of referral, there was

unanimous agreement that intervention, whether provided by pediatricians or physical therapists, was required for all infants presenting with torticollis.

Forty-eight percent of pediatricians decided to initiate intervention without referral to physical therapy at first. The intervention provided generally consists of providing advice on positioning and passive stretching of neck muscles and to monitor the condition from once every 2 weeks to once every 2 months. Failure to improve under such a regimen was consistently reported as an indication to refer to physical therapy. One of the pediatricians noted:

"After many years showing the parents how to do the exercises themselves at home and following them up regularly but finally ending up with residual malformations, I now refer them from the get-go to physiotherapy; they are more motivated."

Table 3.2 presents the factors taken into account by pediatricians when determining the intervention needs, and is structured using the ICF-CY framework. Physical characteristics of the infant, such as neck range of motion limitation, and family factors, such as parental concerns, were identified by pediatricians as important factors prompting to referral. Resources available also appear to play a role, as highlighted by the following quote:

"I work in a nursery, so I can't follow-up the infants. I usually ask that their doctor reassess them around 3 weeks of age, but medical resources are lacking to do so."

Of note, referrals to other health professionals were also reported: chiropractic and osteopathy prompted by parental demand (n=2), and physiatry, neurosurgery and general surgery (n=3) for complicated cases. However, physical therapy remains the primary resource for conservative management of torticollis.

3.2 How often should I see this family? - *Pediatric physical therapists' perspectives on intervention needs*

The focus groups first helped to define the content of the physical therapy intervention which consisted of the assessment and monitoring of the condition, teaching and education to the parents, and procedural interventions (hands-on therapy). The core of the intervention was the home program, which in adjunct to the direct physical therapy intervention, was effectively leading to the resolution of torticollis in most cases.

Physical therapists initiated their clinical decision making by the analysis of the examination data to confirm, through differential diagnosis, the presence of CMT or PT. One physical therapist mentioned:

"Since there are many underlying factors to a clinical presentation of torticollis, there are all the aspects of differential diagnosis that must be eliminated before stating it is a congenital or postural torticollis."

Then, they determined the relative proportion of physical therapy intervention (procedural intervention, parent education, monitoring) and of the home program required by the family depending on various factors that span across all domains of the ICF-CY classification, with personal and environmental factors playing an important role in the decision process. The list of the factors identified through the focus groups along with quotes supporting their importance and ICF-CY codes and descriptions are presented in Table 3.3. Aside of each factor is presented the percentage of therapists participating to the survey rating this item as influencing the determination of intervention needs to "a great or very great extent".

According to physical therapists, and as highlighted by the following quote, either a single factor or a combination of the aforementioned factors may justify increased treatment intensity:

"As soon as one element is severe, we follow-up regularly and when the combination of everything makes the case severe, then we follow-up frequently as well. It is not just the torticollis, it is the scope of the problem that is important."

Most factors were identified as having an important impact on the clinical decision making of a majority of therapists, except for seven factors: mobility of the neck in flexion and extension, strength of flexors and extensors of the neck, mobility of other joints than the neck, personality and irritability of the baby, and the APGAR score.

The participants of the survey were also asked to indicate the five most important factors affecting their decision making regarding intervention needs, without prior knowledge of the factors identified in the focus groups. The responses yielded by this question covered all domains of the ICF-CY, and were comparable to the factors identified during focus groups. Limitation in ROM and severity of torticollis were reported by 77% of therapists, followed by older age at presentation (52%), parental ability to perform the exercises (48%), plagiocephaly (39%) and gross motor function (36%). Factors that were not identified during focus groups brought precision about potential modifiers of the choice of intervention format and included current follow-up by other health professionals, geographical distance between the family and the center where services are provided, financial resources of parents receiving services in private practice, and the complexity of the exercises required by the condition of the infant.

The exploratory analysis of examination and evaluation practices of the participants of the survey revealed that only five of the factors identified (i.e.: gross motor function, craniofacial morphology, ROM of the limbs, reflexes and tone) were examined using a standardized and validated method by more than 10% of therapists. Mobility of other joints was assessed using goniometry, as needed, by 24% of therapists. Tools used in the examination of reflexes and tone included the Ashworth scale (original²³ and modified²⁴), the Amiel-Tison evaluation of the newborn²⁵, and the Movement Assessment of Infants²⁶. Craniofacial morphology was examined using callipers, cephalic index, thermoplastic bands, or the CranialTechnologies Severity Scale²⁷. Different measures of gross motor function were reported: the Alberta Infant Motor Scale²⁸

(n=25), the Talbot battery²⁹ (n=5), Movement Assessment of Infants (n=1)²⁶, Peabody Developmental Motor Scale³⁰ (n=1), and the Amiel-Tison evaluation of newborn²⁵ (n=1). The discrepancy between the level of agreement regarding the importance of a factor and the use of standardized examination tools is presented in Figure 3.1.

None of the environmental factors were formally examined with psychometrically tested tools. There was disagreement as to what approach should be used to assess environmental factors relating to the family. Some reported that they would observe the behaviour of parents to judge a familial characteristic (e.g. level of compliance) while others preferred to ask the parents directly about their concerns and level of confidence in doing the exercises. These results are illustrated in Figure 3.2.

When they were asked to identify facilitators that encourage use of examination tools in clinical practice with infants with torticollis, the physical therapists surveyed reported that a tool that was easy to administer with infants, required little specialized equipment, used diagrams to facilitate interpretation, and took little time to administer were the key elements in implementing the use of these tools in clinical settings. Although practicality was reported as a major issue, physical therapists indicated that the reliability and validity of the tool also influenced their decision to implement a particular tool in clinical practice.

4.0 Discussion

The present study describes the clinical decision making process of pediatricians and pediatric physical therapists in the determination of intervention needs of infants with torticollis. Both pediatricians and physical therapists agreed that formal intervention is required by all infants presenting with torticollis. Even though the natural history of torticollis remains unclear with some case reports of recovery without intervention,³¹ intervention studies show that later age at presentation is associated with poorer outcome^{32, 33} and persistent facial

asymmetries.^{34, 35} Therefore, current management of pediatricians and physical therapists is to provide intervention as soon as the torticollis is diagnosed to avoid such deleterious outcomes.

The factors identified as related to the intervention needs were similar across both disciplines' perspectives, and were confirmed by surveying a larger sample of pediatric physical therapists. For each phase of the study, the factors were encompassing all domains of the ICF-CY. However, the perspective of the pediatricians was restricted to fewer factors. This could be due to the difference in the methods used for the two groups but also to the nature of pediatricians' practice. Pediatricians often have a limited amount of time to diagnose and evaluate the patient's global health condition and to determine the best interventions to provide (average consultation length in UK and Spain ranging from 8 minutes to 23 minutes, the latter in the case of new patients^{36, 37}). Therefore, they likely restrict their assessment to their perceived most pertinent factors. When asked to identify the most important factors influencing their decision making, the physical therapists noted factors closely related to those identified by the pediatricians, supporting this hypothesis. This limited amount of time on the part of pediatricians to provide direct intervention likely contributes to the reported high referral rates to physical therapy.

All of the AP factors, and many of the BS and BF factors identified (mobility and muscle power of the neck, SCM aspects, and craniofacial morphology) were reported in the literature as clinical features of infants presenting with torticollis. The factors that were not previously reported in the literature (sensory integration, alertness, primitive reflexes, tone and weak suck) are all related to a certain extent to the maturation and integrity of the central nervous system. In a study of infants with plagiocephaly, a condition highly associated with torticollis that presents as an oblique deformation of the skull without fusion of cranial sutures, Fowler et al.³⁸ discussed the potential relationship between neurological findings and the development of plagiocephaly. The persistence of reflexes, hypotonia, and

diminished alertness could compromise the ability and intrinsic motivation to move, leading to flattening of the occiput, but also potentially to torticollis. The presence of these features in a subset of infants with torticollis could modify their intervention needs.

Twenty-one environmental factors were identified as determinants of the decision making of physical therapists. This is the ICF-CY domain where there is the biggest gap between available literature (evidence) and current management (practice). Literature to date primarily focussed on the physical components of infants with torticollis, however it appears that a broader perspective is needed in clinical practice to reflect the global needs of this population. Ohman et al. (2009)³⁹ reported on the developmental impact of caregiver positioning practices. Specifically, parents of infants with CMT were less likely than parents in a group of control infants to place their infants in the prone position while awake, even though they received specific recommendations of physical therapists. This highlights the importance of intervention strategies targeting environmental factors such as caregiver attitudes and practices in the management of torticollis.

This important focus on environment, and more specifically on family, is consistent with family-centered care. Family-centred care (FCC) is comprised of a set of values, attitudes, and approaches to health services and recognizes that each family is unique, that the family is the constant in the child's life, and that they are the experts in their child's abilities and needs.⁴⁰ Within this model of care, the family and the service providers collaborate to determine the family's strengths and needs and to decide about the services and supports that the child and family should receive. Supporting evidence of FCC illustrates that families who are involved in clinical decision making are more likely to adhere to treatment regimens, which is crucial in the management of torticollis. Those families therefore experience better health outcomes for their child.^{41, 42}

Within the perspective of FCC, the evaluation of priorities, concerns and resources is used to target strategies that empower the family in meeting the needs of their infants.⁴³ Therefore, the information about the environmental factors should be gathered through discussions with the parents about their needs and concerns rather than on a judgement based on observation of parental behavior. Assessment practices in this area may therefore need to be reoriented towards a more collaborative perspective.

Within the environmental factors identified, a subset of factors related to health professionals and services. The personal attitudes and preferences of health professionals acting as modifiers of decision making reiterates the importance of using scientific evidence when deciding about the optimal interventions to provide. Evidence-based practice would be enhanced through the evaluation of the effectiveness of current management strategies of torticollis.

This research presented the clinical decision making of pediatricians from Montréal, Québec, Canada, and pediatric physical therapists, from Canada, through mixed-methods. Efforts to ensure trustworthiness of data and representativeness of the participants included revision of the codes by the research team, inclusive recruitment strategies of pediatricians and quantitative validation of focus groups data through a national survey.^{44, 45} The lack of information about the population from which the participants were sampled restricted the possibility to compare the respondents to those who decided not to participate.

Further research in this area should focus on the perspective of parents on intervention needs of their infants with torticollis, to complement the understanding gained through this study and facilitate the application of FCC principles to intervention strategies for this population. Also, the determination of the extent to which the presenting factors relate to increased need for intervention

and to outcome of intervention would be beneficial in understanding the intervention needs of infants presenting with torticollis.

An accurate examination using psychometrically tested outcome measures is critical in the determination of intervention needs. However, as revealed by the exploratory results on current examination practices, the use of psychometrically tested tools is not widespread, which is consistent with the findings obtained by Luxford et al. (2009)¹³ in a survey of current management of torticollis by physical therapists in New Zealand reporting that 86% of therapists estimated ROM visually. Facilitators to the use of psychometrically tested outcome measures in clinical practice should be determined to promote their use in the clinical evaluation of infants with PT and CMT.

5.0 Conclusion

Torticollis is often viewed as a simple impairment of the SCM muscle, nonetheless the intervention needs of infants presenting with this condition are complex, and dependent on a spectrum of intrinsic and extrinsic factors. This broader perspective of modifiers of intervention needs in infants with torticollis likely applies to other pediatric populations. The application of such a holistic approach, framed by the ICF-CY, would likely enhance the comprehensiveness and quality of care provided by physical therapists.

Table 3.1 Participants' characteristics to the different phases of the study

Characteristics of paediatricians (n=18)	Mean(min-max) or %
Number of years of experience as a pediatrician	26.4 (10-61)
% who worked predominantly in a community setting (vs hospital)	65%
% who devoted 75% or more of their practice to evaluation of global development of infants	45%
Working in a university affiliated center (% yes)	94%
Number of infants referred to physical therapy over the past month	2.6 (0-10)
Characteristics of physical therapists of focus groups (n=12)	Mean(min-max)
Number of years of experience – total	17.0 (2.5-33)
Number of years of experience with the torticollis clientele	12.4 (2-33)
Number of cases per week (on average over the last month)	10.3 (3-20)
% of weekly practice devoted to the torticollis clientele	58.4 (30-85)
Importance placed on evidence-based practice (scale ranging from 0: not important to 5: very important)	4.4 (3-5)
Characteristics of physical therapists surveyed (n=45)	Mean(min-max) or %
Number of years since graduation of BSc Physical Therapy	17.7 (1-40)
% reporting more than 5 years of experience with the torticollis clientele	67%
% reporting seeing infants with torticollis at least once a week	78%
% reporting treating mainly an orthopedic clientele	62%

Table 3.2 Factors influencing decision making of pediatricians regarding referral to other health professionals

Factors
Health condition: Presence of torticollis itself
Health condition: Absence of improvement with current management
BF - Limitation in ROM and its severity
BS- Palpable mass of the SCM
BS - Associated plagiocephaly and its severity
AP- Maintaining head and body positions
AP - Risk factors or evidence of developmental delay
PF - Feeding problems
EF - Parental ability to perform exercises
EF - Parental concerns
EF- Resources available for health services

Legend : BF=Body function; BS=Body structure, AP= Activities and participation; PF= Personal factors; EF= Environmental factors

Table 3.3 Factors influencing decision making regarding intervention needs identified by physical therapists of focus groups and validated by participants to the survey

Factor identified	ICF-CY code and description	Quotes	% of participants to the survey who considered this factor as having a great or very great impact on their decision making
BODY FUNCTIONS (BF)			
ROM of the neck	B7101: Mobility of several joints	<i>Often we can see infants who will just have a lack of strength or of active range only, but there are infants who will have a difference in passive range and this will have an influence. You will want to see them more quickly [...] but there is a huge difference between active and passive. If an infant has passive limitations versus active limitations, it is really rare that we will interspace follow-up as much.</i>	Rot-100% LF-98% Flex-26% Ext- 23%
Reflexes	B7502 : Reflexes generated by exteroceptive stimuli	<i>Depending on the age, you want to know if there are some persisting reflexes. If you have your protective reactions, postural reactions and righting reactions.</i>	38%
Alertness	B1103 : Regulation of states of wakefulness	<i>If you have a good vigilance state. The alertness of the child.</i>	56%
Sensory problems	B260-265 : Proprioceptive and touch function	<i>For infants that have sensory problems, [...], that is part of our neuro exam as well.</i>	Not assessed
Strength	B7300 : Power of isolated muscles and muscle groups	<i>Muscle imbalance. Then (after ROM) I guess the next big thing is that we also try to look at the strength. General strength of the neck muscles.</i>	Rot-98% LF-93% Flex-19% Ext-30%
Tone	B7355-7356 : Tone of muscles of trunk-all muscles of the body	<i>Or infants with axial hypotonia. When you are sure that it is kind of fortuitous and that it is not explained by a neurological problem, I would feel more comfortable to let them go with an exercise program.</i>	69%

ROM of other joints	B710 : Mobility of joints	<i>And joints' asymmetries. [...] You have to be careful that the shoulder on the side where the child is having fixations that the gleno-humeral range is complete. [...] The same for the hip.</i>	16%
Oral-motor (Breastfeeding) problems	B5100-5105 : Sucking and swallowing	<i>Problems with suction, history of weak suck. [...] Even if it is resolved, if there was a time in the neonatal period where there was important feeding problems. It is not always a good sign of neurological maturity.</i>	50%
BODY STRUCTURES (BS)			
Muscle mass	S7104 : Muscles of the head and neck region – 2 nd qualifier = 7 qualitative change in structure	<i>Generally, when you have a fibromatis colli you pull your hair out because you know it will be hard. So, it is sure that this will increase your frequency. Cause I've seen kids who have a significant hematoma [...] and their range isn't too bad but I've seen others that have no big hematoma and their range is pretty bad.</i>	78%
Muscle tightness	S7104 : Muscles of the head and neck region – 2 nd qualifier = 4 aberrant dimension (shortening)	<i>Do you have capital lateral flexion? Is it more the scalens? [...] So I feel that with time we have to get to these details. The upper trapezium, we often forget about it. The first time I see an infant it is not rare that they have retractions in their upper trapezium.</i>	Not assessed
Plagiocephaly	S7100-01 : Bones of cranium-face	<i>So you know that they are at risk of deteriorating those kids. [...] Once the plagio is installed it is like trying to hold an egg standing, the infant always goes back into it. Cause most of the times, not most but a lot of the times, that's the parents' more pressing concern, the shape of the head. Rather than my baby can't turn one way versus the other.</i>	73%
ACTIVITIES AND PARTICIPATION (AP)			
Head tilt	D4155 : Maintaining head position	<i>I tend to look at the tilt because sometimes you will get the limitation in the rotation and stuff but the tilt isn't quite as bad as others that are really cranked to one side.</i>	83%

Postural alignment	D415 : Maintaining body positions	<i>I explain right away to the parents that this will have an influence on posture. Until the age they are standing you look at the influence of a torticollis on posture.</i>	85%
Global motor development	D410-415-420-450 to 469 : Changing basic body positions; maintaining body positions, transferring oneself; walking and moving	<i>On rare occasions you'll have more than just a torticollis and you may be concerned that there is something that is going on with their development or their quality of movement [...] you'd keep more an eye on these patients. Some of the developmental issues that may go along with torticollis.</i>	93%
Visual tracking	D110 : Watching	<i>And the eyes influence so much on head rotation that for some you just block the vision and the eyes are coming back to the midline and they are able to bring back their head. Yes it is sure that if you don't have visual contact it will take longer.</i>	59%
ENVIRONMENTAL FACTORS (EF)			
Direct environment	E1150 : General products and technology for personal use in daily living	<i>Car seats, swings, this and that...there is too much verticalisation, too early on. Whether it is in the little chair, when they are feeding, breastfeeding, we try to show them how.</i>	Not assessed
Handling and positioning	E410-415 -440 : Individual attitude of immediate/extended family members and personal caregivers	<i>You want to verify parental habits, look at the influence of those habits on what you see in the child, this is essential. I look at how the parents handle the child, how they do their carrying, how do they do their feeding, that type of thing. How is the child positioned at home?</i>	88%

Awareness of the problem by parents	E410 : Individual attitude of immediate family members	<i>And then you get the next group that say: “the only reason I’m here is because my pediatrician said there was a problem and I don’t really see a problem”.</i> <i>The latter family and the plagio or torticollis is severe, then you are thinking that you have to educate these parents on the problem so maybe you would see them more frequently.</i>	85%
Parents’ ability to perform exercises		<i>It is important to try to see how the parents are able to generalize this [home program and advice] to their everyday life.</i> <i>The reaction of the parents with me, with the child, the ability of the parents to do the exercises.</i>	88%
Parental anxiety		<i>If it is a stressed out parent who can’t even handle coping with the everyday life with a newborn, and just changing a diaper is a big thing, then adding on an exercise program is too much for them. Then those parents you would see them more frequently as well just to get them going kind of thing.</i>	63%
Parental motivation		<i>Yes it can make them more participative if they say I worked hard and I had results I will continue to do the exercises. I’m going towards improvement.</i>	85%
Parental coping strategies		<i>And sometimes the parents just can’t cope.</i>	Not assessed
Parent-child interaction		<i>Or just the chemistry that was between the child and the parents.</i>	63%
Parental understanding of the problem		<i>And sometimes, to make the family realize the needs of the infant as well because some families don’t see it necessarily.</i>	95%
Parent’ mental state		<i>And the other example is [...] post-partum depression [...], we’ll have to see each other often!</i>	73%

Parents who are fearful of moving their baby		<i>You have some parents that are able to stretch them, and you have parents who are not even able to touch the head before going in the opposite direction.</i>	80%
Parents' capacity to collaborate		<i>Ask the parents if they are ready to come to the appointments, to which level they are able to collaborate.</i>	83%
Family network	E310-340 : Support and relationship of extended family members- personal caregivers	<i>I feel like we don't always have with us the person who is taking care of the baby the most at home. Not always. And this is important. I always question about who is at home. Apart from the mother obviously who is at home during the first months. But sometimes there is a grand-mother who is caring or a father who is often present. So I often invite these people. Or a baby-sitter. (...) I ask if this significant person can be there at least once so that I can teach them.</i>	77%
Role perception of the physical therapist	E450 : Personal attitudes of health professionals	<i>I think our role is more to assess and see what needs to be done and then to assist the parents in doing it at home.</i>	Not assessed
Experience of the physical therapist		<i>Those are all things, speaking from someone whose been working here for 2 ½ years, at the beginning I found that a lot harder to come up with those things versus now that I've seen a multitude.</i>	
Personal factors of the physical therapist		<i>Why would we do it differently? (readiness to change) X talked a lot about this in his course. (knowledge)</i>	
Other professionals' attitudes		<i>Because we were saying often we get them later because the pediatricians reassure the parents and then Oops it's a little too late. Because I think the philosophy is also still Back to Sleep and you know regardless of the head shape and so what not.</i>	

Available resources	E5800 : Health services	<i>But it is always a question of how to deal with resources versus the needs. We try to find ways to be more efficient but if we could multiply ourselves, we would do so. There is limited time unfortunately.</i>	
Type of workplace	E5801: Health systems	<i>In fact, I work in the home care assistance program which is a bit different. I feel that Community Health and Social Centers should play a great role in that (prevention).</i>	
Protocols, accepted way of providing services	E5802 : Health policies	<i>Canadian Pediatric Society has changed their recommendations because their recommendation were Back to Sleep only and that's it and that's fine they still have that but now what they are saying is alternate the head sides and they are also really pushing play tummy time. Our policy is usually once we get the referral there's a delay of what? 2 weeks?</i>	
Complicated birth	P01.7 : Foetus affected by malpresentation before labour	<i>Relating to history, I would add infants with breech presentation. If there's any complication during the pregnancy, positioning in utero, sometimes the child is positioned a certain way, how the birth went, any problem with that, if the baby required any type of instruments for extraction such as ventouses or forceps, anything that could potentially lead to any type of injury at the neck area, if there was excessive pulling, any type of prolonged labour[...] I had infants with a low APGAR score but which "recover" quickly so the baby had no follow-up regarding this but I like to know about it so that I can make a closer follow-up.</i>	Birth complication 51% APGAR 7%
	P03.2/3 : Foetus affected by forceps/vacuum delivery		
	Others		
PERSONNAL FACTORS (PF) (NB no codes in ICF-CY for this category, for associated health conditions ICD-10⁴⁶ codes were used)			

Age at presentation	-----	<i>We are stuck with a torticollis at 10 months that is way much harder to treat than if we would have seen it at 2 or 3 months.[...] And that's a little harder to address because they've had a few months of being in a specific posture and the muscles get a little tighter.</i>	88%
Baby's response to exercises and baby's personality	-----	<i>How the child responds to us and to their parents. - I think the personality of the baby because if the baby or the child, if you put your hands on their heads and the baby starts screaming, the parents are going to have a really tough time carrying out the home program.</i>	Response to exercises 88% Personality /Irritability 49%
Associated health conditions*	P07 : Disorders related to short gestation and low birth weight	<i>Premature babies. A premature baby who is disorganised.</i>	37%
	Q65 : Congenital deformity of hip Q66 : Congenital deformity of foot	<i>Family history of orthopedic conditions. Because we get to one year of age and we see foot problems. Any associated hip and foot deformities as well, as they are sometimes associated with torticollis.</i>	Musculo-skeletal problem 76%
	P92 : Feeding problems of newborns Others	<i>Those with more important medical problems. If you have gastroesophageal reflux I am not sure it will influence your frequency but it will certainly influence the torticollis. Bronchodysplasia in premature babies. But in fact it was an infant who later had a diagnosis of Autism Spectrum Disorder but this kid was not accepting my hands on his head, on him. I had to work with my hands on his mother's hands.</i>	Other medical condition 63%

* Note: These associated health conditions could be related to the causal pathway of congenital muscular torticollis or postural torticollis, which could interfere with the definition of personal factors. However, formal association between the etiology of torticollis and these health conditions has not been established yet.

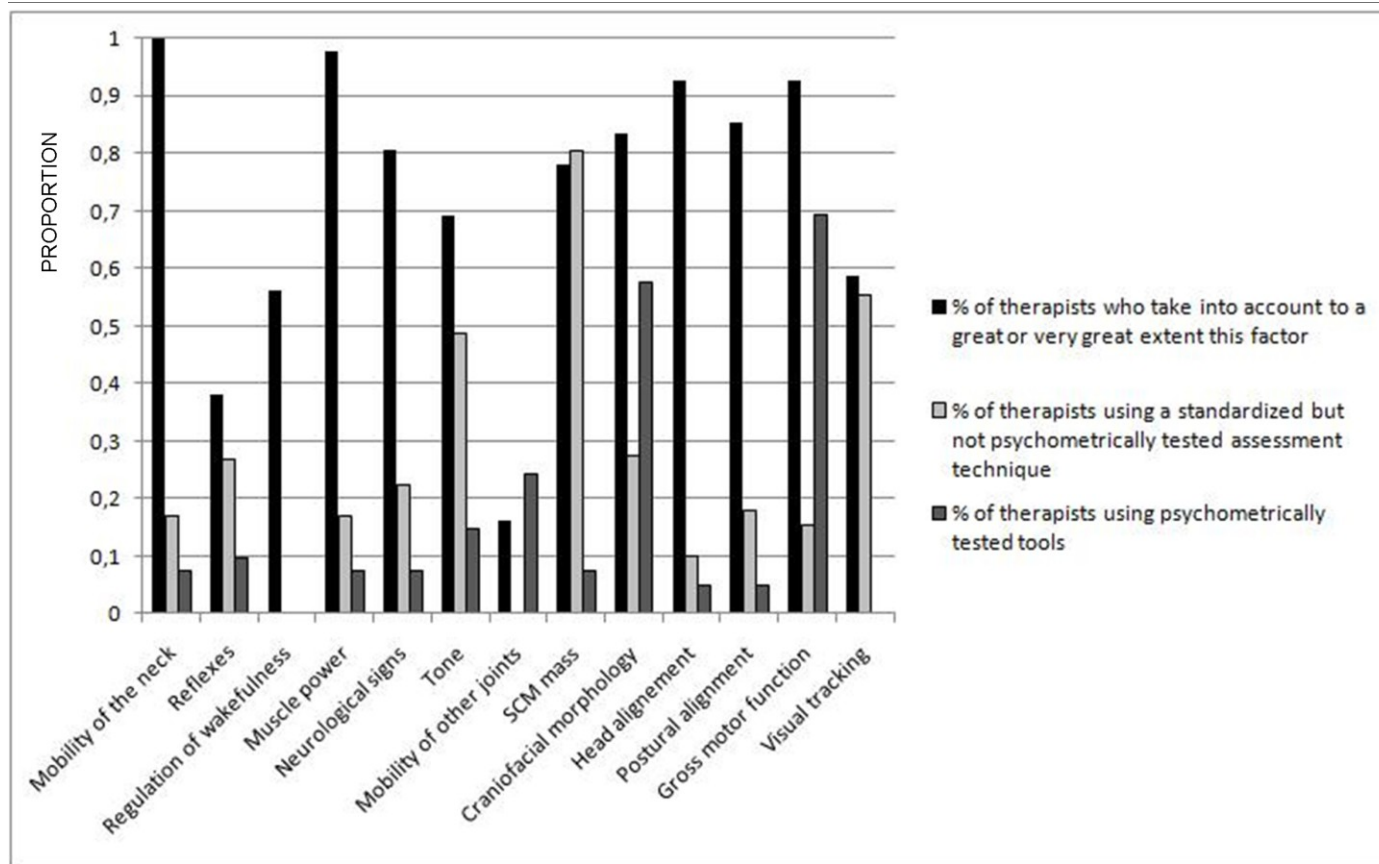


Figure 3.1 Use of standardized techniques or psychometrically tested tools in the determination of intervention needs.

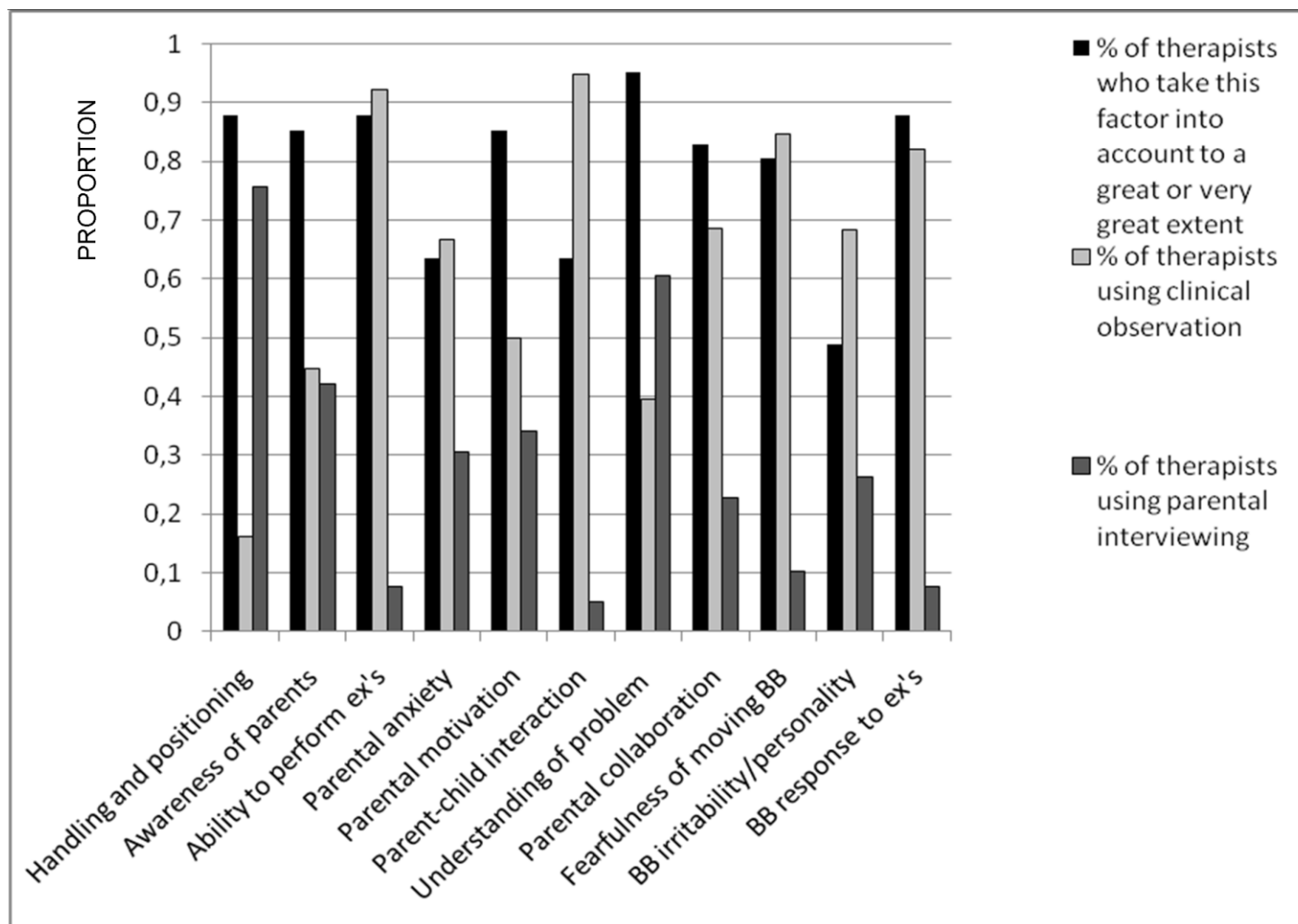


Figure 3.2 Difference in approach to the assessment of environmental factors

APPENDIX 3A – Questionnaire mailed to pediatricians

Which percentage of the infants that you are diagnosing with torticollis are you referring to physical therapy?

On which aspects do you base your decision to refer an infant with torticollis to physical therapy? (physical characteristics, parental habits, medical history...)

Does it happen that you refer infants with torticollis to another health professional than physical therapy? If so, which one and in which particular situation?

Which type of follow-up are you providing when an infant is not referred to physical therapy?

Do you have a preference regarding a reference to: a private clinic, a general hospital, a pediatric hospital, or no preference? Explain why.

During your follow-up, what are the elements that would make you change your treatment plan and opt for a reference?

Socio-demographic information:

Academic training (diplomas obtained, institution and year of obtention)

Which percentage of your practice is dedicated to a) hospital-based practice, b) community-based practice, c) being on call, and d) the evaluation of global development of infants in any of these types of practice?

Are you working in a university affiliated center?

For how long have you been working as a pediatrician?

How many times have you referred infants with torticollis to physical therapy over the past month?

APPENDIX 3B – Questioning route of focus groups with pediatric physical therapists

We will go around the table and I would like you to say your name and a reason why you appreciate working with infants with torticollis.

Do you think that all infants with torticollis should be referred to physical therapy and why?

In which way are you involved in the assessment and follow-up of this clientele?

At the initial evaluation of an infant with torticollis, which key elements of information do you want to know?

Do you use specific tools/methods to measure these aspects?

Are there things that you don't necessarily write down in the file but that affect your decision regarding the delay between the initial assessment and the follow-up?

How is your experience guiding you in the decision regarding the delay between initial assessment and follow-up?

What are the winning conditions that ensure an efficient home program to resolve torticollis?

Which aspects of the therapy can be done by parents as opposed to the aspects that should be handled by the therapists?

How can we be confident that the parents will provide an adequate treatment?

If you would have to teach a student beginning in the evaluation of infants with torticollis, which aspects would you absolutely want to share with him/her regarding the choice of a treatment frequency?

Do you think it is important to measure with precision the items that we mentioned earlier in order to determine treatment frequency?

Our objective was to discuss factors influencing the need for physical therapy intervention in infants with postural or congenital torticollis. Did we miss anything? Are there aspects that we didn't discuss but that you would have liked to?

APPENDIX 3C – Questions of the survey

By checking “Accept”, I declare that I am aware of that the nature of the study and of my participation. I agree to voluntarily participate in this study.

I would like to read the full length consent form, I refuse, I accept (consent form was available but not presented here)

In the past year, were you involved in the assessment of at least 10 infants with torticollis? *Yes, No*

The physical therapy intervention with infants with postural or congenital torticollis can take on many different formats such as:

- education and sporadic monitoring complemented by a home program performed by parents
- frequent direct intervention (hands-on)

As a therapist you have to decide which format/frequency is the most appropriate for each child at a given time. According to you, this decision should be based mainly on the following factors. *(5 blank boxes numbered)*

To what extent is your decision about the treatment format affected by the following factors? For example, if you consider that having blue eyes(factor) influences your choice to provide more direct intervention sessions to an infant, or to rely on parents to handle a home program, then you would choose “To a very great extent”. However, if you feel that having blue eyes has no impact on your decision regarding the format of intervention that you would provide, then you would choose “To no extent at all”

Anchors: To no extent at all, To a very little extent, To some extent, To a great extent, To a very great extent

Factors – see all factors on table 3.2, factors were presented in a randomized order

Are there factors that haven't been mentioned that according to you have an effect on the format of the intervention that you will provide to an infant with torticollis and his family?

Please indicate how you assess the following components in infants with torticollis and their family: - If you don't assess one of the aspects, write NA

- If you use no particular technique, tool or scale, write clinical observation and/or describe what characteristics you observe.
- If you use a particular tool or technique, write its name or describe it if it has no name. You can name more than one if you wish.

Blank boxes were left after each factor: that was measurable in clinical setting (excluded age, prematurity, etc as they don't require a particular assessment tool)

Do you have suggestions regarding the clinical assessment tool to be developed in order for it to be applicable to clinical practice?

What year and from which University did you graduate from your Physical Therapy program?

How many years have you worked with children diagnosed with torticollis? *Less than one year, From 1 to 5 years, From 6 to 10 years, From 11 to 15 years, More than 15 years*

How often have you worked with children diagnosed with torticollis, on average in the last month? *Once a month or less, 2-4 times per month, About once a week, 2-4 times a week, 5 or more times a week*

In which type of setting do you practice most of the times? *Private clinic, Pediatric hospital, General hospital, Pediatric rehabilitation center, General rehabilitation center, Community-based services, Other*

What type of clientele do you mainly treat apart from infants with torticollis? *Orthopedics, Neurology, Cardio-respiratory*

In the last 3 years, how many continuing education activities related to pediatrics have you attended? *None, 1 or 2, 3 to 5, 6 or 7, 8 or more*

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CHAPTER 4 – DISCUSSION AND CONCLUSION

Preface – This chapter will summarize the main findings of our work and relate them to the current literature on infantile torticollis. These findings were discussed separately in each manuscript; we will now highlight important points, their original contribution to the body of knowledge on the topic in a more global perspective, and their clinical implications. We will then propose an approach to the assessment of torticollis based on the current results along with further research directions. The limitations of our work will also be presented.

1.0 Summary of evidence and clinical implications

1.1 Availability of evidence on the clinical presentation of infants with torticollis

The literature review presented in chapter 1 of the thesis allowed us to highlight the lack of evidence regarding the clinical presentation of infants with torticollis, which has primarily been described in terms of the physical impairments that may co-exist with this health condition (i.e. limited mobility of the neck, SCM and peripheral muscles' shortening, craniomorphological changes, and diminished muscle power of the neck, among others).¹ The ICF model, which structured our reflection throughout this work, describes an individual's functioning across a number of other domains and there remains a paucity of knowledge regarding (a) the activity limitations and the participation restrictions that these infants may experience, and (b) the personal and environmental factors that may influence the level of functioning of these infants. Furthermore, knowledge of the long-term outcome associated with torticollis in infancy is also scarce.

These findings are in contrast with what clinicians consider of importance in their clinical decision making. Indeed, the results of chapter 3 suggest that clinicians rely heavily on environmental factors such as support from the family or availability of health care services, yet little information is available in the literature on the influence of these factors on the outcome of infantile torticollis. A more thorough and broad knowledge of the clinical presentation of torticollis

would help identify intervention strategies to limit the impact of torticollis on functioning.

1.2 Intervention for infants with torticollis

The second area of scarce evidence revealed by the literature review of chapter 1 concerns the lack of clarity on the optimal content and format of the intervention that should be applied in the treatment of torticollis. The current literature on intervention predominantly targets the primary impairments of torticollis and fails to provide enough information to truly enlighten families and health care professionals in their decisions. In contrast, the wide range of factors used by professionals to guide their intervention decisions presented in chapter 3 demonstrates the importance of adopting a broad holistic approach when providing intervention for infants with torticollis, as their impairments, activity limitations and participation restrictions span across all domains of the ICF-CY and may be influenced by an important number of contextual factors. For example, family involvement in the intervention, an area largely ignored in current literature on the topic, is thought to be central to the choice of treatment content by the physical therapists in our focus groups. This is in line with what parents themselves express when asked about their needs in the context of pediatric rehabilitation. Indeed, a vast majority of them report that they would benefit from more information about the child's condition and about techniques or ideas to implement more easily the home program in their daily routine, while only 11% of them identified a need for more hands-on intervention from professionals.²

This suggests that a focus towards empowerment of families and environmental changes could facilitate the capacity of parents to participate efficiently in the intervention for their infant. This is a promising avenue that has not been previously explored in this population, and that is consistent with the family-centered model of care, presented in chapter1 (section 4.0). To a certain extent,

physical therapists already appear to integrate the premises of FCC in their reasoning, as denoted by the importance accorded to family factors in chapter 3.

This model is articulated around three key premises: (a) parents know their children best and want the best for their child, (b) families are different and unique, and (c) optimal child functioning occurs within a supportive family and community context as the child is affected by the adaptive and coping strategies of other family members.³

There is evidence that the application of FCC principles to pediatric intervention has multiple benefits. In a literature review of the application of FCC in pediatric settings, Rosenbaum et al. reported different randomized controlled trials that showed benefits of FCC in terms of cognitive and motor gains, parental satisfaction with care, involvement of the parents in the home program, and adaptation of the environment to promote stimulation of the child's development.⁴ Although, FCC has historically been applied to populations with chronic conditions, there is emerging evidence of its effectiveness for more acute conditions such as in pediatric emergency departments and intensive care units.^{5, 6} Because torticollis is often reported as an acute condition, these new applications of FCC should be analysed in order to evaluate the potential implementation of FCC with families of infants with torticollis.

2.0 Future directions

2.1 Improving physical therapy evaluation for torticollis

The second chapter of the thesis provided a literature review of examination and outcome measures specifically designed or validated for the torticollis population. It revealed that: (a) ROM and muscle strength of the neck can be examined using standardized and reliable methods for which normative data are available, (b) techniques to examine asymmetry and the aspect of neck muscles have been described; and (c) four composite measures of outcome are available, but none have been tested for their psychometric properties. The limited available tools

designed or validated specifically for the torticollis population leave clinicians and researchers with few options to gain a comprehensive and accurate description of infants presenting with this condition which could then be used in clinical decision making to determine intervention needs.

We believe that one of the necessary next steps in research for infants with torticollis would be to design an examination strategy that would enable both clinicians and researchers to collect a comprehensive and accurate representation of the functioning of infants with PT and CMT. This examination strategy could be based on FCC principles and use the ICF-CY as a framework, which together would provide a strong basis for the accurate determination of intervention needs.

The clinical implementation of FCC requires the integration of its principles to examination practices. This concretely implies (a) that the priorities, concerns and resources of families are addressed, (b) that the family is taking an active role in providing information, and (c) that the family is provided with the opportunity to engage in the decision making regarding the intervention plan.³

An examination based on FCC principles is likely to foster trusting relationships. Positive relationships with health care professionals, in themselves, have been associated with better physical health outcomes.⁷ For example, lower rates of glycated hemoglobin were observed in those patients with type II diabetes who indicated having a positive relationship with their physician when compared to those with a neutral or negative relationship.⁸

Another advantage to an examination based on FCC is that families have a unique knowledge of their infant's abilities and needs³ and they reliably perform assessment activities⁹ which makes them good partners in the accurate evaluation of the infant. There is also evidence that families who are involved in the assessment process are more satisfied with the process of care and feel more

empowered. They are therefore more likely to follow the recommendations of health professionals,¹⁰ which is crucial in the case of torticollis.

Evaluation of families' priorities, concerns and resources is important so as to target appropriate strategies that meet the needs of infants and families. These needs must be identified during examination and are more likely to be revealed through an assessment based on FCC principles. This is illustrated in Figure 4.1.

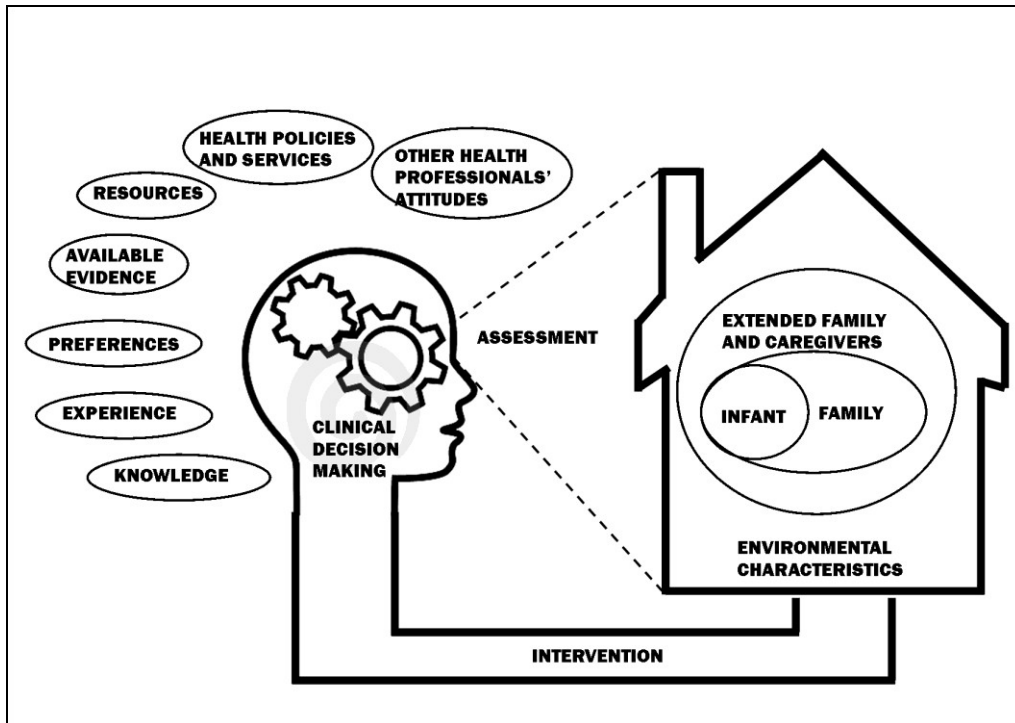


Figure 4.1 Schema of clinical decision making of physical therapists working with infants with torticollis. *Factors lying outside of the assessment target are aspects affecting clinical reasoning of therapists in a more global way, as they will remain similar from a patient to another. Within the assessment target are the environment of the infant, the infant with its personal and physical characteristics, the family and extended family with their unique priorities, concerns and resources. It illustrates that a holistic assessment based on Family-Centered Care could influence to the selection of intervention strategies that are more likely to meet the spectrum of needs of the infant and family. Note that although factors influencing clinical decision making are depicted as separate elements, in practice there is a significant interaction among them.*

Different approaches to the assessment of families' needs exist. The use of proxy variables, such as the education level or socio-economic status, that have been correlated with family functioning could be used.¹¹ Another approach is the use of standardized tests using observation by the therapist of various family characteristics. These two approaches may not be acceptable to the family.¹² Because we do not aim at providing specific interventions to improve parent-child social interaction or socio-economic status, the examination of those characteristics may not be readily justified. Finally, one other option that is more consistent with FCC principles is to interview families regarding parental priorities, concerns and resources, which are aspects that we truly aim to address as health professionals. This assessment can be performed through standardized assessment tools or semi-structured interview formats.¹³

For these reasons, we suggest that an adequate assessment of infants with PT and CMT should incorporate FCC principles through a collaborative questioning of family priorities, concerns and resources.

2.1.1 Suggestions for an examination strategy

In this section we present an examination strategy based on the ICF-CY as a framework and on FCC principles which, with further development and psychometric testing, could be used for clinical and research purposes to provide a comprehensive representation of this health condition.

Throughout this thesis, we used the ICF-CY which provided a comprehensive biopsychosocial approach to intervention needs, revealing the complexity of functioning in infants with torticollis. Literature on assessment in health care shows that a biopsychosocial approach lead to more complete data collection and to more effective intervention.^{14, 15} It could therefore be used to ensure a comprehensive examination of infants with PT or CMT and their families. Although the ICF-CY is a classification system and cannot in itself be used as an assessment tool, innovative applications of the classification for assessment

purposes have recently been proposed. For example, the items included in existing outcome measures have been mapped to ICF codes, so that it would be possible to use the data of these outcome measures to describe the functioning of individuals' in terms of ICF categories.¹⁶ Along the same line, ICF core sets for various health conditions have been developed. ICF core sets are defined as a list of ICF categories salient to a given health condition. The list is established based on the best available evidence, on patients' report and on experts' consensus.¹⁷

Using these ICF core sets, the ICF Research Branch has described a methodology to design categorical profiles of functioning for patients.¹⁸ ICF core sets are used to target categories of the ICF for which assessment is necessary. Based on the assessment of a given category, a qualifier describing the extent of impairment, limitation or restriction is assigned to the ICF category (see Table 4.1).¹⁹

Table 4.1 Application of qualifiers to ICF categories (Adapted from ICF-CY by the WHO, 2007)

Qualifiers for ICF categories of Body Structures, Body Functions, Activities, and Participation			
	Descriptor	Range of body structure and function impairment, activity limitation or participation restriction	
0	No problem	0-4%	
1	Mild problem	5-24%	
2	Moderate problem	25-49%	
3	Severe problem	50-95%	
4	Complete problem	96-100%	
8	Not specified	Not enough information available	
9	Not applicable	Not appropriate or not possible to assign a qualifier	
Qualifiers for ICF categories of Personal Factors, and Environmental Factors			
4	Complete barrier	+4	Complete facilitator
3	Severe barrier	+3	Severe facilitator
2	Moderate barrier	+2	Moderate facilitator
1	Mild barrier	+1	Mild facilitator
0	Not a barrier neither a facilitator		
8	Not specified		
9	Not applicable		

By assigning a qualifier to all relevant areas, a profile of functioning is created which can help identify components of health that are modifiable to target

intervention and the ones that are non-modifiable to guide realistic goal setting.²⁰ It also provides a common language for all health professionals and can facilitate communication with families within the perspective of shared decision making in FCC.

There is no ICF core set specifically developed for PT or CMT. The next steps enabling the development of such a core sets would be a formal literature review of the clinical presentation of torticollis, an evaluation of the parents' perspective and a formal validation through an expert consensus. Meanwhile, the factors identified through our study of clinical decision making regarding intervention needs could be used as relevant ICF-CY categories in the examination of this condition. Each factor, mapped to an ICF-CY code, could be assessed through a standardized examination procedure based on best available evidence. This methodology could therefore be applied for the creation of a categorical profile of functioning specific to infantile torticollis.

Also, in order to integrate FCC principles to this methodology, we propose that areas of concern be identified and added to the functioning profile using colour makers or another technique following the interview with family members to establish their priorities, concerns and resources. This could guide shared decision making on intervention strategies with families.

To illustrate this proposed examination strategy, we selected two categories per domain of the ICF-CY that were identified in the study of clinical decision making (chapter 3) and applied this procedure (presented in Table 4.2 on next page). We also presented an example of the application of the examination strategy to complete a functioning profile that could then be used for the evaluation of torticollis to guide decision making regarding intervention in Figure 4.2 (page 130).

Table 4.2 Application of the proposed strategy for the examination of infants with Postural and Congenital Muscular Torticollis

B7101 Mobility of several joints – neck	
Procedure proposed for assessment	Rotation - Infant in supine, stabilized at the shoulders. Use an arthrodial protractor to measure passive rotation of the neck. ²¹
	Lateral flexion - Infant in supine, stabilized at the shoulders. Use an arthrodial protractor to measure passive lateral flexion of the neck. ²¹
Rationale for the choice of this particular method	From the available methods proposed in the literature (chapter 2), this is the one for which norms have been established using a sample of normal infants. The equipment required is simple and inexpensive which could facilitate its implementation in clinical setting.
Suggestion for applying qualifiers to the aspect	When taking into consideration the norms established, the reliability of the assessment ²¹ , as well as the range of limitation in mobility of the neck presented in the literature ²² , the application of the qualifiers ¹⁸ lead to the following descriptors: 0 = Rotation – 106-110° ; Lateral Flexion 65-70° 1 = Rotation – 91-105°; Lateral Flexion 60- 65 2 = Rotation – 71-90°; Lateral Flexion 51- 60 3 = Rotation – 30-70° ; Lateral Flexion 31-50 4 = Rotation - <30° ; Lateral Flexion<30°
B7300 Power of isolated muscles and muscle groups - neck	
Procedure proposed for assessment	Rotators: Infant in supine, stimulated to rotate the head to the extreme in either direction by jingling a set of keys from side-to-side. Make two attempts. Measure the range of rotation using the protractor using the cervical spine as the axis. (modified from Rogers et al. 2009) ²³
	Lateral flexors: Apply and report the score using the Muscle Function Scale ²⁴
Rationale for the choice of this particular method	These techniques are the ones that were described in the greatest detail in the literature. For the Muscle Function Scale, formal psychometric testing and normative data are available.
Suggestion for applying qualifiers to the aspect	From a chart review of 293 infants treated at the Montréal Children's Hospital the active rotation of the neck ranged from 0 to 90°. Using these values, the application of the qualifiers lead to the following descriptors for rotation: 0 = 86-90°; 1 = 71-85°; 2 = 46-70°; 3 = 6-45°; 4 = 0-5°

(Continued)	<p>For lateral flexors, the score on the Muscle Function Scale is age dependent. According to the age and score of the infant, rank the impairment of muscle power of lateral flexors as no, mild, moderate, severe, or complete. Pilot testing of this assessment method could lead to a better definition of qualifiers.</p> <p>Norms for the 0-4 version (new version has 5 descriptors): Age-mean (range) 2 months - 1.0 (0–2); 4 months - 2.6 (1–4); 6 mths - 3.0 (2–4); 10 mths- 3.4 (3–4)</p>
S7104.7 Muscles of the head and neck region – qualitative change in structure – mass	
Procedure proposed for assessment	Palpate the SCM muscle. In the presence of a mass measure it at its greatest diameter, report measure in millimeters.
Rationale for the choice of this particular method	The lesion/muscle ratio and the echoicity of the SCM muscle, both observed with ultrasound, have been correlated to the outcome of intervention in infants with torticollis. ²⁵ Ultrasonography is not readily available in clinical settings. The only clinical assessment reported in the literature consists of palpation of the muscle and subjective report of qualitative properties of the muscle. ^{26, 27} Therefore, we suggest to use the measure of the diameter of the mass, as a proxy variable for the lesion/muscle ration to quantify the impairment of the SCM muscle. Validation of this method is needed.
Suggestion for applying qualifiers to the aspect	Some studies have measured the diameter of infants with CMT presenting a SCM mass which ranged between 0 and 16 mm. ²⁸ We used this data to describe the following qualifiers: 0=No palpable mass; 1=1-4 mm; 2=5-8 mm; 3=9-15 mm; 4= \geq 16mm.
S7100-7101 Bones of cranium and face – craniofacial morphology	
Procedure proposed for assessment	Using a flexible ruler or a thermoplastic band, trace the cranial circumference on a sheet. Trace diagonals and measure them. (according to the methodology described by Mortenson et al.) ²⁹ Calculate the Cranial Vault Asymmetry (Difference between diagonals)
Rationale for the choice of this particular method	Already 18% of therapists reported using flexible ruler or a thermoplastic band in their current practice. CVA has undergone validity and reliability testing and has been used by many researchers with infants with plagiocephaly. ³⁰ Cut-offs scores were established to describe none, mild moderate and severe asymmetries.
Suggestion for applying qualifiers to the aspect	The cut-off scores were used to assign values to qualifiers 0=<3 mm; 1= 4-8 mm; 2= 9-12 mm; 3=12-16 mm; 4= >16 mm

D4155 Maintaining head position - head tilt	
Procedure proposed for assessment	Take a picture of the child in the position proposed according to the age of the infant (0-3 months supine, 4-5 months prone, 6-10 months sitting, 11 months or older standing supported or not). On the picture, trace a line on the axis of the shoulders and one in the center of the head. Calculate the angle. Note that if it is not possible for legal or cultural reasons to take pictures of the child, the angle can be approximated using the protractor.
Rationale for the choice of this particular method	<p>The choice of the positions for different ages was made according to normal development and aimed at representing developmentally appropriate functional postures in which the activity of maintaining the head position should be performed.</p> <p>Although head tilt may vary during various periods of the day, clinical assessment doesn't allow evaluating the infant multiple times during the day. However, when a good relationship is established with the parents, it is possible to ask them to take picture of their infant in the desired position at different time points to increase reliability.</p>
Suggestion for applying qualifiers to the aspect	<p>From a chart review of 293 infants treated for torticollis at the Montréal Children's Hospital, the maximal head tilt at rest observed was 45°. The desired position of the head, that would represent the absence of a head tilt, is in midline or at 0°. The qualifiers have been defined in relation to these values:</p> <p>0 = 0-3° ; 1 = 4-10°; 2 = 11-25°; 3 = 26-44°; 4 = ≥45°</p>
D410-420-450 to 469 Gross motor function, age dependent; (changing body positions, transferring oneself, walking and moving)	
Procedure proposed for assessment	Use the Alberta Infant Motor Scale ³¹ and report raw score and age. Using the raw score and normative data, find the percentile corresponding to the score according to the corrected infant's age.
Rationale for the choice of this particular method	The AIMS is a psychometrically tested measure for which normative data are available for infants aged 0-18 months old. ³¹ This age range is appropriate for infants with torticollis as the vast majority present within one year of age. Infants below the 10 th percentile were shown to demonstrate a delay in the acquisition of gross motor skills and this is why this cut-off was used in the attribution of qualifiers. ³²
Suggestion for applying qualifiers to the aspect	0= 11th-100th percentile; 1= 10 th percentile; 2 =6-10 th percentile ; 3 = 5 th percentile; 4 =below 5 th percentile

E 410 Individual attitude of immediate family members - towards positioning E1150 General products and technology for personal use in daily living	
Procedure proposed for assessment	When scheduling the appointment with the parents, ask them to record, over a 3 days period, the amount of time spent in various positions (prone while awake, prone asleep, supine awake, supine asleep, sitting, etc.) and in various positioning devices (car seats, orthotic pillows, swings, high chairs, etc.)
Rationale for the choice of this particular method	This technique has been used with infants with plagiocephaly and was found to be reliable. ³³ Involving parents in the assessment, even prior to the first meeting, can facilitate their involvement in decision making further in the process. ¹¹
Suggestion for applying qualifiers to the aspect	Analyze separately the use of positioning devices (E1150) and the attitude of immediate family members towards positioning (E410) Assign a qualifier according to the following descriptors: 4= The positioning/devices completely limit the potential of the child to resolve his torticollis 3= The positioning/devices severely limit (...) 2= The positioning/devices moderately limit (...) 1= The positioning/devices mildly limit (...) 0= The positioning/devices do not affect the potential of the child to resolve his torticollis +1= The positioning/devices mildly facilitate the potential of the child to resolve her torticollis +2= The positioning/devices moderately facilitate (...) +3= The positioning/devices severely facilitate (...) +4= The positioning/devices completely facilitate (...)
E410 Individual attitude of immediate family members – ability to perform home program	
Procedure proposed for assessment	Ask the parents to rate their perspective on a 9 point scale for the two following questions: 1- How convinced are you that applying these advices and exercises will change the condition of your infant? -4 Truly convinced that it will have no effect up to +4 Truly convinced that it will have an effect 2- How confident are you in applying these advices and exercises as recommended by your physical therapist? -4 Confident that I do not have the necessary tools to apply the home program to +4 Confident that I do have the necessary tools to apply the home program
Rationale for the choice of this particular method	In motivational interviewing, these two questions are frequently used to identify the needs for information and support in patients who need to make lifestyle changes. ¹⁴ Considering the FCC approach to assessment that was

(Continued)	discussed, this approach could yield more useful information than observing parents doing the exercises and rate their behaviour. It can also act as a good outcome measure of intervention strategies targeting empowerment of families in the treatment of their infant.
Suggestion for applying qualifiers to the aspect	Combine the results of the 2 question by adding them up. The added score is directly translated in the qualifier (e.g. $(+2)+(-2) = 0$ = neither a facilitator neither a barrier) Clinical observations can be noted down if contradictory with the obtained result.
PF – Age at presentation	
Procedure proposed for assessment	Calculate chronological age at initial visit, report in months. (Date – Date of birth= x months)
Rationale for the choice of this particular method	Chronological age as been chosen since it has been associated with outcome in infants presenting torticollis. ^{34, 35} Chronological age may be more significant than corrected age in the case of torticollis since we want to know the delay between birth and the onset of symptoms.
Suggestion for applying qualifiers to the aspect	From a sample of 293 infants consecutively treated to the MCH, 99% presented before the age of 1 year old. If the infant present at birth with a torticollis, or if he/she presents later than 1-3 months of life, the mobility of the neck is more limited and there is an increased risk for poorer outcome or surgery. (Qualifiers only reported in terms of barrier in this case) 4 = Birth – 10 to 12 months; 3 = 1 week – 8-9 months; 2 = 2 week – 6-7 months; 1 = 3 weeks – 4-5 months; 0 = 1-3 months
PF – Associated medical condition(s)	
Procedure proposed for assessment	Review medical history and systems to ensure appropriate differential diagnosis and to identify necessary referrals. If a medical condition as been identified, use an ICD-10 code to describe it and report its severity if applicable. (http://apps.who.int/classifications/apps/icd/icd10online) ³⁶
Rationale for the choice of this particular method	The report of associated medical conditions in terms of ICD-10 codes could constitute a large database to evaluate the incidence of different medical conditions within the population of infants with torticollis and provide insight for research.
Suggestion for applying qualifiers to the aspect	Consider the impact that the medical condition has on the level of functioning of the infant and the potential impact that it will have on the intervention that you will provide as a therapist. Rank this impact as a Complete/Severe/Moderate/Mild Facilitator or Barrier or score 0 if it has no impact.

	0	1	2	3	4				
BODY FUNCTIONS									
B7101 Mobility of several joints- neck rotation ★									
B7101 Mobility of several joints- lateral flexion									
B7300 Power of isolated muscle groups-neck rotators									
B7300 Power of isolated muscle groups- neck lat. flexors									
B7358 Tone of muscles of all muscles of the body									
B7502 Reflexes generated by exteroceptive stimuli									
B1103 Regulation of states of wakefulness (Alertness)									
B260-265 Proprioceptive and touch function									
B710 Mobility of joints –limbs									
B5100-5105 Breastfeeding and sucking problems ★									
BODY STRUCTURES									
S7104.7 Muscles of the head and neck region SCM mass									
S7104.4 Muscles of the head and neck region – tightness									
S7100-7101 Bones of cranium and face- morphology									
ACTIVITIES AND PARTICIPATION									
D110 Watching ★									
D4155 Maintaining head position – head titlt									
D415 Maintaining body positions – postural alignment									
D410-420-450-469 Gross motor function									
	4	3	2	1	0	+1	+2	+3	+4
PERSONAL FACTORS									
PF Associated medical condition(s) ★									
PF Congenital deformity of the hip					9				
PF Congenital deformity of the foot					9				
PF Foetus/newborn affected by pre-natal history									
PF Baby’s response to exercices and personality									
PF Age at presentation									
ENVIRONMENTAL FACTORS									
E1150 General products and technology for use in daily living									
E410 Individual attitude of family members									
E310-340 Support and relationship of extended family members and caregivers									

Figure 4.2 Functioning profile for Postural and Congenital Muscular Torticollis drawn from the examination strategy Following the examination, therapists could fill in a functioning profile in which the level of impairment, activity limitation, participation restriction, or impact of personal or environmental factors would be identified through shaded areas. The priorities of families could be identified by stars, or another method. This profile could help to guide decision making regarding intervention needs.

2.1.2 Further development of an assessment battery

Focus groups to gather the perspective of parents on intervention needs of their infants and families should be conducted to complement the understanding of intervention needs in this population. Once the perspectives of parents are gathered, the design of an assessment battery applying the proposed examination strategy and principles could be designed. Then, pilot testing could allow assignation of specific values to ICF-CY qualifiers, so that they are representative of what is clinically encountered with this population.

The proposed assessment battery should then undergo psychometric testing before it can be utilized as an evaluative outcome measure in making clinical decisions for individual infants and groups of infants with PT and CMT. Validation of the content and of the ability of the battery to reflect intervention needs would give meaning to the results obtained and ensure that this assessment battery can be used with confidence in making decisions. An assessment of its reliability and responsiveness would verify the extent to which the test can detect true change when it happens and the amount of measurement error that must be taken into consideration when evaluating change in an infant.

On a more long-term perspective, data gathered with this assessment battery could guide the design of innovative intervention strategies to meet the needs of infants with PT or CMT and their families.

The use of such an examination strategy, and eventually of a psychometrically validated assessment battery, could have benefits for clinicians and researchers that are presented in the next subsections.

2.2 Potential benefits to the use of the examination strategy

2.2.1 Guide for clinical decision making

In the survey that was aimed at validating factors identified as influencing determination of intervention needs (chapter 3), an important proportion (36%) of

participants interested in participating did not meet the inclusion criteria (i.e.: assessment or treatment of at least 10 cases in the past year). This suggests that many therapists are in contact with this clientele only occasionally and may have a limited experience in treating this condition.

The Ministry of Health and Social Services of Québec recently made changes in service provision to ensure that required services could be received in a nearby facility.³⁷ This led to a reorganization of services provided by therapists. Physical therapists in remote regions may therefore be required to treat more varied problems that they are exposed to only occasionally, such as torticollis.

Experience is a recognized modifier of clinical decision making,²⁶ there may be a need for more explicit guidance in the determination of intervention needs by novice therapists and therapists who only occasionally see infants with torticollis. During the focus groups, one pediatric physical therapist supported the importance of such guidance:

“From what I understand, the purpose is to objectify our assessment to help us guide the determination of needs of infants and then see to which frequency they need physio. And since we tend to let to local therapists the responsibility to care for these infants, we train a lot of new therapists. And when you are a student, or a novice, it is easier when there are clear guidelines rather than when you have to question yourself to know if it is mild, moderate, or severe, to then judge the frequency needed.”

Binder, in his book entitled *Pediatric Interviewing*,¹⁴ emphasized the importance, mostly for novices, of a clear template that can be used in the assessment so that all relevant data is gathered to make an accurate diagnosis and an appropriate treatment plan.

Furthermore, the use of such an examination strategy could also be beneficial in larger centers where therapists see infants with torticollis on a regular basis. These

centers sometimes have to resort to waiting lists and there are typically not clear guidelines for the prioritization of cases. This strategy could assist in the prioritization of infants according to their intervention needs.

The application of this examination strategy could therefore guide the decision making of therapists in the determination of intervention needs for infants with torticollis.

2.2.2 Use of psychometrically tested outcome measures

The use of psychometrically sound outcome measures in the examination of torticollis is not widespread in the current practice of pediatric physical therapists, as revealed by the exploratory analysis of assessment practices in our national survey, which was consistent with the findings of Luxford et al. in New Zealand.³⁸

The limited use of outcome measures in clinical practice is not particular to the torticollis population. It has been observed in many different settings, with various professionals and clienteles.^{39, 40} Facilitators and barriers to the implementation of outcome measures in clinical practice have been reported in the literature and may include lack of time, human resources, monetary resources, resistance to change by individuals, ease of interpretation of scores and results, and applicability to a wide range of clients within a given clientele, among others.⁴¹

In the exploratory analysis of facilitators in the use of outcome measures with the torticollis population in chapter 3, similar results were obtained. Although practicality was reported as a major issue, physical therapists reported that the reliability and validity of the tool mattered to their decision in implementing a tool in clinical practice. As denoted by Finch et al.: “No matter how quick and cheap tests are to administer, without good measurement properties, they won’t be a strong basis of information for decision making. (p. 21)”,³⁹

Research in the EBP domain support the fact that psychometrically sound assessment optimizes clinical judgement.^{42, 43} By using valid and reliable outcome measures, physical therapists could enhance the accuracy of their perception of the infants' and families' needs which could enhance decision making regarding the intervention strategies that are more likely to meet their needs.

Therefore, the psychometric properties and facilitators to the use of outcome measures should be considered in the design of the assessment battery to facilitate its implementation in the clinical setting.

2.2.3 Research applications

Finally, the use of the suggested examination strategy could also have potential benefits in research. Clinicians using the examination strategy would report the clinical presentation of their patients in terms of ICF-CY codes. Through the common language and precise classification provided by the ICF-CY, this would constitute an important amount of data and a comprehensive representation of the clinical presentation and the natural course of torticollis under conservative care.⁴⁴ These data could also be used in the evaluation of the effectiveness of intervention strategies. These two areas, clinical presentation and optimal intervention strategies, were identified in the literature review as areas where evidence was lacking. It would be beneficial to adopt this examination strategy in the future to gather evidence for research purposes with this population.

3.0 Limitations

This study on clinical decision making of health professionals regarding intervention needs of infants with torticollis presented the current perspective of the literature and of practicing professionals. This step was important in order to identify research avenues to explore in the future. However, further evidence that the factors identified truly influence the needs of infants presenting with PT or CMT is needed. Clinical estimation of the extent to which the identified factors

relate to specific needs and levels of need could be performed to support our current findings.

We presented the perspectives of two categories of health professionals but the perspectives of parents on their infant's and family's needs is lacking. This is important, considering that families were identified by health professionals as important modifiers of the infants' needs. This additional information could also facilitate the implementation of FCC principles to the assessment and intervention with this population.

The limited number of participants is also a limitation of this study. In qualitative research, the heterogeneity of the sample is desired to gather many different perspectives on a given theme. Heterogeneity may have been limited in this study due to the number and geographically limited sampling source for the focus groups and the questionnaires to pediatricians. The participants may have received similar education and adopted a particular philosophy that influenced their clinical decision making, therefore influencing the results.

For both pediatricians and physical therapists participating to the survey, we were unable to compare the respondents to the non-respondents due to the lack of data on the sampling populations. Although efforts have been made to ensure a wide participation of health professionals, this remains an issue in terms of the representativeness of the sample. Those who decided to participate may have a different clinical decision making due to personal or environmental factors.

4.0 CONCLUSION

This thesis work furthered our understanding of the intervention needs of infants with torticollis and their families through an analysis of health professionals' decision making and led to the recommendation of a comprehensive examination strategy for infants with this condition. Although torticollis is often considered as a simple impairment of the SCM muscle, it is apparent that a broader approach to

clinical evaluation of this condition is needed to fully meet the needs and to provide optimal interventions to these infants and families. This concept likely applies to other conditions deemed as “non-complex” that are encountered by rehabilitation professionals. Evidence illustrating the variety of families’ intervention needs and the benefits of using a biopsychosocial approach in health care is needed. Such evidence could inform clinical managers and policy makers about the potential benefits of investing resources to improve the health outcomes and satisfaction with care of citizens, or future citizens in the case of torticollis.

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