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***POSITIONAL PLAGIOCEPHALY:
USE OF CRANIAL ORTHOSIS FOR ITS TREATMENT***

Una revisión bibliográfica

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ABSTRACT

Background: Positional plagiocephaly, a condition characterized by the flattening of an infant’s skull that can potentially alter the infant’s craniofacial development. Its increasing prevalence due to recent changes in infant care practices makes it fundamental to evaluate the use of cranial orthosis as its most adequate form of treatment.

Objectives: The objective of this study is to analyze the efficacy of cranial orthosis as a treatment of positional plagiocephaly, aiming to consolidate the understanding of its effectiveness.

Methods: A systematic review was conducted to evaluate the use of cranial orthoses as a treatment option for positional plagiocephaly through a search conducted through PubMed, Medline, and Web of Science using the medical subject headings “positional plagiocephaly” and “orthosis,” and “positional plagiocephaly” and “helmet.”

Results: In total, 11 articles were selected to carry out this review. There were 2 randomized controlled trials, 5 clinical trials, and 4 observational studies including both comparative and retrospective studies.

Conclusion: Cranial orthoses as a form of treatment for positional plagiocephaly is highly recommended as it can reach the desired skull shape with minimal side effects and therefore avoiding subsequent craniofacial developmental side effects. Early clinical attention and intervention are imperative to address such deformities and offer cranial orthosis as a treatment option right from the start to reach its intended result.

Key words: Positional plagiocephaly, deformational plagiocephaly, cranial orthosis, helmet therapy.

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ACRONYMS INDEX

NICU: Neonatal intensive care unit

PP: Positional plagiocephaly

DP: Deformational plagiocephaly

CA: Cranial asymmetry

CVAI: Cranial vault asymmetry index

CVA: Cranial vault asymmetry

CI: Cephalic index

ODDI: Oblique diameter difference index

PCAI: Posterior cranial asymmetry index

CR: Cephalic ratio

ASR: anterior symmetry ratio

PSR: posterior symmetry ratio

RSI: Radial symmetry index

RDD: Reductions in diagonal difference

DD: Diagonal difference

CT: Computerized tomography

SIDS: Sudden infant death syndrome

CMT: Congenital muscular torticollis

AAP: American Academy of Pediatrics

FDA: Food and Drug Administration

AIMS: Alberta Infant Motor Scale

SD: Standard deviation

1. INTRODUCTION

1.1. Brain and skull development at early age

The brain is one of the most important organs in the human body. Head growth in an infant is majorly determined by brain development. It is protected by the cranium which grows and develops into its final shape throughout a child's life. The head reaches 90% of its growth by 1 year of age; it is 95% of its ultimate adult size by 6 years of age and growth is complete by age 7. (1)

The cranium can be divided in two parts; the neurocranium, which acts as protection for the brain, and the viscerocranium which molds the shape of the individual's face. (2) The neurocranium is made up of the cartilaginous part and the membranous part. The cartilaginous part makes up the basicranium (3), which is the inferior part of the skull, and the membranous part forms the calvarium (3), or the convexity of the skull. The calvarium consists mainly of the flat bones; the frontal, parietal, squamous occipital and squamous temporal bones. In between these bones the fibrous sutures and fontanelles can be found. The fibrous membrane that forms the cranial vault is not ossified at the angles of the parietal bones, producing 6 fontanelles: two medial (anterior and posterior) and two lateral pairs (sphenoidal or anterolateral and mastoid or posterolateral). The anterior fontanelle is the largest and measures approximately 4 cm in anteroposterior and 2.5 cm in its transverse dimension. It occupies the junction between the sagittal and coronal sutures and is therefore triangular. The sphenoid and mastoid are small, irregular and appear at the sphenoid and mastoid angles of the parietal bones respectively. When a child is born the sutures in the cranium are not yet sealed and fused together which allows the skull to be molded during birth. The approximation of the flat bones post birth along the sides of the fontanelles eliminates the open spaces in a short amount of time, but a thin periosteum line remains for a longer period of time. (2) The sutures fully close by the end of the second year when the bones fuse together allowing further growth to happen through accretion and absorption.(3) The calvarium grows in different ways including sutural growth, through remodeling, and displacement by the growing brain.(2) Although this does not occur by the brain pushing the bones outward, instead the growth is stimulated by the growing periosteal membranes which causes the bones to be displaced with them. This draws the flat bones apart and the forces created are believed to be the stimulus that provoke osteogenesis.(2) As the brain expands the two sections of the skull that we previously stated, the cartilaginous or basicranium, and the membranous or calvarium, also expand. The cartilage in the basicranium is able to lengthen and accommodate to this expansion while in the calvarium the sutures that can be found there respond to the stimulus causing new osteogenesis. This production of new bone matter

along the edges of the bones allows the suture to remain relatively the same width while the cranial vault expands (2)

1.2. Skull Deformities

Abnormal skull deformities that develop into craniofacial deformity are a relatively common clinical finding. Craniofacial dysmorphology is categorized into malformations, deformations, and disruptions based on the primary defect in morphogenesis during early pregnancy(3). Malformations, occurring in the first 8 weeks of embryogenesis, result from genetic, environmental, or combined factors and have higher recurrence rates compared to environmentally related disruptions and deformations.

Disruptions arise from destructive processes after organogenesis, affecting morphogenetically normal tissues or organs, such as amniotic banding causing a cleft lip(3). They are predominantly attributed to environmental factors, resulting in low recurrence rates.

Deformations occur post-embryogenesis due to non-disruptive mechanical forces (3), causing abnormal formations or distortions of morphogenetically normal body parts. They may be influenced by local uterine or postnatal positional factors, potentially resolving with a changed environment, and are associated with low recurrence rates (4). There are two main groups of deformities: on the one hand the group of deformities resulting from premature ossification and fusion of the cranial sutures, called craniosynostosis, and on the other hand positional deformities which include brachycephaly, dolichocephaly and deformational plagiocephaly (5)

Accurate identification of an unusual head shape in an infant or child is crucial for effective management of this prevalent condition. Distinguishing between craniosynostosis, which require surgical intervention, and nonsynostotic head deformities, which do not necessitate surgery, is not always a straightforward process (6).

1.2.1. Craniosynostosis

The premature fusion of bones at the sutures, known as craniosynostosis, hinders additional bone development at those sites. This fusion prevents the normal expansion of the neurocranium, resulting in abnormal compensatory changes in the head's overall shape and often leading to craniofacial abnormalities (2). While most researchers have found that craniosynostosis is mainly caused by single-gene genetic mutations, it is believed that environmental and teratogenic causes can also play a role, although less common (7). The

primary genetic mutations associated with craniosynostosis involve fibroblast growth factor receptors, TWIST, and MSX2 genes. The environmental factors that could be involved as well would be rickets, due to vitamin D deficiency or resistance, chronic renal failure, hypophosphatemia and hyperthyroidism (7). Recently, local intrauterine factors affecting fetal constraint have also been identified as risk factors for craniosynostosis. These factors encompass abnormal positioning in utero, overcrowding in multiple pregnancies, oligohydramnios, and prolonged duration of the first stage of labor, all leading to increased fetal head constraint (8). Craniosynostosis can appear as a primary or secondary form. Primary, or isolated craniosynostosis without evidence of a genetic syndrome, can either be syndromic or non-syndromic. It is estimated that 84,665 children are born with craniosynostosis globally each year, including 72,857 with nonsyndromic craniosynostosis (9). Being sagittal synostosis the most common form, 44% (10)

In 1815 Virchow formally outlined the relationship that the calvarial deformity may have with the suture that is fused. He described that the deformity occurs in a parallel plane in concordance to the affected suture (7). This theory is currently still in use.

Sagittal synostosis is the most prevalent form of craniosynostosis, representing 40-60% of all instances (11). As the coronal and metopic sutures remain open, skull growth occurs at these sites, leading to the typical presentation of a prominent forehead with an anterior tilt and a wider gap between the eyes. Around 80% of sagittal synostosis cases manifest as isolated occurrences without any underlying syndromes (11).

Although it is believed that there is an increase in incidence, metopic synostosis correlates to less than 10% of the cases (11). The closure of the metopic suture initiates at the glabella and progresses upwards towards the anterior fontanelle, which commences during the third trimester and is typically finished by 9 months of age. This causes narrowing of the basicranium (11).

Coronal synostoses have a more intricate impact on the craniofacial appearance compared to the midline synostoses, sagittal and metopic. This type of synostoses leads to more significant visual and oculo-motor impairments (11).

These types of deformities need a surgical approach as their treatment to allow the brain growth to develop and avoid neurological effects.

1.2.2. Positional deformities; Brachycephaly, Dolichocephaly, and Plagiocephaly

1.2.2.1. Brachycephaly

Brachycephaly refers to a symmetrical flattening of the occipital bone (12). This positional deformation at the back of the skull, features a reduced anterior–posterior cranial diameter, shorter length, expanded transversal diameter, and noticeable bulging on both sides of the parietal region (4). It is defined by a cephalic index greater than 81% (4). The cephalic index is an objective parameter obtained by taking the biparietal diameter of the head, divided by the occipitofrontal diameter, multiplied by 100% (1). This condition, although normally caused by the same forces that cause positional plagiocephaly, is rarely pathological.

1.2.2.2. Dolichocephaly

Dolichocephaly is also a symmetrical flattening, but in the opposite plane of brachycephaly. A Dolichocephalic infant would be described as having a tall narrow head (1). It is defined as a cephalic index less than 75% (13). This deformity is relatively rare in cultures where infants sleep in a supine position, except preterm babies that have spent a prolonged amount of time in the NICU where they will normally be in a prone or side-lying position (1). Severe dolichocephaly might suggest the presence of sagittal suture synostosis. Differentiating factors include the extent of head elongation, absence of improvement with time in a supine position, and pronounced frontal protrusion along with prominence at the back of the head (1).

1.2.2.3. Plagiocephaly

The word plagiocephaly is derived from the Greek words “plagios”, oblique and “cephalos”, head (1). Plagiocephaly may or may not be associated with a premature sutural closure, synostosis, and when this is the case, it is described as synostotic plagiocephaly, while when there is no association of plagiocephaly with a sutural closure we refer to them as positional, deformational or non-synostotic plagiocephaly (14) the condition that this review is mainly focused on.

Positional or deformational plagiocephaly (DP) is an asymmetrical type of head deformation that can either be anterior or posterior. The deformity is usually due to pre and/or postnatal mechanical forces applied on a malleable and forming skull (12). Deformational plagiocephaly may be the result of external forces prenatally due to the type of infant's presentation in the maternal pelvis, and postnatally such as excessive time in the supine position that have molded the infant's head (8). The cranium adopts a parallelogram shape with the

occipital region flattened unilaterally, possible prominences in the frontal and parietal ipsilateral areas and the pinna and zygomatic arch in an anterior position (15). These characteristics give the head a parallelogram shape when viewed from above, and possible facial asymmetry (14) as seen in *Figure 1*.

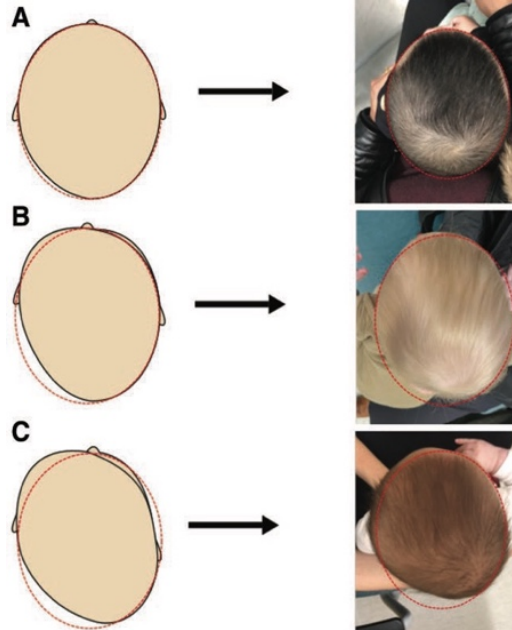


Figure 1 extracted from Watt et al. Occipital flattening, frontal bossing, and ipsilateral ear shift are the hallmark manifestations of deformational plagiocephaly, becoming more pronounced as severity increases. (49)

1.3. Positional Plagiocephaly

1.3.1. History

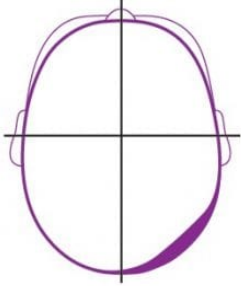
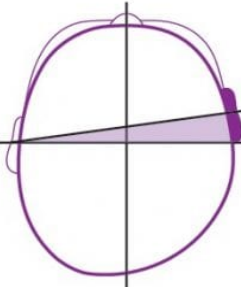
Cranial deformities date back historically to many decades ago. To be more exact the first evidence of artificial cranial deformity would take us to the ancient middle Paleolithic period, being followed by the Inca civilization in Peru, ancient Egypt (12), and every other civilization worldwide. Archaeological evidence reveals various head shapes, indicating that this custom held diverse interpretations, possibly to delineate membership within social or ethnic groups (16). Although nowadays these customs have subsided, cranial deformities are still very much present.

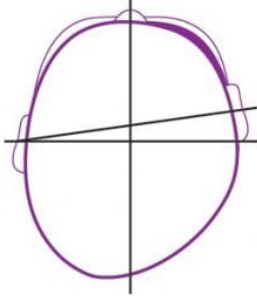
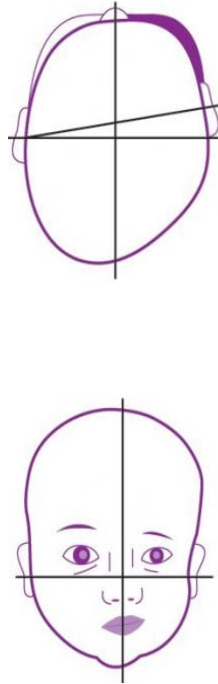
1.3.2. Diagnosis

DP is a very common condition and a recurrent reason why parents visit their pediatrician. As stated previously, deformational plagiocephaly arises from extrinsic molding

rather than intrinsic synostotic events (15). It may be present at birth due to an abnormal intrauterine position most commonly caused in pregnancies of multiples by the pressure that the fetus could place on the other fetus. Although these deformities could debut pre or postnatally, the true definition of positional plagiocephaly is once this deformity persists after the first six weeks of the child's life, since altered skull shape at birth could revert to 'normal' in the early postnatal period (8).

Numerous studies have been undertaken to reach the optimal diagnostic approach for identifying deformational plagiocephaly in infants, aiming to tailor the treatment most suited to their specific condition. Two crucial elements influencing this diagnosis involve effective communication and comprehension on the part of the parents, coupled with the establishment of a measurable criterion to categorize the extent of asymmetry in the infant. Visual assessment is the best way to describe the infants' cranial deformation to the parents, but there is no way to quantify this just visually. Argenta (2004) proposed a classification system for deformational plagiocephaly and brachycephaly exclusively based on visual assessment (17). This classification identifies 5 different types of DP based on the degree of deformation, and 3 different types of brachycephaly (*Tables 1 and 2*).

Type	Description	Visual Representation
Type I	<ul style="list-style-type: none"> - The cranial asymmetry is completely limited to the back of the skull. - The degree of posterior flattening may vary, but all pathological findings are restricted to this area. - There is no asymmetry of the ears as measured from the nose to the ear. - The forehead is straight, the face is symmetrical, and there are no abnormal temporal bulges or vertical elongation of the skull. - Most mild form of plagiocephaly. 	 <p>The diagram shows a top-down view of a skull with a vertical and a horizontal reference line. The posterior part of the skull is flattened, indicated by a shaded purple area at the back. The rest of the skull, including the forehead and ears, is symmetrical.</p>
Type II	<ul style="list-style-type: none"> - Variable degrees of posterior cranial asymmetry. All, however, are significant enough to affect the central cranium and skull base and to allow displacement of the ear on the involved side forward, downward, or in both directions. - This can be visualized from above and while looking at the child in the anterior and posterior views. - The anterior portion of the skull is not affected, and the forehead is symmetrical. - There is no facial asymmetry. - There are no decompressive deformities to the skull. 	 <p>The diagram shows a top-down view of a skull with a vertical and a horizontal reference line. The skull is tilted to the right. The ear on the right side is displaced forward and downward, indicated by a shaded purple area. The forehead and the rest of the skull are symmetrical.</p>

<p>Type III</p>	<ul style="list-style-type: none"> - Includes the triad of posterior cranial asymmetry, malposition of the ipsilateral ear, and ipsilateral frontal bone protrusion causing frontal bone asymmetry. - There are no de- compressive changes in the skull, and the face is symmetrical. - This determination is best made by looking from above and directly anteriorly at the child. 	
<p>Type IV</p>	<ul style="list-style-type: none"> - Deformity of the posterior skull, malposition of the ipsilateral ear, ipsilateral frontal asymmetry, and ipsilateral facial asymmetry. - The facial asymmetry is the result of excess fatty tissue and, less frequently, hyperplasia in the ipsilateral zygoma. - There may be asymmetries visible in the amount of malar and cheek fat. - In most cases, the facial asymmetry is purely soft tissue, although in more severe cases, some bony asymmetry may also develop. 	

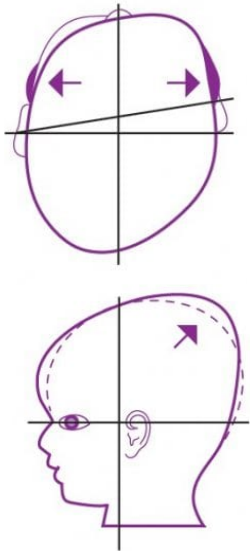

<p>Type V</p>	<ul style="list-style-type: none"> - Deformity of the posterior skull, malposition of the ears, forehead asymmetry, and facial asymmetry. - In addition, these cases demonstrate an anatomical attempt at decompression of the growing brain. - These children have temporal bulging or an abnormal vertical growth of the posterior skull as a result of the cranial constriction to the point that the brain erodes and deforms the skull. - This bulging is most obvious on CT but is readily clinically apparent if the child is viewed from the four proposed positions. 	
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Table 1 Visual classification of Positional Plagiocephaly as described by Argenta 2004 (17).

Type	Description	Visual Representation
<p>Type I</p>	<ul style="list-style-type: none"> - There is a depression of the central skull at the confluence of the lamboids with the sagittal suture. - The position of the ears, forehead, and face is otherwise normal. 	


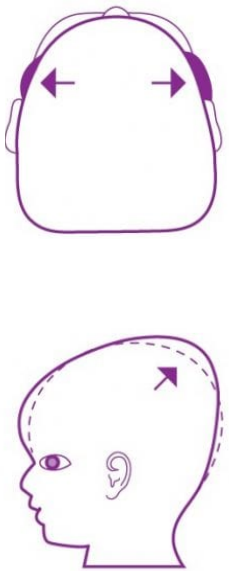
<p>Type II</p>	<ul style="list-style-type: none"> - More severe form of the central deformational plagiocephaly. - There is a central cranial depression in the posterior aspect of the skull; however, in addition, the skull has begun to widen in its posterior half as the brain attempts to decompress. - This widening results in more accentuation of the flatness of the posterior skull. 	
<p>Type III</p>	<ul style="list-style-type: none"> - The most severe form of central plagiocephaly - There is not only flattening and widening of the posterior skull but an attempt at further decompression of the brain by vertical growth of the posterior skull or temporal bulging secondary to erosion of the temporal skull. 	

Table 2 Visual classification of Brachycephaly as described by Argenta 2004. (17)

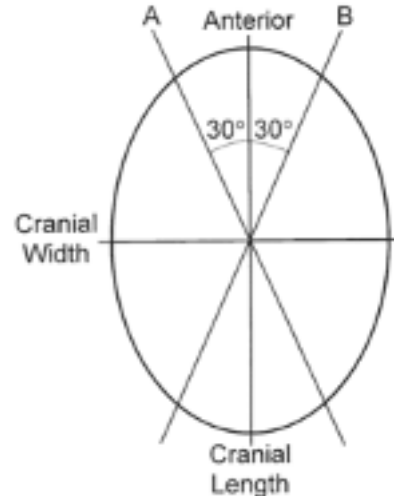
Although this classification has great utility for the discussion of the child's condition with the parents, as stated before this does not include a quantifiable measurement to have a valuable classification. In a study conducted by Wilbrand et al (2012), an alternative clinical classification for infant deformational plagiocephaly was introduced, incorporating anthropometric caliper measurements. This comprehensive analysis, based on a database of over 400 children, aids clinicians in treatment decisions by dividing the forms of DP in a reliable three-level severity categorization determined by their cranial vault asymmetry index (CVAI) (18). This measurement can be obtained by an easily accessible tool, the anthropometric caliper measurement that can be used in clinical settings. The cephalic index (CI) is an objective

parameter obtained by taking the biparietal diameter of the head, divided by the occipitofrontal diameter, multiplied by 100%. CI represents the width of the head as a percent of the length of the head. The average CI in children under 3 years of age is 76% - 81% (1).

$$\text{Cranial Index} = \frac{\text{cranial length}}{\text{cranial width}} \times 100$$

CVAI represents the asymmetry of the cranial vault. A perfectly symmetrical head will obtain a CVAI of 0% (19). If the child has some type of asymmetry, it could be calculated and classified into three different stages of CVAI.

$$\text{Cranial Vault Asymmetry Index} = \frac{\text{diagonal A} - \text{diagonal B}}{\text{diagonal A}} \times 100$$



The mild case would be between the 75th and 90th percentile, the moderate between 90th and 97th percentile and severe above 97th percentile (18).

Figure 2 Head tracings yielded a cranial index and cranial vault asymmetry index. (19)

The visual assessment and the quantifiable measure are normally used simultaneously during a child's diagnosis.

Another method used to quantify skull asymmetry is plagiocephalometry (20). This is a non-invasive, easy-to-apply and accurate method. A high statistical reliability of this type of measurement was given during the Vlimmermen et al study (20). It is performed with a strip of thermoplastic material which is positioned around the infant's head (21). This mold is later traced on a piece of paper to calculate the diagonal diameters. It measures the relation between the transversal shape of the cranium correlated with the mouth and ears' position. The oblique diameter difference index (ODDI) is calculated and given a numerical value (22). It is classified into normal range, mild asymmetry, moderate, and severe. A normal ODDI would be <104, mild asymmetry: ODDI 104–107, moderate: ODDI 108–111, and severe: ODDI ≥112 (22) (**Figure 3**).

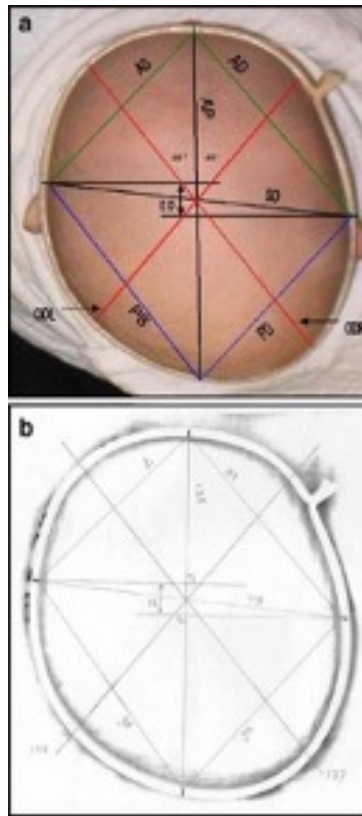


Figure 3 Illustration plagiocephalometry: asymmetry DP left occipital of the skull (boy, aged 4 months old). a Photograph of child with the thermoplastic ring fitted and landmarks. b Paper copy of the same ring with drawn and measured lines. (22)

A laser scanner is arising as a method for head shape measurement, especially when a cranial orthosis is needed for its treatment. A structured light 3D scanner is a device that uses structured light and two cameras to create 3D images.(23) The four points used in these measurements are; the middle point of the glabella, the left and the right junction of the bottom of the helix and the tragus, and the middle point between the nose and upper lip.(23) The laser scanner provides for a rapid measurement that does not expose the child to radiation, has precise quantitative head dimensions and gives the ability to follow changes in head shape over time.(24) This type of measurement is normally used in hospitals that can afford the materials required for it, one that has a large patient count, and for the cranial orthosis measurements.

Computerized tomography and radiographic imaging are also sometimes used as a measurement tool. Although it does give an accurate measurement, with some limitations, they cause unnecessary radiation on the child (24). Therefore, these forms of measurements are normally not used.

Overall, the most common form of head measurement nowadays is the traditional anthropometric caliper measurement, that must be carried out by a trained professional, followed by the 3D laser measurement if there are enough resources.

1.4. Prevalence and incidence of plagiocephaly

Deformational plagiocephaly has been an issue for decades, but it did not receive much attention due to its low prevalence of around 1 in 300 infants (14). This incidence has now increased exponentially. In 1992 the American Academy of Pediatrics released a new study called the “*Back to sleep*” campaign recommending infants to sleep in the supine position to reduce the risk of suffering from sudden infant death syndrome (SIDS) (21). This campaign did reduce significantly the deaths caused by SIDS, by 40%.(21) Although this reduction in the incidence of SIDS was monumental, it caused the exponential *increase* in nonsynostotic deformational plagiocephaly (21). While SIDS is an important issue that should be prevented it is also necessary to increase the awareness of the possibility of the development of nonsynostotic plagiocephaly in infants that sleep in a supine position by alternating the infants head positions while they sleep, amongst other parental habits that should be altered as well (25). Positional plagiocephaly is currently one of the most frequent reasons why parents visit the pediatric neurologist, making it a prevalent issue that should be further studied to obtain a better understanding of the possible catalysts and treatment options.

Due to the recommendation made by the American Academy of Pediatrics in 1992 of placing infants in a supine sleeping position to decrease the risk of SIDS, supine sleeping positioning significantly increased from 13% in 1992 to 70% in 2002 (26). This position is a well-established risk factor for deformational plagiocephaly which likely contributed to the sharp increase in prevalence of PP in infants (26). The noticeable increase in prevalence also caused for PP to be a rising concern among parents, and therefore a common reason to attend to their pediatrician.

Some studies have determined that ethnicity might play a role in PP prevalence. Different cultures have different ways of taking care of a child keeping in mind their surroundings, access to resources, compliance to sleeping position, and cultural habits such as babywearing that could influence the development of positional plagiocephaly. Italian infants have been recorded to be the ethnicity with the highest incidence of PP (25). Being African infants the next most likely, but either way three times less likely than the Italian infants, even after correcting main parental care habits such as sleeping position and tummy-time period (25).

1.5. Risk factors

Many studies consider supine sleeping position as the leading cause of PP, but not all supine sleepers develop positional plagiocephaly (25). Other risk factors that are linked to the development of deformational plagiocephaly are gestational age, intrauterine position, oligohydramnios, assisted delivery, presentation at birth order, male sex, infant neck problem, and numerous infant care practices (25). Although all these factors could contribute to the incidence of DP it is not possible to obtain univocal environmental or lifestyle risk factors that are directly linked to the prevention of DP within the first six months of life (25).

One of the primary observations when studying DP risk factors is the presence of infant torticollis. There are different forms of torticollis in infants, but it is believed that the most common form is congenital muscular torticollis (CMT) which occurs when there is an asymmetric length or strength level of the sternocleidomastoid muscles (27). This causes a predisposition of head turning to a specific side (27). Clinically the head of the infant is tilted to the side of the affected muscle and rotated to the opposite side (27). During a study carried out in 2014 (27), torticollis was considered the most common associating finding in infants with deformational plagiocephaly (27). Rogers et al (28) determined that 90% of infants with PP were noted to have torticollis, but only 24% of these subjects were previously diagnosed or treated for torticollis, showing that the incidence of torticollis is undiagnosed in infants with positional plagiocephaly (27). There is some controversy regarding whether torticollis is the primary disorder, or if positional plagiocephaly is the one that can trigger it, or both (29).

Category	Exposure
Biological infant factors	<ul style="list-style-type: none"> - Male gender - Torticollis, limited head rotation, head rotational asymmetry - Preferred head position/orientation - Higher birth weight - Developmental delay - Head circumference, macrocephaly - Lower level of activity - APGAR score - Others: malformations, prenatal bone mineral density, head shape at birth, temperament, snoring, limitation of head function, zygoty (dizygotic), abnormal cerebrospinal fluid spaces, 'high rate of

	pericerebral fluid collections’, head tilt, neurological problems, ‘lateral strain patterns of spheno-occipital synchondrosis’, pattern of occipito-atlantal motion, asymmetrical movements of trunk, brachycephaly, siblings with plagiocephaly
Obstetric factor	<ul style="list-style-type: none"> - Birth order (parity) - Mode of delivery: forceps, vacuum or assisted delivery - Prematurity - Intrauterine position/cranial immobility - Lower gestational age - Multiple birth - Multiple gestation pregnancies - Others: birth injury, diagnosis of oligohydramnios during pregnancy, other obstetric factors, birth season
Infant care practices, lifestyle of mother/ parents	<ul style="list-style-type: none"> - Supine sleep position - Little time spent prone (‘tummy time’) - Feeding pattern (bottle feeding)/non-varying position during feeding - Use of car seats, swings, carriers, bouncy seats, rockers - Smoking - Others: mother’s holding position, pacifier use, mattress type and softness, pillow use, advice received about plagiocephaly, soft drinks (caffeine), alcohol consumption, attended antenatal classes, late or early begin of prenatal care, chemicals, medication, insufficient vitamin D intake
Sociodemographic factors	<ul style="list-style-type: none"> - Lower parental age - Lower educational level - Others: SES, marital status of mother, maternal number of years lived in Canada, maternal language barriers, ethnicity

Table 3 Exposures that have been identified as potential risk factors of positional plagiocephaly in any of the 22 empirical studies reviewed in De Bock et cols 2017 study. Table extracted from the De Bock et cols 2017 study.(30)

1.6. Effects of plagiocephaly

Positional plagiocephaly affects many aspects of a child's development as seen in various studies. PP can present postural alterations, muscular flexibility, balance, visual, ophthalmologic and, temporomandibular disfunctions, mandibular asymmetries causing aesthetical problems, and possible neurodevelopmental delays. Some of these effects have been more associated to PP than others, but nonetheless all have been correlated to PP in one way or another.

1.6.1. *Plagiocephaly and motor skills*

One of the most important aspects of the motor functions in a child's development is balance. Balance is important in keeping a stable posture and crucial in the functionality of a child, for them to feel secure and independent in their surroundings. It is a key element to develop walking, running, jumping, climbing and other motor abilities in preschoolers (31). In a 2016 study conducted by Cabrera-Martos et al. looking for the repercussions of positional plagiocephaly on posture, muscle flexibility and balance in children aged 3–5years old, it was found that there is indeed a correlation between problems in balance in children with history of plagiocephaly (31). The balance assessment in this study showed that those in the plagiocephaly group, compared to the control group, had a significantly lower balance level (31). Regarding flexibility, also assessed in this study, the PP group had lower thoracic mobility than the control group but both groups had the same lumbar mobility. This lower thoracic mobility and flexibility also affected those in the lower limbs' muscles (31).

1.6.2. *Plagiocephaly and facial asymmetries*

Facial asymmetries are the most obvious effect of positional plagiocephaly since they can sometimes be seen by the naked eye. These asymmetries cause diverse occlusal alterations. Various studies have found a deviation of the dental midline and the chin towards the unaffected side in infants with positional plagiocephaly (32). The cranial asymmetry causes a skull base deformity, a displacement of the glenoid fossa and in consequence a malocclusion seems to be likely (32). Positional plagiocephaly sometimes causes a rotation of the cranial base and an anterior displacement of the temporomandibular joint which causes a mandibular asymmetry (33) Not only a mandibular asymmetry exists but also in correlation to this there could be a certain degree of auricular displacement correlated with the severity of the cranial asymmetry in deformational plagiocephaly (33). A hypothesis exists that unexplained mandibular asymmetry in older patients may be secondary to deformational posterior plagiocephaly (33). Although these facial defects might not always be obvious, when they do become apparent these

differences can be of great concern to the infant's parents (34). The parents are most concerned about their child's physical appearance and the repercussions that these may have in their attractiveness and the possibility that the child may be embarrassed or socially stigmatized by their condition (34). This is therefore the most common first motivation to seek help by parents.

1.6.3. Plagiocephaly and neurological development

The correlation between plagiocephaly and the adequate development of a child's cognitive functions wasn't very clear until recently where there have been some studies that do find a link between the two. One of the first ones that found an association was Panchal et al. (2001) who showed severe cognitive delays in children with plagiocephaly (35). In 2012, Korpilahti et al. supported these findings in the study conducted by them with 21% of the subjects with PP having disorders in speech-language skills, and 30% slight developmental problems (36). These findings were further studied by Collett et al. (2013) that used a population of children up to 3 years of age and coincide with the two previous studies mentioned above (37). The study relates a decrease in gross motor skills with a lower cognitive and language development through the Bayley exam (37). In these tests, by the time the infants were 8 months old the mothers began to notice a that many were not achieving some tasks such as sitting unaided, rolling, and standing supported (37). This does not mean that PP is a cause for underdevelopment of some cognitive functions, but rather could be a marker for developmental risk. The notion of deformational plagiocephaly being a cause for a lower neurological development or a risk for it is a hypothesis presented by some studies. As stated in a Collet et al. study, infants with subtle neuromotor deficits may be less able to reposition themselves and therefore at greater risk for developing a skull deformation (38). Another possible hypothesis would be that a pathology that affects the central nervous system causes an infant to be less mobile and therefore stay in a prolonged position, or causing muscular affection, all causing the development of positional plagiocephaly (38). Due to this possible correlation many authors recommend an early detection and posterior monitorization to achieve optimal treatment.

1.7. Treatment

Over the course of the last decades, the strategy to treat positional plagiocephaly is not certain. Positional plagiocephaly was once considered a purely cosmetic disorder (39), but as stated previously in this article, it has now been suggested that children with this condition may experience as well some sort of developmental difficulties as well (39). This has caused for there to be a search for an appropriate form of treatment. There are a few options on how to approach the treatment such as, repositioning, physical therapy, and cranial orthosis (40,41). It is not certain which type of treatment has the best outcome as many factors must be taken into

consideration, but the treatments reach its maximum benefit if they are installed in the first year of the infant's life (40). Some infants deformational plagiocephaly is believed that it can be resolved spontaneously without any type of intervention over time if the degree of deformity is not too severe. The surgical option has been removed from the options of PP treatment, as it has been proven that nonsynostotic positional plagiocephaly can be cured in a more conservative manner without having an infant undergo surgery.

Timing regarding the beginning of the treatment is crucial in positional plagiocephaly. Many studies have found that the earlier the treatment is placed, the easier it is to treat the deformation and the lower the cost of the treatment.

1.7.1. Repositioning

The most conservative form of treatment of positional plagiocephaly is repositioning. This can be used as a preventative measure, as well as a treatment option for less severe cases. Repositioning consists of positioning the infant to minimize pressure on the back of the head (40). When it is used as a preventative approach, parents are counseled during the newborn period, at around 2 to 4 weeks, when the skull is maximally malleable (42). The preventative repositioning encompasses various exercises that the parent should follow, such as laying the infant down to sleep in the supine position, following the AAP guidelines to reduce the risk of SIDS, but alternating positions in the left and right occiputs (42). On the other hand, while awake the infant should be placed in a prone position to strengthen the neck muscles (42).

Repositioning when used as a treatment would mean conducting these exercises in an infant that already has a head deformation. In this case the parents are guided to position the baby by turning its head to the opposite side of the occipital flattening while sleeping in a supine position, (40) while also placing the infant in "tummy time" while awake.

1.7.2. Physical therapy

Physical therapy and repositioning as two different forms of treatment for plagiocephaly can sometimes be overlapped as there is not a specific, well differentiated physical therapy guideline for positional plagiocephaly. Physical therapy in positional plagiocephaly is centered around the idea of reducing the preferential position in infants and stimulating motor skills development to strengthen the infant's muscle tone (40). Physical therapy is normally used when there is a concomitant diagnosis of torticollis as well as positional plagiocephaly. Physical therapy in patients with positional plagiocephaly is modulated by the severity of the plagiocephaly (43). Normally the exercises included in this treatment involve positional changes

from supine, prone, lateral, and seated positions (43). Some of the exercises included in a Cabrera Martos et al study “Clinical profile and evolution of infants with deformational plagiocephaly included in a conservative treatment program” included a rolled-up towel adapted to the head of the infant. In the supine position the craniums pressure was distributed over a larger surface by adding the rolled-up towel at the base of the occipital bone when the infant is in the supine position. Parents then put objects in the midline and slowly moved them from side to side to stimulate visual displacement (43). In the prone position the rolled-up towel was placed under the armpits, in the lateral position the infants had to support their back on a solid surface on one side and the other, and finally in the seated position the towel was used to maintain a semi flexed position of the head inn zero to four months old, and from four months onward infants were seated with the trunk inclined backward (43). This study concluded that physical therapy is effective modulated by the severity of positional plagiocephaly. Early identification of positional preference in infants is essential for beginning early intervention with physical therapy varying the infant’s position both when laying them to sleep and while awake and supervised during tummy time (44). Klimo et al. conducted a study which concluded that although repositioning is an effective treatment for deformational plagiocephaly, physical therapy is more effective in treating already diagnosed positional plagiocephaly in infants (44). There is also evidence stating that the earlier this physical therapy is introduced the more effective it is.

1.7.3. Cranial orthosis

Orthotic helmets date back to 1979 when they were first described by Clarren (45). This helmet was made with cotton and plaster to make a cast of the patient’s head, finishing the shape with modeling clay or plaster slurry to build up the flattened areas and therefore achieve the desired cranial shape (45).

Since the start of the orthotic helmets, they have become a competitive market, especially with the rise of positional plagiocephaly cases following the 1992 American Association of Pediatrics “*Back to Sleep*” campaign. Internet marketing, news articles, and magazines have also raised the profile of these orthotic devices with many parents now investigating and insisting on the use of these devices (46). Cranial moulding devices now have many interchangeable names including helmet orthosis, cranial orthosis, cranial orthotic device, and orthotic headband (46). The only difference being that helmets cover the whole head and those called headbands have the top portion cut off (*Figure 4*).



Figure 4 extracted from Linz et al. Child wearing cranial remolding orthosis, Arrows show space for growth. (47)

The cranial orthosis is a device placed as a helmet in infants with positional plagiocephaly to correct the deformity. They are built to apply a certain degree of external pressure, much like the one that caused the positional plagiocephaly in the first place, on the protruding area of the skull and allow there to be a space where cranial expansion should take place to return the skull to its adequate shape (47). Nowadays, the orthosis is built by covering the infants head with plaster casting or doing 3D laser imaging to measure the infants head and have a positive mould from it (46,48). This mould, or impression is then filled with plaster to create what is called a positive cast, later corrected by applying a crescent-shaped module over the occipital flattening to create the “typical” shape (48). The mould is designed to fit snugly over the distorted areas and give space where the head should grow to achieve that desired head shape (46). By applying pressure over the areas where further growth is not desired, the cranial orthosis prevents further growth there and stimulates the growth in the flattened areas (19). The orthosis is to be worn 23 hours a day (47). Recent studies have determined a high level of cranial shape correction after the use of cranial orthosis as seen in figure 4 (28).

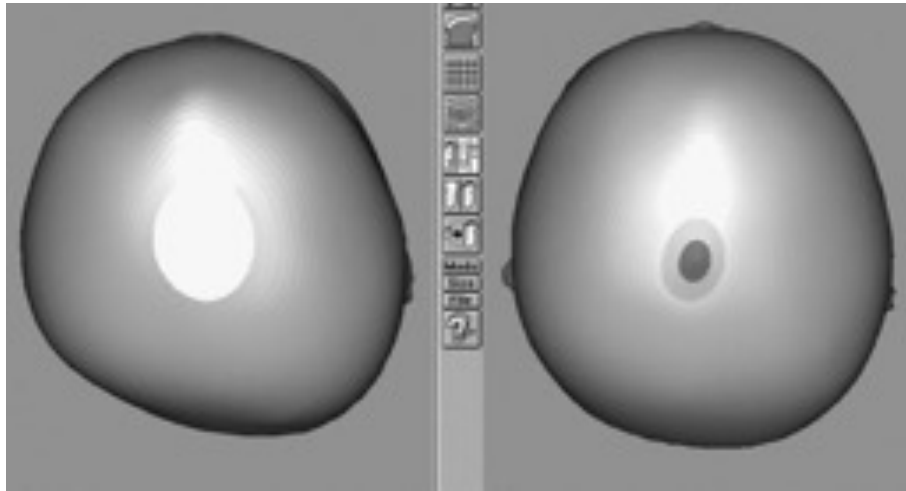


Figure 5 extracted from Rogers et al. Three-dimensional surface laser image of infant with right DP. Left, Before helmet treatment, age 5 months. Right, Correction after 3 months of helmet therapy

1.7.3.1. *Complications of cranial orthosis*

Numerous studies have explored the potential complications that can be associated with the use of cranial orthosis in children. The most obvious complications could be potential skin breakdown and pressure issues if improperly fit (46), skin rashes, and skin pressure ulcers (14). These complications are not considered severe and the rate of them is very low. In a study conducted by Robinson et al. less than 1% of their patients have experienced contact dermatitis or skin breakdown, having used different types of helmet suppliers (14). In addition to these skin related complications some authors in the past have stated head and neck trauma, impairment of brain growth and development, asphyxiation, and eye trauma (14), as other possible more severe complications. None of these complications have been reported in any of the studies conducted around the use of cranial orthosis in its use of over 30 years, therefore it is unlikely that these complications have occurred without having been mentioned.

1.7.3.2. *Parental concern regarding cranial orthosis*

The main decision regarding whether a cranial orthosis will be placed on the infant is up to the parents. A cranial orthosis should not be placed on an infant whose parents have major concerns regarding the treatment as it is a device that must be placed on the child during approximately 23 hours a day for 2 to 6 months depending on the severity of the deformation and the age of the start of the treatment (46). This is a very big commitment that the parents must agree upon. The orthotic treatment is not only for a lengthy time, but it also must have close monitoring by the parents to prevent improper fits that could potentially cause skin damages (46). Routine monitoring and adjustments made by a therapist are also essential in the

correct management of this type of treatment (28). An improper fit would entail an unnecessary compressive force applied on the child's head, which is the moment when the complications could arise. The helmet is not meant to exert a large force of compression on the head, it simply should guide the posterior cranial growth through mere contact (28). This is a reason why cranial orthosis manufacture are FDA regulated and therapist monitored in addition to the parents monitorization.

1.7.3.3. Cost

Another downside that this type of treatment has is its very high cost that makes it an unavailable choice for families that can't afford it. The price range ranges and differs from country to country but in general a cranial orthosis in the United States usually ranges from \$1500 to \$3000, including the helmet and the corresponding follow-ups (49). However, if there is significant cranial growth and a new helmet is necessary, this price would grow exponentially. The degree of insurance coverage of this cost varies widely as well, making the true cost of this treatment versatile, but nonetheless expensive for most families. In a study conducted by Lam et al. it was found that families that had public insurance were less likely to adhere to the treatment rather than those with a private insurance (49), probably due to the lack of coverage for the necessary follow-ups by the public insurance. Overall, the general consensus is that the coverage for low-income families for a cranial orthosis is insufficient for them to continue the treatment plan, affecting the treatment accessibility, and therefor can lead to the development of more severe deformities in low-income households (49). The high cost for cranial orthosis has skewed the perception and discussion of this type of treatment, bringing industry pressure and marketing competition into what should be a medical deliberation (14)

2. HYPOTHESIS

An exponential increase in the prevalence of positional plagiocephaly, due to the 1992 American Academy of Pediatrics “*Back to Sleep*” campaign, has caused it to be an emerging concern among parents. Further investigation regarding positional plagiocephaly is crucial due to the significance of early detection, and prompt treatment initiation to diminish its long-term effects. Positional plagiocephaly is a condition caused by external forces applied to the child’s skull, something that with proper education can be reduced significantly. Currently, treatment options for positional plagiocephaly are wide and lack a consensus to which intervention is the most effective.

This systematic review aims to determine the efficacy of the cranial orthosis to be superior compared to alternative treatments for positional plagiocephaly such as repositioning or physical therapy.

3. OBJECTIVES

Main objective:

- Conduct a systematic review to evaluate the efficacy of cranial orthosis as a treatment of positional plagiocephaly, aiming to consolidate the understanding of its effectiveness.

Secondary objectives:

- Compare the cranial orthosis to other forms of treatment used for positional plagiocephaly.
- Evaluate if the cranial orthosis is a cost-effective form of treatment.
- Determine if the timing of the treatment onset is crucial for the effectiveness of cranial orthosis.

4. METHODS

4.1. Methodological approach

To accomplish the objectives, a systematic review was conducted based upon various articles that evaluated and compared the forms of treatment used to treat positional plagiocephaly retrieved through a literature review. The review delves into the effectiveness, validity and beneficial results of cranial orthosis documented in the databases.

All the data used in this study was obtained from direct consultation and access via internet to the scientific literature in the databases, Medline (PubMed), Web of Science, and Google Scholar according to the following search strategies.

4.2. Search strategy

This systematic review was conducted in accordance with the criteria set out in the 2020 Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) (50). The literature systematic search, conducted from February to March 2024, employed a search strategy which combines the use of MeSH (Medical Subject Headings) thesaurus and free text in the “title” and “abstract” fields. The terms used in the search strategy included “Positional Plagiocephaly”, "Cranial Asymmetry", “Therapeutics”, "Cranial Orthosis", and "Helmet Therapy", combined using the Boolean operators “AND” and “OR”. The search was conducted in two phases to gather the necessary information. First, an unfiltered bibliographic search was carried out to obtain a broad spectrum of data regarding positional plagiocephaly, all of its forms of treatment, and the functionality of the cranial orthosis as a form of treatment. This was essential to have an unrestricted range of articles to compile as much information as possible on the topic. Subsequently, a refined search with specific filters applied to it targeted Randomized controlled trials and Clinical trials that met the inclusion and exclusion criteria outlined in this review within the search results.

4.3. Eligibility criteria

Inclusion Criteria:

- Infants diagnosed with positional plagiocephaly
- Use of cranial orthosis as a form of treatment
- Compared to non-treatment, other forms of treatment, or combined

- Clinical trials and randomized controlled trials

Exclusion Criteria:

- Studies conducted on premature population, or children older than 40 months
- Subjects with true craniosynostosis
- Articles that don't use a cranial orthosis as a treatment option, such as an assistive device, or cranial cup
- Studies that seek out prevention of plagiocephaly, not treatment
- Studies in other languages besides English or Spanish
- Studies carried out before the year 2004

4.4. Article selection

Once the search was completed, the main investigator analyzed the titles, abstracts, and keywords of all the articles to assess their relevance with respect to the defined inclusion criteria.

The articles selected were carefully reevaluated. Those with clear exclusion criteria were discarded. Subsequently articles will be selected after evaluating the full text, those that address the objectives of our review were the ones selected. In addition, the bibliography of the included articles will be reviewed, in case there are any other studies that could be introduced into the review.

4.5. Study limitations

It is possible that, because of the constructed search strategies, there are relevant studies according to the main objective that have not been detected. However, to attempt to reduce this possibility different expressions of the same concept or synonyms have been included in the search.

Given that the selection process involved only the principal investigator, it is possible that some significant studies, important to this research, could have been overseen. However, to minimize this possibility, each of the obtained studies has been analyzed in the most detailed and comprehensive way possible.

5. RESULTS

5.1. Articles included in the review

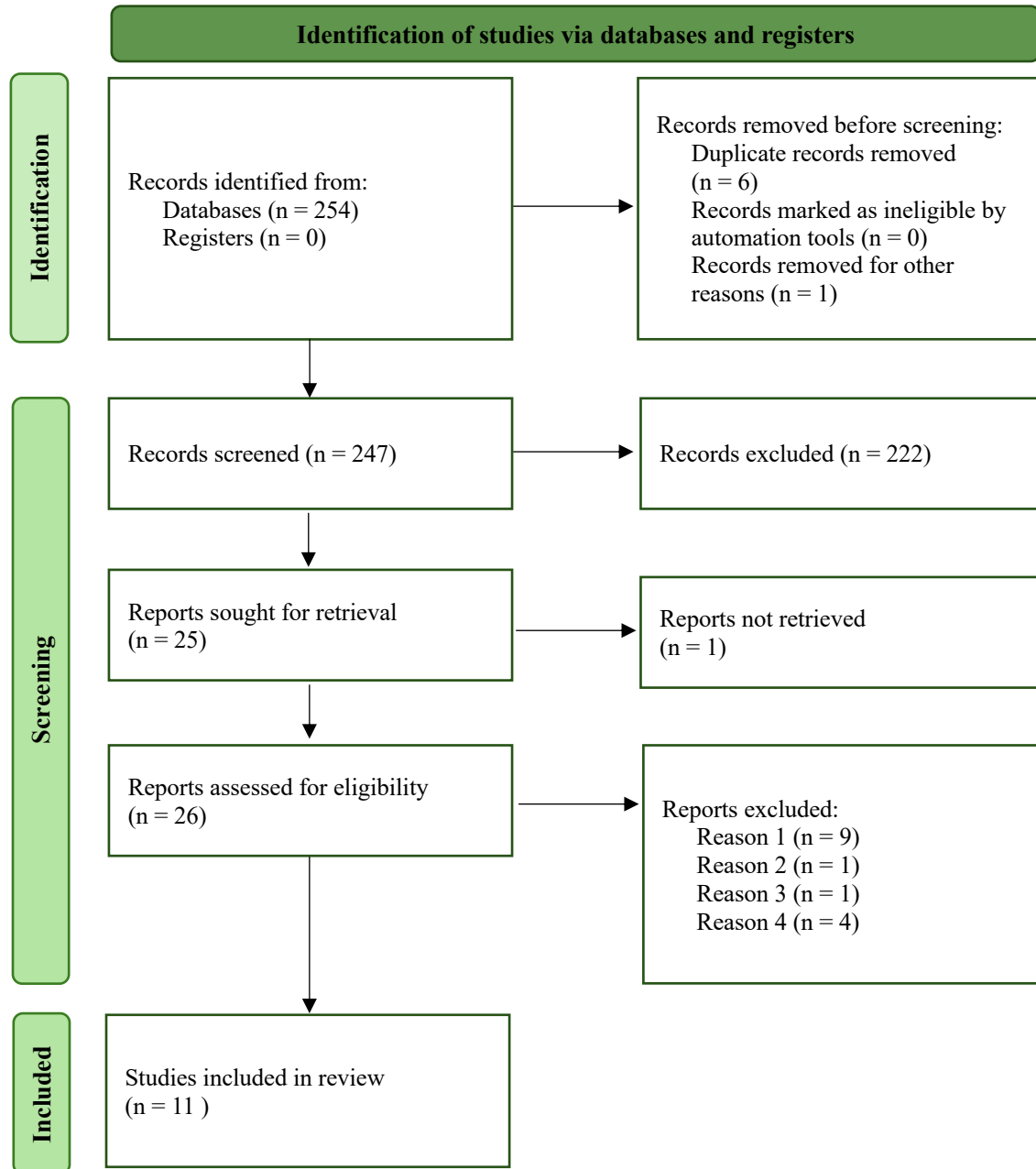


Figure 6 Flow diagram. Outline of gathering of studies via databases based on PRISMA 2020 (50).

5.2. Results table

Title	Authors	Year	Article type	Sample (n)	Results	Conclusions
Helmet therapy in infants with positional skull deformation: randomised controlled trial (51)	Renske M van Wijk, Leo A van Vlimmeren, Catharina G M Groothuis-Oudshoorn, Catharina P B Van der Ploeg, Maarten J Ijzerman, Magda M Boere-Boonekamp	2014	Pragmatic, single blinded, randomized controlled trial	84	The change score for both plagiocephaly and brachycephaly was equal between the helmet therapy and natural course groups, with a mean difference of -0.2 (95% confidence interval -1.6 to 1.2, P=0.80) and 0.2 (-1.7 to 2.2, P=0.81), respectively. Full recovery was achieved in 10 of 39 (26%) participants in the helmet therapy group and 9 of 40 (23%) participants in the natural course group (odds ratio 1.2, 95% confidence	Based on the equal effectiveness of helmet therapy and skull deformation following its natural course, high prevalence of side effects, and high costs associated with helmet therapy, we discourage the use of a helmet as a standard treatment for healthy infants with moderate to severe skull deformation.

					interval 0.4 to 3.3, P=0.74). All parents reported one or more side effects.	
Helmet treatment of deformational plagiocephaly: the relationship between age at initiation and rate of correction (52)	Mitchel Seruya, Albert K Oh, Jonathan H Taylor, Tina M Sauerhammer, Gary F Rogers	2013	Clinical trial	346	Initial transcranial difference was equivalent on all paired-group comparisons. Duration of helmet therapy positively correlated with age at initiation ($r = 0.89$, $p < 0.05$). The rate of change in transcranial difference correlated negatively with age at treatment onset ($r = -0.88$, $p < 0.05$): group 1, 0.93 mm/week; group 2, 0.64 mm/week; group 3, 0.59 mm/week; group 4, 0.56 mm/week; group 5, 0.41 mm/week; group 6, 0.42 mm/week; and	The correction rate of plagiocephaly with helmet therapy decreases with increasing infant age; after 32 weeks, there is a slow and relatively constant rate of change. Improvement can still be achieved in infants older than 12 months.

					group 7, 0.42 mm/week). At the conclusion of therapy, all groups had improved calvarial symmetry, albeit less completely in groups 6 and 7.	
Head orthosis therapy in positional plagiocephaly: longitudinal 3D-investigation of long-term outcomes, compared with untreated infants and with a control group (53)	Felix Kunz, Tilmann Schweitzer, Stephan Große, Nina Waßmuth, Angelika Stellzig-Eisenhauer, Hartmut Böhm, Philipp Meyer-Marcotty, Christian Linz	2019	Controlled clinical trial	45	Symmetry-related variables ($\Delta T1-T3$) improved significantly more in the treated than the control group, whereas these parameters did not differ significantly between the untreated and control group. Comparing the treated and untreated groups between T1 and T3, the reduction in the asymmetry at the treated group was significantly higher for the CVAI and	Head orthosis therapy in patients with DP leads to significantly better long-term outcomes. Facial asymmetries are more frequent in patients with DP who do not receive this treatment.

					<p>PCAI. In follow-up, the untreated group showed higher incidences of facial asymmetries than the treated group. Seventy-five per cent of all lateral crossbites found in patients with DP were contralateral to the posterior flattening.</p>	
<p>Helmet versus active repositioning for plagiocephaly: a three-dimensional analysis (54)</p>	<p>Angelo B Lipira, Shayna Gordon, Tron A Darvann, Nuno V Hermann, Andrea E Van Pelt, Sybill D Naidoo, Daniel Govier, Alex A Kane</p>	<p>2010</p>	<p>Comparative study</p>	<p>70</p>	<p>The helmeted group had a larger reduction than the repositioned group in both maximum (4.0% vs 2.5%; P=.02) and mean asymmetry (0.9% vs 0.5%; P=.02). The greatest difference was localized to the occipital region.</p>	<p>Whole-head 3D asymmetry analysis is capable of rigorously quantifying the relative efficacy of the 2 common treatments of DP. Orthotic helmets provide statistically superior improvement in head symmetry compared with active repositioning immediately after therapy. Additional studies are needed to establish the clinical significance of</p>

						these quantitative differences in outcome, define what constitutes pathologic head asymmetry, and determine whether superiority of orthotic treatment lasts as the child matures.
Effect of helmet therapy in the treatment of positional head deformity (55)	Juan Wen, Jun Qian, Lei Zhang, Chenbo Ji, Xirong Guo, Xia Chi, and Meiling Tong	2020	Retrospective study	376	Before treatment, no significant differences in CR, RSI, CVA and CVAI between groups were found. After treatment, compared with the postural correction training group, the helmet therapy group had significant improvements in CR, RSI, CVA or CVAI (Plagiocephaly: PCVA = 0.017, PCVAI = 0.028; Brachycephaly: PCR	Helmet therapy may be more effective in the treatment of mild–moderate–severe positional head deformity than postural correction training in infants. And helmet therapy may not hinder head circumference growth.

					= 0.002; Asymmetrical brachycephaly: PRSI = 0.002, PCVA < 0.001, PCVAI < 0.001). Moreover, there was no significant difference in head circumference growth between the groups.	
Efficacy of Cranial Orthosis for Plagiocephaly Based on 2D and 3D Evaluation (56)	Hiroki Kajita, Ichiro Tanaka, Hiroaki Komuro, Shigeru Nishimaki, Isao Kusakawa, Koichiro Sakamoto	2024	Retrospective study	1,038	The mean CA, CVAI, and anterior and posterior symmetry ratios improved significantly after cranial orthosis treatment. The improvement in each score was greater in patients with more severe initial deformities and in those who underwent treatment at a younger age.	Cranial orthosis treatment was effective in correcting deformational plagiocephaly in infants, as demonstrated by improvements in both 2D and 3D metrics. Patients with more severe initial deformities and those who underwent treatment at a younger age showed greater improvement.

<p>Three-dimensional analysis of positional plagiocephaly before and after molding helmet therapy in comparison to normal head growth (57)</p>	<p>Tilman Schweitzer, Hartmut Böhm, Christian Linz, Beatrice Jager, Lucia Gerstl, Felix Kunz, Angelika Stellzig-Eisenhauer, Ralf-Ingo Ernestus, Jürgen Krauß, Philipp Meyer-Marcotty</p>	<p>2013</p>	<p>Clinical trial</p>	<p>51</p>	<p>Compared with the control group, the plagiocephalic heads were more brachycephalic, but closely approximated the normal shape under molding therapy. The striking volume difference between the left and right posterior sections in the plagiocephalic children (the mean volume of the flattened side being 21% smaller than the one on the contralateral side) improved as well (to a residual difference of mean 8%) and ended up with a value close to the control group (mean 6%).</p>	<p>There is a broad clinical application area for stereophotogrammetry analyzing skull morphology: In plagiocephalic infants we demonstrate impressive changes of head shape under molding therapy; in normal-looking infants we describe the extent of unperceived asymmetry.</p>
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<p>Head orthosis therapy in infants with unilateral positional plagiocephaly: an interdisciplinary approach to broadening the range of orthodontic treatment (58)</p>	<p>P Meyer-Marcotty, H Böhm, C Linz, F Kunz, N Keil, A Stellzig-Eisenhauer, T Schweitzer</p>	<p>2012</p>	<p>Controlled clinical trial</p>	<p>40</p>	<p>Compared with the control group, infants with positional plagiocephaly demonstrated a reduced maximum length of the head, an increased head height, a shift in the ear axis as well as asymmetric anterior and posterior volumes of the neurocranium in lateral comparisons. Therapy using head orthosis led to a significant improvement of the asymmetry, with a reduction of the diagonal difference and an adjustment of the posterior volumes.</p>	<p>Conservative growth control of extrinsically deformed infant skulls represents an interdisciplinary medical expansion of the orthodontic therapeutic spectrum. To prevent potential effects of positional plagiocephaly on the viscerocranium, head orthosis therapy is advisable in infancy.</p>
<p>Effects of manual therapy on treatment duration and</p>	<p>I Cabrera-Martos, M C Valenza, G Valenza-Demet,</p>	<p>2016</p>	<p>Randomized controlled trial</p>	<p>46</p>	<p>Asymmetry after the treatment was minimal (type 0 or 1 according to the</p>	<p>Manual therapy added to standard treatment reduces the treatment duration in infants with</p>

<p>motor development in infants with severe nonsynostotic plagiocephaly: a randomised controlled pilot study (59)</p>	<p>A Benítez-Feliponi, C Robles-Vizcaíno, A Ruiz-Extremera</p>				<p>Argenta scale) in both groups. A comparative analysis showed that treatment duration was significantly shorter ($p < 0.001$) in the experimental group (109.84 ± 14.45 days) compared to the control group (148.65 ± 11.53 days). The motor behaviour was normal (scores above the 16th percentile of the AIMS) in all the infants after the treatment.</p>	<p>severe nonsynostotic plagiocephaly.</p>
<p>Objective Outcome Analysis of Soft Shell Helmet Therapy in the Treatment of Deformational Plagiocephaly (60)</p>	<p>Terrence W. Bruner, BS Lisa R. David, MD H. Donald Gage, PhD Louis C. Argenta, MD</p>	<p>2004</p>	<p>Clinical trial</p>	<p>69</p>	<p>For children who were compliant with treatment, intracranial volume asymmetry was decreased by 39.1% and 36.2% for the hemispheres and posterior quadrants,</p>	<p>There was a 36% to 54% improvement in asymmetry in the compliant patients over the 6-month study period. Soft shell helmet therapy is an effective technique to decrease cranial asymmetry based on</p>

					<p>respectively. For the hemispheres (left versus right), $P < 0.001$. For the posterior quadrants (back left versus back right), $P < 0.001$. The P value for the front quadrants was not significant at 0.357.</p>	<p>objective outcome measurements. Additionally, it is cost-effective, with the total cost of therapy for the helmet and office visits ranging from \$600 to \$700. This therapy compares favorably with other more expensive and time-consuming therapies that have been reported in the literature.</p>
<p>Management of deformational plagiocephaly: repositioning versus orthotic therapy(61)</p>	<p>John M. Graham, Jr, Md, Scd, Mayela Gomez, Bs, Andy Halberg, Bs, Dawn L. Earl, Cnp, Msn, Jeannie T. Kreutzman, Cnp, Msn, Jinrui Cui, Ms, And Xiuqing Guo, Phd</p>	<p>2005</p>	<p>Comparative Study</p>	<p>176</p>	<p>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</p>	<p>Infants treated with orthotics were older and required a longer length of treatment (4.2 vs 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</p>

					(from an initial mean DD of 1.13 cm)	orthosis was significantly more effective than later orthosis (65% decrease versus 51% decrease in DD).
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Table 4 Results of the 11 articles chosen

6. DISCUSSION

Positional plagiocephaly is a skull deformity caused by the application of external forces to a baby's head that lead to the flattening of the skull. Some other prenatal, perinatal, and postnatal factors also play a role in the development of this deformity. An infant's skull is considered to be more malleable to help through the birth process, as the infant gets older their skull becomes harder. Although this softer skull is a normal part of a child's development, any pressure on the skull can lead to its deformity (62). As a consequence of the 1992 American Academy of Pediatrics "*Back to Sleep*" campaign the prevalence of positional plagiocephaly has skyrocketed. The campaign was developed as a way to reduce the sudden infant death syndrome rates by placing children in the supine position to sleep. As this has significantly reduced the mortality rate due to SIDS, it has increased the positional plagiocephaly incidence. Before the "*Back to Sleep*" campaign, many infants treated themselves by turning their heads while sleeping in the prone position (61).

Treatment options for positional plagiocephaly include repositioning, physical therapy, or cranial orthosis. Repositioning mainly focuses on avoiding pressure in the same area of the skull, the flattened area, and laying the child on the opposite area of the skull. Physical therapy is normally prescribed when the infant has torticollis associated to the positional plagiocephaly, to try and optimize its neck mobility. Cranial orthosis involves the constant wearing of a repositioning helmet that permits the skull to grow in the areas that are desired where the helmet leaves area to grow.

As a result of the increasing incidence of positional plagiocephaly there has also been an increasing number of studies carried out regarding the use of a cranial orthosis as the principal form of treatment for this pathology. The main concerns regarding this treatment method are its high costs, possible side effects, and alternative treatment options that could reach the same outcome. At present, there has not been a definitive agreement on the best treatment as many factors play into the efficacy of the helmet therapy. This review has been conducted to reach a consensus among the different studies present on the advantages of using a cranial orthosis as the primary treatment method for correcting skull deformation in positional plagiocephaly, and to clarify all the long-term benefits that it the best option for treatment. Of the eleven articles chosen in this review, there were 2 randomized controlled trials, 5 clinical trials, and 4 observational studies including both comparative and retrospective studies. The results demonstrate a consistent trend towards significant improvements in cranial asymmetry among infants treated with cranial orthoses, suggesting that this intervention is effective across varying degrees of severity and initial conditions.

Diving into helmet therapy versus the other conservative types of treatments, 8 of the studies directly conclude that the use of cranial orthosis has a significant improvement in the therapeutic outcome compared to the other treatment forms. Of all the studies considered, only those carried out in the last twenty years were considered for this review. In 2004, Terrance et al. conducted a clinical trial including 69 children to analyze the objective outcome of helmet therapy in the treatment of deformational plagiocephaly. This was achieved through CT scanning to retrieve the measurements of the children's skull, eliminating any subjectivity from the measurement process that could be present in other measuring techniques such as anthropometric measurements, head tracings, clinical experience, and parental assessment which are observer dependent (60). This makes the conclusions reached by Terrance et al. more reliable, they concluded that helmet therapy is an effective technique to decrease cranial asymmetry (60). This study also investigated one of the other main concerns regarding this form of treatment, its cost, concluding that there is a cost-effective method to treat positional plagiocephaly, by using a generic-sized soft foam helmet that was fitted to patients by adding extra foam padding. This form of cranial orthosis could be the response to the high-cost concerns of many families when given this treatment option. In subsequent studies, all measured by 3D scanning and comparing helmet therapy directly with repositioning therapy (54,55,61) helmet therapy was considered more effective in the treatment of all degrees of deformity, from mild to severe. Graham et al. concluded that children treated with an orthotic helmet resulted in a 61% decrease in the diagonal difference compared to 52% difference in the group of children treated with repositioning therapy only (61). Lipira et al. demonstrated a significant reduction in treatment duration with the cranial orthosis, 3.1 months, versus repositioning, 5.2 months. Their findings demonstrated that helmeted patients reduced the asymmetry in CVA of 4.5mm while the repositioning group changed in 3.4mm (54), in a shorter period of time.

The skull deformation in positional plagiocephaly causes for there to be different types of facial asymmetries, which is one of the main concerns that parents experience when their child has deformational plagiocephaly. These facial asymmetries include the deviation of the dental midline and chin towards the unaffected side, a displacement of the glenoid fossa, malocclusion (32), anterior displacement of the temporomandibular joint and a certain degree of auricular displacement (33). Kunz et al. analyzed the prevalence of facial asymmetries post treatment in both of the groups in the study, one being the control group and the other the helmeted group. By the end of the treatment period, Kunz determined that the untreated group had higher incidences of these facial asymmetries compared to the treated group (53). Similarly, Meyer-Marcotty et al. established that head orthosis therapy leads to a significant improvement in the neurocranial asymmetry that entailed a decrease in the diagonal difference and correction of the ear shift axis as well (58). The study emphasized the interdisciplinary benefits of cranial orthosis in preventing potential craniofacial developmental issues.

Another important factor that plays into the efficacy of treatment in positional plagiocephaly is timing. Timing as a concept looked at in two different ways, one as the age of initiation of treatment, and also timing as in the duration of the intervention to achieve desired outcomes. Normally the use of cranial orthosis is used as a second treatment option once the other forms of treatment don't reach the preferred results. This makes the age of initiation of cranial orthosis therapy to be later than it should be. With a child's head becoming less malleable as it ages, the length of treatment and desired outcome is affected with the latter start. Seruya et al. conducted a study to investigate the age at initiation and rate of correction. At the time that the study was conducted, there was a general consensus that there is a faster and more complete correction of deformation in younger patients, yet the optimal age was unclear (52). This study concluded that there is an inverse correlation between the age of correction of calvarial asymmetry and the age of initiation (52). Meaning, the older the child gets, the less degree of correction the helmet achieves in the deformation, although significant improvements were still achievable in infants older than 12 months, just taking a longer period of time to achieve it. Helmet therapy relies on remaining brain growth to redirect the head shape. The brain grows to 200% of its birth size by 6 months of age and reduces its growth rate significantly to reach an additional 50% over the next 24 months (52). With this being said, cranial orthosis is not counter indicated in children over 6 months, but it will render a much lower success rate and will take a longer period of time to achieve it.

In a randomized controlled pilot study that took place in 2016, the use of manual therapy as an adjuvant treatment for cranial orthosis was investigated. This was sought out to achieve a lower treatment period, while reaching adequate motor development. Cabrera-Martos et al. concluded that by adding manual therapy once a week to the standard treatment, the duration of the treatment reduces significantly, possibly reducing the concern of a lengthy helmet wear time on young children (59). This possible treatment option should be considered, and studied further, as it could be the resolution to one of the biggest problems that arises with cranial orthosis treatments.

There is only one Class I study on this topic to this day, the randomized controlled trial carried out by van Wijk et al. (51). In this trial, infants with plagiocephaly treated with cranial orthosis were monitored during their first two years of life post treatment at 6 months, compared to a control group in which they left the deformity take its natural course, without treatment. This was the only study present in which the results showed no significant improvement in skull deformity. This randomized controlled trial (RCT) involved 84 infants and reported similar improvements in cranial asymmetry for both groups, with a mean difference for plagiocephaly change scores of -0.2 (95% CI -1.6 to 1.2, $P=0.80$). Full recovery rates were also comparable, at 26% for helmet therapy and 23% for natural course treatment, with an odds ratio of 1.2 (95% CI 0.4 to 3.3, $P=0.74$) (51). There is a slight improvement in the measurements, but the authors

did not consider them as clinically significant enough. No children with severe plagiocephaly (oblique diameter difference index $>113\%$ or cranio-proportional index $>104\%$) were included in the trial, which could alter the results, as it is harder for a severe degree of plagiocephaly to resolve itself overtime without any form of treatment. On the other hand, parent satisfaction scores with results were significantly higher in the helmet group compared to the untreated group. This factor is very important as the parents also presented lower anxiety regarding their children's deformity in the treated group. There has been no significant difference in the quality of life between the two groups, but there seemed to be a better sleep quality in the helmeted group (helmet therapy 5/35, 14%; natural course 10/41, 24%) who also spent less hours crying than the natural course group (helmet therapy mean 1.4 (SD 1.2); natural course mean 1.2 (SD 0.9)) (51). The general consensus on this randomized controlled study, after the two-year follow-up is that cranial orthoses are discouraged as a form of treatment as they will achieve similar results as not treating the deformity at all.

Conversely, in a recent retrospective study conducted on 1,038 patients to analyze the efficacy of the cranial orthosis using both 2D and 3D evaluations, Kajita et al. highlighted significant differences between the group treated with a helmet and the control group (56). They concluded that there is a higher effectivity rate in the correction of skull deformity in both CA, CVAI and anterior and posterior symmetry ratios (ASR and PSR). The improvement in each score was greater in patients with more severe initial deformities and in those who underwent treatment at a younger age (56). The combination of a large sample size, the study's recent advancements, and thorough evaluation of measurements in both 2D and 3D assessments render this study highly reliable when considering its outcomes.

The results in this study correlate with most of the reviews on the use of cranial orthosis for positional plagiocephaly up to date. The general consensus is the positive outcome that a cranial orthosis treatment is effective as a form of treatment and physicians should implement it as soon as possible to reach the best outcomes.

Although the clinical indications for helmet therapy in deformational plagiocephaly are not yet fully defined, a high level of evidence suggests that cranial orthosis treatment is the most adequate and will reach the best outcomes to treat positional plagiocephaly.

As positional plagiocephaly is still a very common skull deformity, additional randomized controlled studies are necessary to reach further Class I evidence to support the use of cranial orthoses. Future research should aim to identify subgroups of infants who are most likely to benefit from helmet therapy. Long-term follow-up studies are needed to assess the durability of treatment outcomes and potential impacts on neurodevelopment. Comparative

studies focusing on cost-effectiveness and quality of life measures will also help refine treatment guidelines.

In conclusion, while helmet therapy appears effective in many cases, its necessity and superiority over natural recovery remain debated. The findings of van Wijk et al. serve as a reminder of the importance of high-quality evidence and the need for balanced, informed decision-making in clinical practice.

7. CONCLUSION

Positional plagiocephaly, an increasingly recognized condition, instills significant concern among parents and therefore reaching a consensus on the use of cranial orthosis for its treatment is crucial.

Through the 11 trials analyzed in this study, 10 of them describe a significant increase in clinical effectiveness on treatment with cranial orthosis. Although the only Class I trial's overarching results deem that using a cranial orthosis as a form of treatment is discouraged, we believe that the helmet should be considered in each individual case as it has been proven by numerous other studies to be beneficial. Considering these results, a helmet may not be necessary for all of the patients that debut with a skull deformity that is not associated to any other pathology, yet it is beneficial to most cases of positional plagiocephaly. A helmet treatment could provide in addition to a correction in the cranial deformity indexes, other types of benefits to the child and visually parents tend to result more satisfied with the outcome.

With the results obtained in this study, we have concluded that the use of a cranial orthosis as a treatment for positional plagiocephaly should be individualized for each patient. It is essential to personalize treatment plans based on the severity of the deformity, the presence of facial asymmetries, and the timing of intervention. Early diagnosis and intervention, ideally before six months of age, are crucial for maximizing the effectiveness of the treatment and preventing irreversible cranial deformities. The importance of avoiding missing valuable opportunities for treatment at an early age is crucial. Skipping this window of time may cause irrecoverable changes in the child's skull shape. Clinical attention and early intervention are necessary to address these types of deformities at an early age and provide cranial orthosis as a treatment option from the beginning. Moreover, it is important to address the variability in treatment accessibility due to cost and insurance coverage disparities. The high cost of cranial orthoses can limit treatment options for low-income families, potentially leading to more severe deformities in these populations. Future research should focus on cost-effectiveness and explore ways to make this treatment more accessible.

Ultimately, cranial orthosis should be considered a viable and often beneficial treatment option for positional plagiocephaly, with the potential to significantly improve cranial symmetry and parental satisfaction. However, its application should be tailored to individual patient needs, and efforts should be made to ensure broader accessibility and affordability. Early clinical intervention remains paramount in achieving the best possible outcomes for infants with positional plagiocephaly.

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Plagiocefalia: Uso de la ortesis craneal para su tratamiento

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INTRODUCCIÓN

La plagiocefalia es una patología caracterizada por el aplanamiento asimétrico del cráneo de un bebé. La prevalencia de esta anomalía ha aumentado considerablemente tras la campaña “Back to Sleep” de la Academia Americana de Pediatría, lo que ha hecho que sea fundamental encontrar la mejor opción de tratamiento. Las opciones actuales de tratamientos se centran en el reposicionamiento, la fisioterapia o la órtesis craneal, un dispositivo similar a un casco que aplica presión para corregir la forma del cráneo.

MATERIAL Y MÉTODOS

Revisión sistemática de la literatura siguiendo las normas de la guía PRISMA. Se realizó una búsqueda en las bases de PubMed y Web of Science con los términos: “positional plagiocephaly” OR “deformational plagiocephaly” AND “cranial orthosis,” OR “helmet therapy.”

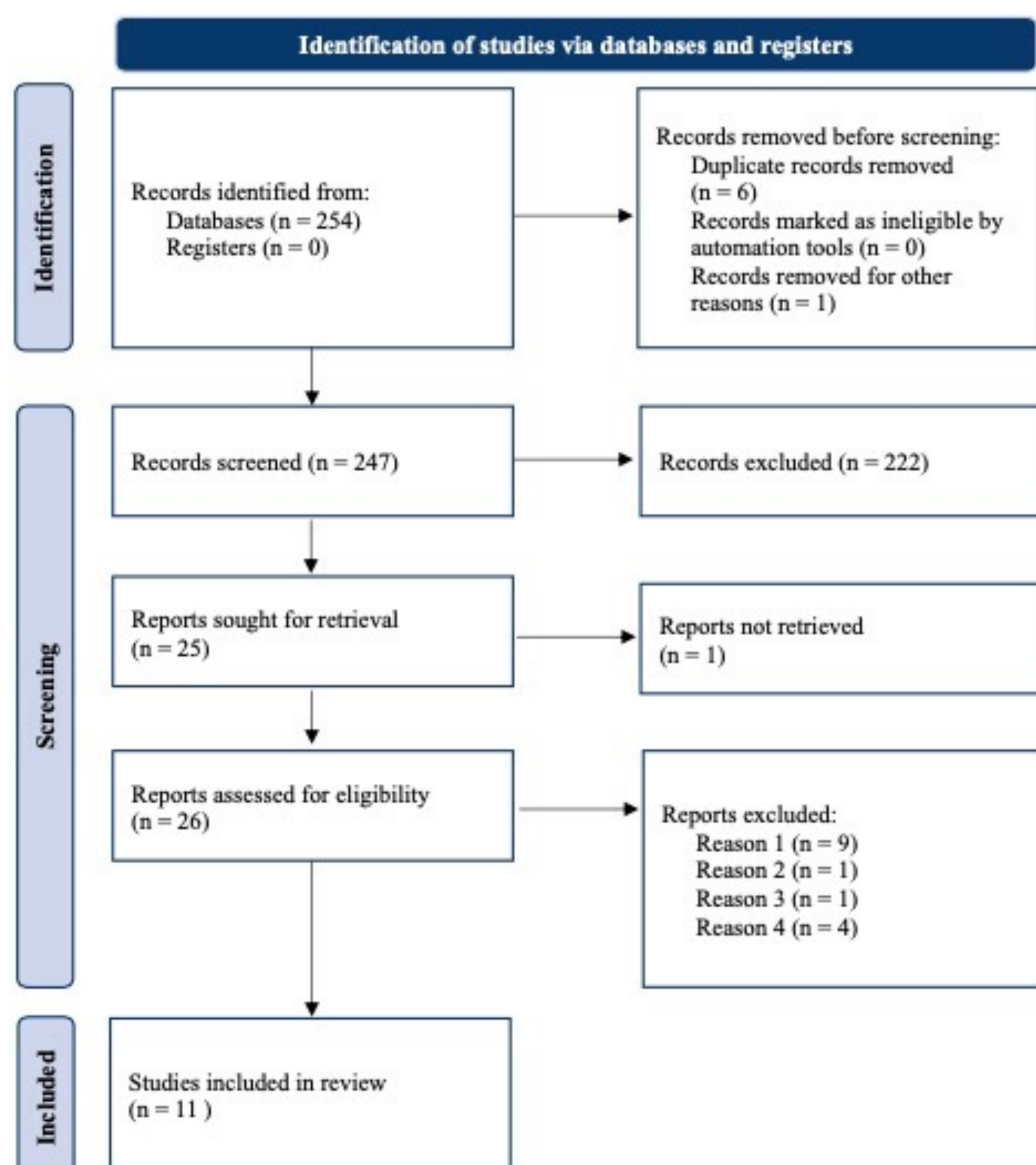


Figura 1: Diagrama de flujo basado en PRISMA 2020. ⁵⁰

HIPÓTESIS Y OBJETIVOS

Evaluar la eficacia de la ortesis craneal como tratamiento para la plagiocefalia posicional, comparar la ortesis craneal con otras opciones de tratamiento como el reposicionamiento y la fisioterapia, y determinar el momento óptimo para iniciar el tratamiento. La hipótesis de partida es que la ortesis craneal es un tratamiento efectivo y superior para la plagiocefalia posicional en comparación con los métodos alternativos, proporcionando una mejora significativa en la simetría craneal con efectos secundarios mínimos.

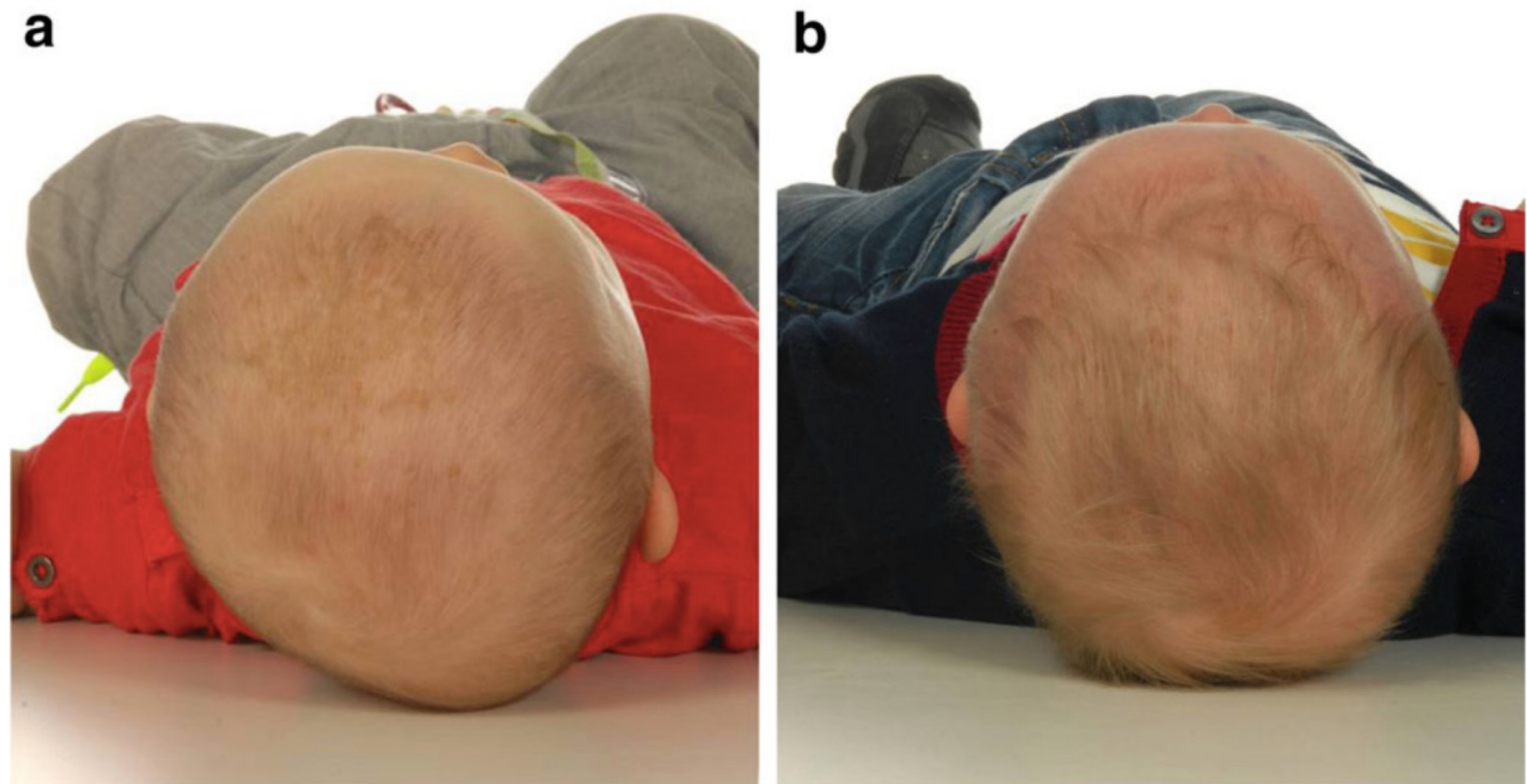


Figura 2: Niño con plagiocefalia. a. Antes de ser tratado con ortesis craneal b. después del tratamiento con ortesis craneal. ³²

RESULTADOS Y DISCUSIÓN

El uso de la ortesis craneal como tratamiento:

1. Mejora en simetría craneal:

- La ortesis craneal mostró una mejora significativa en la simetría craneal comparada con otros tratamientos como el reposicionamiento y la fisioterapia.
- En un estudio, los niños tratados con ortesis presentaron una disminución del 61% en la diferencia diagonal en comparación con el 52% en el grupo tratado solo con reposicionamiento.

2. Reducción en el tiempo del tratamiento:

- En un estudio, el tiempo promedio de tratamiento con ortesis fue de 3.1 meses frente a 5.2 meses con reposicionamiento.

3. Efectividad según la gravedad y la edad:

- Los pacientes con deformidades más severas y aquellos tratados a una edad más temprana mostraron una mayor mejora con tratamiento ortésico.

Los resultados de esta revisión se correlacionan con la mayoría de los estudios sobre el uso de ortesis craneales para la plagiocefalia posicional realizadas hasta la fecha. El consenso general es que el tratamiento con ortesis craneal es eficaz y debe aplicarse lo antes posible para lograr los mejores resultados. Aunque las indicaciones clínicas de la terapia con casco en la plagiocefalia aún no están totalmente definidas, un alto nivel de evidencia sugiere que el tratamiento con órtesis craneal es el enfoque más adecuado. Dado que la plagiocefalia posicional sigue siendo una deformidad craneal común, son necesarios estudios adicionales de Clase I para obtener más evidencia que respalde su uso.

CONCLUSIÓN

- Las ortesis craneales han mostrado eficacia en 10 de 11 ensayos, aunque un ensayo de Clase I desaconseja su uso.
- Es esencial individualizar el tratamiento, considerando la severidad de la deformidad, las asimetrías faciales y la intervención temprana antes de los seis meses.
- Los altos costos pueden limitar el acceso, por lo que la investigación futura debe mejorar la rentabilidad y la accesibilidad.
- En general, las ortesis craneales son una opción de tratamiento viable, mejorando la simetría craneal y la satisfacción de los padres.

BIBLIOGRAFÍA

