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Plagiocephaly in Calgary, Alberta, Canada: Incidence, Risk Factors and Follow-Up

by

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Abstract

Background: The Canadian Foundation for the Study of Infant Deaths (CFSID), the Canadian Paediatric Society (CPS) and the Canadian Institute for Child Health (CICH) released a joint statement in February 1999 recommending that infants be placed to sleep on their backs to prevent sudden infant death syndrome (SIDS). Subsequently, a concern has been raised about a potential consequent increase in positional plagiocephaly across Canada. No literature exists on incidence of positional plagiocephaly in the Canadian context; literature on risk factors is sparse.

Research Questions: (a) What is the incidence of positional plagiocephaly in infants 7-12 weeks of age in Calgary, Alberta, Canada? (b) What are the potential risk factors for positional plagiocephaly in infants 7-12 weeks of age in Calgary, Alberta, Canada?

Methods: Using a prospective cohort design, study participants (n=440 healthy term infants 7 – 12 weeks old from well-child clinics at four community health centres in Calgary, Alberta) were assessed by two Research Nurses using Argenta's (2004) Plagiocephaly Assessment Tool. Parents completed a questionnaire on risk factors.

Results: The incidence of positional plagiocephaly was estimated to be 46.6%. Multiple logistic regression analysis identified the following risk factors: (a) right sided head positional preference (OR: 4.662; p=.000; CI 2.868–7.577), (b) left sided head positional preference (OR: 4.212; p=.000; CI 2.446–7.251), (c) supine sleep position (OR: 2.670; p = .000; CI: 1.582–4.508), (d) vacuum/forceps assisted delivery (OR: 1.883; p=.044; CI 1.016–3.488), and (e) male sex (OR: 1.547; p= .048; CI: 1.004–2.383).

Conclusion: Given that positional plagiocephaly is a preventable condition, an incidence of 46.6% is very troubling. Information to vary infants' head positions needs to be communicated to parents/guardians well before the 2-month well-child clinic visit. Prevention education can be targeted to parents/guardians of those infants that are male and infants that have had assisted deliveries. This could occur in the prenatal period or during the neonatal period by postpartum nurses, and PHNs involved in postpartum care. Further studies are also required in order to better understand the role of family physicians in identifying the condition and follow-up process of such infants.

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Dedication

To my Family

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List of Definitions

Term	Definition
Anthropometry	The science of measuring the human body
Brachycephaly	A skull shape that is short in proportion to its width
Craniosynostosis	Premature closure of skull sutures
Congential Muscular Torticollis	The presence of unilateral fibrosis or shortening of the SCM muscle
Euryon	The extremity, on either side of the head, of the greatest transverse diameter of the head
Frontozygomaticus	The suture between the frontal bone and the zygomatic bone
Incidence	The number of new health-related events in a defined population within a specified period of time
Ipsilateral	Pertaining to the same side of the body
Lambdoid	The suture that separates the parietal and temporal bones of the skull from the occipital bone
Occipital bone	A bone in the lower back part of the skull between the parietal and temporal bones
Parietal bone	One of two bones that together form the roof and sides of the skull
Plagiocephaly	A malformation of the skull producing the appearance of a lopsided head
Prevalence	The proportion of individuals in a population that have a disease or a condition
Sternocleidomastoid	One of two muscles arising from the sternum and inner part of the clavicle
Sclerosis	A hardening within the nervous system especially of the brain and spinal cord resulting from the degeneration of nervous elements such as the myelin sheath
Scoliosis	A lateral curvature of the spine, it usually consists of two curves, the original abnormal curve and a compensatory curve in the opposite direction
Suture	The line of union in an immovable articulation, as those between the skull bones
Torticollis	A deformity of the neck secondary to shortening of neck muscles, which tilts the head to the affected side with the chin pointing to the other side

List of Symbols, Abbreviations and Nomenclature

Symbol	Definition
AAS	Absolute Asymmetry Score
AIMS	Alberta Infant Motor Scale
AS	Asymmetry Score
AUC	Area Under the Curve
BSID	Bayley Scales of Infant Development
CFSID	Canadian Foundation for the Study of Infant Deaths
CHC	Community Health Centre
CIHC	Canadian Institute for Child Health
CMT	Congenital Muscular Torticollis
CPS	Canadian Paediatric Society
CT	Computed Tomography
CVA	Cranial Vault Asymmetry
DD	Diagonal Difference
DOH	Department of Health
HSC	Head Shape Clinic
ICC	Intraclass Correlation Coefficient
IEP	Individual Education Plan
IRC	Infant Repositioning Class
LPFS	Left Posterior Flattening Score
-2LL	Minus Two Times the Log of the Likelihood
OCLR	Oblique Cranial Length Ratio
PDMS	Peabody Development Motor Scales
PDQ – II	Revised Denver II Presecreening Questionnaire
PHN	Public Health Nurse
RDD	Reductions in Diagonal Difference
ROM	Range of Motion
RPFS	Right Posterior Flattening Score
SCM	Sternocleidomastoid
SIDS	Sudden Infant Death Syndrome
TCD	Transcranial Difference
TDD	Transcranial Diameter Difference

CHAPTER ONE: INTRODUCTION AND LITERATURE REVIEW

Background

In 1992, the American Academy of Paediatrics released a statement recommending that healthy infants be placed in the supine position (i.e., on their backs) to sleep (American Academy of Paediatrics Task Force on Infant Positioning and SIDS, 1992). Canada followed suit in February 1999, when the Canadian Foundation for the Study of Infant Deaths (CFSID), the Canadian Paediatric Society (CPS) and the Canadian Institute for Child Health (CICH) released a joint statement, entitled *Reducing the Risk of Sudden Infant Death Syndrome (SIDS) in Canada.* The current version of the statement recommends also that all healthy infants be placed supine to sleep (Government of Canada, CPS, CICH, & CFSID, 2011). Evidence supported the supine sleep position to reduce the incidence of SIDS; indeed SIDS mortality in Canada decreased from 0.6 per 1000 in 1999 to 0.35 per 1000 in 2004 (Smylie & Sauve, 2009). Although the most significant benefit of supine sleeping is reduced infant mortality, it is not without consequence. The supine sleep position has been thought to contribute to an increase in positional plagiocephaly across Canada (Dubé & Flake, 2003; Neufeld & Birkett, 2000). However, no literature exists on the incidence or prevalence of the condition in the Canadian context.

Definition

Plagiocephaly is defined as a malformation of the skull producing the appearance of a lopsided head (Thomas, 1997). The term plagiocephaly has been generically used to describe distortion of the cranium that occurs from both premature fusions of the cranial

sutures (synostotic plagiocephaly) as well as from external moulding forces (deformational plagiocephaly) (Littlefield & Kelly, 2004). These external forces are associated with parents' or caregivers' positioning of infants during sleep and other activities (Collett, Breiger, King, Cunningham, & Speltz, 2005). Only recently has a clear differentiation between these two conditions been made. Various other terms have also been used in the medical literature to refer to the deformational form of plagiocephaly including: (a) positional plagiocephaly, (b) nonsynostotic plagiocephaly, (c) plagiocephaly without synostosis, (d) occipital plagiocephaly, (e) posterior plagiocephaly, (f) benign position moulding, (g) functional lambdoid synostosis, (h) skull moulding and (i) flat head syndrome (Littlefield & Kelly, 2004). Additionally, many early studies mention cranial deformation that occurred as a consequence of congenital muscular torticollis, scoliosis, neurologic issues, and so on, but never specifically use the term plagiocephaly (Littlefield & Kelly, 2004). To avoid confusion, since much of the literature uses the terms positional plagiocephaly or deformational plagiocephaly; in this thesis both terms will be used interchangeably.

According to Kane, Mitchell, Craven, and Marsh (1996), infants with plagiocephaly generally present with unilateral occipital flattening where one side of the occiput is flattened, and contralateral occipital bulging where the other side of the occiput is rounded. Infants with more severe plagiocephaly may also have asymmetric faces that might include: (a) forehead protrusion ipsilateral (same side) to the occipital flattening, (b) forehead flattening ipsilateral (same side) to the occipital rounding, (c) ear displacement where the ear on the side of the occipital flattening is located anteriorly (in

front of) and below when compared to the location of the other ear, and (d) chin deviation where the chin points in direction to the side opposite of the occipital flattening (Kane et al., 1996; Littlefield, Kelly, Pomatto & Beals, 2002). According to Najarian (1999), plagiocephaly is of significant concern because if it is not diagnosed and treated early the associated changes in facial features identified above can be permanent. This permanent chance in facial features may have adverse psycho-social implications for the child that may be at increased risk for teasing and bullying during school years. The helmet approach to treat positional plagiocephaly has been proven effective and is well documented (Bialocerkowski, Vladusic, & Howell, 2005; Bruner, David, Gage & Argenta, 2004; de Ribaupierre et al., 2007; Katzel, Koltz, Sbitany, Emerson & Girotto, 2010; Larsen, 2004; Lee et al., 2008; Lima, 2004; Littlefield, 2004; Losee et al., 2007; McGarry et al., 2008; Najarian, 1999; Robinson & Proctor, 2009; Teichgraeber et al., 2004; Xia et al., 2008;).

Rationale for Conducting the Research

Incidence and risk factors.

No research has been undertaken in the Canadian context to determine the incidence of positional plagiocephaly or the risk factors contributing to its development. Clinicians working at Head Shape Clinics (HSCs) in Canada have reported anecdotally a perception of an increased number of infants attending these specialty clinics (Dubé & Flake, 2003; Neufeld & Birkett, 1999; Neufeld & Birkett, 2000). No surveillance system exists at present in Canada to capture data on plagiocephaly.

Health care systems and plagiocephaly in the Canadian context.

In efforts to understand the magnitude of this issue in the Canadian context, phone calls were made to various children's hospitals across Canada to identify:

- specialty HSCs that have been established to deal with the observed increase in positional plagiocephaly,
- different types of health professions working in the area of plagiocephaly,
- referral mechanisms,
- assessment tools used to determine severity of plagiocephaly, and
- clinicians' perceived increase or decrease in the incidence of positional plagiocephaly based on observations at HSCs.

HSCs in Canada.

Five HSCs were discovered at various Children's Hospitals across Canada: (a) Vancouver, British Columbia; (b) Edmonton, Alberta; (c) Calgary, Alberta; (d) Winnipeg, Manitoba; and (e) Ottawa, Ontario. Although messages were left at the information desk as well as the neurology department at the Montreal Children's Hospital, no response was received and no information exists on their website pertaining to the presence of a HSC.

Of the clinics contacted, clinic days of operation ranged from one day a week to three days a week. Children diagnosed with plagiocephaly in Saskatchewan are referred to the Edmonton clinic. Patients from Northern British Columbia and the North West Territories are referred also to the Edmonton clinic (W. Beaudoin, personal communication, September 18, 2008). Although there is no clinic devoted to head shapes

in Nova Scotia, New Brunswick, PEI and Newfoundland, infants are managed by physiotherapists as the need arises (K. Atkins, personal communication, September 17, 2008). The HSC in Toronto closed after it was unable to keep up with the demand. As a result, the management of positional plagiocephaly in the Toronto area has been decentralized. Family physicians now work with infants identified as having deformational plagiocephaly and their families.

The majority of the clinics contacted were linked to local orthotists that create helmets as indicated/prescribed for treatment (K. Atkins, personal communication, September 17, 2008). In Toronto, referrals are made as required to orthotists at The Hospital for Sick Children if a helmet is the indicated treatment (A. George, personal communication, September 18, 2008; T. DaSilva, personal communication, October 1, 2008). The HSC in Calgary outsources helmet manufacturing to Orthomerica, a company based in the United States of America (U.S.) (L. Walker, personal communication, May 7, 2008).

Key professions working in the area of plagiocephaly.

Across Canada, professionals from various disciplines work in the area of plagiocephaly. Most often nurses, physiotherapists, and occupational therapists manage and run the clinics identified above. The clinic in Calgary was the only clinic identified to have paediatricians directly involved in the day to day assessments of infants.

Referral mechanisms.

Referral mechanisms vary across Canada and include family self-referrals, nurse referrals, and physician referrals. HSC formats vary across Canada from consultations

with individual families to group clinics in Vancouver where four families are seen at one time. Telehealth services are also provided out of the Vancouver clinic to areas where plagiocephaly expertise is minimal (P. Mortenson, personal communication, September 17, 2008).

Assessment tools used to identify plagiocephaly and its severity.

Although many front line health care professionals use subjective assessments to determine the presence or absence of plagiocephaly, once an infant is referred to a specialty HSC, a variety of assessment methods are used that differ across provinces. Assessment techniques identified include subjective observations, manually collecting anthropometric measurements, and the use of Orthomerica's STARscanner. Various rating scales are also used, including one produced by Cranial Technologies, Inc., another U.S. based company. This scale is used by the HSC in Calgary and in Ottawa, while the HSC in Edmonton has adapted this scale and produced a ten-point scale of its own (L. Walker, personal communication May 7, 2008; K. Dubé, personal communication, September 17, 2008; W. Beaudoin, personal communication, September 18, 2008). The STARscanner uses 3-D technology to acquire head shape data through lasers and cameras. Although the clinic in Winnipeg uses the scanner to obtain assessment data, the Calgary clinic uses the scanner after severity has been assessed by physicians to produce measurement data for helmet production (T. Martin, personal communication, September 17, 2008; J. Mair, personal communication, May 21, 2008). These data are e-mailed to Orthomerica, which then manufactures the corresponding helmet.

Although effort has been made in the Canadian context to identify reliable and valid anthropometric methods for quantifying the severity of positional plagiocephaly (Mortenson & Steinbok, 2006), a discussion with one of the authors suggests that these methods, in the end, have been proven unreliable and other more reliable means of quantifying severity are needed (P. Mortenson, personal communication, September 17, 2008).

Perceived increase or decrease of plagiocephaly.

Although no information on the incidence of plagiocephaly in Canada was found in the literature, clinicians at various HSCs across Canada report an increase over the last few years, based on observations of increased referrals, ranging from 100 referrals a year to over 1000 referrals a year (A. George, personal communication, September 18, 2008; K. Atkins, personal communication, September 17, 2008; K. Dubé, personal communication, September 17, 2008; L. Walker, personal communication May 7, 2008; P. Mortenson, personal communication, September 17, 2008; T. DaSilva, personal communication, October 1, 2008; W. Beaudoin, personal communication, September 18, 2008). This perceived increase is considered to be an underestimation in terms of incidence of positional plagiocephaly since clinicians at various HSCs across Canada do not see the infants identified in the community as having the condition. Rather, only those recognized by public health nurses (PHNs) or family physicians as requiring intervention, in the form of helmet treatment, are referred to the clinics, where they exist.

Research Objectives

Since no information exists regarding the incidence of positional plagiocephaly in the Canadian context, and the literature on risk factors is limited, a study is required that

estimates the incidence of plagiocephaly of infants 7-12 weeks of age and identifies potential risk factors for infants aged 7-12 weeks that attend the 2-month well-child clinic. In addition, since there are no agreed-upon assessment methods for positional plagiocephaly in primary care, and the fact that subjective assessments predominate, a study is required to identify and field test an appropriate assessment tool for plagiocephaly for use in well-child clinics. Furthermore, in an effort to understand patterns of care that infants with positional plagiocephaly receive, a study is required to explore how infants identified with positional plagiocephaly are followed at various levels of the health care system. A study is also required in order to explore actions of various health care professionals working with infants identified with positional plagiocephaly. The site chosen for this study is Calgary, Alberta, Canada.

Research Questions

The five research questions for the present study are:

1. What is the incidence of positional plagiocephaly in infants 7-12 weeks of age in Calgary, Alberta, Canada?

2. What are the potential risk factors for positional plagiocephaly in infants 7-12 weeks of age in Calgary, Alberta, Canada?

3. Is Argenta's (2004) Plagiocephaly Assessment Tool appropriate for use in well-child clinics?

4. What intervention and follow-up actions do PHNs take if positional plagiocephaly is identified in healthy infants at the 2-month well-child clinic?

5. What intervention and follow-up actions do clinicians (physiotherapists and occupational therapists) take when healthy infants with positional plagiocephaly are referred to the infant repositioning class after the 2-month well-child clinic visit in Calgary, Alberta, Canada?

Contributions of this Research

The reported incidence of plagiocephaly in the literature varies widely and is frequently based on anecdotal evidence of increase in the number of referrals to specialty clinics (Glasgow, Siddiqi, Hoff & Young; Kane et al., 1996; McGarry et al., 2008). In the U.S., differences in diagnostic criteria and subjective classifications are noted as factors contributing to the lack of clarity in prevalence (and perhaps incidence) information available currently (Hutchison, Hutchison, Thompson, & Mitchell, 2004). It is anticipated that this research will identify the incidence of the condition in Calgary, Alberta, Canada. It is important to know the incidence of the condition to ascertain how wide spread the issue is and if any interventions need to be implemented to prevent the development of the condition.

Second, this research will identify risk factors predictive of positional plagiocephaly in infants 7-12 weeks of age. By understanding the factors associated with the development of positional plagiocephaly, it may be possible to identify potential points for intervention to prevent positional plagiocephaly by the most appropriate health

professionals, thus reducing morbidity due to plagiocephaly and its associated burden on the health system. Morbidity, here, is defined as the number of cases of the condition in relationship to a specific population (Thomas, 1997). Therefore the nature of plagiocephaly morbidity relates to the incidence and prevalence of the condition.

Third, little work has been done to establish reliable and practical assessment tools for plagiocephaly. Argenta (2004) presents a five-point classification scale that is easily reproducible, cost-effective, and easily understood by both families and health professionals. Spermon, Spermon-Marijnen and Scholten-Peeters (2008) found Argenta's five-point rating scale to be moderately reliable ($k = .54$). This research will use Argenta's five point scale to estimate the incidence of plagiocephaly in Calgary at the population level. It is important to have a reliable tool to obtain accurate estimates of incidence and prevalence of the condition. If this tool is widely adopted, it would be possible to determine the geographic spread of positional plagiocephaly and where, geographically, specific interventions need to be implemented.

It must be noted that Argenta's tool is classified as an assessment/diagnostic tool and not a screening tool. The purpose of assessment/diagnostic tools is to establish the presence or absence of a condition, whereas the purpose of a screening test is to detect potential disease indicators (Wilson & Jungner, 1968). Since plagiocephaly does not have a disease process with accompanying changes pathophysiologically, it cannot be detected in asymptomatic infants. Therefore, it is appropriate that an assessment/diagnostic tool be used for its detection.

Fourth, this research will explore actions of various health care professionals working with infants identified with positional plagiocephaly. It is hoped that this research will also raise the awareness of PHNs about positional plagiocephaly prevention as well as the importance of early recognition and early intervention. Finally, this research will explore how infants identified with positional plagiocephaly are followed at various levels of the health care system.

Organization of this Thesis

This thesis is organized into four chapters. In chapter one, the issue is introduced and the literature is critically reviewed. The research objectives and research questions are presented in both chapters one and two. In chapter two, the study methods are discussed including the study design, inclusion criteria, sample size, data collection, data analysis, and rationale for the infant age at which data were collected. In chapter three the research results are presented followed by chapter four in which the results are discussed along with the strengths and limitations of the study, suggestions for future research, implications for PHNs, other health care professionals, and the health system.

LITERATURE REVIEW

A variety of topic areas must be considered in order to understand the process of plagiocephaly development, options for intervention and the importance of its prevention. This chapter will encompass content related to plagiocephaly in the following areas: (a) the epidemiology of plagiocephaly including current evidence of incidence and prevalence of plagiocephaly in infants, (b) various assessment methods currently used to identify plagiocephaly, (c) risk factors predictive of the development of positional plagiocephaly, (d) risk factors predictive of severity (e) plagiocephaly-torticollis co-diagnosis, (f) prevention, (g) interventions for plagiocephaly (h) early recognition of plagiocephaly and what can be done to prevent its development, and (i) potential developmental concerns associated with plagiocephaly.

PubMed, Medline and CINAHL databases were searched during the proposal development phase and every six months subsequently until the research was concluded. The literature search spanned 2007 – 2010. Key terms used to search the databases included: (a) plagiocephaly, (b) positional plagiocephaly, (c) flat head syndrome, and (d) occipital flattening. Reference lists from articles obtained from the database searches were also examined and some publications were obtained from these lists.

Epidemiology of Plagiocephaly

Recently, there has been consensus that the incidence of plagiocephaly has been increasing (Argenta, David, Wilson & Bell, 1996; Biggs, 2003; Glasgow et al., 2007; Kane et al., 1996; Littlefield, Saba & Kelly, 2004; McGarry et al., 2008; Streef & Walker, 2009; White

et al., 2010). Because of different definitions of plagiocephaly and varying methods of assessment, the true incidence and prevalence of positional plagiocephaly remains uncertain.

Incidence.

The incidence rate for positional plagiocephaly is commonly cited as 1 in 300 live births (Argenta, 2004; Hummel & Fortado, 2005; Kane et al., 1996; Liu et al., 2008). However, this rate may have been taken out of context or misinterpreted from the original source as this figure was calculated in the 1970s, well before the increase in positional plagiocephaly was observed as a result of recommendations for infant supine sleep position to prevent SIDS (Clarren, Smith & Hanson, 1979; McGarry et al., 2008). The reported incidence is based frequently on anecdotal evidence of the increase in the number of referrals to specialty clinics (Argenta, David, Wilson & Bell, 1996; Biggs, 2003; Glasgow et al., 2007; Kane et al., 1996; McGarry et al., 2008; Streef & Walker, 2009; White et al., 2010).

Only four population-based studies that report the incidence of positional plagiocephaly were located. Littlefield, Saba et al., (2004) sampled 342 infants less than 10 months old attending well-baby clinics at a single centre in Arizona, U.S. Assessments of study participants were completed by a single physician. Littlefield, Saba et al. (2004) used a clinical observation tool to guide the assessment. Infants were identified as having no plagiocephaly, mild, moderate or severe plagiocephaly. By this method, Littlefield et al. identified 52 of 342 infants (15.2%) as having some form of positional plagiocephaly. While data collection spanned January-December 1999, data were collected at a single centre, limiting the generalizability of the findings to the wider population of Arizona. It must be

noted that the exclusion criteria, the sample size calculation, and the formula used to calculate incidence were not provided.

In their cross-sectional study Peitsch, Keefer, LaBrie and Mulliken (2002) assessed 183 healthy singleton neonates that were examined at two centres 24 – 72 hours after delivery in Massachusetts, U.S. The reported incidence of plagiocephaly was 13.1% of those examined. Conversely, Peitsch et al. (2002) report the incidence of plagiocephaly in infants from multiple births to be much higher at 56% (n=18). Infants that were less than 36 weeks gestation and infants that required prolonged treatment in the intensive care unit were excluded. Anthropometric cranial measurements were taken using a spreading caliper. Two oblique cranial diameters were determined, measuring from the supraorbital point to the parietooccipital scalp at the point of maximal convexivity. These diameters were measured three times and the average was recorded in centimetres and used to calculate the transcranial difference (TCD). All measurements were made by the same person with the same measuring device, lending a high degree of reliability to the data.

In their study of 1001 consecutive healthy newborns 36 – 72 hours after delivery at a single centre over a 6 month period in 2006 in Nice, France, Rubio et al. (2009) report the incidence of neonatal posterior plagiocephaly to be 3.1% as determined by a single paediatrician that conducted subjective assessments of all of the study participants. Infants less than 32 weeks gestation and those requiring prolonged treatment in the intensive care unit were excluded.

Yet another hospital-based prospective cross sectional study of 102 healthy newborns found that 61% of infants presented with head asymmetry (Stellwagen, Hubbard, Chambers &

Jones, 2008). These assessments were made via photo analysis and took place between January-June of 2004 during the newborn physical exam before discharge from the hospital in California, U.S. Photographs were taken by two clinicians and then analyzed by one blinded investigator. The exclusion criteria and the number of centres involved in this study were not stated.

The four studies identified above have produced varying results indicating that the incidence of plagiocephaly ranges from 3.1% to 61%. It is difficult to compare these results based on differences in sampling and sample size, differences in the age range of study participants, and differences in assessment techniques for positional plagiocephaly. Of significance, some infants may have altered skull shape at birth, which van Vlimmeren et al. (2007) have demonstrated to revert to normal early in the postnatal period. In addition, in their systematic review, Bialocerkowski, Vladusic and Ng (2008) assert that the term positional plagiocephaly refers to infants older than 6 weeks of age with altered skull shape. Therefore, the incidence calculations provided by Peitsch et al. (2002), Rubio et al. (2009) and Stellwagen et al. (2008) may not be valid for population measures for the incidence of plagiocephaly. Based on the information provided above there is a clear need for a study that captures incidence data for positional plagiocephaly. The following section will discuss the literature pertaining to the prevalence of positional plagiocephaly.

Prevalence.

Three population-based studies were located that reported the prevalence of positional plagiocephaly. Hutchison et al. (2004) conducted a prospective cohort study in New Zealand of 200 infants that were recruited at birth and followed up to 2 years of age.

Of the 200 infants that were enrolled in the study, 100% were seen at 6 weeks, 198 (99%) were seen at 4 months, 196 (98%) were seen at 8 months, 192 (96%) were seen at 12 months, and 181(90.5%) were followed to 2 years. Head shape assessments of study participants were conducted at 6 weeks, 4 months, 8 months, 12 months and 2 years where each infant's head was photographed using HeadsUp^{TM}, an elastic head circumference band that is photographed from above the head. The photograph is analyzed using a custom-written software program that obtains measurements to quantify the head shape. The cephalic index and oblique cranial length ratio (OCLR) were obtained. The OCLR is the ratio of the long cross-diagonal measurement to the shorted cross-diagonal measurement. If the cephalic index was 93% or above, or the OCLR was 106% or above, the infant was included as a case. The overall prevalence rates for deformational plagiocephaly only of the cohort were 16% at 6 weeks, 19.7% at 4 months, 9.2% at 8 months, 6.8% at 12 months, and 3.3% at 24 months. Although not explicitly stated, it appears that study participants did not receive any form of intervention allowing researcher to track the natural history of plagiocephaly, indicating that head shape changes naturally over time. From this study, it appears that the severity of positional plagiocephaly increases and peaks at 4 months of life and then begins to improve. It would have been helpful to the reader if the ethics of not offering treatment were discussed, along with the long term implications (i.e., emotional costs) of the infants that may have otherwise received treatment.

In their study of 7609 infants under the age of 6 months attending various infant health care centres in The Netherlands, Boere-Boonekamp and van der Linden-Kuiper (2001)

calculated the prevalence of asymmetric flattening of the occiput to be 9.9% in their population (calculation shown). Across various infant health care centres, 167 physicians took part in the assessment of the infants. A limitation of this study is that of the 7609 infants participating in the study, only 623 showed initial signs of positional preference between 1 and 6 months of age. Only those infants were assessed for asymmetric flattening of the occiput between 7–14 months and then again between 2–3 years. Of the 623 infants that showed initial signs of positional preference, 259 infants were found to have asymmetric flattening of the occiput between 7–14 months and 68 between 2–3 years. Infants without positional preference were not assessed for plagiocephaly and hence the reported prevalence of plagiocephaly is likely to be under-estimated. The authors do not discuss training that the 167 physicians may or may not have received in preparation for data collection. The assessment methods used by the 167 physicians were not stated, and the large number of physicians involved in the assessments suggests that the assessments may not have been undertaken systematically thereby jeopardising reliability.

Glasgow et al. (2007) measured the transcranial diameter difference (TDD) using a spreading caliper to assess positional plagiocephaly of 192 infants presenting in two community primary care paediatric practices. Based on a previous validation study, the presence of positional plagiocephaly was determined to be TDD greater than 0.6. The infants ranged in age between 6 and 18 months, and were broken down into 3 age groups: (a) 6-9 months, (b) 9-12 months, and (c) 12-18 months. The prevalence of positional plagiocephaly by age group was found to be 15% in those aged 6-9 months; 20% in those aged 9-12 months; and 19% in those aged 12-18 months. The prevalence in the combined group 6-18 months was

therefore found to be 18.2%. The exclusion criteria and the number of individuals involved with assessing the infants were not stated.

The studies presented above indicate a lack of stability of prevalence results. It is difficult to compare the results of the studies based on differences in age range of study participants, sample considerations, assessment methods. There is clear need for a population-based longitudinal study that uses a standardized assessment method to estimate the prevalence of positional plagiocephaly. The following section will discuss the study aimed at describing the evolution of the shape of plagiocephaly over time.

Evolution in the shape of plagiocephaly.

Between January and April 2003, Pomatto et al. (2006) conducted a prospective, multicentre study in five locations across the U.S. (Arizona, Florida, New Jersey, North Carolina, and Texas). The intent of the research was to study the evolution of the shape of plagiocephaly, comparing head shapes in 39 families. The following measures were used: (a) mean cranial widths, (b) mean cranial lengths, (c) mean cephalic indices, and (d) mean cranial circumferences. Measures taken between infant groups (0-12 months) and parent groups were compared to the age and sex-specific norms. Student *t* test results revealed statistically significant differences between the mean cranial widths of specific sex and age groups when compared to their normative data (females 0-6 months: $p = .0011$; males 0-6 months: $p = .001$; females 6-12 months: $p = .0589$; males 6-12 months: $p = .0017$; adult female: $p = .0001$; adult male: $p = .0006$). Therefore, this study revealed that the crania of these infants were wider than expected. The mean cephalic index was greater for infants when compared to their parents and 14.0% of the infants had a

cephalic index greater than 100, indicating that their heads were wider than they were long. All of the infant age groups had mean circumferences greater than their published norms. Most notable is over a period of 15 years the authors have observed that plagiocephaly shape changed from a parallelogram-like shape to a trapezoidal-like shape, possibly resulting from the extended time infants spend on their backs. Further longitudinal studies are necessary in order to ascertain if the observed results persist into childhood, adolescence and adulthood. Qualitative studies are also required to ascertain if any psychological or social effects of the condition exist and to what extent if they do.

The following section will discuss the various forms of plagiocephaly assessment that are currently being used.

Assessment

Traditional regard for a rounded head continues to be the guide for “normality” in North America and Europe (Habal, Castelano, Hemkes, Scheuerle, & Guilford, 2004). Little work has been done to establish reliable and practical assessment tools for plagiocephaly; therefore, subjective observational assessments of infant skulls continue to be common practice (Biggs, 2003; Cartwright, 2002; Losee & Mason, 2005; Persing et al., 2003). A few assessment methods for plagiocephaly have been presented in the literature with even fewer tested for reliability. In addition, a standard method of measurement has yet to be adopted by clinicians working in this area (McGarry et al., 2008). Furthermore, there is no agreed-upon system to quantify the degree of plagiocephaly severity in order to distinguish infants with a mild to moderate skull deformity from those with a severe skull deformity (Robinson & Proctor, 2009). The four

main assessment methods identified in the literature are: (a) a variety of ways to obtain anthropometric measurements; (b) three-dimensional computer analysis; (c) radiographic assessments including computed tomography; and (d) the use of clinical observation assessment scales. These are presented below.

Anthropometric measurements.

Four methods of anthropometric quantification for positional plagiocephaly have been proposed. The simplest measurement appears to be that proposed by Peitsch et al. (2002) and Glasgow et al. (2007) wherein the transcranial diameter difference is calculated. Using this method, anthropometric cranial measurements are taken using a spreading caliper. In the study by Glasgow et al. (2007), two oblique cranial diameters were determined, measuring from the supraorbital point to the parietooccipital scalp at the point of maximal convexivity. These diameters were measured three times and the average was recorded in centimetres and used to calculate the transcranial diameter difference (TDD). Validity testing of the method was also conducted by a craniofacial plastic surgeon that was blinded to the TDD score. The surgeon conducted the traditional subjective assessment for plagiocephaly, rating the infants' heads from 0-4 (no plagiocephaly to severe plagiocephaly). Spearman's rank correlation coefficient was used to determine if there was a statistically significant relationship between the TDD and the severity score and to assess whether there was a significant relationship between the TDD and the infants' ages or head circumferences. There was a statistically significant positive correlation between the subjective score assigned by the craniofacial plastic surgeon and the TDD (Spearman's rank correlation = .61; $p < .0002$). Every infant whose TDD was

greater than 0.6 cm had a subjective score of 2 or more, and all infants whose subjective scores were 0 or 1 had a TDD of less than 0.6 cm. Neither age (p = .08) nor head circumference (p =.24) was significantly correlated with the presence of deformational plagiocephaly. However, given that there is no gold standard for plagiocephaly assessment, it is unclear why this anthropometric method was compared to the subjective method discussed above.

Various other anthropometric measurements have also been captured in the literature. Ripley et al. (1994) consider 14 measurements of the head and face in concert. These measurements were selected on their ability to reflect plagiocephalic characteristics as well as the dimensional changes accompanying infant growth. Teichgraeber et al. (2002) uses this method as well, although neither group of authors present validity or reliability data of these 14 measures or compared this assessment method with another.

Plagiocephalometry is yet another method that has been developed by van Vlimmeren, Takken et al., 2006) to quantify asymmetry of infant skulls. This method uses thermoplastic material to mould the outline of infants' skulls; a reproduction of the skull shape is performed on paper, allowing for 12 accurate cephalometric measurements to be drawn on the paper copies. Three experienced pediatric physical therapists were trained to measure plagiocephalometry using a standardized protocol. Within one 30 minute session, three moulds were obtained for one child. One pediatric physical therapist performed the first and the third ring tests. One of the other two pediatric physical therapists performed the second ring test. In this way, 150 rings (50 per examiner) were made for 50 children whose age ranged from 0-24 months. Central

estimators were calculated as means and standard deviations. The data regarding intrarater reliability and interrater reliability were analyzed with intraclass correlation coefficients (ICC) with acceptable reliability criteria >0.75. Intraclass correlation coefficients (ICC) regarding the measurements of the drawn lines were all above 0.92 for intrarater reliability and 0.90 for interrater reliability (van Vlimmeren, Takken et al., 2006).

In contrast, Mortenson and Steinbok (2006) chose to investigate the reliability and validity of standardized anthropometric cranial vault asymmetry (CVA). A convenience sample of 71 infants was obtained from volunteering parents referred to the specialty clinic in Vancouver for assessment of plagiocephaly. Two clinicians independently recorded caliper measurements of cranial vault asymmetry (CVA) for infants referred for plagiocephaly or torticollis, and an unbiased observer recorded visual analysis scores via subjective assessments during the same visit. This CVA measurement was obtained by determining two landmark distances: the right frontozygomaticus to the left euryon; and the left frontozygomaticus to the right euryon. CVA scores were then assigned into the three predetermined severity categories (normal CVA < 3mm, mild–moderate CVA ≤ 12 mm, moderate–severe CVA > 12 mm). A 3-point Likert scale was used for the visual analysis for positional plagiocephaly. The measurers were blinded to each other's results. CVA measurements and visual analysis scores were recorded for 71 and 54 infants, respectively. Intrarater reliability was established (k = 0.98, k = 0.99), but inter-rater reliability was not (k = 0.42). In addition, the inter-rater reliability for the severity categories based upon these measures was low among assessors (k = 0.28) and did not

correlate to the visual analysis (k = 0.31). One again, it is unclear why the subjective assessment method (visual analysis) was selected as the comparison for assessment. Therefore, although effort has been made in the Canadian context to identify reliable and valid anthropometric methods for quantifying the severity of positional plagiocephaly, a discussion with one of the authors suggests that these methods, in the end, were proven to be unreliable and other means of quantifying severity are much needed (Mortenson & Steinbok, 2006; P. Mortenson, personal communication, December 1, 2008).

The drawback of the anthropometric methods presented above is the reliance on the assessors' subjective identification of cranial (and/or facial) landmarks used for taking the measurements. The fact that head shape is measured at only one level with anthropometric measurements may be of concern since deformational plagiocephaly is a three-dimensional problem and, therefore, in recording data in this way, some information may be lost (McGarry et al., 2008).

The above discussion provides details of the various methods used to conduct anthropometric assessments for plagiocephaly. The range of difference indicates that clinicians working in the area are unclear about which form of anthropometric assessment is the most useful. The following section will discuss three-dimensional computer analysis as a form of plagiocephaly assessment.

Three-dimensional computer analysis.

Various authors have proposed the use of three-dimensional computer analysis of the skull as a diagnostic tool for plagiocephaly (Donegan, O'Flaherty, & Kernohan, 1996; Glat et al., 1996) although few have been tested for validity and/or reliability. The use of three-

dimensional computer analysis of the skull allows for a quick and non-invasive method to capture the shape of infants' heads with minimal discomfort to the patient (McGarry et al., 2008). According to McGarry et al. (2008) these systems have the potential to support standardized head shape measurements and the use of standard landmarks; they would enable the creation of a database to allow for long-term analysis.

Hutchison, Hutchison, Thompson, and Mitchell (2005) developed a method to quantify the degree of positional plagiocephaly via digital photography. They recruited 31 case patients from outpatient plagiocephaly clinics and 29 control patients from other outpatient paediatric clinics. All case infants that participated in the study had been diagnosed with positional plagiocephaly and were between 2 and 12 months of age. Hutchison et al. (2005) used the digital photograph technique HeadsUp^{TM} wherein infants' head shapes were measured using digital photographs of a head circumference band and a flexicurve ruler. Flexicurve tracings were scanned, and both the digital photos and the scanned flexicurve tracings were analyzed using a custom-written computer program that generated anthropometric calculations. The oblique cranial length ratio (OCLR) was used to quantify cranial asymmetry and the results of the study indicate that an OCLR $\geq$ 106% defines the presence of plagiocephaly. Paired *t* tests on the standard deviations of the photo and flexicurve sets for each infant showed that for both cases and controls, there was less variation in the photo measurements than in the flexicurve measurements for cephalic index, OCLR, and right and left ear angles ($p < .0001$). The training required for obtaining such measurements or the costs associated with obtaining such measurements were not discussed by the authors.

Schaaf, Wilbrand, Boedeker, and Howaldt (2010) conducted a study to determine the accuracy of photographic assessment when compared with anthropometric measurements in deformational plagiocephaly. Standardized digital images in the supracranial view and cranial anthropometric measurements were obtained from 122 children between the ages of 3 and 15 months. The photographs were assessed using Quick Ceph^{TM} software. The cephalic index and cranial vault asymmetry index were the measurements used to indicate the degree of cranial deformity. Children were classified into plagiocephaly, brachycephaly, and the combination of both. To determine the interobserver variability, two clinicians separately measured the cephalic index and cranial vault asymmetry index from digital photographs in 70 infants of the plagiocephalic group. To compare the reliability for these methods of obtaining the cephalic index and cranial vault asymmetry index, the differences between photographically and anthropometrically derived values were plotted against anthropometrically derived values alone (Bland-Altman plots). Comparison between observers revealed excellent agreement, detected by the intraclass correlation coefficient of .982 for the cephalic index and .946 for the cranial vault asymmetry index. This study demonstrated that although digital photography is a rapid and noninvasive tool for quantifying cranial deformities, the reliability of the tool remains in question. Although the authors attempt to quantify reliability of the tool against anthropometric measurements, it remains clear that there is no gold standard currently available for the assessment of plagiocephaly.

Atmosukarto et al. (2010) developed an automated procedure for three - dimensional characterization of positional plagiocephaly that does not depend on human landmark selection. The authors assert that three-dimensional representation is of special importance to the quantification of positional plagiocephaly as it allows for measurement of volume displacement in the parietal and lower occipital regions, areas that are not covered by two-dimensional methods. Data from 90 infants with deformational plagiocephaly and 50 infants without positional plagiocephaly were analyzed. Two-dimensional histograms of surface normal vector angles were extracted from three-dimensional mesh data and used to compute the severity scores.

The outcome measures consisted of the left posterior flattening score (LPFS), the right posterior flattening score (RPFS), the asymmetry score (AS), the absolute asymmetry score (AAS), and an approximation of a previously described two-dimensional measure, and the oblique cranial length ratio (adjusted OCLR). Two-dimensional histograms localized the posterior flatness for each participant. The authors fit receiver operating characteristic curves and calculated the area under the curves (AUC) to evaluate the relative accuracy of positional plagiocephaly classification using the above measures. The AUC statistics were AAS = 91%, LPFS = 97%, RPFS = 91%, AS = 99%, and adjusted OCLR = 79%. Atmosukarto et al. conclude that this novel three-dimensional based posterior severity scores provided better sensitivity and specificity in the discrimination of plagiocephalic and typical head shapes than the two-dimensional measurements provided by a close approximation of OCLR.

Littlefield, Kelly, Cherney, Beals, and Pomatto (2004) advocate for the use of a three-dimensional imaging system to obtain a digital image of an infant's cranium to replace the manual plaster-casting technique used during the process of fabricating cranial remodelling helmets. The system has been proven to be safe and uses 18 triangulated digital cameras and the projection of random infrared patterns to capture a 360° image of an infant's cranium instantaneously, including the face and top of the head (Littlefield, Kelly et al., 2004).

The above representation of the literature on three-dimensional computer analysis demonstrates the variety of techniques that have been developed in order to conduct plagiocephaly assessments in this fashion. However, it is unclear which method is the most accurate and the settings in which these assessment methods would be most appropriate. The following section will discuss radiographic assessment.

Radiographic assessments: Computed tomography.

In their study, Abbott et al. (1998) used computed tomography (CT) scans to determine if the cranial volume of infants with deformational plagiocephaly is different from that of the most commonly cited intracranial volume data from Lichtenberg. They recruited 66 infants, aged 2.5-20.7 months, from California, U.S. They found that the cranial volumes of the infants with deformational plagiocephaly did not significantly differ from that of Lichtenberg. Hence they conclude that the use of CT scans for the purpose of identifying plagiocephaly via measuring cranial volume alone is not considered an appropriate assessment technique.

Sze et al. (2005) identify that there are two causes of posterior plagiocephaly: posterior deformational plagiocephaly (generally presenting as a parallelogram head shape) and

unilateral lambdoid synostosis (generally presenting as a trapezoid head shape). It is important to differentiate between the two as their treatment protocols are very different. True lambdoid fusion occurs in only 2–3% of patients with posterior plagiocephaly, and a CT scan with volumetric data are the next diagnostic step if a synostosis is thought to be the cause. The imaging of lambdoid synostosis will demonstrate true osseous fusion and not a functional fusion, i.e., a "sticky" lambdoid although the suture is open (Peitsch et al., 2002) as well as altered cranial volume. Since the physical findings of synostotic posterior plagiocephaly are not clearly different from those of deformational posterior plagiocephaly, CT is necessary if the physical findings are suspicious for lambdoid synostosis (Ehret, Whelan, Ellenbogen, Cunningham, & Gruss, 2004; Losee & Mason, 2005; Mulliken, Vander Woude, Hansen, LaBrie, & Scott, 1999; Pollack, Losken & Fasick, 1997).

Conversely, Losee et al. (2005) used CT scans of children clinically diagnosed with nonsynostotic occipital plagiocephaly (n = 26) and compared them with CT scans of children diagnosed with lambdoid craniosynostosis (n = 7). Suture and cranial morphology, ear position, and endocranial base angles were qualitatively and quantitatively compared. They found that nonsynostotic occipital plagiocephaly sutures demonstrated areas of focal fusion (25%), endocranial ridging (78%) narrowing (59%), sclerosis (19%), and changes from overlapping to end-to-end orientation (100%). No sutures demonstrated ectocranial ridging. All cases of nonsynostotic occipital plagiocephaly presented with ipsilateral occipital flattening, 85% with ipsilateral frontal, and 95% with contralateral occipital bossing producing parallelogram morphology. In contrast, a greater frequency of sutures in lambdoid craniosynostosis patients

demonstrated nearly complete obliteration ($p < .001$) with ectocranial ridging ($p < .001$); significantly more of these patients presented with ipsilateral occipital flattening with compensatory ipsilateral mastoid ($p < .001$) and contralateral parietal ($p < .01$) bossing, producing a trapezoid morphology. Sutures from nonsynostotic occipital plagiocephaly patients showed endocranial ridging, focal fusions, and narrowing, previously reported as lambdoid craniosynostosis. This study demonstrates that, in contradiction to previous reports, lambdoid craniosynostosis is not radiographically unique among suture fusions. This work establishes the radiographic diagnosis of positional plagiocephaly. van Vlimmeren, Takken, et al. (2006) assert that using three-dimensional computed scanning is the most valid and reliable morphometry to obtain an impression of the shape of the head but serial application of CT scans exposes infants to radiation and risks of complications from general anaesthesia.

Due to the risk of radiation exposure and complications from general anaesthesia, it would appear that although this method may be the most valid and reliable, it would not be practical for use in primary care where assessments need to be obtained quickly in order to determine the appropriate course of action. The following section will discuss the various forms of clinical observation plagiocephaly assessment tools presented in the literature.

Clinical observation assessment tools.

While the above assessment methods appear to be extensive and, perhaps, appropriate for assessment at specialty clinics, the clinical observation scales presented in this section appear to be amenable to assessing for positional plagiocephaly in primary care.

Littlefield, Saba et al. (2004) used set criteria to guide their assessments. Infants were identified as having no, mild, moderate or severe plagiocephaly. The four assessment items used to evaluate infants required clinicians to classify the type of plagiocephaly observed as: (a) mild (posterior asymmetry); (b) moderate (posterior asymmetry, ear malposition with a discrepancy of half an inch or more, and minimal frontal asymmetry and facial asymmetry); (c) and severe (significant posterior asymmetry, ear malposition of one inch or more, frontal asymmetry on the affected side, and facial asymmetry). While this may appear to be feasible to complete in the primary care setting, the criteria developed by Littlefield, Saba et al. (2004) requires the use of a measuring tape or spreading caliper in order to quantify the degree of ear misalignment in terms of inches. It appears that Littlefield's (2004) criteria used to collect data in 1999, is a precursor to the tool developed in 2002 by Cranial Technologies, Inc. Littlefield is identified as one of four Research Leaders employed by Cranial Technologies, Inc. (www.cranialtech.com).

Losee and Mason (2005) speak of a severity assessment scale for plagiocephaly developed by Cranial Technologies, Inc., a U.S. based company that manufactures cranial orthoses. Although not proven to be reliable, the elaborate rating system assesses posterior flattening, ear misalignment, forehead asymmetry, neck involvement and facial asymmetry and four different degrees of severity for each category as a way to classify positional plagiocephaly (Losee & Mason, 2005). The clinical observation tools, published by Littlefield, Saba et al. (2004) and the tool developed by Cranial Technologies, Inc. (2002) introduces a conflict of interest. Since Cranial Technologies, Inc. manufactures helmets used to treat

positional plagiocephaly, there is a vested interest of the company to create these assessment tools.

Argenta (2004) presents a five-point classification scale for positional plagiocephaly that is easily reproducible, cost-effective and easily understood by both families and health care professionals without requiring laborious measurement. The five categories increase in severity depending on the following: (a) posterior asymmetry, (b) ear malposition, (c) frontal asymmetry, (d) facial asymmetry, (e) forehead bossing, and (f) posterior cranial vertical growth. It must be noted that neck involvement is not part of this assessment and the scale does not account for differences in severity observed within each type of identifiable plagiocephaly.

When compared to other plagiocephaly assessment methods presented above (anthropometric measurements, three-dimensional computer analysis, and radiographic assessments in terms of computed tomography), clinical observation guided by assessment tools is the most cost effective assessment method for positional plagiocephaly. This cost-effectiveness of the tool can be considered in terms of the financial costs associated with the assessment procedure and the time associated with conducting the assessment. With respect to anthropometric measurements, although the cost would be minimal in terms of equipment required (spreading calipers), the time associated with taking numerous craniofacial measurements would be substantial. Various three-dimensional computer analyses methods have been proposed. Some using scanners while others use digital cameras to obtain pictures which are then analyzed using custom-written software. Although the time required to obtain scans or photographs

of an infant's head may be minimal, the time required to analyze such data may not be feasible in the primary care context. In addition, the cost associated with having such equipment in each clinic room would be large. Another assessment method identified to assess for positional plagiocephaly is the CT scan. These scans can be very helpful in quantifying the degree of skull deformity, especially because they are pperformed in three dimensions. Programs are available for quantifying volumetric differences in the different areas of the skull. One drawback of this method is that may require considerable wait times (especially if the equipment is required as part of diagnoses for more acute patients). A second drawback is that the equipment necessary for such assessment is very expensive, and thirdly it would subject infants to radiographic exposure and complications from anesthesia may arise (Argenta, 2004).

While the clinical observation scales presented above appear to be amenable to assessing for positional plagiocephaly in primary care, the following question remains: how does one determine which assessment scale is most appropriate for clinical observation? Upon review of the literature, the following criteria, adapted from Wilson and Jungner (1968) will be used to assess the appropriateness of clinical observation assessment tools for the purposes for use in primary care:

1. The suitability/feasibility of the assessment tool. The suitability of the tool refers to its ease of use, the time it takes to conduct an assessment, the level of expertise required to use the tool, the level of intrusiveness of the assessment and its utility as a teaching tool within the primary care context.

2. The acceptability of the assessment tool to the population. The acceptability of the tool refers to the willingness of the target population to agree to have the assessments conducted as proposed.
3. The cost-effectiveness of the assessment tool.
4. The validity of the assessment tool. The validity of the tool refers to both face validity and content validity. Face validity is the validity of a test at face value. Specifically, a test can be said to have face validity if it "looks like" it is going to measure what it is supposed to measure; it is sometimes referred to as a “click of recognition” (Creswell, 2007). Content validity is the extent to which the measurement incorporates the domain of the condition under study (Porta, 2008).
5. The reliability of the assessment tool. Reliability is the degree of stability exhibited when a measurement is repeated under identical conditions (Porta, 2008).
6. The yield of the assessment tool. The yield is described as the ability of the tool to identify a previously unrecognized medical condition.

Selection of an appropriate assessment tool for the current study will be based on the criteria presented above. The rationale for the plagiocephaly assessment tool selected for this study will be discussed in chapter two and will be based on four of the six criteria presented above: (a) the suitability/feasibility, (b) the cost effectiveness, (c) reliability, and (d) validity. The present study will collect information on the suitability/feasibility of using the tool in the well-child clinic context, the acceptability of the tool to the target population, how reliability of the tool was maintained, and the yield.

While it is important to identify an appropriate method of assessment for positional plagiocephaly, it is equally important to understand the risk factors that may contribute to the development of the condition. Understanding of risk factors would assist in identifying points of intervention for plagiocephaly prevention. In the following section, the literature on risk factors for the development of plagiocephaly will be discussed.

Risk Factors

In this section, the risk factors for positional plagiocephaly identified in the research will be outlined. Research articles presented here include only those wherein analysis of the descriptive data were complete.

Kane et al. (1996) conducted a retrospective chart review of 269 infants diagnosed with positional plagiocephaly that presented at a tertiary craniofacial centre in Missouri between 1979 and 1994. In order to identify the risk factors for developing positional plagiocephaly, infants that presented between 1992 and 1994 were compared with those of the 1993 Missouri live birth cohort and analyzed via χ^2 analysis. Risk factors found to be significant included male sex ($p = .04$) and forceps delivery ($p = .05$). A limitation of the study is that neither the age range of the study participants nor those of the comparison group were provided.

As mentioned previously, Peitsch et al. (2002) conducted a cross-sectional study of 183 healthy singleton neonates that were examined at two centres 24 – 72 hours after delivery in Massachusetts, U.S. Infants with a gestational age < 36 weeks and infants that required prolonged treatment in the intensive care unit were excluded. Forceps and vacuum-assisted vaginal delivery ($p < .001$) and prolonged labour ($p = .002$) were

identified as risk factors for the development of plagiocephaly. A limitation of this study is the small sample size.

In their study of 192 infants seen at two community primary care paediatric practices, Glasgow et al. (2007) found that the risk factors found to be associated with deformational plagiocephaly included sleeping supine (OR: 3.5; 95% CI: 1.6–7.5) and infant head position preference (OR: 2.2; 95% CI: 1.0–4.9). They also found that varying infants' sleep position decreased the risk of deformational plagiocephaly (OR: 0.40; 95% CI: 0.2–0.9). The amount of time the infant was reported to spend in either a car seat or in the prone position ("tummy time") was not related to the presence or absence of deformational plagiocephaly. Furthermore, there was no relationship between the presence of deformational plagiocephaly and the parental report of receiving any advice regarding prevention (Glasgow et al., 2007). The information on risk factors was presented for the entire group of 192 infant aged 6-18 months. It appears that data analysis with computation of odds ratios was univariate in nature.

In their prospective cohort study of 200 infants, Hutchison et al. (2004) collected data on risk factors for the development of positional plagiocephaly in New Zealand. The study participants were followed to 2 years. Data were collected at various points (6 weeks, 4 months, 8 months, 12 months and 24 months). Owing to the small number of cases at 8, 12, and 24 months, the authors presented data on risk factors for 6 weeks and 4 months only. At 6 weeks, the variables that remained significant in the final multivariate model were: (a) limited newborn passive head rotation (adjusted OR: 9.51; 95% CI: 2.59–34.94), (b) 6-week sleep position supine only (adjusted OR: 5.27; 95% CI: 1.81–15.39), and (c) surprisingly had tummy time per day greater than 1 hour (adjusted OR: 3.99; 95% CI: 1.42–11.23). At 6

weeks, no significant differences were identified between cases and control study participants for: (a) obstetric factors; (b) socioeconomic factors; (c) sex; (d) newborn head circumference; (e) abnormal head shape at the newborn assessment; (f) the presence of hair loss on the back of the head; (g) snoring; (h) activity level; (i) newborn or 6-week weight, length and head circumference; (j) temperament rating; (k) the amount of reported tummy time per day; (l) breastfeeding; (m) pacifier use; (n) the use of positioning aids or pillows; (o) mattress firmness; (p) maternal hand dominance; and (q) the mother's preferred holding position (Hutchison et al., 2004).

At 4 months of age, the variables that remained significant in the final multivariate model were: (a) limited passive head rotation at birth (adjusted OR: 6.51; 95% CI: 1.85–22.98), (b) limited active head rotation at 4 months (adjusted OR: 3.11; 95% CI: 1.21–8.05), (c) tried but unable to vary head position at 6 weeks (adjusted OR: 4.28; 95% CI: 1.58–11.59), (d) low activity level at 4 months (adjusted OR: 3.28; 95% CI: 1.16–9.29), and (e) average to difficult rating on pictorial temperament test (adjusted OR: 3.30; 95% CI 1.17–9.29). No significant differences were found between the 4-month case and control groups for: (a) obstetric factors; (b) socioeconomic factors; (c) newborn head circumference; (d) abnormal head shape at newborn assessment; (e) weight, length, and head circumference measured at the 3-month well-child check-up; (f) preferential head orientation; (g) developmental delays either reported on the PDQ-II (Revised Denver II Prescreening Questionnaire); (h) number of cautions or delays on the PDQ-II; (i) sleep position at 4 months; (j) hair loss; (k) tummy time and back time at 4 months; (l) upright time; (m) bouncinette time; (n) breastfeeding, (o)

preferred side of feeding; (p) pacifier use; (q) the use of positioning aids; (r) mattress firmness; (s) mother's handedness; and (t) preferred holding position.

In summary, at 6 weeks, the variables that were found to be significant in the final multivariate model were: (a) limited newborn passive head rotation, (b) 6-week sleep position supine only, and (c) tummy time per day greater than 1 hour. At 4 months of age, the variables that were found to be statistically significant were: (a) limited passive head rotation at birth, (b) limited active head rotation at 4 months, (c) tried but unable to vary head position at 6 weeks, (d) low activity level at 4 months, and (e) average to difficult rating on pictorial temperament test. One limitation of this study was that although the authors intended to follow the study participants until 2 years of age, many infants were lost to follow-up making it difficult to obtain sufficient data for risk factors for positional plagiocephaly at 8 months, 12 months and 2 years. Another limitation of this study is the small sample size.

In a case-control study, Hutchison, Thompson and Mitchell (2003) recruited 100 infants that received a diagnosis of nonsynostotic plagiocephaly as cases and compared them with 94 controls that were selected from a citywide database of infants. All infants were between 2-12 months old. Information concerning sociodemographic variables, obstetric factors, infant factors and infant care practices was collected via parental interview. Several variables were significant in the multivariate analysis. With respect to sociodemographic factors, maternal education was significant with an increased risk for the group with no or low qualifications (adjusted OR: 5.61; 95% CI: 2.02–15.56). Regarding obstetric factors the following variables were found to be significant: (a) parity with an increased risk for firstborn infants (adjusted OR: 2.94; 95% CI: 1.46–5.96), (b) gestation with case patients significantly

more likely to be preterm (less than 37 weeks) (adjusted OR: 3.26; 95% CI: 1.02–10.47), and (c) more case mothers had attended antenatal classes for this or a previous infant when compared to control participants (adjusted OR: 6.61; 95% CI: 1.59–27.47). No significant differences were detected between the groups for: (a) the mother's age, (b) multiple births, (c) reduced amniotic fluid, (d) uterine abnormality, (e) position in last month of pregnancy, and (f) the number of weeks that the head was engaged.

Turning now to infant factors, the following variables were found to be significant: (a) male sex (adjusted OR: 2.51; 95% CI: 1.23–5.16), (b) the mother more likely to have reported a perceived developmental delay (head lag problems or late rolling over) (adjusted OR: 3.32; 95% CI: 1.01–10.85), (c) the mother more likely to have reported average levels of activity or below (adjusted OR: 3.23; 95% CI: 1.38–7.56), (d) to have had a preferred head orientation (adjusted OR: 37.46; 95% CI: 8.44–166.32), (e) to have had asymmetric hair loss, (f) to be firstborn (adjusted OR: 2.94; 95% CI: 1.46–5.96), (g) and to have been premature (adjusted OR: 3.26; 95% CI: 1.02–1.47). With respect to infant care factors, the following variables were found to be significant in the first 6 weeks: (a) cases were more likely to have been sleeping in the supine position (adjusted OR: 7.02; 95% CI: 2.98–16.53), (b) not to have had the head position varied when put down to sleep (adjusted OR: 7.11; 95% CI: 2.75–18.37), and (c) to have had less than 5 minutes of tummy time per day (adjusted OR: 2.26; 95% CI: 1.03–5.00). No significant differences were found for mattress firmness, use of a pacifier and mother's hand dominance. One significant limitation of this study is the small sample size. Due to the small sample size, a second limitation of the study is the inability of the researchers to identify if the risk factors vary over time.

In their prospective cohort study comparing risk factors for deformational plagiocephaly at 7 weeks of age, van Vlimmeren et al. (2007) followed 380 neonates born at term in Veghel, Netherlands. Assessments for plagiocephaly were undertaken at birth and 7weeks using plagiocephalometry parameter oblique diameter difference index. Data regarding obstetrics, sociodemographics, asymmetry of the skull, anthropometrics, motor development, positioning, and care factors were collected. Only 9 of the 23 children that presented with deformational plagiocephaly at birth still had the condition at 7 weeks. In 75 other children, deformational plagiocephaly was not present at birth but developed between birth and 7 weeks. One variable was identified as a risk factor at birth: male sex (adjusted OR: 5.4; 95% CI: 1.92–15.28). The following variables were found to be significant at the multivariate level at 7 weeks age: (a) male sex (adjusted OR 2.0; 95% CI: 1.12–3.41), (b) first born birth order (adjusted OR: 2.4, 95% CI: 1.36–4.22), (c) positional preference when sleeping (adjusted OR: 7.5; 95% CI: 3.49–14.37), and (d) positioning to the same side when bottle feeding (adjusted OR: 1.8; 95% CI: 1.01–3.30). No significant differences were found with respect to tummy time, side lying positioning when awake, only bottle feeding, positioning on chest of drawers (change table), sleeping supine after 2 weeks of age, delivery type, presentation at birth, parity, education level of father and education level of mother.

Littlefield, Kelly, Pomatto and Beals (1999), conducted a retrospective review of their database of Cranial Technologies, Inc. located in Arizona, U.S. in order to determine if infants of multiple pregnancies were at higher risk of developing deformational plagiocephaly. They phoned the parents of identified multiples and asked them to complete a survey regarding prenatal and postnatal history. Similar information was obtained from vital statistics

information from the state of Arizona. The authors compared the incidence of multiple birth infants presenting with deformational plagiocephaly with the incidence of multiple births in the Arizona population. A χ^2 analysis of 60 patients between 1993 and 1996 confirmed that a statistically significant number of multiple-birth infants presented with deformational plagiocephaly (1993: $\chi^2 = 28.292$; $p < .001$; 1994: $\chi^2 = 15.162$; $p < .001$; 1995: $\chi^2 = 26.574$; $p < .001$; 1996: $\chi^2 = 23.956$; $p < .001$).

In another study conducted by the same authors (Littlefield, Kelly, Pomatto & Beals, 2002), they detailed the medical histories were detailed on 140 sets of twins (280 study participants) that had presented for treatment at 1 of 9 specialty clinics across the United States. Information about prenatal and postnatal history of the infants was obtained through a chart review as well as review of patient databases maintained on all infants that received treatment. Follow-up interviews were completed to verify the information recorded as well as to obtain additional information about the twin that had not received treatment. Statistical analyses of pairs of twins demonstrated that the lower in utero infant was significantly more likely to be diagnosed with deformational plagiocephaly ($\chi^2 = 17.391$; $p < .001$); the more severely affected infant was significantly more likely to have neck tightness or torticollis ($\chi^2 = 46.380$; $p < .006$), as well as to have been carried in the vertex position ($\chi^2 = 7.408$; $p < .002$).

In a case-control study, McKinney, Cunningham, Holt, Leroux & Star (2009) identified cases born 1987–2002 in Washington State and diagnosed with plagiocephaly at the Craniofacial Centre at Seattle Children's Hospital. The case definition required a clinical diagnosis noted in the chart of positional or deformational plagiocephaly or brachycephaly before 18 months of age. Their sample consisted of 2764 cases and 13,

817 controls. A list of cases was provided to the Washington State Department of Health (DOH), where it was cross referenced with birth certificate and hospital discharge data by using each case's name, date of birth and sex. Five controls per case were randomly selected, matching on hospital and birth year. Birth certificate information was linked by the DOH to hospital discharge data for the mother and infant. Risk factor information was abstracted from birth certificate and hospital discharge data. The variables found to be significant at the multivariate level are presented. Cases (n = 2764) were more likely than controls (n = 13 817) to be male (adjusted OR: 2.0; 95% CI: 1.8–2.2) or to have been low birth weight (1500–2499 vs. 2500–3999 g) (adjusted OR: 1.6; 95% CI: 1.3–1.9). Infants with plagiocephaly were at greater risk of having been delivered preterm (32-36 vs. 37-41 weeks' gestation) but at no greater risk of extreme prematurity (<32 weeks) (adjusted OR: 1.7; 95% CI: 1.5–1.9). Cases were also more likely than controls to: (a) have been small for gestational age (adjusted OR: 1.7; 95% CI: 1.5–1.9), (b) a twin or higher order multiple birth (adjusted OR: 3.2; 95% CI: 2.7–3.7), and (c) first-delivered multiple birth infants were more likely than singletons to have plagiocephaly (adjusted OR 4.5; 95% CI: 3.2–6.3). Infants with plagiocephaly were more likely to have been injured at birth (adjusted OR: 1.4; 95% CI: 1.2–1.7). Cases were also more likely to have been diagnosed with a congenital anomaly (adjusted OR: 2.0; 95% CI: 1.8–2.3). Associations between plagiocephaly and craniofacial conditions such as torticollis (adjusted OR: 57.7; 95% CI 16.5–202.1), cleft lip and palate (adjusted OR: 6.9; 95% CI: 3.4–14.1), hydrocephalus (adjusted OR: 5.5; 95% CI: 2.2–13.8), and skull and bone anomalies (adjusted OR: 12.9; 95% CI: 6.6–25.3) appeared remarkably strong. Mothers

of case infants were more likely to have been ≥35 years old at the time of the infant's birth (adjusted OR: 1.3; 95% CI: 1.1–1.5). One limitation of the study is that the age range of the study participants was not disclosed.

Losee, Mason, Dudas, Hua, and Mooney (2007) conducted retrospective medical chart reviews and telephone interviews of 105 infants diagnosed and treated for deformational plagiocephaly. A standardized questionnaire was used during the telephone interview to obtain information and confirm data obtained from the chart review. The severity of positional plagiocephaly, on initial presentation and at final follow-up or completion of treatment, was scored subjectively by a single craniofacial surgeon using a positional plagiocephaly severity score ranging from 1 to 9, where 1 represents no identifiable deformity and 9 represents the most severe imagined deformity. Data from the cohort of positional plagiocephaly patients were then compared with national statistics regarding breastfeeding, torticollis, multiple gestation pregnancies, and prematurity. When comparing the prevalence of breast-fed children in the cases with that of the general population (controls) a statistically significant difference was found at all ages the infants in the cohort with cases having been breast-fed at a significantly lower rate than in the general population (χ^2= 11.02; $p < .05$). Cases demonstrated a significantly greater incidence of torticollis than the controls (χ^2 = 184.0; $p < .05$). Cases also demonstrated a higher rate of multiple gestation pregnancies than controls (χ^2 = 37.61; $p < .05$). No significant difference was found between the cases and controls for prematurity.

Due to the variability in study design, age of the infants, inclusion criteria and sample size, it is difficult to compare the results of the studies presented above. However, it is clear that risk factors for the development of positional plagiocephaly vary by infant age. The research presented above indicates that only two studies have been published that speak to the risk factors for developing positional plagiocephaly between 6-7 weeks of age (Hutchison et al., 2004; van Vlimmeren et al. 2007) . This is a significant gap if we are to determine how to prevent the development of the condition. This demonstrates that there is clear need for additional research that identifies the risk factors for the development of positional plagiocephaly for infants aged 6-7 weeks. There is also need for a longitudinal study to track changes in risk factors over time.

While it is important to identify risk factors for positional plagiocephaly in order to identify prevention strategies, it is equally important to consider the predictors of severity of plagiocephaly. The following section will discuss the predictors of severity of plagiocephaly.

Predictors of Severity

Management strategies for deformational plagiocephaly rely on the 'mild', 'moderate', or 'severe' classifications that are often subjective and vary between clinicians (McGarry et al., 2008). Although an objective classification system that grades the severity of deformational plagiocephaly is yet to be developed and agreed upon, Oh, Hoy and Rogers (2009) conducted a prospective cohort study to determine if various identified risk factors could be predictive of the severity of deformational plagiocephaly. A total of 434 patients diagnosed with deformational plagiocephaly were evaluated between January 2006 and December 2007. The assessment method included

anthropometric calculation of the transcranial difference in oblique cranial lengths (TCD), evaluation of head tilt and rotational asymmetry. Parents of infants also completed a standardized questionnaire assessing potential risk factors for deformational plagiocephaly before the assessment. Pearson correlation coefficient, one way analysis of variance, and the two sample t-test were used to quantify the relationship between identified risk factors and TCD. Risk factors associated with more severe deformational plagiocephaly included: (a) multiple gestation ($p < .05$), (b) lower gestational age ($p < .05$), (c) male sex ($p < .05$), (d) head positional preference ($p < .01$), (e) pre-existing diagnosis of torticollis by a paediatrician ($p < .05$), (f) head tilt ($p < .005$), and (g) head rotational asymmetry ($p < .000$). This study suggests that although a standardized method to evaluate degree of severity has yet to be established, the relationship between the severity of deformational plagiocephaly and certain risk factors can be quantified.

Much of the research above indicates that torticollis is a risk factor for both the development of positional plagiocephaly and a predictor of severity of the condition. The following section will discuss the current literature that considers the plagiocephaly – torticollis co-diagnosis and its implications.

Plagiocephaly – Torticollis Co-Diagnosis

Torticollis is the clinical term for a twisted neck, caused by the shortening of the sternocleidomastoid (SCM) muscle located on either side of the neck (van Vlimmeren, Helders, van Adrichem, & Engelbert, 2006). Do (2006) describes it as a spectrum of disease while Golden, Beals, Littlefield & Pomatto (1999) make a distinction between weakness of the sternocleidomastoid (SCM) muscle (SCM imbalance) and true torticollis

(congenital muscular torticollis - CMT). SCM imbalance and CMT generally present at birth. van Vlimmeren, Helders, et al. (2006) use the term positional torticollis to describe weakness in the SCM that can develop secondary to positional plagiocephaly. The presence of unilateral fibrosis or shortening of the SCM muscle defines CMT (Pivar & Scheuerle, 2006). In many cases, infants with CMT present with a tumour or mass in the SCM during the first 3 months of life (Pivar & Scheuerle, 2006). CMT of the right or left SCM muscle causes the ipsilateral side of the neck to lose range of motion, staying tight and contracted, pulling the ear down and forward and causing the chin to tilt to the contralateral side (Golden et al., 1999). Infants with CMT generally present with the head tilted towards the shortened SCM (Golden et al., 1999). Unlike the severe decreased range of motion of the neck typical of CMT, SCM imbalance presents with less severe characteristics. Infants with SCM imbalance also constantly prefer rotating their heads to one side while supine and upright (Golden et al., 1999). In contrast, infants with SCM imbalance can have normal passive range of motion in the neck and spine, the head tilt may seem intermittent and may vary in degree of severity, and there is no tumour or mass associated with it (Golden et al., 1999).

The reported incidence of torticollis presented in the literature varies. de Chalain and Park (2005) report the incidence of torticollis to be 8.58 per 1000 live births, while according to Do (2006), CMT is a common finding in the newborn period with an overall incidence of 4 per 1000 live births. In their study, Chen, Chang, Hsieh, Yen and Chen (2005), studied 1021 consecutive infants born within 24–72 hours at a regional hospital in Taiwan from January–August 2002. Each infant underwent ultrasonography examination

to assess the condition of soft tissue around bilateral sternocleidomastoid muscle and the functional activity of the head and neck. They found abnormalities of the soft tissue in 40 infants and calculated the overall incidence rate for CMT at 392 per 1000. Stellwagen et al. (2008) examined 102 healthy newborn infants prospectively during the newborn physical exam for torticollis that included neck range of motion (ROM) assessment and for facial, mandibular, and cranial asymmetry by photographic analysis and found the incidence rate for CMT to be 160 per 1000.

The aetiology of CMT is poorly understood with little research evidence available on the topic. Several hypotheses exist including, birth trauma, intrauterine crowding, vascular injury to the SCM, infection, and heredity (de Chalain & Park, 2005; Do, 2006). Some cases of CMT have been attributed to a pseudotumor in infancy (Golden et al., 1999). It is also believed that damage during birth may give rise to a mass of fibrotic tissue in a previously abnormal muscle (Rogers, Oh, & Mulliken, 2009). Alternatively, intrauterine conditions such as malpositioning of the head in utero may cause injury, and consequent fibrosis to the SCM muscle (van Vlimmeren, Helders, et al., 2006). An infant born with this type of torticollis may present with an asymmetry of the head at birth, which can be exacerbated when the infant is laid in a supine position, giving rise to positional plagiocephaly (van Vlimmeren, Helders, et al., 2006). Although perceived to be relatively benign on its own, when CMT is considered a co –morbidity with plagiocephaly, its level of severity may need to be re-evaluated.

Torticollis is a common anomaly that has been linked with plagiocephaly (Clarren, 1981; Golden et al., 1999; Pivar & Scheuerle, 2006; Rogers et al., 2009; van Vlimmeren, Helders, et al., 2006). The specific head tilt, described above, will create a supine position in

which the contralateral side of the head has contact with the sleeping surface (Pivar & Scheuerle, 2006). Due to the limited range of motion caused by the torticollis, infants' preferential use of the contralateral side of the occiput while supine is thought to cause or perpetuate plagiocephaly (Pivar & Scheuerle, 2006). In their study of 18 hospitals or birthing centres in Texas, Pivar and Scheuerle (2006) found that plagiocephaly cases between 1994–2004 ranged from 22–376, and 5–67% were co-diagnosed with torticollis. The total number of infants assessed was not provided. Furthermore, in their study of 1021 infants, Chen et al. (2005) found that infants that had plagiocephaly had a 22-fold greater risk of abnormal findings in the SCM than those without (adjusted OR: 22.30; 95% CI: 7.01–70.95). Rogers et al. (2009) conducted a prospective cohort study between 2002-2003 and evaluated 202 infants between 3-16 months of age with cranial asymmetry. They found that although 92% of parents recalled a preferential head position at birth, only 24% of infants had been previously diagnosed or treated for torticollis. Rogers et al. (2009) assert that CMT and SCM imbalance is underreported in deformational plagiocephaly. Because the condition improves quickly in infancy, the findings may be more subtle and evidenced only by a history of preferential head position (Rogers et al., 2009). A preferential head position can be common; of 380 healthy neonates assessed at 7 weeks, 68 (17.9%) exhibited positional preference (van Vlimmeren et al., 2007).

In their retrospective chart review of 83 patients diagnosed with positional plagiocephaly that were referred to a private orthotic clinic for treatment in 1996, Golden et al. (1999) found that over three-quarters of their plagiocephalic population suffered from some form of neck dysfunction: 64% (n = 53) of the patients had sternocleidomastoid (SCM)

imbalance and 12% (n = 10) of the patients had congenital muscular torticollis (CMT). A χ^2 analysis was used to compare the SCM imbalance group to the CMT group with respect to sex and etiological factors related to positional plagiocephaly. Due to the small sample sizes, the significance of the cross-tabulated data were analyzed using the Fisher's exact test. No statistical significant difference was found between the groups for the following variables: (a) sex, (b) head shape at birth, (c) left or right sided occipital-parietal flattening, (d) left or right SCM tightness, (e) caesarean delivery, (f) *in utero* positioning, (g) breech positioning, (h) multiple birth prematurity, (i) birth trauma, and (j) sleep position at birth. The only observed difference between the groups was related to symptoms of the neck itself. Therefore, this study verified that the symptoms of SCM imbalance are consistently different from CMT. Golden et al. (1999) found that although the two conditions manifest into different symptomology, SCM imbalance does not differ in aetiology leading to positional plagiocephaly, which suggests that any degree of neck dysfunction should be considered a potential precursor to the development of positional plagiocephaly.

Torticollis may also develop secondary to plagiocephaly in many cases, although no research evidence of this proposition was found. Several authors have hypothesized that acquired deformational plagiocephaly is thought to occur when infants are placed supine and assume a "position of comfort" that most likely corresponds to their previous intrauterine lie (Losee & Mason, 2005). With this "position of comfort", an unvarying sleep position develops (Losee & Mason, 2005). In the case of a persistent positional preference of the head, positional torticollis will develop (van Vlimmeren, Helders. et al., 2006). This type of torticollis is not evidenced by morphologic changes in the SCM and

may be induced by deformational plagiocephaly at birth or one-sided persistent positioning after birth during the first 1-5 months of life (van Vlimmeren, Helders, et al., 2006).

In light of the risk factors for the development of positional plagiocephaly discussed above, and the significant plagiocephaly-torticollis co-morbidity, it is important to consider what can be done to prevent the condition. In the following section various strategies that have been brought forward in the literature are discussed.

Prevention

Several techniques for the prevention of plagiocephaly have been proposed in the literature and are presented in this section. However, it must be noted that no research evidence was found to support these methods. This lack of evidence is likely due to the lack of incidence and prevalence data available, lack of agreed-upon assessment methods used to identify plagiocephaly, and the lack of a surveillance system in place to capture any changes over time in the epidemiology of the condition.

The Alberta Infant Cranial Remodelling Program takes a community approach to prevention of plagiocephaly (Neufeld & Birkett, 1999). PHNs and nurses working in post-partum and paediatric areas have important roles to play in preventing plagiocephaly (Neufeld & Birkett, 1999). To reduce the risk of positional plagiocephaly, families are often counselled to ensure infants are experiencing a variety of positions, other than supine, while they are awake and supervised (Bialocerkowski et al., 2008; Persing et al., 2003). It is suggested that “tummy time” begin early in the postpartum period, as soon as the umbilical cord falls off (Neufeld & Birkett, 1999). Graham (2006) suggests that

infants should be on their stomachs whenever they are awake and under direct adult supervision in order to develop prone motor skills and to encourage the full range of neck rotation. Littlefield, Kelly, Reiff, & Pomatto (2003) conducted a chart review of 636 infants with plagiocephaly that were referred for treatment between 1998–2000. Littlefield at al. (2003) found that 56.6% of the infants spent less than 1.5 hrs per day; 28.6% spent 1.5 to 4 hrs per day; and 14.8% of infants spent more than 4 hrs per day in car seats, infant carriers, bouncy seats, and infant swings. Therefore, parents were encouraged to limit the time infants spend sitting in these devices (Littlefield et al., 2003; Losee & Mason, 2005 Neufeld & Birkett, 1999). Streef and Walker (2009) assert that such prevention teaching for positional plagiocephaly is effective for infants less than 4 months of age. One technique that has been proposed to prevent plagiocephaly is to alternate infants' head orientation in the crib such that the heads are positioned toward the head of the crib on even calendar days and toward the foot of the crib on odd calendar days (Dubé & Flake, 2003). Clearly, research studies are required to determine the effectiveness of the prevention methods identified above.

While the various prevention options presented above exist, the need for treatment is frequently inevitable. In the following section, the current research available on interventions to treat plagiocephaly will be presented and discussed.

Interventions for Plagiocephaly

The treatment of positional plagiocephaly generally includes two interventions: active counter-positioning and the use of orthotic helmets or headbands. Treatment for infants with a co-diagnosis of torticollis generally also includes physiotherapy. Two factors affecting the

treatment plan are age at diagnosis and severity of the deformity (Losee & Mason, 2005). As the age of the infant at age of diagnosis increases and the severity of the deformity worsens, the potential for successful correction of head shape decreases substantially (Losee & Mason, 2005; Loveday & de Chalain, 2001). In this section the mechanisms of active counter-positioning and orthotic helmets will be discussed and the effectiveness of these two methods will be compared.

The mechanism of active counter-positioning.

In active counter-positioning, parents are directed to position infants "back to sleep", but to turn babies' heads to the side opposite to the occipital flattening (Steinbok, Lam, Singh, Mortenson, & Singhal, 2007). As sleeping space is often placed against a room wall, the crib should be positioned so that the infant is required to lie on its unaffected side to obtain auditory and visual stimuli from toys and activity in the room (Losee & Mason, 2005; Persing et al., 2003). During feeding, parents are taught to position the infant so that the arm that cradles the infant eliminates pressure from the flattened side of the head rather than contributing to it (Losee & Mason, 2005). Supported upright positioning and supervised tummy time are recommended as ideal awake and play positions. However, in Calgary, clinicians at the HSC stress that side lying on the opposite side to the flattening is the most effective position for applying counter-pressure to the awake and supervised child's head, thus reshaping the head (B. Mikkelsen, personal communication, June 29, 2009). Active counter-positioning is the simplest therapy to implement, is the least traumatic to the child, and has no related cost.

The mechanism of orthotic helmets.

A custom-made helmet or headband is another treatment option that is often used. Its effectiveness was first demonstrated in 1979 in four patients (Clarren et al., 1979). Ten patients were referred for evaluation by Clarren et al. (1979); four followed the recommended protocol, wearing the helmets continuously for 2 to 3 months, removing them only for daily bathing. In these cases cranial symmetry improved as the brain grew and the head filled out the helmet (Clarren et al., 1979). The six untreated or incompletely treated patients were found to have the same degree of craniofacial asymmetry 4 to 9 months after the initial evaluation (Clarren et al., 1979). The success of helmet treatment since has been well documented (Bialocerkowski et al., 2005; Bruner et al., 2004; de Ribaupierre et al., 2007; Katzel et al., 2010; Larsen, 2004; Lee et al., 2008; Lima, 2004; Littlefield, 2004; Losee et al., 2007; McGarry et al., 2008; Najarian, 1999; Robinson & Proctor, 2009; Teichgraeber et al., 2004; Xia et al., 2008;). A custom-made helmet is designed so that, when worn, it applies pressure to the prominent aspects of the asymmetric head and has room to grow for the flattened parts (Biggs, 2003; Steinbok et al., 2007). The rationale for using this type of cranial orthotic device is that the prominent areas of the occiput will become flatter, and with growth, the flattened parts will round out, and that this form of treatment may be more helpful than counter-positioning (Steinbok et al., 2007). This finding is significant as the skull undergoes 85% of its postnatal growth within the first year of life (Biggs, 2003; Pollack et al., 1997). The device is sometimes not well tolerated by some infants and may also cause irritation to the child and anxiety to the family (Steinbok et al., 2007). In addition, there is a sizeable cost associated

with the manufacturing of the helmet, ranging from $1000–$3000 US (Steinbok et al., 2007), and monitoring of treatment progress.

Furthermore, correct production of the headband or helmet requires an orthotist with specific expertise. An inner foam liner is incorporated to allow the device to be adjusted weekly to biweekly, thereby achieving control of the correction (Littlefield, 2004). The limited number of qualified orthotists may create difficulties in initial access in obtaining appropriate follow-up to have adjustments made in the device, and to ensure that undue pressure is not being placed on infants' delicate skin (Steinbok et al., 2007). To date, 23 different types of cranial remodelling devices have been approved by the US Food and Drug Administration (Littlefield, 2004). A regulatory body that approves cranial orthotic devices in Canada was not found.

The orthotic treatment protocols for plagiocephaly indicate that cranial orthoses (helmets) are approved for use on infants, aged 3 to 18 months with moderate to severe deformational plagiocephaly. Infants considered for helmet treatment must first have undergone at least 6 to 8 weeks of repositioning therapy before the age of 6 months and have not demonstrated improvement (Larsen, 2004). The best results of helmet therapy have been observed in infants aged 4 to 12 months as a result of greater malleability of the skull and rapid brain growth during that period. The average length of treatment is 3 to 5 months. Regardless of the design, cranial orthoses are safe and effective in reducing cranial asymmetries and show no evidence of regression after treatment. Cranial orthoses are typically worn 23 hours per day for a period of 6 weeks to 6 months, with an average duration of 4.3 months. The average age range for initiating treatment is 4 to 12 months.

The optimum age range is 4 to 8 months. Infants are followed up 1 week after the fit and every 2 weeks thereafter. Goals of a fit include maintaining good suspension with room for growth, appropriate trims, a lack of evident redness after 15 minutes of wear time, and control of rotation. Age and severity have the most significant of effect on treatment outcomes (Larsen, 2004).

The effectiveness of active counter-positioning vs. orthotic helmet treatment.

Xia et al. (2008) conducted a systematic review of seven cohort studies of otherwise healthy infants comparing the effectiveness of moulding helmet therapy with that of head repositioning therapy (Clarren, 1981; Graham et al., 2005; Loveday & de Chalain, 2001; Moss, 1997; Mulliken et al., 1999; Pollak et al., 1997; Vles et al., 2000). Due to the methodological issues associated with allocation of treatment groups, measurement of observed effect, analysis, and loss to follow up, confidence in the treatment effect was only achieved in one study (Graham et al., 2005).

In their study, Graham et al. (2005) compared active counter-positioning with orthotic helmet therapy in 298 consecutive infants diagnosed with plagiocephaly and referred for treatment. Of these infants, 176 were treated with active counter-positioning, 159 were treated with orthotic helmets, and 37 were treated with initial counter-positioning followed by helmet therapy when counter-positioning failed. Cranial diagonal differences (DD) were compared before and after treatment. Graham et al. (2005) compared reductions in diagonal difference (RDD) between repositioning and cranial orthotic helmet therapy. Helmets were routinely used for infants older than 6 months with DD >1 cm. For infants treated with repositioning at a mean age of 4.8 months, the mean RDD was 0.55 cm (from an initial mean DD of 1.05 cm).

For infants treated with cranial orthotics at a mean age of 6.6 months, the mean RDD was 0.71 cm (from an initial mean DD of 1.13 cm). Infants treated with orthotics were older and required a longer length of treatment (4.2 versus 3.5 months). Infants treated with orthosis had a mean final DD closer to the DD in unaffected infants (0.3 ± 0.1 cm), orthotic therapy was more effective than repositioning (61% decrease versus 52% decrease in DD), and early orthosis was significantly more effective than later orthosis (65% decrease versus 51% decrease in DD).

In their study, Rogers, Miller & Mulliken (2008) assessed the effectiveness of a modifiable cranial cap (a modifiable concave resting surface) with that of active counter-positioning and cervical stretching to correct deformational plagiocephaly in infants younger than 4 months. An adjustable concave cranial cup was used in the treatment group (n = 24). The control children were managed by repositioning and cervical stretching exercises (n = 23). Infants were examined at the initial and final visits by a single examiner. Calvarial asymmetry was measured using a large cranial caliper. Active head rotation and degree of rotational asymmetry were estimated to the nearest 10 degrees. This nonrandomized, prospective clinical trial used historical controls. The treatment and control groups consisted of infants younger than 4 months referred for deformational posterior plagiocephaly. There were no statistically significant differences between the control and treatment groups with respect to gestational age at birth, age at initial visit, initial head rotational asymmetry, initial transcranial difference, final head rotational asymmetry, and age at final evaluation. Mean time between initial and final evaluation was 56.3 days for the treatment group and 61.6 days for the control group. The

mean transcranial difference decreased from 11.2 to 3.5 mm in the treatment group, and from 9.0 to 8.0 mm in the control group ($p < .000$). The authors conclude that a cranial cap is significantly more effective than repositioning for correcting early deformational plagiocephaly.

Lipira et al. (2010) provide the most robust evidence to date on the effectiveness of helmet treatment versus active counter-positioning for positional plagiocephaly. Whole-head three-dimensional surface scans of 70 infants with deformational plagiocephaly were captured before and after treatment by using stereophotogrammetric imaging technology. Helmeted ($n = 35$) and non-helmeted/actively repositioned ($n = 35$) infants were matched for severity of initial deformity. Surfaces were spatially registered to a symmetric template, which was deformed to achieve detailed right-to-left point correspondence for every point on the head surface. A ratiometric asymmetry value was calculated for each point relative to its contralateral counterpart. Maximum and mean asymmetry values were calculated. The outcome measure was the change in mean and maximum asymmetry with the two forms of treatment. There was no statistically significant difference in average head growth during treatment. Treatment decision was guided by parental preference. All parents of infants referred for treatment for deformational plagiocephaly were presented both treatment options and invited to participate in the study. The results of this study indicate that the helmeted group had a larger reduction than the repositioned group in both maximum (4.0% versus 2.5%; $p < .02$) and mean asymmetry (0.9% versus 0.5%; $p < .02$). The greatest difference was localized to the occipital region. While the authors conclude that three-dimensional

asymmetry analysis is capable of quantifying the relative efficacy of the two common treatments of deformational plagiocephaly, the age of the infants participating in the study is not presented, which may have a bearing on the findings. The authors conclude that additional studies are necessary in order to: (a) establish the clinical significance of these quantitative differences in outcome, (b) define what constitutes pathologic head asymmetry, and (c) determine whether the results of orthotic treatment last as the child matures.

Early Recognition

Early treatment may be necessary for infants with neck problems and/or strong head positioning preference in order to prevent the development of positional plagiocephaly (Bialocerkowski et al., 2008). In their double-blind randomized control trial, van Vlimmeren et al. (2008) recruited 65 infants to assess if physiotherapy reduced the risk of severe deformational plagiocephaly in infants that exhibit a preferred head position while in the supine position. Infants in the study were 7 weeks of age and exhibited head rotation to either the right or left side for approximately three-quarters of the time of observation. Infants in the study did not have active range of motion of the head over the full range of 180 degrees. Infants with torticollis, dysmorphisms and syndromes were excluded. Thirty-three infants were randomized to the physiotherapy groups and 32 to the control group. The parents of both groups of infants received routine information including a pamphlet describing preventive measures for plagiocephaly and advice at well-child clinics. The control group did not receive any additional intervention.

The physiotherapy group received eight physiotherapy sessions at least one week apart until the infant reached 6 months of age. The physiotherapy sessions included: (a) exercises to reduce positional preference and to stimulate motor development; (b) parental counselling about counter-positioning, handling, tummy time, and nursing; and (c) the causes of positional preference. The sessions ceased when the positional preference disappeared, parents had incorporated the advice about handling and there were no signs of motor developmental delay or asymmetry. The primary outcome was severe deformational plagiocephaly assessed by plagiocephalometry. The secondary outcomes were positional preference, motor development and cervical passive range of motion. All outcomes were measured at 6 and 12 months of age. Findings of the study indicate that physiotherapy reduced the risk for severe deformational plagiocephaly by 46% at 6 months (RR: 0.54; 95% CI: 0.30–0.98) and 57% by 12 months of age (RR: 0.43; 95% CI: 0.22–0.85). The number of infants with positional preference that needed to be treated were 3.85 and 3.13 at ages 6 and 12 months, indicating that for every three infants with positional preference treated with physiotherapy, one case of severe deformational plagiocephaly between ages 7 weeks and 6 or 12 months will be prevented.

If torticollis is diagnosed, neck exercises are performed and taught to the parents of affected infants as part of the management (Losee & Mason, 2005). These exercises entail gently rotating an infant's head so that the chin touches the shoulder, holding for ten seconds, and repeated three times on each side to stretch the SCM muscle (Losee & Mason, 2005). According to Losee and Mason, chin-to-chest stretches should also be included in this management program. Parents are encouraged to perform neck exercises with each diaper

change, a process that can be completed in a few minutes. Although treatment is available for infants with severe positional plagiocephaly, prevention and early recognition are optimal. PHNs are in excellent position to educate parents about infant positioning, screen for plagiocephaly, implement repositioning strategies, and provide referrals for those that may have a more serious skull deformity (Neufeld & Birkett, 1999).

While effective treatment options exist, the question of long term consequences must also be addressed. In the following section the research available about potential developmental concerns associated with plagiocephaly will be discussed.

Potential Developmental Concerns Associated with Plagiocephaly

Positional plagiocephaly was once considered – and still is, by many – a purely cosmetic disorder. As a result, the management approach has traditionally been based on "normalizing" skull shape. However, there is beginning research that suggests that infants with the condition may also experience long term consequences. This section will discuss the current research available on the long term consequences associated with plagiocephaly.

Steinbok et al. (2007) conducted a retrospective review of 65 infants in order to describe developmental outcomes at a minimum of 5 years after initial diagnosis. Steinbok et al. (2007) found that of patients that were diagnosed with plagiocephaly, 33% had received learning assistance and 14% were in a special class in school 5 years after initial diagnosis. There were no differences with respect to age at initial diagnosis, overall cosmetics at initial visit or use of a helmet (Steinbok et al., 2007). Five study participants (8%) had a co-morbid diagnosis consistent with developmental delay at the time of their initial assessments (Steinbok et al., 2007). Upon further investigation, Steinbock et al. (2007) attribute these

developmental delays to confounding variables including prematurity, neurological impairment including microcephaly, atrophic brain development, hydrocephalus, anoxia at birth and genetic syndromes. As this study was a retrospective review, standardized developmental information was not available for the study participants. While the study by Steinbok et al. (2007) was not designed to answer questions concerning the nature of the relationship between developmental delays and plagiocephaly, it does raise questions of whether plagiocephaly is in fact related to developmental delay and if so, which is the precursor. Does a diagnosis of plagiocephaly place infants at higher risk for developmental delays, or are children with pre-existing delays and risk factors at risk for developing plagiocephaly? In either case, it is clear that more research is necessary in order to answer the questions posed above and to determine the exact causes of the developmental delays.

Miller and Clarren (2000) conducted a retrospective medical record review of 254 infants diagnosed with plagiocephaly from 1980–1991 in the U.S. Researchers were able to notify 181 of those families found by the review; 63 agreed to participate in a telephone interview. Miller and Clarren (2000) used the patients' siblings as controls in order to compare the need for special services during the school-age years. The need for any special services was identified by school documentation on an Individual Education Plan (IEP), and included special education assistance, speech therapy, physical therapy or occupational therapy. Study findings revealed that there was a statistically significant difference between the study participants and their siblings with respect to engaging in an IEP ($p = .000$). Controlling for sex, a significant difference was found for males ($p = .000$) but not for females. ($p = .13$). Overall, the percentage of study participants on an

IEP with plagiocephaly was 34.9% and their siblings 6.6%. In contrast, the percentage of children qualifying for special services on an IEP in the state of Washington for 1997 was 11.9%. A limitation of this study was the number of patients included in the telephone interview compared to the number identified in the chart review because of an inability to contact families previously evaluated in the Craniofacial Centre. Ultimately, Miller and Clarren (2000) concluded that males with abnormal head shapes at birth associated with uterine constraint, need to be followed closely for potential developmental delay presenting as subtle problems of cerebral dysfunction during the school-age years.

Kordestani, Patel, Bard, Gurwitch and Panchal (2006) studied 110 infants with plagiocephaly between 1997–2003 in the U.S. in order to identify any demonstrated cognitive and psychomotor developmental delays. Additional information for potential confounders was found for 63 of the study participants. It is the analysis of this data which will be presented here. The infants ranged from 3 to 10 months at the time of initial evaluation. Mental development index and psychomotor development index scores were compared with the Bayley Scales of Infant Development-II standardized sample and were found to be significantly different ($p < .000$). When the mental development index scores of this sample of 63 infants were examined, no infants were found to be in the accelerated group, 57 patients (91%) were in the normal group, four patients (6%) were in the moderately delayed group, and two patients (3%) were in the severely delayed group.

When the psychomotor development index scores were examined, no patients were found to be in the accelerated group, 47 patients (75%) were in the normal group, 12 patients (19 %) were in the moderately delayed, and four patients (6%) were in the severely delayed

group. Upon multivariate analysis of confounding factors, the stepwise selection method for prediction of the mental development index severity chose a four-variable model that included congenital defects, family history of congenital defects, early sickness (requiring intensive care unit admission), and evidence of torticollis. The model prediction of psychomotor development index demonstrated similar results. The final model included five predictors: (a) low birth weight, (b) premature birth status, (c) congenital defects, (d) family history, and (e) sex. This study concludes that children without any of the confounding factors described above do not have an increased incidence of developmental delays despite having a deformational plagiocephaly.

Panchal et al. (2001) studied 42 infants diagnosed with plagiocephaly (mean age of 8.4 months) in order to determine whether infants with positional plagiocephaly displayed cognitive and psychomotor delays when compared with a standardized population sample. Data were collected over a 2 year period. Each infant was assessed using the Bayley Scales of Infant Development-II (BSID-II) for cognitive and psychomotor development before therapeutic intervention (moulding-helmet therapy for plagiocephaly without synostosis). The infants were categorized as accelerated, normal, mildly delayed and significantly delayed according to the Mental Developmental Index and Psychomotor Developmental Index. The distributions of the Mental Developmental Index and the Psychomotor Developmental Index were then compared with a standardized Bayley's age-matched population, using Fisher's exact chi-square test. A standardized distribution of the Mental Developmental Index of infants in the general population demonstrates that 16.5% of infants would be in the accelerated category,

68.7% would be classified as normal, 12.5% would be identified as mildly delayed and 2.3% would be in the severely delayed category (Panchal et al., 2001). In comparison, Panchal et al. (2001) found that none of their study participants were classified as accelerated, 82.6 % were classified as normal, 8.7% were in the mildly delayed category and 8.7% were classified as severely delayed. In the same way, a standardized distribution of the Psychomotor Developmental Index of infants in the general population demonstrates that 14.8% of infants in the general population would be in the accelerated category, 72.6% would be classified as normal, 11.1% would be identified as mildly delayed and 1.6% would be in the severely delayed category (Panchal et al., 2001). In their study, Panchal et al. (2001) found 67.4% to be classified as normal, 19.6% to be in the mildly delayed category and 13.0% to be classified as severely delayed. Within the group with plagiocephaly, both the Psychomotor Developmental Index and Mental Developmental Index scores were significantly different from the normal curve distribution ($p < .001$). This study indicates that before any intervention, subjects with single-suture syndromic craniosynostosis and deformational plagiocephaly demonstrated delays in cognitive and psychomotor development. The authors also indicated that the BSID-II is predictive of future outcome with respect to development, but the long term consequences of these findings need to be studied. Continued post-intervention assessments are needed to determine whether these developmental delays can be ameliorated with intervention.

Kennedy, Majnemer, Farmer, Barr & Platt (2009) conducted a study to compare motor development between infants with positional plagiocephaly and their matched

peers without positional plagiocephaly. Differences in infant positioning practices when asleep and awake were also examined between the two groups. Seventy seven infants with positional plagiocephaly, ranging from 3-8 months of age were matched to infants without positional plagiocephaly with respect to age, sex and race. Motor performance was evaluated using the Alberta Infant Motor Scale (AIMS) and the Peabody Developmental Motor Scales (PDMS). For 3 days, parents completed a diary that recorded infant positioning. Mean AIMS percentile score for infants with positional plagiocephaly was 31.1 ±21.6 and 42.7 ± 20.2 in infants without positional plagiocephaly ($p = .06$). Better performance on the AIMS was positively correlated with the amount of time in prone position when awake for both groups of children (infants with positional plagiocephaly $r = .52$; infants without positional plagiocephaly $r = .44$; $p < .05$). This study demonstrates that perhaps it is not the positional plagiocephaly that may be associated with motor developmental delays, but rather the misunderstanding of the importance of prone positioning while infants are awake.

Speltz et al. (2010) conducted a case-control study in order to assess assessed the neurodevelopment of infants with and without deformational plagiocephaly, at an average age of 6 months. The Bayley Scales of Infant Development-III (BSID-III) were administered to 235 case participants and 237 demographically similar, control participants. Exclusions included: (a) prematurity (<35 weeks of gestation); (b) a known neurodevelopmental condition (e.g., Down syndrome), brain injury, or significant vision or hearing impairment; (c) major malformations or ≥ 3 “typical” extracranial anomalies (e.g., extra digits, skin tags, or ptosis); (d) hemifacial microsomia; (e) a non-English

speaking mother; (f) a history of adoption or out-of-home placement; and (g) current family plans to move out of state before completion of the research project. Infants were eligible for participation in the control sample if they had not been diagnosed as having deformational plagiocephaly or any other craniofacial anomaly and they did not meet any of the exclusionary criteria for case participants described above. Three-dimensional head photographs were randomized and rated for severity of deformation by two craniofacial clinicians that were blinded to case status. With age, sex, and socioeconomic status controlled, case participants performed worse than control participants on all BSID-III scales and subscales. Case participants' average scores on the motor composite scale were approximately 10 points lower than control participants' average scores ($p < .001$). Similarly approximately 5 points of difference on average were demonstrated for the cognitive and language composite scales ($p < .001$ for both scales). With further analyses, Speltz et al. (2010) found that case participants' gross-motor deficits were greater than their fine-motor deficits. Furthermore, among case participants, no association was found between BSID-III performance and the presence of torticollis or infant age at diagnosis. Speltz et al. (2010) indicate that the presence of deformational plagiocephaly seems to be associated with early neurodevelopmental disadvantage, which is most evident in motor functions. After follow-up evaluations of this cohort at 18 and 36 months, Speltz et al. (2010) intend to assess the stability of this result. The authors conclude that these data do not necessarily imply that deformational plagiocephaly causes neurodevelopmental delay; rather the results indicate only that deformational plagiocephaly is a marker of elevated risk for delays.

Research by Fowler et al. (2008) evaluated the neurologic profiles of infants with deformational plagiocephaly. Forty-nine infants with deformational plagiocephaly between the ages of 4 and 13 months (mean age, 8.1 months) were recruited over a 2 month period and evaluated, along with 50 age matched controls (mean age, 8.1 months). Parents were asked to complete the age-appropriate Ages and Stages Questionnaires. The Ages and Stages Questionnaires consist of a series of age-appropriate surveys about psychomotor development of children aged 4 to 48 months. A modified version of the Hammersmith infant neurologic assessment was performed also on each infant. A caregiver completed a questionnaire regarding the infant's prematurity, development, and health to date. Data collected were analyzed using the *t* test. The findings of this study reveal that there is a statistically significant difference in overall neurologic assessment scores of infants with deformational plagiocephaly in comparison to their healthy peers ($p=.002$). This difference was predominately observed in muscle tone, whereby infants with deformational plagiocephaly have significantly more abnormal muscle tone than non-plagiocephalic infants ($p = .003$). This abnormality is not one of decreased tone but one of variable tone, reflecting abnormally high and low tone. Infants with deformational plagiocephaly are more likely to have altered tone but not exclusively decreased tone.

Siatkowski et al. (2005) conducted a study to determine if visual field abnormalities occur in infants with deformational plagiocephaly and to assess whether there is a relationship between the severity and laterality of the visual field abnormalities with the severity and laterality of skull asymmetry. They conducted a retrospective chart review on 40 consecutive infants with deformational plagiocephaly. Each was tested with

standardized binocular arc perimetry in the horizontal plane. Sixteen patients also had 3-dimensional computed tomography of the skull. Hemifield asymmetry of ≥ 20 degrees and/or a decrease in hemifield values by ≥ 20 degrees from established normal patients was considered abnormal. Visual field data from study patients was plotted against previously published normative data. Graphs comparing visual field defects and laterality to cranial asymmetry were then generated. The study results indicate that 35% of infants with deformational posterior plagiocephaly had constriction of one or both hemifields by at least 20 degrees from established normative data. Hemifield asymmetry of 20 degrees or more was found in 17.5% of infants tested. There was a significant difference in the worse hemifield values measured in each patient and the standard visual fields obtained from normative data ($p = .036$). Using analysis of covariance, no statistically significant relationship was found between the laterality of the visual fields to the laterality of the defects ($p = .209$). All data from the study population showed a seemingly delayed progression of visual field compared with the standard curve (i.e., the slopes of all cohort data curves were shallower than the slope of the standard data group). The authors conclude that although this difference was not statistically significant ($p = .147$), it may suggest a trend of delay in visual field maturation in patients with posterior plagiocephaly.

It must be acknowledged that although research is beginning in this area, none of the results to date have proven plagiocephaly to be the causative factor of developmental delays. In addition, the studies presented above use various methods to assess for developmental delay, use differing samples, and the sample sizes of all are relatively small for any

comparisons to be made. It is clear that further studies are required to clarify the relationship, if any, between plagiocephaly and developmental delays.

Based on the information that is presented in this chapter, it is clear that there are gaps in the literature pertaining to the incidence of positional plagiocephaly, the risk factors for the development of positional plagiocephaly and the long term implications of positional plagiocephaly. This research will begin to address some of these gaps. The purposes of the present research are:

- To explore the incidence of positional plagiocephaly in Calgary, Alberta, Canada
- To identify points of prevention for positional plagiocephaly in Calgary, Alberta, Canada

The primary research objective is to contribute to the body of knowledge on positional plagiocephaly incidence and risk factors. The secondary research objectives are:

- To identify and field test an appropriate plagiocephaly assessment tool for use in well-child clinics
- To explore how infants identified with positional plagiocephaly are followed at various levels of the health care system
- To explore actions of various health care professionals working with infants identified with positional plagiocephaly.

In chapter two, the research questions will be presented along with the study design, inclusion criteria and sample size calculation.

CHAPTER TWO: METHODS

The study methods are presented in this chapter. The chapter begins with a discussion of the research objectives and research questions followed by the study design, inclusion criteria and sample. The recruitment process is outlined followed by a discussion of how data are collected to answer each of the five research questions. The rationale for conducting the study at the 2-month clinic visit is explored followed by a discussion of how data were analysed to answer the research questions. The significance of the study and ethical considerations are provided at the end of the chapter.

Research Objectives and Research Questions

Based on the literature review provided in chapter 1, several gaps exist in the area of positional plagiocephaly including: (a) the incidence of positional plagiocephaly, (b) risk factors for its development at various ages in infancy, and (c) the long term developmental implications of infants identified as having positional plagiocephaly. The present study will begin to fill some of these gaps. The primary research objective of the present study is: To contribute to the knowledge base on positional plagiocephaly incidence and risk factors. The primary research questions are:

1. What is the incidence of positional plagiocephaly in infants 7-12 weeks of age in Calgary, Alberta, Canada?

2. What are the potential risk factors for positional plagiocephaly in infants 7-12 weeks of age in Calgary, Alberta, Canada?

Because data presented above are being collected, this provided an opportunity to collect additional data pertaining to which assessment method is appropriate for use in well-child clinics. In addition, this study presented an opportune time to collect data relevant to interventions and follow-up actions of various clinicians working in the area of plagiocephaly across the health care continuum. As a result, the secondary research objectives are:

1. To identify and field test an appropriate plagiocephaly assessment tool for use in well-child clinics
2. To explore how infants identified with positional plagiocephaly are followed at various levels of the health care system
3. To explore actions of various health care professionals working with infants identified with positional plagiocephaly

The secondary research questions are:

- Is Argenta's (2004) Plagiocephaly Assessment Tool appropriate for use in well-child clinics?
- What intervention and follow-up actions do PHNs take if positional plagiocephaly is identified in healthy infants at the 2-month well-child clinic?
- What intervention and follow-up actions do clinicians (physiotherapists and occupational therapists) take when healthy infants with positional plagiocephaly

are referred to the infant repositioning class (IRC) after the 2-month well-child clinic visit in Calgary, Alberta, Canada?

Study Design

A prospective cohort design was used in this study. This design is consistent with that presented in the literature; Hutchison et al. (2004) used a prospective cohort study in New Zealand in order to assess the prevalence and natural history of plagiocephaly in otherwise healthy infants in the first 2 years of life and to identify potential factors that may contribute to the development of the condition. Similarly, van Vlimmeren et al. (2007) used a prospective cohort study in the Netherlands in order to identify risk factors for plagiocephaly within 48 hours of birth and at 7 weeks of age. In their systematic review, Bialocerkowski et al. (2008) identified that using cohort study designs are appropriate when studying the epidemiology of plagiocephaly since these designs are able to determine causation, can provide evidence about prevalence and natural history of the condition, and have fewer sources of bias when compared to many other quantitative research designs.

The sources of outcome information for cohort studies include surveys and physical examinations (Aschengrau & Seage, 2008). The main strengths of prospective cohort studies are that they can directly measure disease incidence or risk, and they can provide good information on exposures (Aschengrau & Seage, 2008; Koepsell & Weiss, 2003). On the other hand, prospective cohort studies tend to be expensive and time consuming and are inefficient when evaluating conditions that have long induction and latent periods (Aschengrau & Seage, 2008). Since the onset of plagiocephaly does not

include a long induction or latent period, and because the objectives of the study include identifying incidence and predictive factors, using a prospective cohort study would be most appropriate.

Incidence is the number of new health-related events in a defined population within a specified period of time (Porta, 2008). The incidence rate is the most appropriate measure of disease frequency as the study population is open or dynamic with study participants having a changeable characteristic; in this study, the type of care the infant is receiving in terms of positioning (Aschengrau & Seage, 2008).

Inclusion Criteria

Study participants included full term infants (born at ≥ 37 weeks of gestation) ranging from 7-12 weeks of age that presented at 2-month well-child clinics at four sites in the city of Calgary, Alberta. Neufeld and Birkett (1999) assert that assessing for positional plagiocephaly is essential at the 2-month well-child clinic visit since teaching about infant positioning at that time is crucial for the prevention of plagiocephaly. In addition, parents/guardians of these infants were also available to complete a short questionnaire on risk factors in order to be included in the study. Infants presenting with brachycephaly only were excluded.

Sample

A cohort study involving incident cases is concerned with first occurrence of plagiocephaly; therefore the candidate population is composed of all individuals in the study population that are eligible to be identified as cases (Kleinbaum, Kupper, & Morgenstern, 1982). There were 15,520 healthy births in 2007 in Calgary, including High

River, Canmore, Strathmore and Banff (Alberta Health Services, 2007). However, access was given in four community health centres (CHCs) from July – September 2010 to complete the data collection. As shown in Table 1, 1712 infants were eligible to participate in the study (C. MacLeod, personal communication, February 4, 2011).

Table 1: Number of Infants that met the Inclusion Criteria at the Four CHCs

CHC	Month	Count
Clinic 1	July	73
Clinic 1	August	83
Clinic 1	September	79
Clinic 2	July	155
Clinic 2	August	143
Clinic 2	September	143
Clinic 3	July	170
Clinic 3	August	219
Clinic 3	September	205
Clinic 4	July	141
Clinic 4	August	148
Clinic 4	September	153
Total		1712

A sample of 471 was consecutively recruited at 2-month well-child clinics in four CHCs in Calgary. Because the participants were consecutively recruited they are representative of the larger population and hence the results may be generalizable to the larger population (van Vlimmeren et al., 2008). The four CHCs were selected based on their location, each CHC representing one quadrant of the city. Selecting one CHC from each quadrant of the city ensures also that the results are representative of the larger population in the city of Calgary.

The following formula was used to calculate the sample size:

$$n = \frac{z^2 [p\ (1-p)]}{e^2}$$

The convention is to use p = ½ and the margin of error (e) = 0.05 = 0.025. The confidence interval in this study was 95% (Daniel, 1999; Food Safety Research and Response Network, 2007).

$$n = \frac{1.96^2 [0.5\ (0.5)]}{(0.05)^2} \qquad n = 384$$

It was assumed that at the most, there would be a 20% attrition rate in the study, therefore, 77 (20% of 384) were added to the sample size for a total recruitment of 461.

Clinical Observation Tool

Three clinical observation tools were presented in chapter one (Argenta, 2004; Littlefield, Saba et al., 2004; Losee & Mason, 2005). These three clinical observation methods will be compared according to four of the six criteria, as indicated in chapter one: (a) cost-effectiveness, (b) validity, (c) reliability, and (d) suitability/feasibility. These are presented, in turn, below. The discussion below indicates the rationale for selecting Argenta's (2004) plagiocephaly assessment tool.

Cost effectiveness.

Two of the three assessment methods (Argenta, 2004; Losee & Mason, 2005) do not require specific equipment, costly procedures or tests, or software programs; they are both cost effective forms of assessing for plagiocephaly. The main associated cost with the assessment methods developed by Argenta (2004) and Cranial Technologies, Inc.,

(Losee & Mason, 2005) would be photocopying of the one page tools if the PHNs wish to include this as part of the patients' hard copy chart. For each specific assessment method incorporating associated check boxes for each assessment item could be easily integrated into the current electronic charting system, with a minimal up-front set-up cost. The plagiocephaly assessment method proposed by Littlefield, Saba et al. (2004) requires the use of a spreading caliper or measurement tape in order to determine the degree of ear misalignment in inches, in order to categorize the plagiocephaly as identified.

Validity.

With respect to content validity of the tools, Littlefield, Saba et al. (2004) used set criteria to guide their assessment. Infants were identified as having no plagiocephaly, mild, moderate or severe plagiocephaly. Although the criteria provided were not in the form of a formal tool with associated pictures for ease of identification, four criteria are identified for assessment and used to categorize the plagiocephaly as mild, moderate or severe. The four criteria used to evaluate infants were guided by definitions of what was meant by mild (posterior asymmetry); moderate (posterior asymmetry, ear malposition with a discrepancy of half an inch or more, and minimal frontal asymmetry and facial asymmetry); and severe (significant posterior asymmetry, ear malposition of one inch or more, frontal asymmetry on the affected side, and facial asymmetry). The tool produced in 2002 by Cranial Technologies, Inc., and presented by Losee and Mason (2005) indicates five categories of assessment: (a) posterior flattening, (b) ear misalignment, (c) forehead asymmetry, (d) neck involvement, and (e) facial asymmetry. In addition, four

levels of severity (0-4) are proposed within each category. Based on this, an overall score (out of 20) is given to the infant.

Both tools above present 4 items of assessment that are specific to plagiocephaly (posterior flattening, ear misalignment, forehead asymmetry, and facial asymmetry). In contrast, Argenta's (2004) plagiocephaly assessment tool provides six items of assessment (posterior asymmetry, ear malposition, frontal asymmetry, facial asymmetry, temporal bossing and posterior vertical cranial growth) and instead of identifying the plagiocephaly as mild, moderate or severe, classifies the plagiocephaly into five different types that increase in severity. Porta (2008) defines content validity as the extent to which the measurement incorporates the domain of the condition under study. Since there are more items of assessment that are specific to plagiocephaly, Argenta's (2004) tool can be said to have a greater degree of content validity than the other two clinical observation methods.

Face validity is defined as the validity of a test at face value. Specifically, a test can be said to have face validity if it "looks like" it is going to measure what it is supposed to measure; it is sometimes referred to as a "click of recognition" (Creswell, 2007). Both the criteria developed by Littlefield, Saba et al. (2004) and Argenta's (2004) plagiocephaly assessment tool can be said to have a high degree of face validity as every assessment item pertains specifically to plagiocephaly. The tool developed by Cranial Technologies, Inc. includes one item that relates to a torticollis assessment. Although torticollis is often co-diagnosed with plagiocephaly, torticollis assessments involve more than clinical observation of a head tilt. Therefore, the inclusion of an assessment item that

does not specifically relate to plagiocephaly – a malformation of the skull producing the appearance of a lopsided head (Thomas, 1997) – decreases the face validity of this plagiocephaly assessment tool.

Reliability.

No reliability studies for the assessment criteria developed by Littlefield, Saba et al. (2004) or the tool developed by Cranial Technologies, Inc. (Losee & Mason, 2005) were found in the literature. As previously mentioned Spermon et al. (2008) conducted a study in the Netherlands in order to determine the reliability of Argenta's (2004) five-point rating scale. They used three paediatricians, three physiotherapists and three manual therapists that had no experience in the assessment of head deformity. The nine raters examined 20 patients, and k scores were used to measure agreement for classification type as well as specific clinical features. Spermon et al. (2008) found Argenta's five-point rating scale to be moderately reliable in terms of plagiocephaly classification type among all three sets of health professionals ($k = .54$). The k coefficient can range from -1.0 to +1.0, with .4 to .6 indicating moderate agreement. Spermon et al. (2008) found the interrater agreement for the first four clinical features of deformational plagiocephaly (occipital flattening, ear malposition, frontal bossing and facial asymmetry) to range from 0.6 – 0.85, indicating substantial to almost perfect agreement. This finding is important since PHNs working in well child clinics may come across Types 1 to 4 more frequently than they would the most severe form, Type 5. It is important to note that the clinicians participating in Spearmon's study had no prior experience in conducting head assessments. As a result, it would be reasonable to assume that the interrater reliability

could be higher for health care professionals, such as PHNs, that have experience with conducting head assessments and that are engaged in this frequently as part of their practice.

Suitability/feasibility.

The suitability/feasibility of the tools will be presented here according to: (a) ease of use, (b) time required to complete the assessment, (c) level of expertise required for an accurate assessment, (d) level of intrusiveness, and (e) utility as a teaching tool.

Ease of use.

When compared to the other two tools that provide pictures for ease of use, the criteria put forward by Littlefield, Saba et al. (2004) may be more difficult to use. Simply writing out the criteria may require the PHN to visualize what the criteria mean in terms of changes to the infant's head before determining if the infant being assessed has the condition. In addition, the criteria developed by Littlefield, Saba et al. (2004) requires the use of a measuring tape or spreading caliper in order to quantify the degree of ear misalignment in inches. No assessment guidelines accompany the tool developed by Cranial Technologies, Inc. (Losee & Mason, 2005).

Argenta (2004) provides guidelines for assessing plagiocephaly based on four positions of examination. The first examination position is looking directly anteriorly at the infant. This allows the PHN to determine if there are asymmetries of the forehead and of the face. The second position of examination is to view the infant's head directly from above. The index fingers of the examiner are placed in each external auditory canal. This allows evaluation of forehead asymmetry, posterior cranial asymmetry, and malposition

of the ears. Viewing from directly above also allows for identification of abnormal bulging of the temporal fossa. The third clinical examination position is posterior to the infant's posterior skull. This position allows for the confirmation of ear position and posterior asymmetry. It allows also for the evaluation of widening of the posterior skull. The fourth position of examination is a direct lateral view. This position allows the evaluator to determine any degree of abnormal vertical growth of the skull, which can occur in severe plagiocephaly when the restrained brain attempts to decompress. When using Argenta's (2004) plagiocephaly assessment tool, abnormalities are clinically visible or classified according to whether they are present or absent. No anthropometric measurements are taken and minimal abnormalities that require precise measurements are not considered clinically present. Because Argenta (2004) provides assessment guidelines along with the assessment tool, this increases ease of use of the tool.

Time required to complete the assessment.

Although both Argenta's (2004) tool and the tool developed by Cranial Technologies, Inc. (Losee and Mason, 2005) have pictures associated with each item of assessment, the tool developed by Cranial Technologies appears more complex, requiring the PHN to categorize each assessment item into four potential levels of severity. As a result, there are 4 pictures that the PHN is required to look at for each assessment item (20 pictures in total) before making a conclusion. This may take more time than the items of assessment required to be completed for Argenta's (2004) assessment tool, which has 6 assessment items and 7 associated pictures. Since some manual measurement is required for the plagiocephaly assessment criteria proposed by Littlefield, Saba, et al.

(2004), this approach would also require more time, depending on how easy or difficult it may be to obtain such measurements on an infant. Therefore, Argenta's (2004) plagiocephaly assessment tool would require the least amount of time to complete.

Level of expertise required for an accurate assessment.

Since the assessment criteria developed by Littlefield, Saba et al. (2004) require expertise to conduct some degree of anthropometric measurement in order to quantify the degree of ear misalignment, those using this tool would require training in order to identify various cranial landmarks and to take the measurements accurately. Therefore, this would require a higher level of expertise than the other two methods that use simple clinical observation. With respect to the plagiocephaly assessment tool developed by Cranial Technologies, Inc., it appears that a higher level of expertise would be required in order to distinguish between the different levels of severity according to each item of assessment. Furthermore, because this tool identifies the head tilt as part of the assessment, a complete torticollis assessment would be required in order to put this information into context. Currently, PHNs do not conduct torticollis assessments and would need to be trained in order to use this tool, thereby requiring a higher level of expertise. In contrast, Spermon et al. (2008) used clinicians that had no prior experience in conducting head assessments to prove the reliability of Argenta's (2004) plagiocephaly assessment tool. As indicated above, the results of the study by Spermon et al. (2008) indicated that the plagiocephaly assessment tool developed by Argenta (2004) is reliable, thus demonstrating that no specific training is required to use the tool.

Level of intrusiveness.

The plagiocephaly assessment tools developed by Argenta (2004) and Cranial Technologies, Inc. (Losee & Mason, 2005) are not intrusive as they both use clinical observation as the method of assessment, whereas, the plagiocephaly assessment criteria proposed by Littlefield, Saba et al. (2004) may be viewed as minimally-moderately intrusive as some anthropometric measurements are required.

Utility as a teaching tool.

The criteria proposed by Littlefield, Saba, et al. (2004), may not be the best teaching tool as there is no tool *per se* and there are no visual aids to accompany the assessment criteria. The visual aids would need to be developed. The plagiocephaly assessment tool developed by Cranial Technologies, Inc. also may not be useful as a teaching tool as the plagiocephaly assessment items are not as complete as those developed by Argenta (2004). In addition, the numerous pictures may cause confusion and anxiety among parents. With respect to the neck involvement assessment item, parents may not fully understand the extent of the torticollis unless additional teaching is completed. Argenta's (2004) plagiocephaly assessment tool is easier to understand, providing six items of assessment accompanied by seven pictures. Based on the prevention or repositioning teaching received, parents could be educated to monitor their infant's head shape and report either the progress made or worsening of the condition.

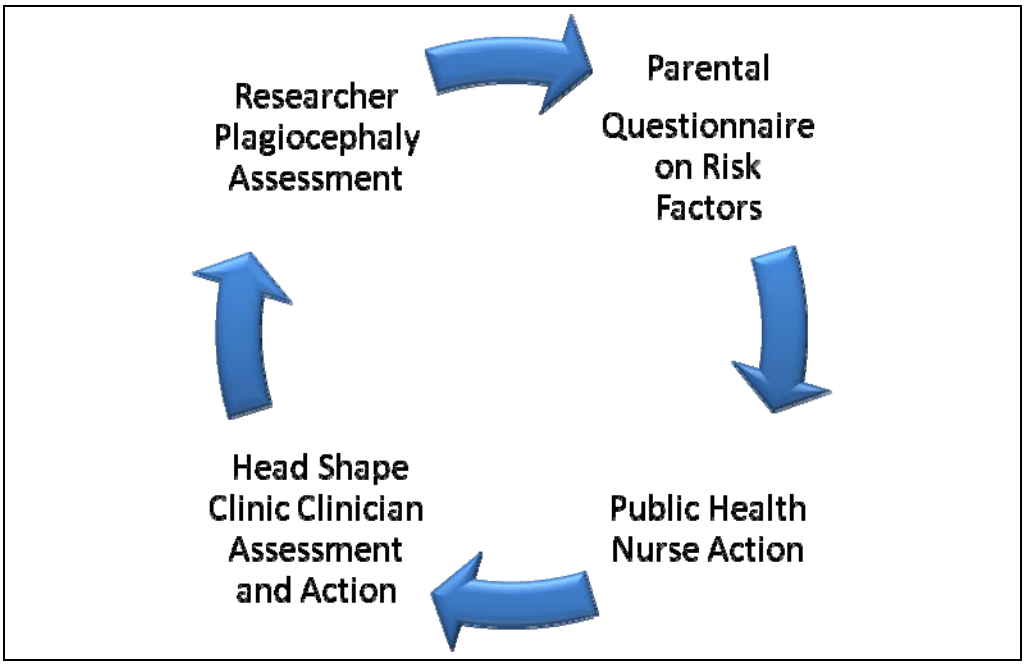
Based on the evaluation of the three assessment tools, according to the criteria presented in chapter one, Argenta's (2004) plagiocephaly assessment tool was considered to be most appropriate for use in the present study. After a review of the literature, of all

clinical observation tools, Argenta's (2004) plagiocephaly assessment tool was the only one found to be reliable. The discussion above indicates that it is also the most valid. The assessment guidelines provided allows the tool to be used easily with minimal time required for the assessment to be completed. The assessment, as proposed by Argenta (2004) is minimally intrusive using only clinical observation as the assessment method. Lastly, Argenta's (2004) plagiocephaly assessment tool shows the most promise for use as a teaching tool. Based on this information, Argenta's (2004) plagiocephaly assessment tool was selected for use in the present study.

Recruitment

In Figure 1 four points of data collection are identified. Posters (Appendix A) were displayed at the four clinics identified above in order to assist with the recruitment process. One of the Research Nurses approached parent(s)/guardian(s) of each term infant before or after their 2-month well-child appointment. Details of the study were explained and parent(s)/guardian(s) were offered an opportunity to ask questions. Parent(s)/guardian(s) were then invited to participate in the study and written informed consent was obtained (Appendix B).

Figure 1. Data Collection Process Diagram



Data Collection

The data collection process included four components with multiple professionals collecting the data (Figure 1). One of the Research Nurses has Masters and PhD level training in public health while the other Research Nurse has a background in neurology and was completing her Nurse Practitioner program at the time of data collection. Data were collected to answer the five research questions. This section will outline which data were collected to answer each question.

What is the incidence of positional plagiocephaly in infants 7-12 weeks of age in Calgary, Alberta, Canada?

After receiving informed consent, one of the Research Nurses conducted the plagiocephaly assessment using Argenta's (2004) five point scale (Appendix C). Assessments were completed as the parent(s)/guardian(s) held the infant. In addition to

the type of plagiocephaly recorded on the data collection tool, the severity of plagiocephaly observed in terms of mild, moderate or severe was recorded also. Additionally, the side of the infant's head on which the plagiocephaly was observed and whether the infants identified with plagiocephaly also demonstrated accompanying features of brachycephaly were recorded.

What are the potential risk factors for positional plagiocephaly in infants 7-12 weeks of age in Calgary, Alberta, Canada?

Parent(s)/guardian(s) were asked to complete the questionnaire on risk factors. Infant risk factors included: sex, birth order, infant age, multiple gestation, feeding position, sleep position, head positional preference, and tummy time. Maternal risk factors included: age, education, delivery type, and number of years lived in Canada. If one of the Research Nurses noted that the mother had a language barrier, this was also captured as a risk factor and was noted on the questionnaire (Appendix D).

Is Argenta's (2004) Plagiocephaly Assessment Tool appropriate for use in well-child clinics?

With respect to Argenta's (2004) plagiocephaly assessment tool, data were collected pertaining to the following criteria: (a) the suitability/feasibility in terms of ease of use and time required, (b) the acceptability of the tool to the target population, (c) measures taken to ensure reliability, and (d) the yield of the tool.

What intervention and follow-up actions do PHNs take if positional plagiocephaly is identified in healthy infants at the 2-month well-child clinic?

PHNs assessed infants for positional plagiocephaly according to their usual

protocol using subjective assessments during the 2-month well-child clinic visit, which occurred, for the most part, after the Research Nurses' assessment. According to this protocol, PHNs assessed for plagiocephaly by standing above and behind the infant and observed for asymmetry in the infant's head or facial features, or any asymmetrical range of motion when moving the head to either side (Alberta Health Services, 2009). As negotiated with Alberta Health Services, the PHNs were blinded to the plagiocephaly assessment information acquired from the Research Nurses, by folding and stapling the data collection tool closed. The PHNs recorded their portion of the data on the same data collection sheet. As a result, the only remaining information that the PHNs could see was the information they recorded pertaining any intervention that took place relating to positional plagiocephaly if identified (Appendix C). The options used by the PHNs in terms of plagiocephaly intervention included: (a) repositioning teaching, (b) referral to an IRC, (c) referral to the family doctor, and (d) any follow-up activities advised/planned (Alberta Health Services, 2009). If a referral was made either to the family physician or to the IRC, a space was provided for the PHNs to provide their reason for referral. A space was also provided on the data collection tool for the PHNs to record any additional comments they may have had. Furthermore, the parent(s)/guardian(s) of infants that were referred to the infant repositioning class were given a copy of the completed data collection tool to present to the clinicians at the IRC in order to be identified as study participants. Whether or not a copy was given to the parent(s)/guardian(s) was also recorded on the data collection tool.

What intervention and follow-up actions do clinicians (physiotherapists and occupational therapists) take when healthy infants with positional plagiocephaly are referred to the infant repositioning class (IRC) after the 2-month well-child clinic visit in Calgary, Alberta, Canada?

Data collection tools were provided to the clinicians (two physiotherapists and one occupational therapist) at the IRC. Data were collected by these clinicians at the IRC once the infants that were referred there arrived (Appendix E). Data collection included: (a) clinician assessments of the infant according to Argenta's (2004) five-point positional plagiocephaly assessment scale; (b) the severity of plagiocephaly observed in terms of mild, moderate and severe; (c) the presence or absence of torticollis; (d) whether the parent/guardian and infant attended the IRC; (e) the appropriateness of the PHN referrals made; and (f) the type of treatment offered (e.g., cranial orthosis (helmet), physiotherapy or repositioning teaching). A space was also provided for the clinicians to offer additional comments.

The following section will discuss the rationale for collecting data at the 2-month well-child clinic.

Rationale for Collecting Data at the 2-month Well-Child Clinic Visit vs. 4-month Well-Child Clinic Visit

This section will examine the rationale for conducting this study at the 2-month well-child versus at the 4-month well-child clinic visit respectively. One of the data collection points will be recommended and a rebuttal to the potential arguments posed by the proponents of the other will be presented.

Rationale for collecting data at the 2-month well-child visit.

There are many reasons why this study should be conducted at the 2-month well-child visit. This section will discuss five reasons for collecting data at the 2-month mark. The first reason is that asymmetrical infant heads identified at birth may revert back to normal within the first few weeks of life. This was demonstrated by van Vlimmeren et al. (2007), who found that of the 23 infants that had plagiocephaly at birth, 14 had demonstrated reversion to normal by 7 weeks of age. In addition, 75 of 380 infants in the study were found not to have plagiocephaly at birth but presented with the condition at 7 weeks of age (van Vlimmeren et al., 2007). As a result of this evidence, in their systematic review, Bialocerkowski et al. (2008) assert that the term positional plagiocephaly should be used to describe infants older than 6 weeks of age with altered skull shape. Therefore, in order to calculate incidence of positional plagiocephaly, the number of new health related events in a defined population within a specified period of time (Porta, 2008), it would make sense to collect data at the 2-month mark, rather than at the 4-month mark when it would be unclear if it was incidence data or prevalence data that was being collected.

Secondly, it is more difficult to reposition and correct plagiocephaly in infants' when sleeping preference is more established at the four month mark. Compliance with the "back to sleep" campaign has resulted in infants spending most of their time supine. Accordingly, the infant skull generally comes to rest on one side of the occiput, and with this "position of comfort", a persistent sleep position develops (Losee & Mason, 2005). As a result, plagiocephaly can develop very quickly, typically within the first 6-8 weeks

of life (Bialocerkowski et al, 2008; van Vlimmeren, Helders, et al., 2006). Clinically, it is more difficult to reposition infants' "position of comfort" the more established the sleeping preference is (L. Walker, personal communication, August 27, 2009). Therefore, if a sleeping preference is identified at the 2-month clinic, early intervention in terms of repositioning can be implemented thereby preventing the development of plagiocephaly. In infants that have already developed plagiocephaly by 2-months, early identification would enable early intervention in terms of active counter-positioning thereby correcting the plagiocephaly.

Thirdly, since one potential outcome of the study is increased awareness of PHNs about early intervention, it is hoped that beginning the study at the 2-month mark would increase parental education on: repositioning if a sleeping preference is identified; and the need for active counter-positioning to correct an identifiable flat spot. It is important to begin this process early when these prevention and correction options exist. If assessment and intervention take place at the 4-month mark, the plagiocephaly may have progressed to the point where there may be permanent facial deformation and when intervention options are limited to invasive cranial orthoses (helmet) treatment.

Fourthly, as mentioned above, torticollis is a significant co-morbidity with plagiocephaly (Pivar & Scheuerle, 2006; Rogers et al., 2009; van Vlimmeren, Helders et al., 2006). If symptoms of torticollis are recognized at the earliest opportunity, namely the 2-month well-child visit, then physiotherapy treatment can be offered early, thereby avoiding the progressive development of plagiocephaly and accompanying cranio-facial deformation. In essence, the earlier torticollis is identified and treated correctly, the better

the likelihood for repositioning to be effective. Identifying torticollis in newborns prior to the presentation of plagiocephaly and craniofacial deformation provides an advantage when treating infants afflicted with or at risk for developing positional plagiocephaly (Golden et al., 1999).

Finally, conducting standardized assessments at the 2-month mark will lead to clear identification of plagiocephaly early, resulting in more appropriate interventions offered. Currently a variety of assessment methods for plagiocephaly have been presented, although there is no agreement among clinicians as to which method is the most accurate. This study will field test Argenta's (2004) plagiocephaly assessment scale (Appendix C). Use of this standardized scale at the 2-month mark would significantly increase the identification of infants whose parents require prevention messages and infants whose parents require active-counter positioning teaching. The use of this standardized tool at the 2-month mark would also increase the level of appropriate referrals to the IRC. Thus, the use of a standardized plagiocephaly screening tool at the 2-month mark would not only increase the effectiveness of services provided, but it would ensure that the right intervention is being offered to the right infant depending on the results of the plagiocephaly assessment.

Rationale for conducting the study at the 4-month well-child visit.

Conversely, there are also reasons why the study should take place at the 4-month well-child visit.

The first reason why it may be better to collect the data at the 4-month clinic visit is the heavy workload of PHNs at the 2-month well-child clinic. PHNs spend close to an hour

during the first well-child clinic visit explaining vaccination information, conducting formal postpartum depression screening, discussing infant health assessments, informing parents/guardians about the services offered at the clinics, and referring families to services offered at other agencies (B. George, personal communication, August 21, 2009). Because of the crucial developmental stage of infants at 2-months, parents are also given the opportunity during the 2-month clinic to ask questions that they may have about vaccination, feeding and nutrition, infant crying and infant sleep. At the 4-month clinic visit, there are typically fewer concerns to be addressed, fewer formal assessments to complete, and families are typically more familiar with the services provided at the CHC (B. George, personal communication, August 21, 2009). Furthermore, at different points in the year, the 2-month clinic time may be reduced from 60 to 40 minutes to accommodate additional services that PHNs provide to the public, such as influenza clinics during the fall season (M. Nimmock, personal communication, September 17, 2009). Hence it may be increasingly difficult to accommodate data collection at the 2-month clinic visit if it moves into the autumn.

Secondly, regardless of when plagiocephaly is identified (2-months or 4-months) the current treatment guidelines indicate that helmetting is generally recommended between 3-18 months of age with the optimum range of treatment between 4- 8 months (Larsen, 2004). Therefore if plagiocephaly is identified at 4-months, there still may be ample time to correct it.

Reasons for the decision to collect data the 2-month clinic.

It was decided that data collection should begin at the 2-month mark. This section will provide a rebuttal to the potential arguments posed by the proponents of why the study should begin at the 4-month well-child visit.

In response to the first argument, that PHNs already have their hands full at the 2-month well-child visit, it must be noted that as the age of the infant at diagnosis increases, the severity of the deformity may get worse and the potential for successful head shape correction through non-invasive means decreases significantly (Losee & Mason, 2005; Loveday & de Chalain, 2001). Clinically, active counter-positioning is an effective method for correcting plagiocephaly before 4-months of age (L. Walker, personal communication, August 27, 2009). Since PNHs are one of the main sources of health services for families during the first few post-natal months, it is vital that the appropriate prevention messages and early correction messages are passed on to parents by PHNs as early as possible.

The HSC in Calgary provides IRCs that have proven to be a cost-effective means of delivering hands-on education regarding repositioning, as well as torticollis screening (L. Walker, personal communication, August 27, 2009). This early intervention has resulted in a decreased percentage of infants requiring treatment with a cranial orthotic device. The classes are offered to infants between the ages of 0-4 months and require a verbal referral from a physician or PHN. Collecting data at the 4-month mark would likely decrease referral to these classes and result in families not receiving the appropriate early correction messages and would likely result in an increased rate of potential harm being caused to these infants. During IRCs, much time is spent with parents advising them of various active counter-positioning techniques. Although well-child clinic time may present opportunities to engage in active-counter positioning teaching, referral to these classes may be more appropriate for parents that require extensive teaching and reinforcement of these techniques.

Secondly, although treatment in terms of helmetting is available after the 4-month mark there are drawbacks to this option in terms of wait times, cost, and the time commitment required. Due to the large volume of referrals received at the HSC in Calgary (over 1000 annually) it is often a few months before infants are seen. The average age of the babies seen in clinic over the past several months has been is 7 to 7.5 months (L. Walker, personal communication, August 27, 2009). As a result, some families are not receiving adequate information on repositioning prior to this appointment and are missing very valuable time to correct their infant's plagiocephaly through active counter-positioning (L. Walker, personal communication, August 27, 2009). Because the trend of increasing wait times at specialty clinics is being observed, infants that need treatment in terms of helmetting may actually begin the treatment after the 6 to 8 month mark, when helmet treatment has been found to be less effective (Losee & Mason, 2005). This time frame for appropriate treatment is significant as most head growth is completed by the age of 2 years and when cranial moulding helmet therapy is started after the age of 6 months, plagiocephaly correction is less successful (Graham et al., 2005; Lee et al., 2008; Pollack et al., 1997; Teichgraeber et al., 2004).

There is a sizeable cost associated with helmet manufacturing, ranging from $1000 - $3000 US (Steinbok et al., 2007). In addition the devices must be worm 23 hours per day and families must return to the clinic weekly or biweekly in order to have the inner foam liner of the helmet adjusted to accommodate for head growth and plagiocephaly correction (L. Walker, personal communication, August 27, 2009). The limited number of qualified orthotists may create difficulties in initial access and in obtaining appropriate follow-up to have adjustments made in the device and to make sure

that undue pressure is not being placed on infants' delicate skin (Steinbok et al., 2007). Furthermore, the device is sometimes not well tolerated by some infants resulting in irritability that may cause families anxiety (Steinbok et al., 2007). These inconveniences in terms of costs and time commitment can be minimized through early recognition of plagiocephaly and the timely delivery of prevention messages or correction messages in terms of active counter-positioning in the early post-natal period.

Ultimately it is recommended that data collection begin at the 2-month mark. Two factors that affect the intervention options available, and thus ultimate outcome, are the age at presentation and the severity of deformity. As the age at presentation increases and the severity of the deformity worsens, the potential for successful correction with behaviour modification alone decreases significantly (Losee & Mason, 2005). The high prevalence of positional preference in infancy, the persistency of torticollis co-morbidity, the large number of children referred for further diagnostic and/or treatment, and the resulting high medical expenses call for a primary prevention approach (Boere-Boonekamp & van der Linder-Kuiper, 2001). This primary prevention approach would be better suited at the 2-month mark when non-invasive, yet effective interventions, can be implemented and result either in prevention of plagiocephaly or early correction of plagiocephaly.

Data Cleaning

Before data were analysed, the accuracy of data input was ensured by checking the data that was entered four times. Frequency descriptives were run in order to identify outliers. Interval data (infant age, maternal age, and the number of maternal years lived in Canada) were described by frequencies, proportions, means, medians, modes, standard

deviation, minimum value and maximum value. The dichotomous independent variables (sex, multiple gestation, and maternal language barrier), nominal independent variables (infant feeding position, infant sleep position, head positional preference, and maternal delivery type) and ordinal independent variables (infant tummy time, maternal education, and infant birth order) were described by frequencies and proportions. Implausible values, significant departures from the normal distribution of interval variables, gaps in values, and outliers were assessed. Using this method, data that are in some sense "far" from what one would expect based on the rest of the data were identified and corrections were made. In addition, missing values were defined for each category.

Data Analysis

Data analysis was conducted in order to answer the research questions of this study. The analysis is presented below according to the research questions posed.

What is the incidence of positional plagiocephaly in infants 7-12 weeks of age in Calgary, Alberta, Canada?

The incidence of plagiocephaly was calculated using the formula: (number of new events in the time period/number of persons exposed to the risk during the time period) x 10^n (Porta, 2008). This translates to (# of infants with plagiocephaly/# of infants in the sample) x 100. The incidence was reported as a percentage.

What are the potential risk factors for positional plagiocephaly in infants 7-12 weeks of age in Calgary, Alberta, Canada?

SPSS software (PASW version 17) was used to perform the analysis. Two data sets, one with eight sets of twins and one without were analyzed according to the process

outlined below. The population of infants and mothers was described and summarized. The outcome variable was described by frequencies and proportions according to the five types of plagiocephaly in Argenta's (2004) assessment scale. These data were also described by the side of the infant's head on which the plagiocephaly was observed, the severity of the plagiocephaly observed, whether or not the plagiocephaly had accompanying brachycephaly, and how the condition was distributed across the four data collection sites.

Since the outcome variable was captured in terms of the five types of plagiocephaly according to Argenta's (2004) assessment scale, it was re-coded in order to display either the presence or absence of plagiocephaly. In order to determine whether the observed frequencies (counts) differ markedly from the frequencies that would be expected by chance, contingency tables (chi-square) were computed for the outcome variable (the presence or absence of plagiocephaly) and the dichotomous and nominal variables in turn. Similarly, the independent sample t-test was performed for the outcome variable and the interval variables. Levene's test for equality of variances was performed and the results indicated that equal variances were assumed. The Mann-Whitney test was performed for the outcome variable and the ordinal variables. In all cases, a probability of .05 or less was considered to be significant (i.e., that the results were not due to chance).

In order to proceed with the multivariable analysis, both ordinal and nominal groups of data were transformed into dichotomous data (Katz, 2006). Data were grouped into categories based on the distribution of the variables and the relationship between the different categories of the ordinal and nominal variables and the outcome variable (Katz,

2006). This ensured the transformed variables adequately represented the data in order to have clinical significance. The χ^2test described above was repeated.

In order to ascertain if any of the independent variables were multicollinear, every combination of independent variables was tested using either the χ^2 test, Pearson correlation or Spearman Rank correlation; the test was chosen based on its appropriateness given the type of data. In order to assess for multicollinearity, the magnitude of the p value was assessed. Three combinations of variables had $\chi^2 \geq 30$ and a p=.000. When the presence of multicollinearity was identified, the variables that were theoretically less important or had missing data (maternal age and maternal number of years lived in Canada) were not included in the logistic regression analysis (Katz, 2006). The other two combinations of variables (birth order x sleep position and maternal education x maternal number of years lived in Canada) had smaller χ^2 values and $p > .01$. These combinations of variables were not treated as highly correlated and therefore, in both instances, both variables were included in the logistic regression analysis. Multiple logistic regression is able to handle situations in which the independent variables are correlated with each other to some degree (Norman & Streiner, 2000). The problem arises when these variables are highly correlated. In order to ensure that the two combinations of variables are not multicollinear, four additional regression models were produced (Appendix F). The presence of multicollinearity results in high standard errors and changes in the signs as well as in the magnitudes of the partial regression coefficients from one sample to another sample (Statistics Solutions, 2011). Since none of these changes were observed, it is assumed that a high degree of multicollinearity does not

exist between birth order and sleep position; and maternal education and head positional preference. Assessing for multicollinearity is necessary because when two variables are so closely related to one another, the model may not be able to assess reliably the independent contribution of each variable (Katz, 2006). Independent variables that did not satisfactorily capture the specificity sought in terms of its plausible contribution to the development of positional plagiocephaly were also not included in the logistic regression analysis (i.e., feeding position and infant age). The dichotomous variable that yielded cell counts less than five was also not included in the logistic regression analysis (i.e., maternal language barrier).

Simple logistic regression analysis was performed to understand the association of each predictive factor/independent variable (infant sleep position, infant tummy time, sex, multiple gestation, head positional preference and maternal age) and the outcome variable (i.e., the presence or absence of plagiocephaly). The dependent outcome variable was regressed separately with each independent variable. Simple logistic regression analysis using the logistic regression model below was used to assess each risk factor: $p = [\exp (\beta_o + \beta_1 x)]/[1 + \exp (\beta_o + \beta_1 x)]$ where $p = P(Y = 1)$; Y is the dependent variable (the presence or absence of plagiocephaly); x is the independent variable (risk factor), the β_1 coefficient gives the change in ln [p/1-p] associated with a change of one unit of x (T. Fung, personal communication, January 26, 2010). If x is binary, with the values of 0 or 1, the equation, the equation yields the ratio associated with x (Elwood, 2007). The statistical analysis software provided as part of its output the point estimate of β, p-value,

95% confidence interval and the numerical value of the odds ratio. A variable with $p < .05$ was considered to be statistically significant at a 95% confidence interval.

As a convention for planning multiple logistic regression, for every independent variable in the model, at least 10 cases (i.e., number of infants identified as having plagiocephaly) are needed (Katz, 2006). Since 204 infants that participated in this study were observed to have positional plagiocephaly, this convention would allow for up to 20 variables to be included in the multiple regression model. Therefore, the 10 variables identified below, were an acceptable number to be included in the model.

Multiple logistic regression is an accepted statistical method for assessing association between a risk factor and the probability of disease occurrence while statistically adjusting for potential confounding effects of other covariates (Lee, 1985). Therefore, the following predictive factors of plagiocephaly were used in this analysis: (a) infant sleep position, (b) infant tummy time, (c) birth order, (d) sex, (e) multiple gestation, (f) left head positional preference, (g) right head positional preference, (h) vacuum/forceps delivery, (i) caesarean section delivery, and (j) maternal age were used in this analysis.

The following multiple logistic regression model was used in this study:

$\ln [p/1-p] = \beta_o + \beta_1 x_1 + \beta_2 x_2 + \beta_3 x_3 + ... \beta_{10} x_{10}$ (Elwood, 2007). $p = P(Y = 1)$; Y is the dependent variable (the presence or absence of plagiocephaly); x_1 are the independent variables (risk factors), the β coefficient gives the change in the log odds ratio of p associated with a change of one unit of x (Elwood, 2007). If x is binary, with the values of 0 or 1, the equation, the equation yields the ratio associated with x (Elwood, 2007). The statistical analysis software, provided as part of its output, the point estimate of β, p-

value, 95% confidence interval and the numerical value of the odds ratio. A variable with $p < .05$ was considered to be statistically significant.

This type of analysis assists in understanding whether the model including a given independent variable provides more information about the dependent variable than the model without this variable. According to Daniel (1999), the multiple logistic regression model is used in health sciences research to identify the probability (risk) that an individual will acquire a medical condition during some specified time period during which he/she is exposed to the risk factor known to be (or suspected of being) associated with that medical condition. The objective of analysis of data that meet the criteria identified above is a statistic known as the odds ratio (Daniel, 1999). In this case it is the measure of how much greater (or less) the odds are for study participants possessing a risk factor to be diagnosed with plagiocephaly.

The multiple logistic regression method of analysis has several variable selection techniques that are available for model selection (Katz, 2006). For the purposes of exploratory research, the “all variables in” method was used in order to avoid running the risk of eliminating or not selecting a variable that is on the causal pathway to the outcome in favour of a variable that is a confounder (Katz, 2006). Using this method, all variables were entered simultaneously (Katz, 2006). Seven interaction terms (the interaction of sex with each independent variable in the model) were tested. The overall significance of the model with the interaction terms in the model versus out of the model was observed. The change in -2 times the log of the likelihood (-2LL) was observed and since the model with the interaction terms was not found to have a $p < .05$, it was not considered to be

statistically significant, and therefore all interaction terms were excluded from the model (T. Fung, personal communication, January 26, 2010).

After the model was developed, the goodness of fit of the model was assessed in order to examine how "likely" the sample results were given the parameter estimates (Norusis, 1990). The probability of the observed results given the parameter estimate is known as the Likelihood (Norusis, 1990). Since the likelihood is a small number less than 1, it is common to use -2LL as a gauge of how well the estimated model fits the data (Norusis, 1990). A good model is one that produces a high likelihood of the observed results (Norusis, 1990). This translates to a small value for -2LL (Norusis, 1990). If a model fits perfectly, then the likelihood is 1 and - 2 times the log likelihood is 0. To test the null hypothesis that the observed likelihood does not differ from 1 (the value of the likelihood for a model that fits perfectly), the value of - 2LL can be used (Norusis, 1990). Assuming that the model fits perfectly, -2LL has a chi-square distribution with N – p degrees of freedom, where N is the number of cases and p is the number of parameters estimated (Norusis, 1990). If the observed significance level is large, the hypothesis that the model fits is not rejected (Norusis, 1990). Furthermore, the Cox and Snell and Nagelkerke R square value that is large indicates a model that has a good fit (T. Fung, personal communication, April 21, 2011).

The Hosmer-Lemeshow goodness-of-fit test is another test that was used to compare the estimated to observed likelihood of outcomes for groups of study participants; it is a qualitative assessment of how well the model *accounts* for the outcome (Katz, 2006). The groups are formed by dividing the sample into approximately

10 groups based on the range of estimated probability of the outcome (Katz, 2006). The first group contains the 10 percent of study participants with the lowest estimated likelihood of the outcome and the second group contains 10 percent of study participants with the next lowest estimated likelihood of the outcome, etc. (Katz, 2006). In a model that is well-fitting, the estimated likelihood will be close to the observed likelihood of the outcome and this will result in a small chi-square and a non-significant p-value.

What intervention and follow-up actions do PHNs take if positional plagiocephaly is identified in healthy infants at the 2-month well-child clinic?

Patterns of PHN intervention (i.e., repositioning teaching, referral to the IRC or referral to the family physician, and follow-up planned) were described using frequencies and proportions. In addition, reasons for referral and any additional comments that the PHNs provided were described.

What intervention and follow-up actions do clinicians (physiotherapists and occupational therapists) take when healthy infants with positional plagiocephaly are referred to the infant repositioning class (IRC) after the 2-month well-child clinic visit in Calgary, Alberta, Canada?

The following discussion pertains to the group of infants identified as having positional plagiocephaly that presented at the IRC. The outcome variable was described by frequencies and proportions according to the five types of plagiocephaly on Argenta's (2004) assessment scale, as observed by the clinicians. The severity of the positional plagiocephaly was presented. Descriptive statistics were run to identify patterns of clinician actions in terms of cranial orthosis (helmet), physiotherapy or repositioning teaching. The presence or absence

of torticollis was also analyzed descriptively according to the type of positional plagiocephaly identified. The appropriateness of the PHN referral as well as whether the parent/guardian and infant attended the IRC were captured.

Significance of the Study

This research will provide a baseline estimated incidence of positional plagiocephaly in Calgary and can be used to inform future PHN and primary care physician practice about positional plagiocephaly assessment and action (intervention). It is important to determine the incidence of the condition so that appropriate resources can be allocated for the management of infants with positional plagiocephaly. From an epidemiological perspective, the modifiable risk factors that place infants at risk of positional plagiocephaly can be addressed. Using knowledge of incidence and risk factors, the incidence and prevalence of positional plagiocephaly and the cost of its management would potentially be reduced.

Ethics

The study was approved by the University of Calgary Conjoint Health Research Ethics Board on June 3, 2010 (Ethics ID E-23061). Issues pertaining to consent and privacy and confidentiality as outlined by to the Tri-Council Policy Statement for Ethical Conduct for Research Involving Humans (Canadian Institutes of Health Research, Natural Sciences and Engineering Research Council of Canada, and Social Sciences and Humanities Research Council of Canada, 2010) will be discussed.

With respect to consent, four general principles were adhered to: (a) consent shall be given voluntarily, (b) consent shall be informed, (c) consent should be an ongoing process, and (d) consent shall precede collection of, or access to, research data (Canadian

Institutes of Health Research, Natural Sciences and Engineering Research Council of Canada, and Social Sciences and Humanities Research Council of Canada, 2010). With respect to voluntary consent**,** undue influence and manipulation may arise when prospective participants are recruited by individuals in a position of authority (Canadian Institutes of Health Research, Natural Sciences and Engineering Research Council of Canada, and Social Sciences and Humanities Research Council of Canada, 2010). The Research Nurses that were obtaining informed consent were not employees of the public health branch of Alberta Health Services and not involved in service delivery at the CHCs. Therefore, the potential study participants did not have any reason to feel obligated to follow the wishes of the Research Nurses obtaining informed consent. Potential study participants were not coerced to participate in the study in any way, shape or form. Potential study participants were informed that participation in the study was completely voluntary and the decision to participate or not would not affect any services offered by Alberta Health Services. No incentives, monetary or otherwise, were offered for participation in the research study.

Informed consent was obtained. Parent(s)/guardian(s) of infants enrolled in the study signed a consent form that was approved by the University of Calgary Conjoint Health Research Ethics Board (Appendix B). The consent form indicated in plain language the purpose of the research and the expected duration and nature of participation. The consent form indicated that the individual and infant are being invited to participate in a research project but there is no obligation to participate and they are free to withdraw at any time without prejudice to pre-existing entitlements. A description

of all reasonably foreseeable risks and potential benefits, both to the participants and in general, that may arise from research participation was also provided on the consent form in plain language. Informed consent was obtained prior to the collection of any data.

With respect to privacy and confidentiality, five key concepts were considered: (a) privacy, (b) confidentiality, (c) security, (d) identifiable information, and (e) types of information (Canadian Institutes of Health Research, Natural Sciences and Engineering Research Council of Canada, and Social Sciences and Humanities Research Council of Canada, 2010). Privacy is defined as an individual's right to be free from intrusion or interference by others (Canadian Institutes of Health Research, Natural Sciences and Engineering Research Council of Canada, and Social Sciences and Humanities Research Council of Canada, 2010). Privacy was respected by giving individuals the opportunity to exercise control over personal information by consenting to, or withholding consent for, the collection, use and/or disclosure of information as outlined for the purposes of the present study.

The ethical obligation of confidentiality refers to the responsibility of an individual or organization to safeguard entrusted information (Canadian Institutes of Health Research, Natural Sciences and Engineering Research Council of Canada, and Social Sciences and Humanities Research Council of Canada, 2010). With respect to confidentiality, the data collected were protected from unauthorized access, use, disclosure, modification, loss or theft.

Security refers to measures used to protect information (Canadian Institutes of Health Research, Natural Sciences and Engineering Research Council of Canada, and

Social Sciences and Humanities Research Council of Canada, 2010). Raw data collected for the purposes of this study were stored in a locked filing cabinet in a locked office. The computer containing research data was located in a locked office away from public areas. The computer containing research data was password protected, and had the appropriate firewalls and anti-virus software programs installed, as per the University of Calgary Information Technologies Protocol.

Information that may reasonably be expected to identify an individual, alone or in combination with other available information, is considered identifiable information (Canadian Institutes of Health Research, Natural Sciences and Engineering Research Council of Canada, and Social Sciences and Humanities Research Council of Canada, 2010). In order to adhere to this ethical obligation, consent forms with the name of the parent/guardian providing consent and the name of the infant were kept separate from data collection forms. In addition, data were reported in aggregate forms.

With respect to types of information, researchers may seek to collect, use, share and access different types of information about participants. Such information may include personal characteristics or other information about which an individual has a reasonable expectation of privacy (Canadian Institutes of Health Research, Natural Sciences and Engineering Research Council of Canada, and Social Sciences and Humanities Research Council of Canada, 2010). In order to limit the types of identifiable information collected, various measures were taken. The only directly identifying information that was collected was the name of the infant and the name of the parent/guardian that was providing consent for the purposes of obtaining informed

consent. No social insurance numbers or Alberta health care numbers were collected. No indirectly identifying information – the information can reasonably be expected to identify an individual through a combination of indirect identifiers (e.g., date of birth, place of residence or unique personal characteristic) – were collected. In addition, all data entered into the password protected computer was coded. The code key was kept in a locked filing cabinet.

In the following chapter, the results of the study are presented.

CHAPTER THREE: RESULTS

The results of the study are presented in this chapter. The chapter begins with a discussion of the response rate in proportion to all eligible participants at the four CHCs. The sample obtained is then discussed in relation to the distribution of data collected according to each data collection site. The remainder of this chapter is devoted to the analysis undertaken in order to answer each of the five research questions, identified as main headings in this chapter.

Response Rate

Of the 486 parents/guardians of infants that were approached to participate in the study, 471 agreed to participate. The response rate for this study was 96.7% (471/486 X 100). Since there were 1712 infants eligible to participate in the study from the four data collection sites during the duration of the study (July – September 2010), the number of eligible infants whose parents/guardians agreed to participate represents 27% of all eligible participants. In Table 2, the distribution of the sample across the four CHCs is presented.

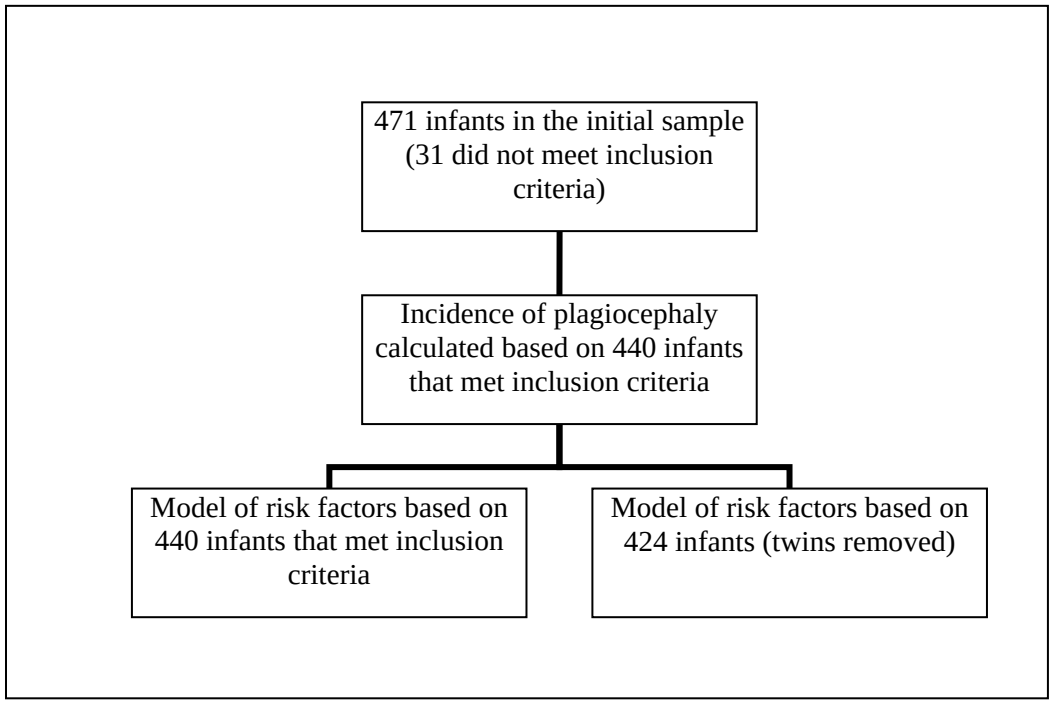
Table 2: Distribution of Sample across Sites

Clinic Number	Frequency	Percent	Cumulative Percent
Clinic 1	23	5.2	5.2
Clinic 2	130	29.5	34.7
Clinic 3	179	40.7	75.4
Clinic 4	108	24.5	100
Total	440	100	

The Sample

The initial sample consisted of 471 infants. Thirty one infants did not meet the inclusion criteria and were removed. Seventeen of the 31 infants were at the clinic for reasons other than immunization and were removed. Ten infants were older than 12 weeks, one infant was premature, and three infants presented with brachycephaly only and had no signs of plagiocephaly. The incidence rate was calculated based on 440 infants that met the inclusion criteria. The sample used to model risk factors was 440. Because there were 8 pairs of twins, in order to demonstrate that the data obtained from the 8 pairs of twins did not affect the results with respect to the issue of independence, a second model was developed with the twins removed (Figure 2).

Figure 2. The Sample



What is the Estimated Incidence of Positional Plagiocephaly in Infants 7-12 Weeks of Age in Calgary, Alberta, Canada?

In this section, the calculated estimated incidence of positional plagiocephaly is presented. The calculated incidence is followed by the distribution of each type of plagiocephaly across the data collection sites. Descriptive information pertaining to the type of plagiocephaly observed, the side of the head on which the plagiocephaly was observed, the severity of plagiocephaly observed and whether or not those infants identified as having plagiocephaly also demonstrated signs of accompanying brachycephaly is also provided.

Incidence of positional plagiocephaly (n=440).

As demonstrated in Table 3, 205 of 440 infants assessed were observed to have some form of plagiocephaly. The incidence of plagiocephaly in infants that ranged from 7-12 weeks of age was 46.6%.

Table 3: Number of Infants Identified With Positional Plagiocephaly According to Type

Plagiocephaly Type	Frequency	Percent	Valid Percent
Type 1	119	27.0	58.0
Type 2	38	8.6	18.5
Type 3	43	9.8	20.9
Type 4	2	0.5	1.0
Type 1 and 5 (vertical growth)	1	0.2	0.5
Type 1 and 3 (frontal asymmetry)	1	0.2	0.5
Missing	1	0.2	0.5
No observed plagiocephaly	235	53.4	100.0
Total	440	100.0	

In Table 4 the distribution of data collected at the four sites is displayed.

Table 4: Distribution of Positional Plagiocephaly across Data Collection Sites

Plagiocephaly Type	Clinic 1	Clinic 2	Clinic 3	Clinic 4	Total
Type 1	3	34	51	31	119
Type 2	3	7	13	15	38
Type 3	3	8	17	15	43
Type 4	0	0	2	0	2
Type 1 and 5 (vertical growth)	0	0	1	0	1
Type 1 and 3 (frontal asymmetry)	0	0	1	0	1
Total	9	49	85	61	204

In Table 5 the type of plagiocephaly and the corresponding side of the infants' head on which the plagiocephaly was observed is presented. Of all infants indentified as having plagiocephaly, 63.2% were affected on the right side.

Table 5: Side of Head on which Positional Plagiocephaly was Observed According to Type

Plagiocephaly Type		Side of Head that Plagiocephaly was Observed		
		Left	Right	Total
Type 1				
	Frequency	40	77	117
	Percent	19.8	37.7	57.9
Type 2				
	Frequency	13	25	38
	Percent	6.4	12.3	18.8
Type 3				
	Frequency	17	26	43
	Percent	8.3	12.7	21.3
Type 4				
	Frequency	2	0	2
	Percent	1.0	0.0	1.0
Type 1 and 3				
	Frequency	1	0	1
	Percent	0.5	0.0	0.5
Type 1 and 5				
	Frequency	0	1	1
	Percent	0.0	0.5	0.5
Total				
	Frequency	73	129	202
	Percent	36.0	63.2	100.0
Missing				2

All infants were assessed subjectively for severity of positional plagiocephaly. The severity of plagiocephaly was identified as either mild, moderate or severe. Of all infants identified as having plagiocephaly, 78.3% had a mild form, whereas 18.8% had a moderate form and 3.0% had a severe form (Table 6).

Table 6: Distribution of Observed Positional Plagiocephaly According to Severity and Type

		Severity of Plagiocephaly that was Observed			
Plagiocephaly		Mild	Moderate	Severe	Total
Type 1					
	Frequency	104	12	2	118
	Percent	51.5	5.9	1.0	58.4
Type 2					
	Frequency	29	8	1	38
	Percent	14.4	4.0	0.5	18.9
Type 3					
	Frequency	23	16	3	42
	Percent	11.4	7.9	1.5	20.8
Type 4					
	Frequency	0	2	0	2
	Percent	0.0	1.0	0.0	1.0
Types 1 and 3					
	Frequency	1	0	0	1
	Percent	0.5	0.0	0.0	0.5
Types 1 and 5					
	Frequency	1	0	0	1
	Percent	0.5	0.0	0.0	0.5
Total					
	Frequency	158	38	6	202
	Percent	78.3	18.8	3.0	100.0
Missing					2

As presented in Table 7, of all infants observed to have plagiocephaly, 3.9% also showed the characteristic symmetrically flattened occiput associated with brachycephaly.

Table 7: Distribution of Infants with Positional Plagiocephaly also Demonstrating Signs of Brachycephaly According to Type of Plagiocephaly

		Brachycephaly Observed		
Plagiocephaly Type		Yes	No	Total
Type 1				
	Frequency	6	113	119
	Percent	2.9	55.4	58.3
Type 2				
	Frequency	1	37	38
	Percent	0.5	18.1	18.6
Type 3				
	Frequency	1	42	43
	Percent	0.5	20.6	21.1
Type 4				
	Frequency	0	2	2
	Percent	0.0	1.0	1.0
Type 1 and 3				
	Frequency	0	1	1
	Percent	0.0	0.5	0.5
Type 1 and 5				
	Frequency	0	1	1
	Percent	0.0	0.5	0.5
Total				
	Frequency	8	196	204
	Percent	3.9	96.1	100.0

Argenta's (2004) plagiocephaly assessment tool was used to complete the assessments. It was important to conduct the assessments systematically in order for the incident calculation to be precise. The question that remains to be answered is: Is Argenta's (2004) plagiocephaly assessment tool appropriate for use in well-child clinics? The rationale for selecting the tool was provided in chapter 2 and in the section below, this question will be explored further.

Is Argenta's (2004) Plagiocephaly Assessment Tool Appropriate for Use in Well-Child Clinics?

With respect to Argenta's (2004) plagiocephaly assessment tool, information was collected pertaining to the following criteria: (a) the suitability/feasibility in terms of ease of use and time required, (b) the acceptability of the tool to the target population, (c) measures taken to ensure reliability, and (d) the yield of the tool.

Suitability/feasibility.

Based on Argenta's (2004) assessment guidelines providing four examination positions (described in chapter two), plagiocephaly assessment data were relatively easy to collect. Data were recorded in terms of which type of plagiocephaly was identified according to Argenta's (2004) plagiocephaly assessment tool. Although not part of the tool, data were also collected subjectively about the severity of plagiocephaly identified within each category, in terms of whether the observed plagiocephaly was mild, moderate or severe. Each plagiocephaly assessment took less than five minutes to complete and a few seconds to record.

Acceptability of the tool to the target population.

When parents/guardians of potential infants were approached, informed of what the study would entail, and invited to participate in the study, the majority of parents/guardians agreed to participate. As stated above, the response rate for the study was very high at 96.7%. This high response rate indicates the willingness of the target population to participate in the study, indicating that the data collection method, in terms of the plagiocephaly assessment was very acceptable to the target population.

Measures taken to ensure reliability.

Various measures were taken in order to ensure that reliability of the study was maintained in terms of the plagiocephaly assessments. First, both Research Nurses spent eight or more hours conducting plagiocephaly assessments with clinicians at the HSC in order to gain awareness of the range of severity of plagiocephaly in Calgary. Second, for the first week, assessments were conducted together to ensure that the same classification, according to the five types of plagiocephaly in Argenta's (2004) plagiocephaly assessment tool, was obtained. Lastly, both Research Nurses connected half way through data collection and again towards the end of data collection in order to ensure the plagiocephaly assessments were conducted in a consistent fashion.

The yield of the tool.

As described in chapter one, the yield of the tool is defined as the ability of the tool to identify a previously unrecognized medical condition (Wilson & Jungner, 1968). Using Argenta's (2004) plagiocephaly assessment tool, 205 infants out of 440 infants were identified as having some form of plagiocephaly.

Given the information collected pertaining to four criteria presented above, Argenta's (2004) plagiocephaly assessment tool appears to be appropriate for conducting plagiocephaly assessments in well-child clinics, where accurate assessment data are required within a relatively short time frame.

Although the estimated incidence of positional plagiocephaly is presented above, it is also important to determine which factors put infants aged 7-12 weeks at increased

risk for developing the condition. In the following section the results obtained around risk factors is presented.

What are the Potential Risk Factors for Positional Plagiocephaly in Infants 7-12 Weeks of Age in Calgary, Alberta, Canada?

The information in this section pertains to the analysis that took place in order to arrive at a model identifying risk factors predictive of positional plagiocephaly. For ease of understanding, the section is divided into non-modifiable and modifiable risk factors and further subdivided into infant and maternal risk factors within each section.

Non-modifiable risk factors (n=440).

This study looked at seven non-modifiable risk factors generated from the literature review: (a) infant birth order, (b) maternal delivery type, (c) infant sex, (d) multiple gestation, (e) infant age, (f) maternal age, and (g) maternal number of years lived in Canada.

Infant risk factors.

As demonstrated in Table 8, almost 50% of all infants assessed were the first born into the family.

Table 8: Non Modifiable Infant Risk Factors for Positional Plagiocephaly According to Birth Order

Variable		Frequency	Percent	Cumulative Percent
Birth order				
	First child	214	48.6	48.6
	Second child	161	36.6	85.2
	Third child	45	10.2	95.5
	Fourth child or more	20	4.5	100.0
	Total	440	100.0	

There was no statistically significant difference in the incidence of plagiocephaly between those infants that were first born compared to those that were not first born (Table 9).

Table 9: Occurrence of Positional Plagiocephaly According to Infant Birth Order

		Birth Order					
Plagiocephaly		Not First Child	First Child	Total	χ^2	df	p^a
No							
	Frequency	123	112	235			
	Percent	54.4	52.3	53.4			
Yes							
	Frequency	103	102	205			
	Percent	45.6	47.7	46.6			
Total							
	Frequency	226	214	440			
	Percent	100.0	100.0	100.0			
					.193	1	.661

Note. df = degrees of freedom.
[a]two-tailed *p*-value.

Almost 60% of the infants assessed were male (Table 10).

Table 10: Non Modifiable Infant Risk Factors for Positional Plagiocephaly According to Infant Sex

Variable		Frequency	Percent	Cumulative Percent
Sex				
	Male	261	59.3	59.3
	Female	179	40.7	100.0
	Total	440	100.0	

According to Table 11, there was no statistically significant difference in positional plagiocephaly by sex.

Table 11: Occurrence of Positional Plagiocephaly According to Infant Sex

		Sex					
Plagiocephaly		Female	Male	Total	χ^2	df	p^a
No							
	Frequency	102	133	235			
	Percent	57.0	51.0	53.4			
Yes							
	Frequency	77	128	205			
	Percent	43.0	49.0	46.6			
Total							
	Frequency	179	261	440			
	Percent	100.0	100.0	100.0			
					1.549	1	.213

Note. df = degrees of freedom.
[a]two-tailed *p*-value.

The proportion of infants born of multiple gestation pregnancies is shown in Table 12.

Table12: Non Modifiable Infant Risk Factors for Positional Plagiocephaly According to Multiple Gestation Pregnancy

Variable		Frequency	Percent	Cumulative Percent
Multiple				
	Yes	17	3.9	3.9
	No	419	95.2	96.1
	Sub-total	436	99.1	100.0
	Missing	4	0.9	
	Total	440	100	

As presented in Table 13, there was no statistically significant difference in the incidence of positional plagiocephaly between multiple gestation and singleton pregnancies.

Table 13: Occurrence of Positional Plagiocephaly According to Multiple Gestation Pregnancies

		Multiple					
Plagiocephaly		Yes	No	Total	χ^2	df	p^a
No							
	Frequency	222	11	233			
	Percent	53.0	64.7	53.4			
Yes							
	Frequency	197	6	203			
	Percent	47	35.3	46.6			
Total							
	Frequency	419	17	436			
	Percent	100.0	100.0	100.0			
					.902	1	.342

Note. df = degrees of freedom.
[a]two-tailed *p*-value.

In Table 14, the mean, median, mode, standard deviation, range, minimum and maximum for infant age are presented.

Table 14: Age of Infants Participating in the Study in Months

Statistic	Infant Age
N	440
Mean	2.251
Median	2.250
Mode	2.00
Standard deviation	0.300
Range	1.25
Minimum	1.75
Maximum	3.00

Maternal risk factors.

There are several important results from the analysis of maternal risk factors. As is evident in Table 15, more than half of the infants in the study were delivered vaginally without assistance.

Table 15: Maternal Delivery Type

Delivery Type	Frequency	Percent	Cumulative Percent
Vaginal (no assistance)	234	53.2	53.2
Cesarean	133	30.2	83.4
Vacuum	54	12.3	95.7
Forceps	19	4.3	100
Total	440	100	

As demonstrated in Table 16, there was no statistically significant difference in positional plagiocephaly by delivery type.

Table 16: Occurrence of Positional Plagiocephaly Compared with Maternal Delivery Type

Plagiocephaly		Maternal Delivery Type			Total	χ^2	df	p^a
		Vaginal	Vacuum/Forceps	Cesarean Section				
No								
	Frequency	124	34	77	235			
	Percent	53.0	46.6	57.9	53.4			
Yes								
	Frequency	110	39	56	205			
	Percent	47.0	53.4	42.1	46.6			
Total								
	Frequency	234	73	133	440			
	Percent	100.0	100.0	100.0	100.0			
						2.462	2	.292

Note. df = degrees of freedom.
[a]two-tailed *p*-value.

With regard to maternal age and length of time in Canada, in Table 17 the mean, median, mode, standard deviation, range, minimum and maximum for these variables are presented. The age of mothers participating in the study ranged from 17-47 years. The average age of mothers was 30.86 years. The large variation in age may provide some indication about whether or not maternal age is a risk factor for the development of positional plagiocephaly. In terms of the number of years lived in Canada, some mothers have lived in Canada for less than a year, whereas others have lived in Canada for much longer periods, some having never lived in another country.

Table 17: Maternal Age and Maternal Number of Years Lived in Canada

Statistic	Maternal Age	Number of Years Lived in Canada
N	440	440
Mean	30.86	22.13
Median	31.00	27.00
Mode	30.00	30.00
Standard deviation	4.94	12.09
Range	30.00	43.70
Minimum	17.00	.3
Maximum	47.00	44.00

According to Table 18 and 19, there is a statistically significant difference in the mean age of mothers with respect to the presence or absence of positional plagiocephaly. The Levene's test for equality of variances was performed and the results indicated no significant differences (p=.385) in variances. As a consequence, equal variances were assumed for the t-test. However, there was no statistically significant difference in the incidence of positional plagiocephaly with respect to the number of years mothers of the infants in the study have lived in Canada.

Table 18: Occurrence of Positional Plagiocephaly According to Maternal Age and Number of Years lived in Canada

Variable	Plagiocephaly	Frequency	Mean	SD	SEM
Mother's age	No	235	31.323	4.856	.317
	Yes	205	30.332	4.991	.349
Years in Canada	No	235	22.637	12.019	.784
	Yes	205	21.556	12.181	.851

Note. SD = standard deviation; SEM = standard error of mean.

Table 19: t- test for Equality of Means for Maternal Age and Number of Years Lived in Canada

Variable	t	df	p^a	Mean Difference	Standard Error of the Difference	95% CI of the Mean Difference	
						LL	UL
Maternal Age	2.109	438	.035	0.992	0.470	0.068	1.916
Number of Years in Canada		438	.350	1.082	1.156	-1.190	3.354

Note. df = degrees of freedom; CI = confidence interval; LL = lower limit; UL = upper limit.
[a]two-tailed *p*-value.

As indicated above, one of the non-modifiable risk factors proved to be statistically significant in bivariate analyses. There is a statistically significant difference in the mean age of mothers with respect to the presence or absence of positional plagiocephaly. The following section will present the results of the bivariate analyses of the modifiable risk factors.

Modifiable risk factors (n=440).

Six modifiable risk factors were generated from the literature and considered in this study under two broad headings: infant risk factors (infant sleep position, infant head positional preference, infant feeding position, and infant tummy time); and maternal risk factors (maternal education, and maternal language barrier).

Infant risk factors.

Over three-quarters of infants in the study sample slept in the supine position (Table 20).

Table 20: Infant Usual Sleep Position

Sleep Position	Frequency	Percent	Cumulative Percent
Back	335	76.1	76.1
Side	42	9.5	85.6
Tummy	25	5.7	91.3
Back and side	25	5.7	97.0
Tummy and side	4	0.9	97.9
Back and tummy	3	0.7	98.6
Back, tummy, and side	5	1.1	99.7
Missing	1	0.2	100.0
Total	440	100.0	

As indicated in Table 21, there was a statistically significant difference in the incidence of plagiocephaly in the infants that were placed supine to sleep compared to those that were placed to sleep in other positions (tummy, side or a combination of back, tummy or side).

Table 21: Occurrence of Positional Plagiocephaly According to Infant Usual Sleep Position

		Infant Sleep Position					
Plagiocephaly		Other	Back	Total	χ^2	df	p^a
No							
	Frequency	71	164	235			
	Percent	68.3	49.0	53.5			
Yes							
	Frequency	33	171	204			
	Percent	31.7	51.0	46.5			
Total							
	Frequency	104	335	439			
	Percent	100.0	100.0	100.0			
					11.901	1	.001*

Note. df = degrees of freedom.
[a]two-tailed *p*-value.
*Significant at 0.05 level.

When asked if they noticed a head positional preference (i.e., the side of the head on which the infant prefers to rest his/her head), 58% of parents noticed a positional preference while their infant(s) were positioned on their backs and 42% reported no demonstrated preference (Table 22).

Table 22: Infant Head Positional Preference While Laying Supine

Preference	Frequency	Percent	Cumulative Percent
Right Side	154	35.0	35.0
Left Side	101	23.0	58.0
No Preference	185	42.0	100.0
Total	440	100.0	

There was a statistically significant difference in the incidence of positional plagiocephaly in the infants that had a positional preference when compared to those that had no positional preference (Table 23).

Table 23: Occurrence of Positional Plagiocephaly According to Infant Head Positional Preference While Laying Supine

		Head Positional Preference						
Plagiocephaly		Right	Left	No Preference	Total	χ^2	df	p^a
No								
	Frequency	62	37	136	235			
	Percent	40.3	36.6	73.5	53.4			
Yes								
	Frequency	92	64	49	205			
	Percent	59.7	63.4	26.5	46.6			
Total								
	Frequency	154	101	185	440			
	Percent	100.0	100.0	100.0	100.0			
						52.173	2	.000*

Note. df = degrees of freedom.
[a]two-tailed *p*-value.
*Significant at 0.05 level.

As presented in Table 24, the majority of infants were fed while being held in the caregiver's arms.

Table 24: Infant Usual Feeding Position

Feeding Position	Frequency	Percent	Cumulative Percent
In my arms	404	91.8	91.8
Infant laying on another surface	16	3.6	95.4
Infant sitting in a chair	2	0.5	95.9
In my arms and infant sitting in a chair	2	0.5	96.4
In my arms and infant laying on another surface	14	3.2	99.6
Infant sitting in a chair and infant laying on another surface	1	0.2	99.8
Missing	1	0.2	100.0
Total	440	100.0	

There was no statistically significant association between plagiocephaly and infant feeding position (Table 25).

Table 25: Occurrence of Positional Plagiocephaly According to Infant Usual Feeding Position

Plagiocephaly		Infant Feeding Position: Other	Infant Feeding Position: In Arms	Total	χ^2	df	p^a
No							
	Frequency	18	217	235			
	Percent	51.4	53.7	53.8			
Yes							
	Frequency	17	187	204			
	Percent	48.6	46.3	46.5			
Total							
	Frequency	35	404	439			
	Percent	100.0	100.0	100.0			
					0.068	1	.759

Note. df = degrees of freedom.
[a]two-tailed *p*-value.

As presented in Table 26, over half of the infants received tummy time less than three times per day.

Table 26: Infant Tummy Time Per Infant Per Day

Tummy Time (per day)	Frequency	Percent	Cumulative Percent
0 times	11	2.5	2.5
1 time	103	23.4	25.9
2 times	139	31.6	57.5
3 times	76	17.3	74.8
4 times	29	6.6	81.4
More than 4 times	80	18.2	99.6
Missing	2	0.5	100
Total	440	100.0	

There was no association between positional plagiocephaly and tummy time (Table 27).

Table 27: Occurrence of Positional Plagiocephaly According to Infant Tummy Time (Per Infant per Day)

Plagiocephaly		Infant Tummy Time ≤ 2 times	Infant Tummy Time ≥3 times	Total	χ^2	df	p^a
No							
	Frequency	134	101	235			
	Percent	53.0	54.0	53.4			
Yes							
	Frequency	119	86	205			
	Percent	47.0	46.0	46.6			
Total							
	Frequency	253	187	440			
	Percent	100.0	100.0	100.0			
					0.047	1	.828

Note. df = degrees of freedom.
[a]two-tailed *p*-value.

Of the modifiable infant risk factors presented above, the following were found to be statistically significant during bivariate analyses: (a) supine sleep positioning, (b) right-sided head positional preference, and (c) left-sided head positional preference. The following section will present the modifiable maternal risk factors.

Maternal risk factors.

Two modifiable maternal risk factors were examined: maternal education and maternal language barrier. With respect to maternal education, the majority of mothers of the infants in the study had completed some high school, college or trade school, or completed an undergraduate university degree (Table 28).

Table 28: Highest Level of Maternal Education Completed

Level of Education	Frequency	Percent	Cumulative Percent
Less than grade 6	4	0.9	0.9
Grade 7 – 9	1	0.2	1.1
Grade 10 – 12	99	22.5	23.6
College and trade school	121	27.5	51.1
Undergraduate degree	182	41.4	92.5
Masters degree	29	6.6	99.1
PhD	3	0.7	99.8
Missing	1	0.2	100
Total	440	100	

As demonstrated in Table 29, when compared with the presence or absence of positional plagiocephaly in infants whose mothers' level of education ranged from grade 1-12, there is a statistically significant difference with respect to the presence or absence of plagiocephaly in infants whose mothers completed some form of higher education.

Table 29: Occurrence of Positional Plagiocephaly According to Highest Level of Maternal Education Completed

		Maternal Education					
Plagiocephaly		Grade 1-12	Higher Ed	Total	χ^2	df	p^a
No							
	Frequency	46	189	235			
	Percent	44.2	56.4	53.5			
Yes							
	Frequency	58	146	204			
	Percent	55.8	43.6	46.5			
Total							
	Frequency	104	335	439			
	Percent	100.0	100.0	100.0			
					4.739	1	.029*

Note. df = degrees of freedom.
[a]two-tailed *p*-value.
*Significant at 0.05 level.

Of all mothers participating in the present study, 2% were found to have a language barrier while communicating with them (Table 30).

Table 30: Maternal Language Barrier

Language Barrier	Frequency	Percent	Cumulative Percent
Yes	9	2.0	2.0
No	431	98.0	100
Total	440	100	

There was no statistically significant difference in the incidence of positional plagiocephaly of infants of mothers that had a language barrier when compared to those that did not (Table 31).

Table 31: Occurrence of Positional Plagiocephaly According to the Presence or Absence of Maternal Language Barrier

		Maternal Language Barrier					
Plagiocephaly		No	Yes	Total	χ^2	df	p^a
No							
	Frequency	233	2	235			
	Percent	54.1	22.2	53.4			
Yes							
	Frequency	198	7	205			
	Percent	45.9	77.8	46.6			
Total							
	Frequency	431	9	440			
	Percent	100.0	100.0	100.0			
					3.591	1	.089

Note. df = degrees of freedom.
[a]two-tailed *p*-value determined from Fisher's exact test.

Of the modifiable maternal risk factors presented above, maternal education was found to be statistically significant. The following sections will address multicollinearity, and present a statistical model predictive of the development of positional plagiocephaly.

Multicollinearity.

The combinations of variables that demonstrate multicollinearity are presented in Table 32. Due to the low strength of association between birth order and sleep position; and maternal education and infant head positional preference, these four variables were included in the model. Appendix F confirms that no mulitcollinearity exists between these variables. Maternal age and maternal number of years lived in Canada were excluded from the model.

Table 32: Variables Demonstrating Multicollinearity

Variable Combinations	χ^2	df	p^a
Birth order x sleep position	5.520	1	0.024*
Birth order x maternal age	30.962	3	0.000*
Maternal education x maternal age	62.099	3	0.000*
Maternal education x number of years lived in Canada	57.576	8	0.000*
Maternal education x infant head positional preference	7.933	2	0.019*

Note. df = degrees of freedom.
[a]2-tailed p-value.
*Significant at 0.05 level.

Statistical model of risk factors predictive of positional plagiocephaly.

The results of the simple logistic regression analyses are displayed in Table 33. According to these results, infants that slept in the supine sleep position had 2.2 times the odds of developing positional plagiocephaly than infants that did not sleep in the supine position. Maternal education at the post secondary level serves as a protective factor for the development of positional plagiocephaly. When compared to infants that have no head positional preference, infants with a preferred side have more than 4 times the odds of developing positional plagiocephaly.

Table 33: Unadjusted Statistical Model of Risk Factors Predictive of Positional Plagiocephaly

Variable	β.C.	S.E.	Wald	df	*p*	OR	95% CI
Sleep position	0.808	0.237	11.588	1	.001*	2.245	[1.409, 3.572]
Tummy time	-0.042	0.193	0.047	1	.828	0.959	[0.656, 1.401]
Birth order	0.084	0.191	0.193	1	.661	1.088	[0.748, 1.582]
Infant sex	0.243	0.195	1.547	1	.214	1.275	[0.870, 1.869]
Multiple	-0.487	0.517	0.887	1	.346	0.615	[0.223, 1.693]
Maternal education	-0.490	0.226	4.696	1	.030*	0.613	[0.393, 0.954]
Delivery type	0.257	0.269	0.915	1	.339	1.293	[0.764, 2.190]
Delivery type (c-section)	-0.199	0.219	0.822	1	.365	0.820	[0.534, 1.260]
Head positional preference (right)	1.415	0.234	36.589	1	.000*	4.118	[2.603, 6.515]
Head positional preference (left)	1.569	0.265	34.953	1	.000*	4.801	[2.854, 8.076]

Note. β.C. = beta regression coefficient; S.E = standard error; df = degrees of freedom; OR = odds ratio; CI = confidence interval.
*Significant at 0.05 level.

The results of the multivariate logistic regression analysis are displayed in Table 34. Ten variables were entered into the model simultaneously to produce the adjusted model: (a) infant sleep position, (b) infant tummy time, (c) birth order, (d) infant sex, (e) multiple gestation pregnancy, (f) maternal education, (g) delivery type (vacuum/forceps), (h) delivery type (c-section), (i) infant head positional preference (right), (j) and infant head positional preference (left). As presented in Table 34, when compared to infants that did not sleep in the supine position, infants that sleep supine had 2.7 times the odds of developing positional plagiocephaly. Males are at increased risk, as are those infants that have had an assisted delivery (forceps or vacuum). In the adjusted model, when

compared to infants that have no head positional preference, infants that have either a left or right sided head positional preference have more than 4 times the odds of developing positional plagiocephaly, with a slightly higher risk for those infants that have a right-sided preference.

Table 34: Adjusted Statistical Model of Risk Factors Predictive of Positional Plagiocephaly (n=435)

Variable	β.C.	S.E.	Wald	df	*p*	OR	95% CI
Sleep position	0.982	0.267	13.509	1	.000*	2.670	[1.582, 4.508]
Tummy time	-0.075	0.215	0.123	1	.726	0.927	[0.608, 1.414]
Birth order	-0.147	0.221	0.444	1	.505	0.863	[0.560, 1.330]
Infant sex	0.436	0.221	3.914	1	.048*	1.547	[1.004, 2.383]
Multiple	-0.325	0.566	0.329	1	.566	0.723	[0.238, 2.191]
Maternal education	-0.349	0.254	1.893	1	.169	0.705	[0.429, 1.160]
Delivery type (vacuum/forceps)	0.633	0.315	4.046	1	.044*	1.883	[1.016, 3.488]
Delivery type (c-section)	-0.229	0.247	0.865	1	.352	0.795	[0.490, 1.289]
Head positional preference (right)	1.539	0.248	38.593	1	.000*	4.662	[2.868, 7.577]
Head positional preference (left)	1.438	0.277	26.907	1	.000*	4.212	[2.446, 7.251]
Constant	-1.715	0.400	18.366	1	.000	.180	

Note. β.C. = beta regression coefficient; S.E = standard error; df = degrees of freedom; OR = odds ratio; CI = confidence interval.
*Significant at 0.05 level.

The summary of the adjusted model is presented in Table 35, indicating that the model with the ten variables is statistically significant. However, as indicated in Table 35, the model is not a great fit.

Table 35: Summary of the Adjusted Statistical Model (n=435)

χ^2	*p*	-2 Log likelihood	Cox and Snell R^2	Nagelkerke R^2
75.798	.000*	523.493	.160	.214

*Significant at 0.05 level.

The Hosmer-Lemeshow test displayed in Table 36 shows that the model is a good fit. This conflicting information was not unexpected given the exploratory nature of the research and differences in the level of rigour of the goodness of fit tests used.

Table 36: Contingency Table for Hosmer-Lemeshow Test

	No Plagiocephaly		Plagiocephaly					
Step	Observed	Expected	Observed	Expected	Total	χ^2	df	*p*
1	36	36.858	7	6.142	43			
2	39	37.127	9	10.873	48			
3	34	32.120	11	12.880	45			
4	26	28.648	18	15.352	44			
5	26	23.878	17	19.122	43			
6	20	19.887	23	23.113	43			
7	12	16.370	29	24.630	41			
8	16	15.172	27	27.828	43			
9	14	14.149	32	31.851	46			
10	10	8.792	28	29.208	38			
						8	4.298	.829

Note. df = degrees of freedom.

As demonstrated in Table 37, of all infants assessed for plagiocephaly, 68.2% of cases were classified correctly

Table 37: Percentage of Cases Classified Correctly

	Predicted Plagiocephaly		Percentage Correct
Observed Plagiocephaly	No	Yes	
No	167	66	71.7
Yes	64	137	68.2
Overall percentage			70.0

While the section above presents a statistical model of risk factors predictive of positional plagiocephaly, in the section below, another statistical model will be presented with the twins removed to demonstrate that the answers to the questions regarding maternal risk factors were not an issue in terms of independence, given that the entire sample consisted of only 8 pairs of twins.

Statistical Model of risk factors predictive of positional plagiocephaly (twins removed).

The unadjusted results of the simple logistic regression analyses of the data set with the twins removed are displayed in Table 38. According to these results, when compared to infants that did not sleep in the supine position, infants that slept supine had 2 times the odds of developing positional plagiocephaly. Maternal education at the post-secondary level serves as a protective factor for the development of positional plagiocephaly. When compared to infants that have no head positional preference, infants that prefer either the left or right side have 4 times the odds of developing positional plagiocephaly.

Table 38: Unadjusted Model of Risk Factors Predictive of Positional Plagiocephaly

Variable	β.C.	S.E.	Wald	df	*p*	OR	95% CI
Sleep position	0.699	0.241	8.399	1	.004*	2.012	[1.254, 3.229]
Tummy time	-0.053	0.197	0.073	1	.786	0.948	[0.645, 1.394]
Birth order	0.103	0.195	0.280	1	.597	1.109	[0.757, 1.624]
Infant sex	0.241	0.199	1.474	1	.225	1.273	[0.862, 1.879]
Maternal education	-0.465	0.227	4.180	1	.041*	0.628	[0.402, 0.981]
Delivery type (vacuum/forceps)	0.320	0.273	1.375	1	.241	1.377	[0.807, 2.349]
Delivery type (c-section)	-0.156	0.225	0.483	1	.487	0.855	[0.550, 1.329]
Head positional preference (right)	1.458	0.239	37.279	1	.000*	4.297	[2.691, 6.861]
Head positional preference (left)	1.483	0.267	30.789	1	.000*	4.408	[2.610, 7.443]

Note. β.C. = beta regression coefficient; S.E = standard error; df = degrees of freedom; OR = odds ratio; CI = confidence interval.
*Significant at 0.05 level.

The results of the multivariate logistic regression analysis are presented in Table 39. Nine variables were entered into the model simultaneously to produce the adjusted model: (a) infant sleep position, (b) infant tummy time, (c) birth order, (d) infant sex, (e) maternal education, (f) delivery type (vacuum/forceps), (g) delivery type (c-section), (h) infant head positional preference (right), and (i) infant head positional preference (left). As indicated in Table 39, when compared to infants that did not sleep supine, infants that sleep supine had 2.3 times the odds of developing positional plagiocephaly. Male infants are no longer at increased risk. When compared to infants that were delivered vaginally with no assistance, infants that had an assisted delivery (forceps or vacuum) had almost 2 times the odds of developing positional plagiocephaly. When compared to infants that did

not have a head positional preference, infants that had a right-sided head positional preference had 4.7 times the odds of developing positional plagiocephaly. Finally, when compared to infants that had no head positional preference, infants that had a left-sided preference had almost 4 times the odds of developing positional plagiocephaly.

Table 39: Adjusted Model of Risk Factors Predictive of Positional Plagiocephaly (n=424)

Variable	β.C.	S.E.	Wald	df	*p*	OR	95% CI
Sleep position	0.863	0.270	10.217	1	.001*	2.370	[1.396, 4.023]
Tummy time	-0.092	0.217	0.178	1	.673	0.912	[0.596, 1.396]
Birth order	-0.148	0.222	0.441	1	.507	0.863	[0.558, 1.334]
Infant sex	0.427	0.223	3.687	1	.055	1.533	[0.991, 2.371]
Maternal education	-0.345	0.250	1.907	1	.167	0.708	[0.434, 1.156]
Delivery type (vacuum/forceps)	0.648	0.314	4.251	1	.039*	1.911	[1.032, 3.538]
Delivery type (c-section)	-0.209	0.249	0.707	1	.401	0.811	[0.498, 1.321]
Head positional preference (right)	1.545	0.249	38.604	1	.000*	4.686	[2.879, 7.629]
Head positional preference (left)	1.377	0.277	24.623	1	.000*	3.961	[2.300, 6.823]
Constant	-1.816	0.440	17.052	1	.000	.163	

Note. β.C. = beta regression coefficient; S.E. = standard error; df = degrees of freedom; OR = odds ratio; CI = confidence interval.
*Significant at 0.05 level.

The summary of the adjusted model is presented in Table 40, indicating that the model with the nine variables is statistically significant. The results displayed in Table 40 also indicate that the model is not a great fit.

Table 40: Summary of the Adjusted Model (n=424)

χ^2	*p*	-2 Log likelihood	Cox and Snell R^2	Nagelkerke R^2
69.582	.000*	513.832	.152	.203

*Significant at 0.05 level.

However, the Hosmer-Lemeshow test presented in Table 41 shows that the model is a good fit. This conflicting information was not unexpected given the exploratory nature of the research and differences in the level of rigour of the goodness of fit tests used.

Table 41: Contingency Table for Hosmer-Lemeshow Test

	No Plagiocephaly		Plagiocephaly					
Step	Observed	Expected	Observed	Expected	Total	χ^2	df	*p*
1	35	35.354	7	6.646	42			
2	34	33.072	9	9.928	43			
3	33	31.460	11	12.540	44			
4	24	27.134	18	14.866	42			
5	28	22.874	14	19.126	42			
6	19	19.240	23	22.760	42			
7	11	17.175	32	25.825	43			
8	17	14.925	25	27.075	42			
9	10	11.632	27	25.368	37			
10	11	11.132	32	33.868	45			
						8	8.845	.356

Note. df = degrees of freedom.

Of all infants assessed for plagiocephaly, 69.7% of cases were classified correctly (Table 42). These results are very similar with those above, indicating that the presence of the eight sets of twins does not have a large impact in terms of the pieces of maternal information that are duplicated.

Table 42: Percentage of Cases Classified Correctly

	Predicted Plagiocephaly		Percentage Correct
Observed Plagiocephaly	No	Yes	
No	154	70	68.8
Yes	60	138	69.7
Overall percentage			69.2

While it is important to identify which factors put infants aged 7-12 weeks of age at increased risk of developing positional plagiocephaly, it is equally important to gain an understanding of what actions can be taken once plagiocephaly is identified. The following section will present the actions taken by PHNs at the 2-month well-child clinic once plagiocephaly was identified by their usual method of subjective assessment.

What Intervention and Follow-Up Actions do PHNs Take if Positional Plagiocephaly is Identified in Healthy Infants at the 2-Month Well-Child Clinic?

The PHN actions discussed in this section pertain to subjective assessments performed by each individual PHN. According to Alberta Health Services (2009), each PHN must complete a plagiocephaly assessment at the 2-month, 4-month and 6-month well-child clinic appointment. Nurses are instructed to assess for plagiocephaly by standing above and behind the infant and observe for asymmetry in the infant's head or facial features, or any asymmetrical range of motion when moving the head to either side (Alberta Health Services, 2009). After this assessment is completed, PHNs have four courses of action: (a) referral to the IRC, (b) referral to the family physician, (c) engage in repositioning teaching, and (d) follow-up at the 4-month well-child clinic. The data are presented in this section in terms of

the Research Nurses' plagiocephaly assessment performed using Argenta's (2004) plagiocephaly assessment tool for information purposes only.

Almost three-quarters of parent(s)/guardian(s) of infants in the study were given repositioning teaching during their well-child clinic visit (Table 43).

Table 43: Proportion of Infants Receiving Repositioning Teaching by PHN According to the Presence or Absence of Positional Plagiocephaly

		Positional Plagiocephaly		
		No	Yes	Total
Repositioning Teaching				
Yes				
	Frequency	136	180	316
	Percent	57.7	87.8	71.8
No				
	Frequency	99	25	124
	Percent	42.1	12.2	28.2
Total				
	Frequency	235	205	440
	Percent	100.0	100.0	100.0

As displayed in Table 44, of all infants assessed for positional plagiocephaly by a PHN, 93 infants were referred to the IRC at the Alberta Children's Hospital. The Research Nurses identified 86 of these 93 infants with positional plagiocephaly.

Table 44: Proportion of Infants Referred to the IRC by PHN According to the Presence or Absence of Positional Plagiocephaly

		Positional Plagiocephaly		
		No	Yes	Total
Referred to Infant Repositioning Class				
Yes				
	Frequency	7	86	93
	Percent	3.0	42.0	21.1
No				
	Frequency	228	119	347
	Percent	97.0	58.0	78.9
Total				
	Frequency	235	205	440
	Percent	100.0	100.0	100.0

As indicated in Table 45, 40 infants were referred to their family physician for further assessment. Of the 93 infants that were referred to the IRC, 38 were simultaneously referred to their family physicians. Two infants were referred only to their family physicians (Table 45).

Table 45: Proportion of Infants Referred to their Family Physician by PHN according to the Presence or Absence of Positional Plagiocephaly

		Positional Plagiocephaly		
		No	Yes	Total
Referred to Family Physician				
Yes				
	Frequency	4	36	40
	Percent	1.7	17.6	9.1
No				
	Frequency	231	169	400
	Percent	98.3	82.4	90.9
Total				
	Frequency	235	205	440
	Percent	100.0	100.0	100.0

PHNs' reasons for referral to either the infant repositioning class or to the family physician are provided in Table 46.

Table 46: PHN Reasons for Referral to IRC and/or Family Physician

Reason	Frequency	Percent	Cumulative Percent
No reason noted	43	9.8	45.3
OF	29	6.6	75.8
SP	2	0.5	77.9
ES and SP	1	0.2	78.9
ES and OF	8	1.8	87.4
OF and SP	1	0.2	88.4
OF, ES, and FB	6	1.4	94.7
OF and FB	2	0.5	96.8
Reinforce PHN teaching	2	0.5	98.9
Parental interest	1	0.2	100.0
Total	440	100.0	

Note. OF = occipital flattening; SP = sleeping preference; ES = ear shift; FB = forehead bossing.

PHNs noted that 64 infants will be followed-up with respect to plagiocephaly at the 4-month well-child clinic visit (Table 47).

Table 47: Proportion of Infants that will be followed up by PHN at 4-Month Well-Child Clinic According to the Presence or Absence of Positional Plagiocephaly

		Positional Plagiocephaly		
		No	Yes	Total
Follow-up Planned				
Yes				
	Frequency	13	51	64
	Percent	5.5	24.9	14.5
No				
	Frequency	222	154	376
	Percent	94.5	75.1	85.5
Total				
	Frequency	235	205	440
	Percent	100.0	100.0	100.0

Additional information provided by PHNs is offered in Table 48 based on the space provided on the data collection tool.

Table 48: PHN Comments Providing Additional Information About Individual Infant Plagiocephaly Assessments

Comment	Frequency	Percent	Cumulative Percent
None	223	50.7	50.7
No concerns	149	33.9	84.6
Prevention teaching	27	6.1	90.7
No concerns and prevention teaching	8	1.8	92.5
OF	13	3.0	95.5
Refused referrals to IRC and FP	5	1.1	96.6
Already referred to IRC by FP	6	1.4	98.0
Physiotherapy info given	3	0.7	98.7
Trouble with TT due to FB	1	0.2	98.9
Mom will come to drop in to check once a week	1	0.2	99.1
Unsure about IRC due to time	1	0.2	99.3
No FP	1	0.2	99.5
Mom will have FP assess	2	0.5	100
Total	440	100.0	

Note. OF = occipital flattening; IRC = infant repositioning class; FP = family physician TT = tummy time; FB = fussy baby.

In this section, the actions that PHNs can be taken once plagiocephaly is identified, is presented. One of these actions is to refer the infant to the IRC. The section below presents information on what actions are taken by clinicians (physiotherapists and occupational therapist) at the IRC once an infant is referred to the IRC by a PHN.

What Intervention and Follow-Up Actions do Clinicians (Physiotherapists and Occupational Therapists) Take When Healthy Infants with Positional Plagiocephaly are Referred to the Infant Repositioning Class (IRC) After the 2-Month Well-Child Clinic Visit in Calgary, Alberta, Canada?

Of the 93 infants that were referred to the IRC, only 19 attended (Table 49).

Table 49: Proportion of Infants with each Type of Plagiocephaly That Presented at the IRC According to IRC Clinician (Physiotherapists and Occupational Therapist) Assessment

Plagiocephaly	Frequency	Percent	Cumulative Percent
Type 1	4	21.1	21.1
Type 2	3	15.8	36.9
Type 3	10	52.6	89.5
Type 4	1	5.3	94.8
Brachycephaly only	1	5.3	100.0
Total	19	100.0	

Over half of these 19 infants presented with Type 3 plagiocephaly and over half presented with a mild form of plagiocephaly (Table 50).

Table 50: Severity of Plagiocephaly of Infants that presented at the IRC According to IRC Clinician (Physiotherapists and Occupational Therapist) Assessment

	Severity		
Plagiocephaly	Mild	Moderate	Total
Type 1	4	0	4
Type 2	3	0	3
Type 3	4	6	10
Type 4	0	1	1
Brachycephaly only	0	1	1
Total	11	8	19

In Table 51 the outcomes of the plagiocephaly assessment of these 19 infants between the CHC and the IRC are compared. All assessments used the same plagiocephaly assessment scale (Argenta, 2004). Eight infants were classified the same way by the researchers and the clinicians at the IRC. Eleven were classified differently. Five of these 11 infants had lower scores while six had higher scores when compared to the initial assessment that took place at the 2-month well-child clinic visit. None of the infants with plagiocephaly that presented at the IRC had signs of accompanying brachycephaly.

Table 51: Comparison of Assessment of infants assessed at both CHC and IRC

Infant	Plagiocephaly Type at CHC	Plagiocephaly Type at IRC
1	Type 1	Type 1
2	Type 2	Type 1
3	No Plagiocephaly	Type 1
4	Type 3	Type 3
5	Type 2	Type 3
6	Type 3	Type 2
7	Type 3	Type 3
8	Type 3	Type 3
9	Type 3	Type 4
10	Type 3	Type 3
11	Type 1	Type 2
12	Type 2	Type 1
13	Type 3	Type 1
14	Type 1	Type 2
15	Type 3	Type 3
16	Type 3	No Plagiocephaly
17	Type 3	Type 3
18	Type 3	Type 3
19	No Plagiocephaly	Type 3

As presented in Table 52, three infants were assessed to have torticollis. Clinicians also found that one infant that presented with Type 1 did not need to be referred to the IRC. Since all infants attended the IRC, they all received repositioning teaching.

Table 52: Frequency of Torticollis Assessed in Infants that presented at the IRC According to IRC Clinician (Physiotherapists and Occupational Therapist) Assessment

	Torticollis		
Plagiocephaly	Present	Absent	Total
Type 1	1	3	4
Type 2	1	2	3
Type 3	1	8	9
Type 4	0	1	1
Brachycephaly only	0	1	1
Missing			1
Total	3	15	19

No infant was referred for helmet treatment. Finally, as indicated in Table 53, three infants were referred for physiotherapy treatment.

Table 53: Frequency of Infants that presented at the HSC that were Referred for Physiotherapy by IRC Clinicians (Physiotherapists and Occupational Therapist)

	Physiotherapy		
Plagiocephaly	Yes	No	Total
Type 1	1	2	3
Type 2	0	3	3
Type 3	2	8	10
Type 4	0	1	1
Brachycephaly only	0	1	1
Missing			1
Total	3	15	19

The most significant piece of data obtained by this part of the research appears to be the difficulty accessing the IRC, as indicated by very low attendance. Changes in head shape over time were also observed. Unfortunately due to the low attendance observed at the IRC, sufficient data were not obtained to conduct a meaningful analysis of the results.

Conclusion

In conclusion, this study was able to estimate the incidence of positional plagiocephaly at 46.6% in infants aged 7-12 weeks. The majority of infants that presented with positional plagiocephaly were identified to have Type 1, a mild form of the condition. A larger portion of infants with plagiocephaly were affected on the right side than the left. This high incidence indicates that intervention in the form of parental education about how to prevent the development of positional plagiocephaly is warranted. In this study, seven non-modifiable risk factors (infant birth order, maternal delivery type, infant sex, multiple gestation, infant age, maternal age and maternal number of years lived in Canada) and six modifiable risk factors (infant sleep position, infant head positional preference, infant feeding position, infant tummy time, maternal education and maternal language barrier) were considered. During bivariate analyses to test for group differences, the following variables were found to be statistically significant in relation to the incidence of positional plagiocephaly: (a) maternal age ($p=.035$), (b) infant sleep position ($p=.001$), (c) infant head positional preference ($p=.000$), and (d) maternal education ($p=.033$). Simple logistic regression analysis revealed that: (a) when compared to infants that did not sleep in the supine position, infants that slept supine had 2.2 times the odds of developing positional plagiocephaly, (b) post secondary maternal level of education served as a protective factor, and (c) when compared to infants that had no head positional preference, infants that had either a left or right sided head positional preference had more than 4 times the odds of developing positional plagiocephaly. Multiple logistic regression analysis identified the following

risk factors: (a) right sided head positional preference (OR: 4.662; p=.000; CI 2.868–7.577), (b) left sided head positional preference (OR: 4.212; p=.000; CI 2.446–7.251), (c) supine sleep position (OR: 2.670; p = .000; CI: 1.582–4.508), (d) vacuum/forceps assisted delivery (OR: 1.883; p=.044; CI 1.016–3.488), and (e) male sex (OR: 1.547; p= .048; CI: 1.004–2.383).These results suggest that further intervention in terms of prevention education can be targeted to parents/guardians of those infants that are male, infants that have had an assisted delivery or both. Further studies are necessary in order to better understand the roles of PHNs around plagiocephaly assessments and intervention. In addition, further studies are also required in order to better understand the follow-up process of infants referred to the IRC and HSC for additional intervention. In the next chapter the results of this study will be discussed in depth as outlined by the five research questions.

CHAPTER FOUR: DISCUSSION

In this chapter the implications of the results of the analysis will be discussed, including for the incidence of positional plagiocephaly at the population level, and risk factors predictive of positional plagiocephaly. There will also be a discussion of the practical elements of follow-up after identification of positional plagiocephaly by PHNs and clinicians at the IRC at Alberta Children's Hospital, followed by an examination of the strengths and limitations of the study and suggestions for future research.

Incidence of Positional Plagiocephaly

The results of this study indicate that the incidence of positional plagiocephaly in infants that range from 7-12 weeks of age is 46.6% (n=440). Given that positional plagiocephaly is a condition that can be prevented, this high incidence rate is troubling. Simple techniques to prevent the development of positional plagiocephaly include: alternating the end of the crib a baby's head is placed at every night; and frequent position changes, including the early introduction of prone positioning while the baby is awake and supervised, to foster motor development and to prevent head shape concerns (B. Mikkelsen, personal communication, June 13, 2011; Dubé & Flake, 2003; Glasgow et al., 2007). However, it must be noted that no research evidence exists to support these prevention techniques.

Comparison with other incidence studies.

Four population-based incidence studies were found in the literature (Littlefield, Saba et al., 2004; Peitsch et al., 2002; Rubio et al., 2009; Stellwagen et al., 2008), none of which were Canadian. Littlefield, Saba et al. (2004) estimated the incidence of

plagiocephaly to be 15.2%. Peitsch et al. (2002) reported a similar incidence at 13.1%. Rubio et al. (2009) reported the incidence of plagiocephaly to be much lower at 3.1%, while Stellwagen et al., (2008) reported a much higher rate at 61.0%. In order to account for these differences in incidence, the results of the four studies identified above will be compared to the results of the present study. Three areas of focus will guide this comparison: (a) sampling and sample size, (b) age range of study sample, and (c) assessment techniques for plagiocephaly. The section on assessment techniques will end with a discussion about the criteria used to assess the merits of using Argenta's (2004) tool for assessing for positioning plagiocephaly.

Sampling and sample size.

Three groups of authors reported that their studies took place at a single centre (Littlefield, Saba et al., 2004; Peitsch et al., 2002; Rubio et al., 2009). Stellwagen et al. (2008) simply indicated that their study was hospital-based. What distinguishes the present study from past work is that it was designed to ensure the broadest representation of participants. While the studies identified above were undertaken at a single centre, the present study took place across four CHCs representing each quadrant of the city. Elwood (2007) indicates that for a prospective cohort study that has one outcome and the confounders are not fully known, it is important for the sample to be representative of the eligible population. Addressing the limitations of previous studies, the eligible population for the present study comprised four CHCs each located in one of the four quadrants of Calgary and the present study ensured representation from each CHC. As indicated in chapter three, the number of eligible infants whose parents/guardians agreed to

participate represents 27% of all eligible participants. This increases the likelihood that the results of the present study can be generalized to the eligible population.

Three of the four studies identified above (Littlefield, Saba et al., 2004; Peitsch et al., 2002; Stellwagen et al., 2008) had sample sizes smaller than that of the present study (n=342; n=183; and n=102, respectively). Sample size calculations were not provided by the authors listed above. As a result, it is unclear whether the results of their studies are accurate or dependable. The sample size calculation provided in chapter two indicates that in order for the results to be interpreted with 95% confidence, with a margin of error of plus or minus 5%, a sample of 384 study participants is required (Daniel, 1999; Food Safety Research and Response Network, 2007). The present study used a sample of 440 to calculate the incidence rate of positional plagiocephaly. In order to determine the accuracy of the incidence estimate of the present study provided above, the margin of error was calculated. The margin of error is the difference between the proportion of the population that is estimated to have plagiocephaly (N=15,520) and the proportion of the sample that is estimated to have plagiocephaly (n=440). According to Cochran (1977), the formula that connects *n* with the desired degree of precision is:

$$e = z[\sqrt{(N-n)/(N-1)}][\sqrt{(pq)/n}]$$

$$e = 1.96[\sqrt{(15{,}520 - 440)/15{,}519}][\sqrt{(0.5)(0.5)/440}]$$

$$e = (1.96)(0.98575)(0.02383)$$

$$e = 0.0460$$

Based on this calculation, the margin of error (the difference between the proportion of the population that is estimated to have plagiocephaly and the proportion of the sample

that is estimated to have plagiocephaly) is very small. Therefore, it can be said with confidence that the results of the present study, in terms of incidence, is accurate within plus or minus 5% 19 times out of 20 (at a 95% confidence interval). As a result, the estimate of incidence of plagiocephaly obtained in this study can be generalized to the population level (N=15, 520).

Age range of study participants.

Three of the comparison studies were undertaken during the newborn physical exam before discharge from the hospital, ≤ 72 hours after birth (Peitsch et al., 2002; Rubio et al., 2009; Stellwagen et al., 2008). However, as previously mentioned, van Vlimmeren and colleagues (2007) found that of the 23 infants that had plagiocephaly at birth, 14 reverted to normal by 7 weeks of age. In addition, while 75 of 380 infants in the van Vlimmeren study were found not to have plagiocephaly at birth, they presented with the condition at 7 weeks of age (van Vlimmeren et al., 2007). As a result of this evidence Bialocerkowski et al. (2008) assert, in their systematic review, that the term "positional plagiocephaly" should be used to describe infants older than 6 weeks of age with altered skull shape. Therefore, the incidence calculations provided by Peitsch et al. (2002), Rubio et al. (2009) and Stellwagen et al. (2008) may not be valid for population-level measures for the incidence of positional plagiocephaly. Littlefield, Saba et al. (2004) sampled infants 10 months and younger that attended routine well-child clinics. According to the Centers for Disease Control and Prevention (1999), there may have been a portion of infants that began their immunizations at 4 weeks of age in 1999, the year Littlefield, Saba et al. (2004) collected data. Since it is unclear what the lower age

limit was for the study by Littlefield, Saba et al. (2004) in terms of inclusion criteria, there may have been 4 week old infants attending the “routine well-child clinics in the sample. As a result, these infants may have been misclassified.

Porta (2008) defines incidence as the number of new health-related events in a defined population within a specified period of time (p. 124). Given the information provided by van Vlimmeren et al. (2007) regarding evolving head shapes in the first few weeks of life and the correct of the use of the term positional plagiocephaly to describe infants with altered skull shape that are older than 6 weeks of age (Bialocerkowski et al., 2008), it can be proposed that any incidence study that begins when the infants are younger than 6-7 weeks would not be a good measure of incidence at the population level. In comparison to the four incidence studies found in the literature (Littlefield, Saba et al., 2004; Peitsch et al., 2002; Rubio et al., 2009; Stellwagen et al., 2008), the present study addresses this issue by providing a more accurate measure for population level incidence of positional plagiocephaly in Calgary. The 2-month well-child clinic visit would be the first time that positional plagiocephaly could be identified by any PHN. Therefore, the ideal age for calculating incidence of plagiocephaly would be when the infant is 7-8 weeks of age. Currently, there is no evidence to support that the head shapes of infants with plagiocephaly at 7-8 weeks spontaneously reverted to normal at 9-12 weeks. As a result, infants that attend their first well-child clinic between the ages of 9-12 weeks, and have plagiocephaly, are also likely to have had some form of the condition at 7-8 weeks of age. Therefore, the inclusion criteria of the present study, in terms of age (infants that were 7-12 weeks), is fitting. The present study articulates clearly its

inclusion and exclusion criteria, including how multiple births, premature infants and older infants attending the 2-month well-child clinics were handled. In terms of infant age, the inclusion criteria for the present study were clear in identifying that only those infants attending the 2-month well-child clinic visits and whose ages range from 7-12 weeks would be eligible to participate in the study. Based on the above statements, the current study is rendered appropriately a study of the incidence of plagiocephaly. The present study will be useful in developing future comparative research.

Assessment techniques for positional plagiocephaly.

As discussed in chapter one, Littlefield, Saba et al. (2004) used set criteria to guide their assessments. Infants were identified as having no, mild, moderate or severe plagiocephaly. The four assessment items used to evaluate infants required clinicians to classify the type of plagiocephaly observed as: (a) mild (posterior asymmetry); (b) moderate (posterior asymmetry, ear malposition with a discrepancy of half an inch or more, and minimal frontal asymmetry and facial asymmetry); and (c) severe (significant posterior asymmetry, ear malposition of one inch or more, frontal asymmetry on the affected side, and facial asymmetry). Argenta's (2004) plagiocephaly assessment tool provides six items of assessment (posterior asymmetry, ear malposition, frontal asymmetry, facial asymmetry, temporal bossing and posterior vertical cranial growth) and instead of identifying the plagiocephaly as mild, moderate or severe, classifies the plagiocephaly into five different types that increase in severity. Furthermore, Littlefield, Saba et al. (2004) indicate that only infants with moderate and severe forms of plagiocephaly were included in their study, giving the reason that these are the forms

most commonly observed at specialty HSCs. It can be argued that the incidence of plagiocephaly provided by Littlefield, Saba et al. (2004) is likely to be an underestimation of what one may actually see at the population level. Although the present study used Argenta's (2004) plagiocephaly assessment tool, information pertaining to severity was also subjectively captured, based on the relative extent of skull malformation. The type of plagiocephaly that was identified was further classified subjectively as mild, moderate or severe, indicating that an infant with a severe form of Type 1 may also require intervention. This approach is significant because not only does the severity of plagiocephaly increase as the classification type increases in number, as found in the present study, there is also a range of severity that can be identified within each type of plagiocephaly.

Assessment techniques used by the three studies undertaken during the newborn physical exam varied. Each will be discussed in turn. In the study by Peitsch et al., (2002), one single person took anthropometric cranial measurements using the same spreading calliper. Two oblique cranial diameters were determined, measuring from the supraorbital point to the parietooccipital scalp at the point of maximal convexivity. The transcranial diameter difference (TDD) was then calculated. As discussed in chapter one, this method was subsequently tested for validity by Glasgow et al. (2007). Validity testing of the method was conducted by a craniofacial plastic surgeon that was blinded to the TDD score. The surgeon conducted the traditional subjective assessment for plagiocephaly, rating of the infants head from 0–4 (no plagiocephaly to severe plagiocephaly). Spearman's rank correlation coefficient was used to determine if there

was a statistically significant relationship between the TDD and the severity score resulting in a statistically significant positive correlation between the subjective score assigned by the craniofacial plastic surgeon and the TDD (Spearman's rank correlation = .61; $p < .0002$). However, this test for validity may in fact be invalid, since there is no accepted gold standard for assessing positional plagiocephaly. Using subjective assessments as the comparison assessment to test for validity poses an additional question: Since there are a variety of ways to subjectively assess for plagiocephaly, the question of Glasgow et al. (2007) decided which subjective assessment method was suitable for comparison in order to establish validity of the TDD method is unclear. In addition, one drawback to taking manual anthropometric measurements is that measurement error may be inherent in subjectively identifying craniofacial landmarks. This may produce systematic error and could result in the internal validity of the study being called into question. Furthermore, Mortenson and Steinbok (2006) did not find taking anthropometric measurements to be a reliable form of conducting plagiocephaly assessments.

Rubio et al. (2009) used subjective assessments, conducted by the same physician, to assess for positional plagiocephaly. Since both Argenta (2004) and Littlefield, Saba et al. (2004) indicate that positional plagiocephaly can be classified into three or more categories, both the reliability and validity of assessing positional plagiocephaly subjectively without a tool (as was done by Rubio et al., 2009) can be called into question. If plagiocephaly assessments are not conducted in a systematic fashion, it can be difficult to identify the level of plagiocephaly severity and therefore justify which

form of intervention is required (Losee & Mason, 2005). Using Argenta's (2004) plagiocephaly assessment tool, clinicians can conduct plagiocephaly assessments in a systematic fashion.

Stellwagen et al. (2008) chose to analyze photographs as their method of assessment. Two clinicians took photographs of infants' heads that were subjectively analyzed by one blinded investigator (Stellwagen et al., 2008). Although the use of photographs with corresponding computer analyses using a custom-written computer program has been proposed by other authors (Hutchison et al., 2005), Stellwagen et al. (2008) do not discuss the reliability of their proposed assessment method. Furthermore, funding and equipment for their study was provided by Cranial Technologies Inc., a U.S. based company that primarily manufactures helmets used to treat positional plagiocephaly, suggesting some conflict of interest. Recognizing the limitations of these approaches the present research sought to identify an appropriate plagiocephaly assessment tool by testing the usefulness of Argenta's (2004) plagiocephaly assessment tool.

Argenta's (2004) Plagiocephaly Assessment Tool.

The present study used Argenta's (2004) plagiocephaly assessment tool to capture the incidence of plagiocephaly of infants arriving at the 2-month well-child clinic. As indicated in chapter one, six criteria were used to determine the appropriateness of clinical observation tools for plagiocephaly assessments. As described in chapter two, four of the six criteria were used to provide rationale for selecting Argenta's (2004) plagiocephaly assessment tool to collect data for the present study: (a) cost-effectiveness,

(b) validity, (c) reliability, and (d) suitability/feasibility. During the data collection process, factors pertaining to the usability of Argenta's (2004) plagiocephaly assessment tool were also collected. This included: (a) the suitability/feasibility in terms of ease of use and time required, (b) the acceptability of the tool to the target population, (c) measures taken to ensure reliability, and (d) the yield of the tool. These will be discussed below.

Based on Argenta's (2004) assessment guidelines providing four examination positions (described in the methods), data were relatively easy to collect. Data were recorded in terms of which type of plagiocephaly was identified according to Argenta's (2004) plagiocephaly assessment tool. Although not part of the tool, data were also collected subjectively about the severity of plagiocephaly identified within each category, in terms of whether the observed plagiocephaly was mild, moderate or severe. This was based on the relative extent of skull malformation. Each plagiocephaly assessment took less than five minutes to complete and a few seconds to record. Given that no equipment was required to complete the assessments and that the assessments were done quickly, Argenta's (2004) assessment method is suitable and feasible for use by PHNs in the well-child clinic setting.

Argenta's (2004) plagiocephaly assessment method was acceptable to the target population, as demonstrated by the high response rate of 96.7%. This high response rate indicates the willingness of the target population to participate in the study, after they were informed of what the assessment would involve.

As indicated in chapter three, in order to ensure reliability was maintained in terms of the plagiocephaly assessments, various measures were taken. First, both Research Nurses spent eight or more hours conducting plagiocephaly assessments with clinicians at the HSC in order to gain awareness of the range of severity of plagiocephaly in Calgary. Second, for the first week of the study period, assessments were conducted together to ensure that the same classification, according to the five types of plagiocephaly in Argenta's (2004) plagiocephaly assessment tool, was obtained. Lastly, both Research Nurses consulted half way through data collection and again towards the end of data collection in order to ensure the plagiocephaly assessments were conducted in a consistent fashion. During this process, a few of the assessments were conducted together to ensure consistency. Both Research Nurses agreed on the assessment results for all infants that were assessed by both. The three measures taken above ensured that data were collected consistently throughout the data collection period. This approach enhanced the reliability of the plagiocephaly assessments. In turn this contributed to the internal validity of the study.

As described in chapter one, the yield of a test is defined as the ability of the tool to identify a previously unrecognized medical condition (Wilson & Jungner, 1968). Using Argenta's (2004) plagiocephaly assessment tool, 205 infants out of 440 infants were identified as having some form of plagiocephaly. Given that about half of the infants assessed were identified to have some form of positional plagiocephaly, it appears that Argenta's (2004) assessment method has the potential to produce a high yield.

However, given that the present study was not able to acquire PHN plagiocephaly assessments, it is difficult to quantify the yield.

Given that Argenta's (2004) plagiocephaly assessment tool met all of the criteria proposed in chapter one to assess the appropriateness of a plagiocephaly assessment tool in the primary care context, the use of this tool in well-child clinics is most suitable. Further studies are required in order to ascertain the validity of Argenta's (2004) plagiocephaly assessment method as a teaching tool for parents/guardians. The tool could assist parents/guardians to track progress or worsening of the condition after repositioning strategies are implemented in the home environment. Further studies are also required to assess the utility of the tool by family physicians.

Out of all of the assessment methods discussed in the literature review, Argenta's (2004) plagiocephaly assessment tool would be most appropriate for use in primary care. It is cost effective, highly reproducible and easy to understand by health care professionals and families (Argenta, 2004). In addition, Argenta (2004) presents six items of assessment that can then be used to classify plagiocephaly into five potential categories. The criteria to assess the appropriateness of plagiocephaly assessment tools, discussed in chapter two, demonstrate that Argenta's (2004) tool is more valid in terms of content than the criteria proposed by Littlefield, Saba et al. (2004). Using Argenta's (2004) plagiocephaly assessment tool, clinicians would be better equipped to gauge the severity of the condition, if present. This method assists clinicians in identifying what type of intervention, if any, is required for an individual infant.

However, Argenta's (2004) plagiocephaly assessment tool may not be appropriate for use in specialty HSCs that may require more sophisticated assessment methods to quantify degree of severity. In addition, other assessment methods may be more appropriate for use in specialty HSCs whereby progress of interventions can be tracked. Of course, further studies are required in order to identify which assessment method is best for use in specialty clinics.

In comparison with the other incidence studies of positional plagiocephaly found in the literature, the present study produced a multi-site sample and exceeds the optimal sample size in order for the study to be 95% confident in the results produced. As previously indicated, infants that attend the 2-month well-child clinic are in the age range of 7-12 weeks, and thus form an ideal sample for collecting incidence data. In addition, the incidence calculations provided by Peitsch et al., (2002), Rubio et al. (2009), and Stellwagen et al. (2008) may not be valid for population measures for the incidence of positional plagiocephaly. In addition, the present study used an assessment tool that met all of the inclusion criteria proposed for an appropriate assessment tool for use in primary care. As a result, it can be strongly argued that the present study is the most robust when compared to the four incidence studies presented in the literature (Littlefield, Saba et al., 2004; Peitsch et al., 2002; Rubio et al., 2009; Stellwagen et al., 2008).

While a standard method of measurement has yet to be adopted by clinicians working in this area (McGarry et al., 2008), it is important to ascertain which method is best for quick assessment in primary care, where competing priorities may be present. Taking anthropometric measurements and photographs may not be feasible in well child

clinics where it is necessary to obtain a valid and reliable assessment quickly. In addition, by using Argenta's (2004) plagiocephaly assessment tool, clinicians can conduct plagiocephaly assessments in a systematic fashion that can assist in determining the severity of the condition and the appropriate intervention required. Lastly, Argenta's (2004) plagiocephaly assessment tool is easy to use, has been demonstrated to be reliable by Spermon et al. (2008) and is easily understood by clinicians and family members.

Severity of Plagiocephaly Observed

The present study demonstrated that of all infants identified as having positional plagiocephaly, 58.0% of infants were classified as Type 1 (occipital flattening only). In addition, 77.1% of all infants assessed had a mild form of plagiocephaly when classified according to Argenta's (2004) classification scale. The type of plagiocephaly most identified (Type 1) is not surprising, given the previous definition identifying that the term positional plagiocephaly be reserved for infants 6 weeks or older (Bialocerkowski et al., 2008). However, as mentioned above, the proportion of infants identified as having plagiocephaly is troubling. Given that the majority of plagiocephaly identified in the present study was classified as Type 1, the present study confirms that plagiocephaly had just begun to develop in infants whose ages ranged from 7-12 weeks at the time of assessment. Given that milder forms of plagiocephaly were identified in the present study, less invasive intervention methods (i.e., counter-positioning) may be used if plagiocephaly is identified at the 2-month well-child clinic. Again this information is significant since repositioning/active counter-positioning techniques to reverse mild forms of plagiocephaly are most effective from birth to 4 months of age (B. Mikkelsen,

personal communication, June 13, 2011; Larsen, 2004; Losee & Mason, 2005; Persing et al., 2003). As previously mentioned, active counter-positioning is the simplest therapy to implement, is the least traumatic to the child, and has no related cost.

After the plagiocephaly assessment at the 2-month well-child clinic visit, if the PHN refers the family to IRC at the HSC, clinicians at the IRC will conduct an in-depth assessment including an assessment for torticollis. This in-depth assessment is important since PHNs in Calgary do not conduct torticollis assessments in well-child clinics. Similar to active counter positioning, Celayir (2000) identifies that torticollis is best treated with physiotherapy within the first 4 months of age. Celayir (2000) indicates that if torticollis is identified early and treated with physiotherapy such as passive stretching exercises before 4 months of age, the average length of treatment is 3.2 months. Passive stretching exercises include 3 stretches, lateral flexion and lateral rotation and anterior flexion–extension of the neck (Celayir, 2000). If torticollis persists after 6 months of physiotherapy, surgical intervention is likely to be indicated (Do, 2006). Therefore, in order for infants affected with plagiocephaly and torticollis to obtain less invasive intervention options, it is imperative for plagiocephaly to be identified early and referrals made as appropriate.

Accompanying Brachycephaly

Eight infants in the study (3.8%) were found to have signs of both plagiocephaly and brachycephaly. After a review of the literature, this is the first incidence study to capture information about infants that were identified to demonstrate characteristics of both conditions simultaneously. However, the results from the present study are similar to

the prevalence study undertaken by Hutchison et al. (2004) who found that 1% of their study population also had signs of both plagiocephaly and brachycephaly.

The Model

This prospective cohort study was designed to develop a statistical model that would identify risk factors predictive of positional plagiocephaly in infants aged 7-12 weeks. The results discussed here refer to the adjusted statistical model (n=435) wherein the eight sets of twins were included in the analyses. The model as a whole was found to be significant. The model consists of five significant variables and four non-significant variables.

Risk factors predictive of positional plagiocephaly.

Five variables that were found to be significant in the adjusted multivariate logistic regression model will be presented in this section: (a) supine sleep positioning, (b) sex, (c) delivery type, (d) head positional preference (right), and (e) head positional preference (left). These variables will be discussed followed by a comparison with the literature on the risk factors for positional plagiocephaly.

Supine sleep positioning.

The present study demonstrates that infants that sleep in the supine position had about 2.7 times the odds of developing positional plagiocephaly than infants that were not placed supine. Similar results were found with respect to supine sleep positioning in the literature (Glasgow et al., 2007; Hutchison et al., 2004). Given the recommendation of supine sleep position to reduce the incidence of SIDS (Government of Canada, CPS, CICH, & CFSID, 2011), supine sleep position is not viewed as a modifiable risk factor.

The importance in continuing to recommend that all healthy infants be placed supine (on their backs) to sleep in order to prevent SIDS cannot be overemphasized. The supine sleep position has been reported to reduce the risk of SIDS, and indeed deaths reported to be from SIDS decreased from 0.6 per 1000 in 1999 to 0.35 per 1000 in 2004 (Smylie & Sauve, 2009). As a result, although supine sleep positioning increases the risk of developing positional plagiocephaly, its benefit in terms of decreasing the risk of infant mortality far outweighs potential gains in reducing the incidence of positional plagiocephaly. In addition, reliable prevention techniques and treatment options exist to correct positional plagiocephaly. What parents need to be most aware of are the positioning techniques that need to be implemented at home in order to prevent the development of the condition.

Sex.

The present study found that sex is a risk factor with males at an increased risk of developing positional plagiocephaly. Male infants had 1.5 times the odds of developing positional plagiocephaly than females. In the present study, males were slightly over represented and comprised 59.3% of the sample. Male sex is a plausible risk factor that can be explained by the occurrence of larger male head circumferences. The larger and less flexible male head makes them more susceptible to compression in utero and deformational forces during delivery (Losee et al., 2007). Furthermore, the more rapidly growing male head (van Vlimmeren et al., 2007) may increase gravitational forces post delivery that contribute to the development of positional plagiocephaly, especially when a position of comfort has already been established.

Similar results were found in the literature. In their cross sectional study of 201 healthy newborns aged 24-72 hours, Peitsch et al. (2002) found male sex to be a significant risk factor for the development of positional plagiocephaly. The results presented by Rubio et al. (2009) are consistent with that of the present study. van Vlimmeren et al. (2007) followed 380 neonates and also found that sex was a significant risk factor for the development of positional plagiocephaly and that males were more likely to develop positional plagiocephaly both at birth and at 7 weeks of age.

In addition, two case control studies found that infants with positional plagiocephaly were more likely to be male (Hutchison et al., 2003; McKinney et al., 2009). Infants in the study by Hutchison et al. (2003) ranged in age from 2-12 months of age while the study by McKinney et al. (2009) required case infants to be identified as having positional plagiocephaly before 18 months of age.

Delivery type.

The present study compared unassisted vaginal deliveries with assisted deliveries (forceps and vacuum) and cesarean section deliveries. As indicated in chapter three, 30.2% of mothers had a cesarean delivery. This is higher than the national average rate of cesarean deliveries, which is 25.6 per 100 deliveries (Liu, Liston, & Lee, 2008). In addition, 16.6% of mothers in the present study had assisted deliveries including either forceps or vacuum. Again this is higher than the national average wherein 14.8% of deliveries included forceps or vacuum extraction of the infant (Liu, Young, & Liston, 2008). As indicated in chapter three, infants that had had an assisted delivery (forceps or vacuum) had 1.9 times the odds of developing positional plagiocephaly than infants that

were delivered vaginally with no assistance. Delivery type is also a plausible risk factor as the instruments used may compress the skull of the infant being delivered. Positional plagiocephaly may develop if the infant is continuously placed on the side of the head that had been compressed during assisted vaginal delivery.

Similar results were found in the literature. In their univariate analysis of 201 healthy newborns, delivery type (vacuum and forceps) was found to be a significant risk factor for the development of positional plagiocephaly 24-72 hours after birth (Peitsch et al., 2002). Likewise, Rubio et al. (2009) found that babies that had an instrumental delivery also had an increased risk of developing positional plagiocephaly.

Head positional preference and the side of head that plagiocephaly was observed.

The present study indicated that when an infant had a right-sided head positional preference (i.e., an infant that prefers to rest his/her head on the right side while supine, with his/her right side coming into contact with the surface on which he/she is laying) he/she had 4.7 times the odds of developing positional plagiocephaly than an infant that did not have a head positional preference. Similarly, if an infant has a left-sided head positional preference, he/she had 4.2 times the odds of developing positional plagiocephaly than an infant that did not have a head positional preference. In the present study, right-sided flattening was present in 63.8% of plagiocephaly cases whereas 36.2% were observed to have left-sided flattening. The right sided preference may result from events toward the latter period of pregnancy. During this period, the foetus turns and engages the birth canal, usually head down. Most often, the vertex of the head lies within

the birth canal with a left occipital anterior presentation (Losee et al., 2007). In this position, the infant's right occiput is compressed against the maternal pelvis and the left forehead against the lumbosacral spine (Losee et al., 2007; Peitsch et al., 2002). This position may initiate a process that may be perpetuated by postnatal supine sleep positioning, resulting in the preference to right sided head turning and allowing for a position of comfort to be established (Losee et al., 2007). It is this position of comfort that contributes to the compressive force on the right occiput, resulting in positional plagiocephaly. Other studies have also reported that occipital cranial flattening is more frequently observed on the right side (Hutchison et al., 2003; Losee et al., 2007; Peitsch et al., 2002).

Positional preference may develop as a result of not varying infant head positions when putting them down to sleep or offering feeding consistently from either the left or right side (Boere-Boonekamp & van der Linden-Kuiper, 2001). In addition, infants with plagiocephaly are significantly less likely to have their head positions varied when put down to sleep (Boere-Boonekamp & van der Linden-Kuiper, 2001; Glasgow et al., 2007; Hutchison et al., 2004; Hutchison et al., 2003; van Vlimmeren et al., 2007). Glasgow et al. (2007) found that varying infants' sleep position served as a protective factor and decreased the risk of deformational plagiocephaly. This evidence suggests that varying infants' head positions after they have been placed supine to sleep would contribute to preventing the development of positional plagiocephaly.

Non-significant variables.

The following variables were found to be not significant: (a) tummy time, (b) birth order, (c) multiple gestation pregnancy, and (d) maternal education. Each will be discussed in turn.

Tummy time.

The present study found that the amount of tummy time (prone positioning while awake) was not found to be a significant risk factor for the development of positional plagiocephaly. In their multivariate analysis, van Vlimmeren et al. (2007) obtained similar results that indicated that the amount of tummy time received was not found to be a risk factor for the development of positional plagiocephaly at birth or at 7 weeks. Glasgow et al. (2007) obtained comparable results from univariate analysis during their study of 192 infants at two primary care practices.

The use of tummy time as a prevention technique is plausible as the benefits would be two-fold: it would simply decrease the amount of time the infant sleeps supine, thereby decreasing prolonged periods of pressure on one area; and it assists the infant in developing head control and upper body strength, which promotes reaching important developmental milestones such as supine-to-prone rolling (Dubé & Flake, 2003). Interestingly, Hutchison et al. (2004) found that infants that had developed plagiocephaly by 6 weeks were found to also have had tummy time per day greater than 1 hour. This may have been due to the fact that parents/guardians of infants with plagiocephaly were more aware of it and hence attempting to alter the head shape by engaging in activities

such as tummy time to assist in changing their infant's head shape. However, this risk factor did not appear to be significant at 4 months of age.

The value of tummy time used as a technique to prevent positional plagiocephaly remains to be quantified. Currently, there are no guidelines in the literature about how much tummy time an infant should receive. Dubé & Flake (2003) identify that as soon as the umbilical cord falls off, infants should be placed in the prone position several times a day while awake and supervised, gradually increasing the length of time spent in the prone position. Future research is necessary to determine how much tummy time is appropriate for an infant to receive in the first few weeks of life. It is particularly important to show that the benefits of prone positioning while awake outweigh the potential risks of parents/guardians misinterpreting the advice. Parents/guardians may initiate prone positioning as a sleep position or increase the amount of time that their infants sleep in the prone position if they are already engaged in this type of sleep positioning (Glasgow et al., 2007). This may potentially increase the risk of SIDS in these infants.

Birth order and maternal age.

The present study did not find birth order to be a significant risk factor for the development of positional plagiocephaly. Similarly, Hutchison et al. (2003) did not find maternal age to be a significant risk factor for the development of positional plagiocephaly. It is plausible that birth order may be a function of maternal age. Rubio et al. (2009) considers this to be attributed to changes that occur in a woman's uterus with age. It can be conceived that the uterus becomes fibrous and less flexible with increasing

age, thereby placing less pressure on the infant during the birthing process. It is plausible that other factors may also impact uterine muscle tone including maternal height and weight, the size of the pelvic bone as well as the size of the infant in relation to the size of the pelvis. The relationship between these factors and that of the development of positional plagiocephaly warrants further study. This study looked at maternal age in order to capture similar information. Maternal age was found to be significant during bivariate analysis. However, it was also discovered to be multicollinear with birth order and maternal education and therefore was removed from the multivariate analysis.

Multiple gestation pregnancy.

The present study did not find multiple gestation pregnancy to be a risk factor predictive of the development of positional plagiocephaly. It must be noted that multiple gestation pregnancies were under represented in the study sample, comprising only 17 infants and 8 pairs. Similar results were found also in the literature with the proportion of multiples in relation to singletons being very small (Hutchison et al., 2003; Rubio et al., 2009; van Vlimmeren et al., 2007). Therefore additional studies are required to examine multiple gestation pregnancy as a risk factor.

The restrictive intrauterine environment has been postulated to contribute to the development of positional plagiocephaly in infants that are products of multiple gestation pregnancies (Littlefield et al., 2002). More specifically, Littlefield et al. (2002) found that it was the position and orientation in utero that were the main conditions during pregnancy that appear to increase one twin's risk of developing positional plagiocephaly. One plausible explanation that has been proposed is that the lower in utero infant is

commonly reported to become engaged in the maternal pelvis. This infant habitually bears the weight of the second infant, thereby decreasing the opportunity to change orientation due to less mobility (Littlefield et al., 2002). This positioning is most notable during the last several weeks of pregnancy. The inability to change orientation may predispose the lower in utero infant to develop positional plagiocephaly by allowing the head to become deformed prenatally by the amount of contact time with the maternal pelvis and spine. In addition, the lower in utero infant may also be predisposed to developing CMT or other neck dysfunction as a result of the inability to change orientation (Littlefield et al., 2002).

Maternal education.

As indicated in chapter three, the simple logistic regression analysis indicated that maternal education was a significant protective factor with decreased risk to infants whose mothers had post secondary education. However, this did not hold true in the multivariate logistic regression analysis. What this result indicates is that in the presence of other variables, maternal education is no longer significant. Similar results to the present study were found by van Vlimmeren et al. (2007). In addition, in their multivariate analysis, Hutchison et al., (2003) found that maternal education was significant with an increased risk for the group with no or low qualifications. It is plausible for maternal education to be a risk factor for the development of positional plagiocephaly. Lower levels of maternal education have been found to be a risk factor for poor birth outcomes in the Canadian context (Luo, Wilkins, Kramer & The Fetal and Infant Health Study Group of the Canadian Perinatal Surveillance System, 2006).

Education is one of the most powerful determinants of health. Education is closely linked to health literacy, which improves people's ability to access and understand information to help keep them healthy (Public Health Agency of Canada, 2003).

Contradictions Found in the Literature

Conflicting information about sex as a risk factor for positional plagiocephaly is also found in the literature. In their prospective cohort study of 200 infants, Hutchison et al. (2004) did not find sex to be a significant risk factor at 6 weeks and at 4 months. Males were also over represented in their sample at 53%. Similarly, 70% of the 105 infants assessed in the study by Losee et al. (2007) were male, although sex was not identified as a risk factor. Differences observed in conflicting information about sex as a risk factor in the literature may be attributed to differences in sampling techniques and the relatively small sample of these studies.

As with the other risk factors presented in this section, differing information was also found in the literature with respect to delivery type. Various studies did not find a significant relationship between delivery type and the occurrence of positional plagiocephaly (Kane et al., 1996; Hutchison et al., 2003; Hutchison et al., 2004; van Vlimmeren et al., 2007). This discrepancy may be due to differing study designs and sample size considerations.

Hutchison et al. (2003) found tummy time was a significant risk factor for the development of positional plagiocephaly. Infants identified as having plagiocephaly were found to have had less than 5 minutes of tummy time per day during the first six weeks of life. This is the only study demonstrating the effectiveness of promoting tummy time as a

technique in preventing positional plagiocephaly at home in healthy term infants. Differences in study results may be due to variations in study designs. The study by Hutchison et al. (2003) is a case control study whereas the studies mentioned above that did not find tummy time to be a significant risk factor were cohort studies. Furthermore, the sample in the study by Hutchison et al. (2003) consisted of infants whose ages ranged from 2-12 months and whose mothers were asked to recall information pertaining to when their infant was 6 weeks of age, potentially introducing recall bias on the part of case parents.

Many studies have identified parity as a risk factor, with an increased risk for firstborn infants (Hutchison et al., 2003; Hutchison et al., 2004; Rubio et al., 2009; van Vlimmeren et al., 2007). This discrepancy may be due to differing study designs and sample size considerations. Conversely, McKinney et al. (2009) indicate that mothers of case infants were more likely to have been ≥35 years old at the time of the infant's birth. However, the inclusion criteria of infants in the study by McKinney et al. (2009) appear to be quite broad, with significant risk factors identified to be associated with the sequelae of pre-term birth and/or congenital anomalies. Due to the broad nature of inclusion criteria in the study by McKinney et al. (2009), the risk factors identified, as they pertain to healthy term infants, cannot be ascertained.

Five studies identified multiple gestation pregnancies as a significant risk factor for the development of positional plagiocephaly (Kane et al., 1996; Littlefield et al., 1999; Littlefield et al., 2002; Losee et al., 2007; McKinney et al., 2009). Two of these studies were designed specifically to compare the risk of developing positional

plagiocephaly if the infant was part of a multiple gestation pregnancy (Littlefield et al., 1999; Littlefield et al., 2002).

The present study provides additional support for the associations identified above: (a) supine sleep position, (b) male sex, (c) assisted delivery, (d) right-sided infant head positional preference, and (e) left-sided infant head positional preference. Although the model presented was found to be statistically significant, conflicting data regarding the goodness of fit of the model was obtained. Since this study was exploratory in nature, and the first study undertaken in the Canadian context to identify risk factors predictive of the development of positional plagiocephaly, this result is not surprising. Undertaking further studies regarding the risk factors for the development of positional plagiocephaly at the population level may assist in identifying additional risk factors. Future research may produce models that have a better fit than that of the present study.

Implications for Practice Based on Incidence and Risk Factor Data

Given that plagiocephaly is a preventable condition, the high incidence (46.6%) is very troubling. This section will focus on what can be done to decrease this high incidence. The majority of the discussion will focus on head positional preference as a risk factor, as this is the only modifiable risk factor that was identified in the present study.

It appears that the information received by parents regarding varying their infant's head position consistently every time they put him/her to sleep is received for the first time at the 2-month well-child clinic visit. This lack of prior knowledge to vary head positions was demonstrated by the present study, which indicates that 58% of all infants

demonstrated either a left-sided or right-sided head positional preference. In addition, the present study identified head positional preference as a risk factor for the development of positional plagiocephaly during both simple and multiple logistic regression. This study demonstrates that the information to vary infants' head positions when putting them to sleep needs to be communicated to parents/guardians well before the 2-month well-child clinic visit. In addition, because identified risk factors for the development of positional plagiocephaly include assisted delivery and male sex, parents/guardians of male infants or those infants that have had assisted deliveries could be made aware of this increased risk and the appropriate prevention teaching could be offered.

Various avenues for communicating prevention techniques for plagiocephaly exist across different groups of health professionals. Registered Nurses are in frequent contact with their patients/clients and, as a result, have more opportunities for health promotion than any other health professional group. It would be ideal if parents/guardians could receive this information first from Registered Nurses involved in delivering prenatal classes. Post partum nurses could then reinforce this information before discharge from the hospital and PHNs involved in postpartum care could also explain the importance of such prevention activities and demonstrate how this can be done in the home environment. It would be ideal if information pertaining to the prevention of the development of a head positional preference could also be delivered by family physicians and paediatricians that see infants during first few weeks of life, when appropriate. Since every new mother receives the Alberta Health Services (2010) publication entitled *From Here through Maternity* during pregnancy it would also be useful to include information

about the importance of varying infants' head positions along with the "back to sleep" information. This publication currently indicates only to place infants supine to sleep to avoid SIDS with no mention of varying head positions of infants from left to right and vice versa when they are placed supine to sleep to prevent the development of positional plagiocephaly.

If the process of communicating prevention techniques were carried out as recommended by the present study, then by the time the infant and parent(s)/guardian(s) arrive at the 2-month well-child clinic visit, they would have received the messaging about varying the infant's head position. This process would open opportunities for PHNs to reinforce teaching that has already occurred or refer the infant to the IRC if required. These opportunities could include PHNs during the 2-month appointment focussing on strategies to vary head position for parents/guardians that are having difficulty with repositioning due to the development of a head positional preference. As a result, referrals to IRC would likely result in clinicians there focussing more on torticollis/SCM imbalance assessments and interventions. With multiple health care professional providing the same information, parent(s)/guardian(s) may be more likely to implement the strategy to vary infants head positions when placing them supine to sleep.

One method to deliver the same health information to a target population would be via social marketing campaigns (Grier & Bryant, 2005). As plagiocephaly is not a life-threatening condition, and because the long term consequences in terms of developmental delays has not been demonstrated, conducting a cost-benefit analysis of implementing a

social marketing campaign, which delivers various messages about how to prevent the development of positional plagiocephaly, is recommended.

PHN Actions and Follow-up by the IRC at Alberta Children's Hospital

The secondary research questions of the study were to understand follow-up action by PHNs and clinicians at the IRC. Because the study method that was proposed in terms of PHN assessment could not be applied, data could not be obtained to fully understand PHN actions as they relate to plagiocephaly assessments. Hence the results obtained are not as robust as they might have been. Nevertheless, they offer a certain degree of insight to develop future research.

With respect to positional plagiocephaly, PHNs in Calgary generally have four courses of action: (a) repositioning teaching, (b) referring an infant to the IRC at the Alberta Children's Hospital, (c) referring an infant to the family physician for further assessment, and (d) follow-up at the 4-month well-child clinic visit. Each of these actions will be discussed in turn and implications related to follow-up at the IRC will be integrated.

Repositioning teaching.

As indicated in chapter three, almost three-quarters of parent(s)/guardian(s) of infants in the study were given repositioning teaching during their well-child clinic visit. This may be an underestimation of the repositioning teaching that actually occurs as the PHNs may have misunderstood the data collection tool. It may have been unclear to the PHNs whether the tool was designed to collect information about routine prevention teaching that was offered to prevent the development of positional plagiocephaly or if

additional repositioning teaching was provided if plagiocephaly was identified. This was evident in the comments section wherein some PHNs identified the reason behind the repositioning teaching. Other nurses may not have indicated if prevention teaching was offered if this was a routine part of their practice. To clarify, prevention teaching may not have been fully captured if PHNs were under the impression that data collection was meant only to capture the repositioning teaching that was done if plagiocephaly was identified. Clarifying the data collection tool would help capture this information more fully in future studies.

Referral to the IRC.

As indicated in chapter three, 93 infants were referred to the IRC. This number represents 21% of all infants that participated in the study. This information provides corroboration for the recommendations above to include all health care professionals with whom the parent(s)/guardian(s) and infants come into contact prior to arriving at the 2-month well-child clinic visit. These health care professionals can engage in prevention teaching around the development of a head positional preference that may lead to the development of positional plagiocephaly. Unfortunately, only 19 of the 93 infants referred actually attended a repositioning class. This low attendance rate may have been due to the fact that referral to the IRC is not a formal process, as it is with other specialty clinic referrals, where appointments are made only by physicians' offices. When referred to the IRC, parent(s)/guardian(s) are presented with a phone number to call and make an appointment themselves. The fact that only a small percentage of study infants were able to make their way to the IRC generates questions about the efficacy of introducing

another level of health system that parent(s)/guardian(s) are required to navigate in order to obtain information. Part of the information received at the IRC could easily be delivered by other health care professionals with whom they would already be in contact. Altering the mechanism of information delivery whereby information on prevention is delivered by health care professionals with whom most parents come into contact with (i.e. postpartum nurses, PHNs, or family physicians), would enable parent(s)/guardian(s) to obtain the information they require to promote the health of their infant in a timely and efficient manner. Other reasons for low attendance could be obstacles faced by parent(s)/guardian(s). These could include transportation and child care challenges and time constraints.

Another reason for the data gathered on low follow-up at IRC could be due to the study design. Parents of infants that were referred to the IRC were given a copy of the data collection tool indicating the results of the assessment that had taken place and the PHN action. Ethical approval was not obtained for the Research Nurses to share this information directly with the clinicians at the IRC. Therefore, parents that arrived at the IRC, but who may have forgotten to bring and/or present the data collection tool they were asked to pass on, were lost to follow-up. This may, in part, explain the small number of study participants that were part of the follow-up.

As presented in chapter 3, there were differences in head shape assessments in 11 of 19 infants between the initial and clinic assessment. Six infants scored higher on Argenta's (2004) plagiocephaly assessment tool at the IRC when compared to the initial assessment at the 2-month well-child clinic visit. Reasons for this are two-fold. First,

infants with plagiocephaly that have a head positional preference and continue to be placed supine to sleep in the same position would be likely to present with an increasingly severe form of plagiocephaly over time. The date of the final assessments were not recorded therefore the time span between the initial and final assessments were unclear. Second, the assessment of positional plagiocephaly must be placed within the context of the population with which the Research Nurses and PHNs were working versus the population with which the IRC clinicians are working. It is important to note that the assessments were likely made with subjective reference to the type of population with which a particular health care professional is used to working. For instance, the Research Nurses were more likely to see infants with milder forms of plagiocephaly, whereas the clinicians, as part of their daily practice, are more likely to see infants that have moderate to severe forms of plagiocephaly. Therefore, assessments made would reflect this and may explain some of the variations observed. This may also have implications for the reliability of the tool. Future studies are necessary to determine the differences in results of assessments obtained between PHNs and clinicians working at specialty HSCs.

Five infants had lower scores on Argenta's (2004) plagiocephaly assessment tool at the IRC when compared to the initial assessment at the 2-month well-child clinic visit. This may be a result of parent(s)/guardian(s) heeding the recommendations regarding repositioning received by the PHNs at the initial 2-month well-child clinic visit. As noted above, it is important to note that the exact length of time between the initial clinic visit and the visit to the IRC as this may also be a factor in observed differences in

assessments made. In future studies, it would be important to note this exact length of time in order to gain a better understanding of the role that time plays in changes in head shape that may occur. This information also signifies to the importance of ensuring inter-rater reliability of the tool with PHNs and clinicians at the IRC/HSC. This study result is in line with the literature that indicates that an infant's head shape evolves over time (Hutchison et al., 2004; van Vlimmeren et al., 2007).

Referral to the family physician.

Forty infants were referred to their family physician for further assessment for positional plagiocephaly in the present study. If a referred infant is under 4 months of age, the family physician's course of action would be to engage in repositioning teaching and/or refer the infant to the IRC (Appendix G; L. Walker, personal communication, May 7, 2008). Thirty eight of these 40 infants were concurrently referred to the IRC. This process is in line with current protocols that indicate that both PHNs and family physicians can refer infants up to 4 months of age to the IRC (Appendix G; B. Mikkelsen, personal communication, June 13, 2011). Research is required to determine whether referral from both types of health care professionals increases attendance at the IRC. Research is also required to answer the following question: Do the uptake of recommendations vary if they are provided by a PHN or a family physician? Furthermore, studies are also required to determine the outcome of infants whose parent(s)/guardian(s) receive conflicting information from PHNs and physicians regarding the necessity of either repositioning teaching after plagiocephaly onset or helmet treatment.

Follow-up planned at the 4-month well-child clinic visit.

As presented in chapter three, 64 infants (14.5%) were tagged for follow-up at the 4-month well-child clinic visit. This number may be an underestimation of the follow-up that actually occurs as the PHNs may have misunderstood the data collection tool. The tool was unclear about whether it was designed to collect information about routine follow-up for all infants or if the follow-up pertained to infants identified as having positional plagiocephaly only. PHNs in Calgary routinely conduct plagiocephaly assessments at the 2 month, 4 month and 6 month well-child visits (Alberta Health Services, 2009). Therefore, it may be appropriate to conclude that all infants would be followed-up and reassessed regardless of whether or not they were identified as having positional plagiocephaly at the 2-month clinic appointment.

Strengths of the Present Study

The strengths attributed to this study will be presented and discussed in this section. Strengths will be discussed in terms of: (a) methods, (b) tools, and (c) data collection.

Methods.

The first strength of this study was the support that was garnered for the involvement of four CHCs as data collection sites. The four CHCs were purposively chosen based on their ability to participate as well as their geographic locations, which represented the four quadrants of the city.

Secondly, the Research Nurses were able to secure the participation of 440 infants and their parent(s)/guardian(s). Since the sample size calculations indicate that in order

for the results to be interpreted with 95% confidence, with a margin of error of plus or minus 5%, a sample of 384 study participants is required. Therefore, due to the fact that 440 infants were recruited for the present study, this relatively large sample size, allows the incidence estimate to be generalized with 95% confidence to the source populations, the four CHCs, with a margin of error of 4.6%.

Thirdly, the inclusion criteria for participation in the study were in line with current definitions of positional plagiocephaly; that it refers to infants older than 6 weeks of age (Bialocerkowski et al., 2008). Since the infants that attend the 2-month well-child clinic visit would be the closest in age, this is where it was agreed data collection would take place. As a result, setting the lower limit of the inclusion criteria in terms of age at 7 weeks decreased the chance of misclassifying a younger infant that may have presented with plagiocephaly at birth, but whose head shape may have normalized within the first few weeks of life, as demonstrated by van Vlimmeren et al. (2007). Additionally, based on the inclusion criteria, specifically conducting the study at the 2-month well-child clinic visit, other researchers will be able to replicate the study.

Fourthly, two Research Nurses completed all of the plagiocephaly assessments, serving as a major strength of the study in terms of maintaining consistency in how the assessments were conducted. Using two experienced health care professional as the individuals responsible for completing the assessments may have decreased information bias in the way the assessments were made, thereby increasing the internal validity of the study (Elwood, 2007).

Tool.

The fifth strength of this study was that Argenta's (2004) plagiocephaly assessment tool was used to identify and classify infants with plagiocephaly that presented at the 2-month well-child clinic visit. Using an assessment tool may have decreased the potential for observation bias, since observation bias is less likely when objective assessment methods are used (Elwood, 2007).

Data collection.

The sixth strength of the study was that the parent(s)/guardian(s) of all infants that met the inclusion criteria were approached to participate in the study during the time frame of data collection at all four CHCs. This served to decrease selection bias (Elwood, 2007). This also improved the external validity of the study as there was no difference in who was approached and chose to participate or chose not to participate. This process ensured an equal opportunity for all infants that met the inclusion criteria to participate in the study, regardless of whether they had plagiocephaly or not. This lack of selection bias is also reflected in the response rate of 96.7%.

Limitations of the Present Study

The limitations attributed to this study will be presented and discussed in this section. Limitations will be discussed in terms of: (a) methods, (b) data collection, and (c) data analysis.

Methods.

One limitation of this study was that although support for data collection was garnered across four CHCs, there were difficulties in setting up data collection schedules

such that equal representation from all four sites could be obtained. The majority of study participants were recruited from Clinics 2 and 3, which were located in more affluent neighbourhoods. Furthermore, Clinic 4 served higher proportions of visible minority groups, which may have affected the external validity of the study and the incidence of plagiocephaly identified in the study may have been an underestimation. Indeed, it has been widely recognized that socially disadvantaged people experience higher rates of illness, disability and death (Public Health Agency of Canada, 2003). Lower levels of neighbourhood income have been found to be a risk factor for poor birth outcomes in the Canadian context (Luo et al., 2006).

Data collection.

Due to difficulties in negotiation of site access, data collection spanned 3 months. The fact that data collection spanned only 3 months may affect the external validity of the study. Incidence studies generally collect data over the course of one year in order to ensure stability of the data. To have increased confidence in the results of the present study, it is recommended that future incidence studies on positional plagiocephaly span one year.

One significant limitation of this study was that torticollis assessments were not completed during data collection. This again was due to the access issue, whereby difficulties were faced in obtaining adequate amount of time available for data collection with each study participant and space for data collection (i.e., private room with a flat surface on which to examine for torticollis). This information would have provided insight into the proportion of infants with true torticollis versus SCM imbalance (Golden

et al., 1999). Its impact could then be assessed in terms of a risk factor for the development of positional plagiocephaly for this study. Since torticollis is a common anomaly that has been linked with plagiocephaly, if this information had been captured, this study would have been able to ascertain the rate of plagiocephaly-torticollis co-diagnosis (Clarren, 1981; Golden et al., 1999; Pivar & Scheuerle, 2006; Rogers et al., 2009; van Vlimmeren, Helders, et al., 2006). This information would have provided insight into the benefits and drawbacks of introducing torticollis assessments into the well-child clinic visits. As previously mentioned, PHNs do not generally engage in torticollis assessments.

Another limitation of the study was that specific data were not collected to calculate the *kappa* – the degree of nonrandom agreement between the assessments (Porta, 2008). In the future, in order to ensure the reliability of the plagiocephaly assessments that were conducted by the two Research Nurses, data would need to be collected in order to calculate the kappa. It would also be useful to do the same between the Research Nurses and the clinicians conducting assessments with the same tool to ascertain the degree of similarity or difference in the assessments.

Data analysis.

Data collection in terms of the lack of PHN disclosure about their subjective plagiocephaly assessments served as a limitation. Since it was the Research Nurses that completed the assessments, and PHNs did not disclose their assessments, the PHN action could not be linked to the specific type of plagiocephaly with which the infant was identified. If the PHN assessment and PHN action could be linked, this could have

provided richer information. Again, due to difficulties in negotiating access, PHNs were not permitted to conduct the assessments using Argenta's (2004) plagiocephaly assessment tool or disclose the results of their subjective assessments. If PHNs' subjective assessments had been reported, they could have been compared with the Research Nurses' assessments, thus providing more insight into the number of infants identified as having plagiocephaly with the two assessment methods.

Directions for Future Research

The present study is the first of its kind in Canada to: estimate the incidence of positional plagiocephaly; identify potential risk factors for the development of positional plagiocephaly; identify PHN actions when an infant is identified as having the condition at the 2-month well-child clinic visit; and track the follow-up of infants that are referred to specialty HSCs. Future studies are required to corroborate the findings of the present study. Research is required to assess the incidence of plagiocephaly using Argenta's (2004) plagiocephaly assessment tool across more CHCs and at different infant age groups. A longitudinal study could be completed at the various well-child clinic visits (2 month, 4 month, and 6 month) and could provide useful information about changes in incidence and prevalence over time, and across various age ranges. In addition, credibility for using Argenta's (2004) plagiocephaly assessment scale in well-child clinic visits could be established. Similar studies are also required that identify risk factors as they change with differing infant age groups. The use of Argenta's (2004) plagiocephaly assessment tool as a teaching tool for parents to track progress or worsening of the

condition after repositioning strategies are implemented in the home environment needs to be established.

Further studies are also required to understand referral to the IRC by PHNs and follow-up by PHNs at subsequent well-child clinic visits. Qualitative studies are also required to ascertain reasons behind the surprisingly low attendance rate at the IRC at the Alberta Children's Hospital and follow-up by clinicians at the IRC and HSC. In addition, the professional roles of PHNs, family physicians and other clinicians working with infants with plagiocephaly need to be understood more clearly.

Studies are required to understand how parents view head shape in the broader context of the issues faced in the first year of life. It would also be useful to capture information on the impact of early intervention, in terms of repositioning teaching.

Conclusion

This was the first study undertaken in the Canadian context that estimated the incidence of positional plagiocephaly in infants ranging from 7-12 weeks in age. The estimated incidence of positional plagiocephaly was found to be 46.6%. This study was able to identify the following risk factors that increased the risk of developing positional plagiocephaly: supine sleep positioning, male sex, assisted delivery (forceps, vacuum), and either a left or right head positional preference. This study also demonstrated that parents of infants referred to the IRC may not access this service. Knowledge of the risk factors identified in this study, and of low rates of attendance to the IRC, will assist health professionals to anticipate which infants may be at higher risk of developing positional plagiocephaly. Support for implementing interventions aimed at decreasing

this risk, such as the importance of prevention teaching by a variety of health professionals during the neonatal period and before the infant arrives at the 2-month well-child clinic visit, needs to be garnered.

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APPENDIX B

CONSENT FORM

TITLE: Plagiocephaly in Calgary, Alberta, Canada: Incidence, Public Health Nursing roles, and follow-up by the Head Shape Clinic, Alberta Children's Hospital

INVESTIGATOR: Dr. Ardene Robinson Vollman
This consent form is only part of the process of informed consent. It should give you the basic idea of what the data collection process is about, and what your baby's participation will involve. If you would like more detail about something mentioned here, or information not included here, please ask. Take the time to read this carefully. You will receive a copy of this form.

BACKGROUND AND PURPOSE
As a result of putting babies to sleep on their backs to prevent SIDS, some babies develop flat heads. Little is known in Canada about the occurrence of flat heads and what is done about it. This research will help calculate how many babies develop flat heads in Calgary as well as if there are any factors that help predict which babies develop flat heads and which do not. The information gathered will be used to inform future public health nurse practice around how they identify babies with flat heads and how this is managed. It is important to find out how many babies develop flat heads so that appropriate resources can be allocated to help these babies.

WHAT WOULD MY CHILD HAVE TO DO?
If you choose to participate in the study, a research nurse will look at your baby's head shape and record the information at his/her first immunization clinic visit. This may take place either before or after your baby's immunization. You will also be asked to fill out a survey that asks some questions about your age and highest level of school you finished, how you feed your child, how much "tummy time" your child receives and some questions about your pregnancy. If the research nurse thinks your child has a flat head,

she may give you suggestions or refer you to a repositioning class or your family doctor. If your family doctor thinks your child needs more care, he or she will refer to the Head Shape Clinic at the Alberta Children's Hospital. You will be given a form that the nurse fills out to take with you to your doctor, the repositioning class and/or the Head Shape Clinic. If you do not wish to participate in the study and the nurse thinks your child has a flat head, you may still be referred to the repositioning class or your family doctor who may refer you to the Head Shape Clinic.

WHAT ARE THE RISKS?

There are no known risks to your child by participating in the study.

ARE THERE ANY BENEFITS FOR MY CHILD?

If your child has a flat head, participating in this study will help to better diagnose the extent of the flat head. Participating in the study will also help you to understand the issue of flat heads. Your participation will also help health professionals to understand babies with flat heads better and how to prevent it. In the future, this study will help to improve how children are referred for treatment.

DOES MY CHILD HAVE TO PARTICIPATE?

If you or your child is uncomfortable with the assessment, you are free to interrupt and withdraw your child from the assessment. If you choose not to participate in the study, it will in no way affect other care your child may receive at the clinic.

WILL MY CHILDS RECORDS BE KEPT PRIVATE?

We will assign a number to the data collection form. Any information collected will not be linked to you or your child. Any data collected will be coded and entered in a data base which will be used for statistical analysis of grouped data. Database information will be treated as confidential and anonymous.

The information from the database will be summarized in a report which will be given to public health nurses and the clinicians at the Head Shape Clinic. They will use the information to better diagnose and manage children with flat heads.

SIGNATURES

Your signature on this form means that you have understood to your satisfaction the information regarding your child's participation in the research project and agree to their participation as a subject. In no way does this waive your legal rights nor release the investigators, or involved institutions from their legal and professional responsibilities. You are free to withdraw your child from the study at any time without jeopardizing their health care. If you have further questions concerning matters related to this research, please contact:

Dr. Ardene Robinson Vollman (403) 239-3180

Or

Dr. Jennifer Hatfield (403) 220-8323

If you have any questions concerning your rights as a possible participant in this research, please contact The Chair of the Conjoint Health Research Ethics Board at the Office of Medical Bioethics, 403-220-7990.

Parent/Guardian's Name	Signature and Date
Investigator/Delegate's Name	Signature and Date
Witness' Name	Signature and Date
Infant's Name	Infant ID_____

The University of Calgary Conjoint Health Research Ethics Board has approved this research study.

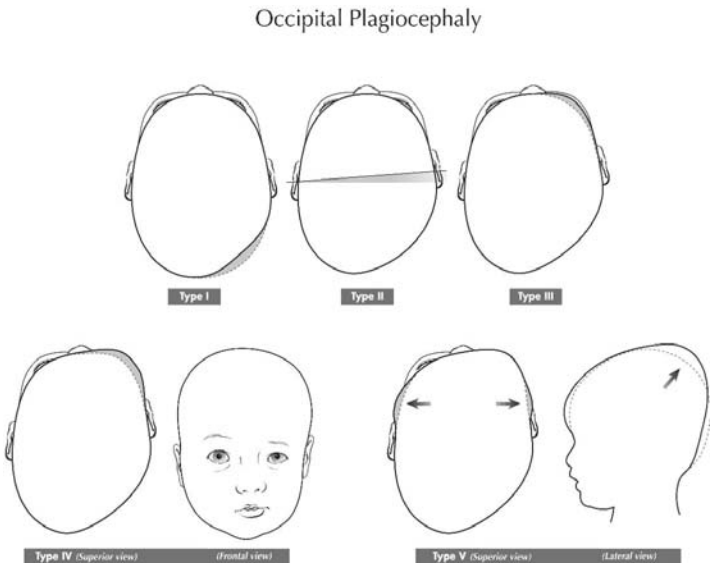
A signed copy of this consent form has been given to you to keep for your records and reference.

APPENDIX C

Plagiocephaly Data Collection Tool – Research Nurse and Public Health Nurses

Directions: Please circle the type of occipital plagiocephaly you observed, check appropriate boxes below and fill in answers in space provided

Infant ID _____



From Clinical Classification of Positional Plagiocephaly by L. Argenta *Journal of Craniofacial Surgery, 15*(3), 368-372. Reprinted with permission.

Table 54: Clinical Classification of Positional Plagiocephaly

Clinical Finding	Type I	Type II	Type III	Type IV	Type V
Posterior asymmetry	Present	Present	Present	Present	Present
Ear malposition	Absent	Present	Present	Present	Present
Frontal asymmetry	Absent	Absent	Present	Present	Present
Facial asymmetry	Absent	Absent	Absent	Present	Present
Temporal bossing or posterior vertical cranial growth	Absent	Absent	Absent	Absent	Present

Note. From Clinical Classification of Positional Plagiocephaly by L. Argenta *Journal of Craniofacial Surgery, 15*(3), 368-372. Reprinted with permission.

No observed plagiocephaly ☐

Torticollis present ☐

Torticollis absent ☐

Torticollis not assessed ☐

PHN intervention:

Repositioning teaching ☐

Referral to Infant Repositioning Class ☐

Referral to the family physician ☐

Follow-up planned ☐

Reason for referral _____

Comments _____

APPENDIX D

Infant ID _____

Baby's sex: male ☐ female ☐

Was this baby part of twins or triplets? Yes ☐ No ☐

This is my

First Child ☐ Second Child ☐ Third Child ☐

I have 4 children or more ☐

My baby age (in months and weeks) is _____

Most of the time (about 80% of the time) while my baby is awake he or she is on his or her tummy ("tummy time"):

1 time a day ☐

2 times a day ☐

3 times a day ☐

4 times a day ☐

More than 4 times a day ☐

Most of the time (about 80% of the time) my baby sleeps on his or her

Back ☐

Tummy ☐

Side ☐

Most of the time (about 80% of the time) when my baby is asleep, his or her head rests on the

Right side ☐

Left side ☐

I have not noticed a resting preference ☐

Most of the time (about 80% of the time) when I feed my baby, he or she is:

In my arms ☐

Laying on another flat surface ☐

Sitting in a chair ☐

How concerned are you about your child's head shape?

Very concerned ☐

A little concerned ☐

Not concerned ☐

Delivery type

Forceps ☐

Vacuum) ☐

C-section ☐

Vaginal delivery with no equipment ☐

Mother's highest level of education completed _____

Mother's Age _____

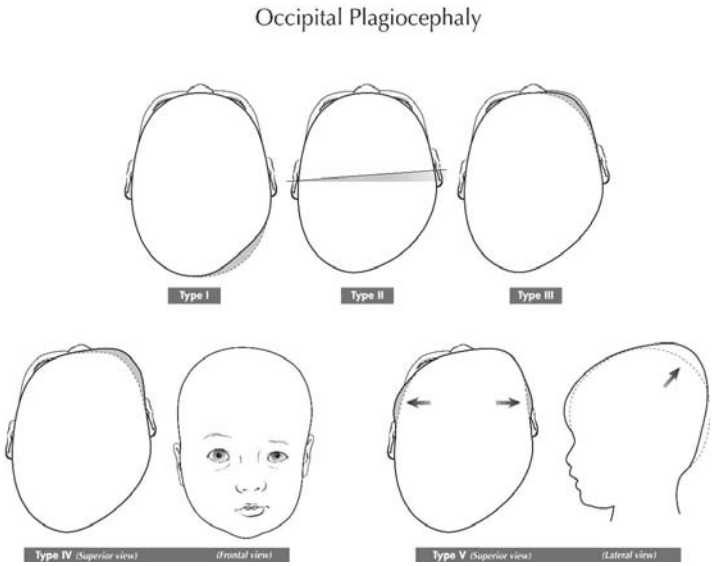
Number of years lived in Canada _____

APPENDIX E

Plagiocephaly Data Collection Tool – Head Shape Clinic

Directions: Please circle the type of occipital plagiocephaly you observed

Infant ID _____



From Clinical Classification of Positional Plagiocephaly by L. Argenta *Journal of Craniofacial Surgery, 15*(3), 368-372. Reprinted with permission.

Table 55: Clinical Classification of Positional Plagiocephaly

Clinical Finding	Type I	Type II	Type III	Type IV	Type V
Posterior asymmetry	Present	Present	Present	Present	Present
Ear malposition	Absent	Present	Present	Present	Present
Frontal asymmetry	Absent	Absent	Present	Present	Present
Facial asymmetry	Absent	Absent	Absent	Present	Present
Temporal bossing or posterior vertical cranial growth	Absent	Absent	Absent	Absent	Present

Note. From Clinical Classification of Positional Plagiocephaly by L. Argenta *Journal of Craniofacial Surgery, 15*(3), 368-372. Reprinted with permission.

Attended Infant Repositioning Class ☐

Within the type of plagiocephaly you've identified above, how would you classify the severity?

Mild ☐

Moderate ☐

Severe ☐

Appropriateness of PHN referral

Appropriate referral ☐

Inappropriate referral ☐

Type of treatment offered:

Cranial orthosis (helmet) ☐

Physiotherapy referral ☐

Repositioning teaching ☐

Comments_____

APPENDIX F

Table 56: Adjusted Model of Risk Factors Predictive of Positional Plagiocephaly (n=435)

Variable	β.C.	S.E.	Wald	df	*p*	OR	95% CI
Sleep position	0.982	0.267	13.509	1	.000*	2.670	[1.582, 4.508]
Tummy time	-0.075	0.215	0.123	1	.726	0.927	[0.608, 1.414]
Birth order	-0.147	0.221	0.444	1	.505	0.863	[0.560, 1.330]
Infant sex	0.436	0.221	3.914	1	.048*	1.547	[1.004, 2.383]
Multiple	-0.325	0.566	0.329	1	.566	0.723	[0.238, 2.191]
Maternal education	-0.349	0.254	1.893	1	.169	0.705	[0.429, 1.160]
Delivery type (vacuum/forceps)	0.633	0.315	4.046	1	.044	1.883	[1.016, 3.488]
Delivery type (c-section)	-0.229	0.247	0.865	1	.352	0.795	[0.490, 1.289]
Head positional preference (right)	1.539	0.248	38.593	1	.000*	4.662	[2.868, 7.577]
Head positional preference (left)	1.438	0.277	26.907	1	.000*	4.212	[2.446, 7.251]
Constant	-1.715	0.400	18.366	1	.000	0.180	

Note. β.C. = beta regression coefficient; S.E. = standard error; df = degrees of freedom; OR = odds ratio; CI = confidence interval
*Significant at 0.05 level

Table 57: Adjusted Model of Risk Factors Predictive of Positional Plagiocephaly, Sleep Position Removed (n=435)

Variable	β.C.	S.E.	Wald	df	*p*	OR	95% CI
Tummy time	-0.098	0.211	0.215	1	.643	0.907	[0.600, 1.370]
Birth order	-0.013	0.214	0.004	1	.951	0.987	[0.648, 1.502]
Infant sex	0.321	0.215	2.237	1	.135	1.378	[0.905, 2.099]
Multiple	-0.545	0.551	0.980	1	.322	0.580	[0.197, 1.706]
Maternal education	-0.307	0.247	1.543	1	.214	0.735	[0.453, 1.194]
Delivery type (vacuum/forceps)	0.451	0.304	2.204	1	.138	1.570	[0.865, 2.849]
Delivery type (c-section)	-0.227	0.242	0.876	1	.349	0.797	[0.496, 1.282]
Head positional preference (right)	1.482	0.242	37.511	1	.000*	4.403	[2.740, 7.075]
Head positional preference (left)	1.532	0.273	31.498	1	.000*	4.628	[2.710, 7.903]
Constant	-1.161	0.376	9.537	1	.002	0.313	

Note. β.C. = beta regression coefficient; S.E. = standard error; df = degrees of freedom; OR = odds ratio; CI = confidence interval
*Significant at 0.05 level

Table 58: Adjusted Model of Risk Factors Predictive of Positional Plagiocephaly, Birth Order Removed (n=434)

Variable	β.C.	S.E.	Wald	df	*p*	OR	95% CI
Sleep position	0.957	0.264	13.103	1	.000	2.604	[1.551, 4.372]
Tummy time	-0.068	0.215	0.990	1	.753	0.935	[0.613, 1.424]
Infant sex	0.424	0.220	3.725	1	.054	1.528	[0.993, 2.349]
Multiple	-0.356	0.562	0.401	1	.527	0.700	[0.233, 2.101]
Maternal education	-0.351	0.254	1.911	1	.167	0.704	[0.428, 1.158]
Delivery type (vacuum/forceps)	0.580	0.304	3.638	1	.056	0.787	[0.984, 3.239]
Delivery type (c-section)	-0.240	0.246	0.950	1	.330	0.787	[0.486, 1.275]
Head positional preference (right)	1.530	0.247	38.343	1	.000	4.618	[2.845, 7.495]
Head positional preference (left)	1.440	0.277	26.977	1	.000	4.220	[2.451, 7.266]
Constant	-1.987	0.436	20.797	1	.000	0.137	

Note. β.C. = beta regression coefficient; S.E. = standard error; df = degrees of freedom; OR = odds ratio; CI = confidence interval
*Significant at 0.05 level

Table 59: Adjusted Model of Risk Factors Predictive of Positional Plagiocephaly, Maternal Education Removed (n=435)

Variable	β.C.	S.E.	Wald	df	*p*	OR	95% CI
Sleep position	0.922	0.263	12.245	1	.000	2.514	[1.500, 4.213]
Tummy time	-0.081	0.214	0.141	1	.707	0.923	[0.606, 1.404]
Birth order	-0.156	0.220	0.500	1	.480	0.856	[0.556, 1.318]
Infant sex	0.427	0.219	3.798	1	.051	1.533	[0.998, 2.355]
Multiple	-0.413	0.563	0.359	1	.463	0.662	[0.219, 1.994]
Delivery type (vacuum/forceps)	0.632	0.313	4.067	1	.044	1.881	[1.018, 3.477]
Delivery type (c-section)	-0.266	0.245	1.177	1	.278	0.766	[0.474, 1.240]
Head positional preference (right)	1.542	0.247	38.968	1	.000	4.675	[2.881, 7.588]
Head positional	1.514	0.274	30.524	1	.000	4.544	[2.656, 7.775]
Constant	-2.188	0.387	31.961	1	.000	0.112	

Note. β.C. = beta regression coefficient; S.E. = standard error; df = degrees of freedom; OR = odds ratio; CI = confidence interval
*Significant at 0.05 level

Table 60: Adjusted Model of Risk Factors Predictive of Positional Plagiocephaly, Head Positional Preference Removed (n=434)

Variable	β.C.	S.E.	Wald	df	*p*	OR	95% CI
Sleep position	0.969	0.252	14.823	1	.000	2.634	[1.609, 4.313]
Tummy time	-0.033	0.202	0.027	1	.869	0.967	[0.650, 1.438]
Birth order	-0.093	0.208	0.202	1	.653	0.911	[0.606, 1.369]
Infant sex	0.415	0.207	4.020	1	.045	1.515	[1.009, 2.273]
Multiple	-0.078	0.540	0.021	1	.886	0.925	[0.321, 2.665]
Maternal education	-0.468	0.238	3.880	1	.049	0.626	[0.393, 0.998]
Delivery type (vacuum/forceps)	0.435	0.292	2.225	1	.136	1.545	[0.872, 2.737]
Delivery type (c-section)	-0.204	0.231	0.778	1	.378	.815	[0.518, 1.283]
Constant	-0.939	0.385	5.959	1	.015	0.391	

Note. β.C. = beta regression coefficient; S.E. = standard error; df = degrees of freedom; OR = odds ratio; CI = confidence interval
*Significant at 0.05 level

APPENDIX G

Management of Positional Plagiocephaly (Head Shape Clinic, March 21, 2011

Head Shape Assessment

- Assess the infant's head from above, looking for areas of flatness
- Assess ear symmetry
- Assess facial symmetry (forehead, eyes, cheek size and level

Any Visible Concerns (Non-Synostotic)

- Educate on repositioning to achieve counter-pressure on the non-flattened area of the head, and alleviate pressure from the flattened area
- Provide a repositioning handout for plagiocephaly or brachycephaly
- Screen for torticollis/neck muscle imbalance red flags and refer to physiotherapy as appropriate
- Refer for further evaluation of head shape and teaching regarding repositioning, depending on the age of the infant and the severity of the concern
- Fax referral to Head Shape Clinic at (403) 955-7609

Birth up to 4 Months of Age

- Refer to Head Shape Clinic or verbal referral to an Infant Repositioning Class
- Provide booking information sheet
- If verbal referral, parents to call (403) 955-5437 to register

Infant Repositioning Class

Possible outcomes:

- Continue with repositioning with ongoing monitoring by the referring physician (discharge from clinic)
- Continue with repositioning with follow-up visit at the Early Identification and Treatment (EIT) Clinic
- Letter to the referring physician to request referral to physiotherapy
- Referral to the Pediatrician Assessment Clinic (PAC), if one has not already been made

Management of Positional Plagiocephaly (Head Shape Clinic, March 21, 2011

Head Shape Assessment

- Assess the infant's head from above, looking for areas of flatness
- Assess ear symmetry
- Assess facial symmetry (forehead, eyes, cheek size and level

Any Visible Concerns (Non-Synostotic)

- Educate on repositioning to achieve counter-pressure on the non-flattened area of the head, and alleviate pressure from the flattened area
- Provide a repositioning handout for plagiocephaly or brachycephaly
- Screen for torticollis/neck muscle imbalance red flags and refer to physiotherapy as appropriate
- Refer for further evaluation of head shape and teaching regarding repositioning, depending on the age of the infant and the severity of the concern
- Fax referral to Head Shape Clinic at (403) 955-7609

4-6 Months of Age

- Refer to the Head Shape Clinic at the Alberta Children's Hospital using the Head Shape Clinic Referral Form
- Fax referral to Head Shape Clinic at (403) 955-7609

Early Identification and Treatment (EIT) Clinic

Possible outcomes:

- Continue with repositioning with ongoing monitoring by the referring physician (discharge from clinic)
- Continue with repositioning and return to the EIT Clinic for re-assessment within a specified time frame
- Referral to physiotherapy for treatment of torticollis and/or
- Referral to the Pediatrician Assessment Clinic (PAC)for further assessment and possible prescription of a corrective helmet (Starband by Orthomerica)

Management of Positional Plagiocephaly
(Head Shape Clinic, March 21, 2011

Head Shape Assessment

- Assess the infant's head from above, looking for areas of flatness
- Assess ear symmetry
- Assess facial symmetry (forehead, eyes, cheek size and level

Any Visible Concerns (Non-Synostotic)

- Educate on repositioning to achieve counter-pressure on the non-flattened area of the head, and alleviate pressure from the flattened area
- Provide a repositioning handout for plagiocephaly or brachycephaly
- Screen for torticollis/neck muscle imbalance red flags and refer to physiotherapy as appropriate
- Refer for further evaluation of head shape and teaching regarding repositioning, depending on the age of the infant and the severity of the concern
- Fax referral to Head Shape Clinic at (403) 955-7609

6-12 Months of Age

- Refer to the Head Shape Clinic at the Alberta Children's Hospital using the Head Shape Clinic Referral Form
- Fax referral to Head Shape Clinic at (403) 955-7609

Pediatrician Assessment Clinic

Possible outcomes:

- Continue with repositioning with ongoing monitoring by the referring physician (discharge from clinic)
- Continue with repositioning and return to the PAC for re-assessment within a specified time frame
- Referral to physiotherapy for treatment of torticollis and/or
- Prescription of a corrective helmet (Starband by Orthomerica)
- Referral to Neurosurgery if indicated