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EARLY DIAGNOSIS AND TREATMENT OF CHILDREN WITH SKULL DEFORMATIONS. THE CHALLENGE OF MODERN MEDICINE

WCZESNE ROZPOZNAWANIE I LECZENIE DZIECI ZE ZMIANAMI DEFORMACYJNYMI CZASZKI. WYZWANIE WSPÓŁCZESNEJ MEDYCYNY

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Abstract

Skull deformations affect approximately 45% of newborn babies, the most common ones being: plagiocephaly, brachycephaly and scaphocephaly. The first symptoms can be observed 4 to 8 weeks after birth. The causes of skull deformation in newborns can be divided into congenital ones and those acquired after birth. An increase in the incidence of acquired head deformations can be attributed to the "BACK TO SLEEP" campaign, carried out in 1992 by the American Academy of Pediatrics (AAP), which was aimed to reduce the frequency of sudden infant death syndrome (SIDS) by placing babies to sleep in the supine position. By the year 2000, the number of SIDS incidents had been significantly lowered, however, it seems that this improvement was achieved at the cost of an increased number of head deformations [3, 4, 5, 34].

Skull deformations, if left untreated, may have consequences for the future. Plagiocephalic deformations may be associated with delayed intellectual and motor development [2].

Early recognition of the condition and the appropriate classification of each skull deformation are crucial for the success of the treatment [8]. Treatment choice depends on the etiology of the problem and its severity, as well as on the age of the infant. Available options include training for the parents/ caregivers, physical therapy, custom head orthosis and surgical intervention.

Key words: *"oblique head", plagiocephaly, brachycephaly, scaphocephaly, rehabilitation, orthotic equipment, helmet therapy*

Streszczenie

Deformacje czaszki występują u około 45% niemowląt. Najczęściej stwierdzane są: skośnogłowie (plagiocefalia), krótkogłowie (brachycefalia) i łódkogłowie (skafocefalia). Pierwsze objawy zauważane są najczęściej pomiędzy 4 a 8 tygodniem życia. Przyczyny deformacji czaszki można podzielić na wrodzone i nabyte po urodzeniu. Wzrost częstości nabytych deformacji czaszki można powiązać z prowadzoną od roku 1992 przez Amerykańskie Towarzystwo Pediatryczne (AAP) kampanią społeczną "BACK TO SLEEP", mającą na celu zapobieganie nagłej śmierci niemowląt (SIDS) poprzez kładzenie dzieci w pozycji na wznak. Do roku 2000 liczba przypadków SIDS istotnie się obniżyła, zwiększyła się jednak częstość deformacji czaszki [3, 4, 5, 34].

Deformacje czaszki, pozostawione bez leczenia, mogą mieć konsekwencje w przyszłości. Deformacje plagiocefaliczne mogą być związane z opóźnionym rozwojem intelektualnym i motorycznym [2]. Wczesne rozpoznanie nieprawidłowości i ich prawidłowa klasyfikacja są punktem wyjścia skutecznego leczenia. Wybór metody leczenia zależy od etiologii deformacji oraz jej ciężkości, a także od wieku dziecka. Opcje terapeutyczne obejmują szkolenie rodziców/opiekunów w prawidłowej pielęgnacji dziecka, fizjoterapię, indywidualnie przygotowywane ortezy głowy oraz leczenie chirurgiczne.

Słowa kluczowe: *skośnogłowie, plagiocefalia, krótkogłowie, brachycefalia, łódkogłowie, skafocefalia, rehabilitacja, orteza, kask terapeutyczny*

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INTRODUCTION

The problem of skull shape accompanied men since time immemorial, the oldest records dating from Peru 30,000 years ago [1]. In those days it was believed that shaping the skull in a particular way would guarantee the acquisition of specific skills required for high positions in society, so skulls were deliberately disfigured. Today we face the opposite situation: we try to protect the physiological "round" shape of the child's head and any deformations are unwelcome.

The first publications identifying skull deformities as a problem in newborn children date back to 1936. It took another forty years until the first attempts to supply children with head orthoses took place in 1970 [1].

Nowadays skull deformations are diagnosed in 45% of newborn babies, the most common ones being: plagiocephaly, brachycephaly and scaphocephaly. The first symptoms are observable between the 4th and 8th week of life [2, 3, 4, 5].

ETIOLOGY

The causes of skull deformation in newborns can be divided into congenital ones and those acquired after birth. Risk factors for deformities *in utero* include: genetic syndromes, multiple pregnancy, high fetal weight, a small uterus, uterine malformations, traumatic birth, male gender and metabolic disorders during pregnancy [3, 6]. Risk factors for postnatal deformities include: prematurity, intensive care of the newborn, the high weight of the child, low activity, muscle tone asymmetry, *torticollis*, disorders of bone mineralization, decreased muscle tone and "favorite position" [7].

In the 1990's developed countries all over the world started reporting an increasing frequency of head deformations in children. This turned out to be closely related to the "back to sleep" campaign, initiated in 1992 by the American Academy of Pediatrics (AAP) in order to reduce the incidence of sudden infant death syndrome (SIDS) by placing babies to sleep in the supine position [4,8]. Before the introduction of this program, the number of children with "oblique head" was estimated at 1: 300 births. Current estimates are much higher, as high as 1: 60 births [9]. This is because the baby's skull is very soft and plastic. Positional head deformity is caused by external pressures on the rapidly developing skull at work when the same lying position is kept for a longer time [7].

By 2000, the number of SIDS had significantly decreased. However, in many cases the recommendations of the AAP were understood too literally, so that more and more caregivers refrained from putting their children in positions other than lying supine. In consequence, the flattening of the skull in children became a growing issue [7, 8].

EPIDEMIOLOGY AND CLASSIFICATION

The most frequently observed deformities of the skull are: plagiocephaly, brachycephaly and scaphocephaly.

Plagiocephaly, or "oblique head", is a deformity characterized by the flattening of one side of the posterior section of the head with compensatory ipsilateral frontal bossing.

The clinical symptoms of plagiocephaly include: craniofacial asymmetry, the ear on the affected side is situated ventrally, a higher setting of the eye on the flattened side. Plagiocephaly was diagnosed in 1: 300 live births in 1976, nowadays it is found in 1:60 live births [10].

The deformity is more common among males and is more frequently right-sided [10, 11, 12].

Around 62% of all children have flattening of the right side [9]. The main cause of oblique head after delivery is repeated external pressure to the one side of a rapidly growing skull; the most common reason (88%) for this being a fixed, unchanged position during sleep [7, 9, 11, 12].

Positional plagiocephaly should be differentiated from torticollis, which may also cause head deformity [9, 13]. Torticollis is a congenital muscular dystrophy, which is a consequence of abnormal blood flow through the muscles, leading to their fibrosis. Reduced muscle length puts the baby's head in a forced position, resulting in the unilateral flattening of the posterior section of the head. This problem may affect up to 85% of the children with plagiocephaly.

Plagiocephaly could also be related to the premature closure of the lambdoid suture, resulting in a type of craniostenosis (fig. 1).

Brachycephaly, the second most common deformity, is called short head syndrome [6]. It is characterized by flattening of the entire occipital part of the skull, resulting

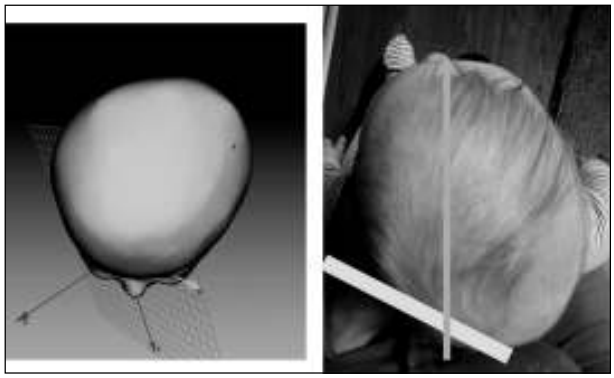

Fig. 1. Plagiocephaly- top view; scan (left), photo of a 6 months boy (right) (courtesy of VIGO Ortho Sp. z o.o.).

Fig. 1. Plagiocefalia/Skośnogłowie- widoku z góry, skan (po lewej), zdjęcie chłopca w 6 miesiącu życia (po prawej) (udostępnione dzięki uprzejmości firmy VIGO Ortho Sp. z o.o.).

in the shortening of the anterior-posterior dimension and the compensatory expansion of the medial-lateral dimension. "Top of the head" is a phenomenon commonly associated with brachycephaly, when the cranial vault at the rear is in a higher position [2]. (fig. 2).

Scaphocephaly is another kind of skull deformation. It is also known as narrow head and is characterized by the lengthening of the anterior-posterior dimension with the reduction of the temporal dimension. Scaphocephaly prevalence is 1: 2000 of live births, and the condition is three times more common in boys. The premature closure of the sagittal suture seems to be the main cause of this type of craniostenosis. The next cause of craniostenosis can be the wedging of the child's head in utero, under the mother's ribs [13]. (fig. 3).

The term craniostenosis refers to the situation when one or multiple sutures of the skull close too early. Closing of the sutures can be observed even during

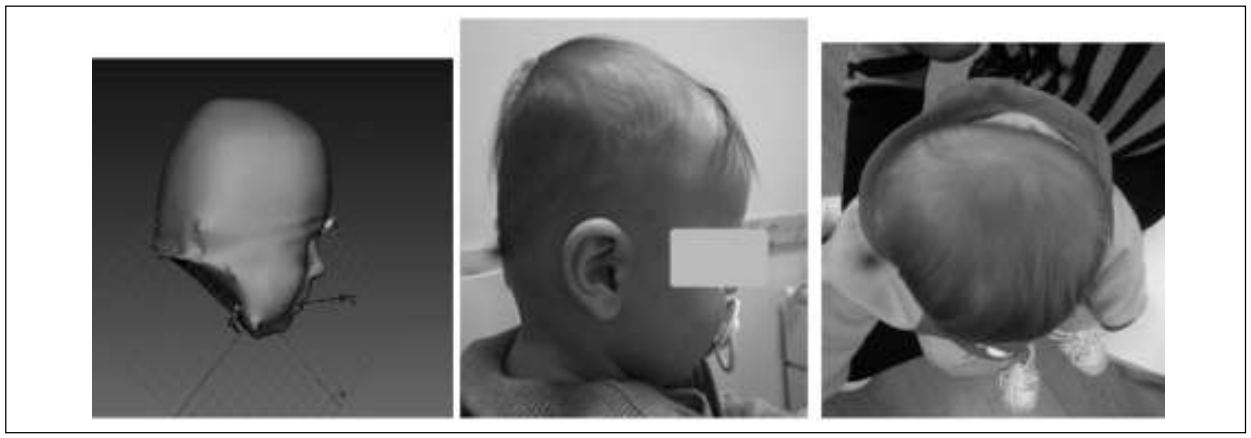

Fig. 2. Brachycephaly; from the left: 5 months boy scan; 7 months boy; 5 months girl (courtesy of VIGO Ortho Sp. z o.o.).

Fig. 2. Brachocefaila/Krótkogłowie; od lewej: skan 5 miesięcznego chłopca; 7 miesięcznego chłopca; 5 miesiecznej dziewczynki (udostępnione dzięki uprzejmości VIGO Ortho Sp. z o.o.).

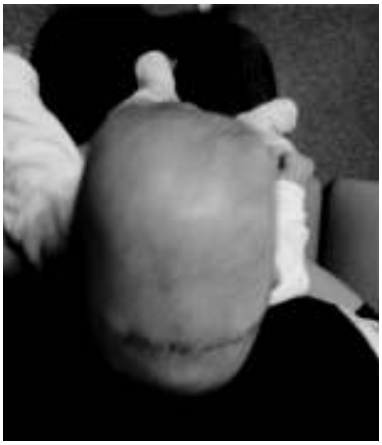

Fig. 3. Scaphocephaly due to craniostenosis. 3 months boy (courtesy of VIGO Ortho Sp. z o.o.).

Fig. 3. Scaphocephalia/Łódkogłowie spowodowane kraniosynostozą. 3 miesięczny chłopiec (udostępnione dzięki uprzejmości VIGO Ortho Sp. z o.o.).

pregnancy or in the first months of life. Plagiocephaly or scaphocephaly develop resulting from the premature closure of the lamboid or saggital suture, accordingly. 13-16% children with craniostenosis develop plagiocephaly [13]. Craniostenosis is the most dangerous deformation, as it can generate pressure on the brain with ensuing brain hypo-vascularity, increased intracranial pressure, developmental/ neuropsychiatric problems or problems with social adaptation. (fig. 4).

CONSEQUENCES

Neglected skull deformations may have unfavorable consequences in the future. Children suffering from plagiocephaly without adequate support were reported to have a five times higher risk of abnormal speech development and language deficits. Plagiocephalic deformations were recently found to be related to delayed intellectual and motor development [9, 13]. School-age children often need supportive education, speech therapy, physiotherapy

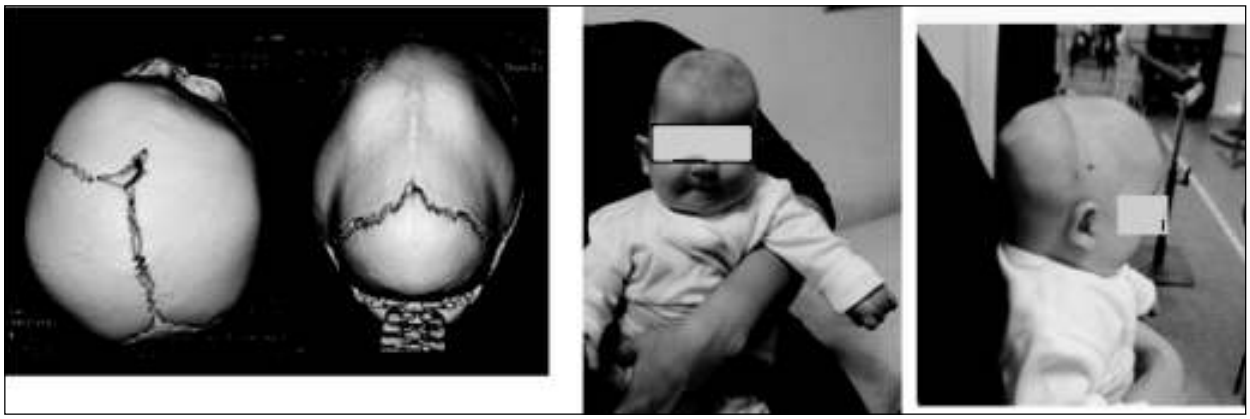

Fig. 4. Craniostenosis after surgery (courtesy of VIGO Ortho Sp. z o.o.).

Ryc. 4. Kraniosynostoza po korekcie chirurgicznej (udostępnione dzięki uprzejmości VIGO Ortho Sp. z o.o.).

or occupational therapy. Astigamtism is also frequently diagnosed in children with plagiocephaly. The aesthetic aspects of this condition include problems with fitting prescription glasses or difficulties with fitting protective helmets for sports activities, such as snowboarding or cycling.

TREATMENT

Our practice as rehabilitation medicine consultants shows that neonatologists and paediatricians do not refer patients to us due to skull deformities, but rather diagnose psychomotor developmental delay, positional asymmetries or torticollis.

The early recognition of skull deformation and precise classification are crucial factors for successful treatment [8]. Several treatment approaches can be considered, depending on the etiology, severity of the problem and the age of the patient, namely: training for the parents/caregivers, physical therapy, custom head orthosis and surgical intervention [8, 14, 15].

Caregivers training/behavioral training

The first step in approaching plagiocephaly is to teach caregivers how to carry the child correctly and what are the correct child-handling techniques, as well as to introduce a home-based exercise program of corrective positioning. In the literature, therapies that do not involve physical therapy *per se*, are usually based on everyday activities performed by the caregivers in a way which reshapes the head deformation [13]. Caregivers are also taught how to feed the child [7]. They are informed about the necessity of changing the position of the child quite often, with special regard towards lying prone and they learn how to arrange the positions in a proper way [16]. Sitting, e.g. in a car seat, should usually be avoided [8, 9]. Home-based exercise programs put stress on using the side of the child which is spontaneously less used [17]. Some reports found that caregiver training alone proves to be an effective intervention for plagiocephaly [18]. However, other studies indicate that full improvement can be achieved in only up to 30 percent of the cases and that intervention needs to be continued with a custom-made head orthosis [7]. In fact, repositioning itself and teaching the caregivers the appropriate techniques of handling, seems to resolve the problem only partially [14]. At the same time, training is a very useful preventive tool against the development of plagiocephaly. Raising the awareness of the caregivers about the importance of symmetric stimulation and supervised prone lying seems to be a reasonable precaution for position-induced plagiocephaly. It may also affect the child's development in a positive way, as the prone position is more and more often reported to be irrelevant for typical motor development and is found dangerous by some researchers due to the choking hazard [19].

Physical therapy

Physical therapy programs concentrate on stimulation of the motor development with a strong focus on the symmetric performance according to the child's age, increasing elasticity of sternocleidomastoid muscles and achieving the full, active cervical range of motion followed by positioning [8, 15, 16, 17, 20]. Manual therapy manipulations and ostheopathy are also accepted as means of therapeutic intervention [21, 22]. The effectiveness of physical therapy is considered to be rather good, especially when the condition is recognized at an early stage [6, 15, 22, 23, 24]. However, according to van Vlimmeren, 30% of the children who had received physical therapy, still presented asymmetry in skull architecture at the age of 6 months [15]. Physical therapy is often recommended to caregivers of infants with skull asymmetry, as there are some reports that indicate motor delay within this group of patients [16, 17]. On the other hand, there are also studies which did not find a correlation between gross motor development and skull deformation [15]. Still, the prevalence of torticollis within the plagiocephaly group is significantly higher and hence physical therapy seems to be necessary [7].

Physical therapy remains the main therapeutic approach in plagio- and brachycephaly but the term comprises a lot of different interventions which may vary depending on their time of introduction.

Custom-made head orthosis

A custom-made head orthosis may be indicated when all the previous approaches are found to be insufficient [2, 7, 8, 20]. Helmet therapy is the method most often recommended in the literature [14, 25, 26, 27]. Some studies even see it as the therapy of choice for moderate and severe cases of plagiocephaly [14].

The principle of using a head orthosis is to redirect the process of growing into the areas where the head is flattened [28, 29]. These locations are exposed to less or no resistance compared to the areas that are round. Hence the head, with time, changes its shape in a desired manner [6]. Usually the process of forming the orthosis begins with plaster casting or 3D scanning in order to form a positive mould [6]. The helmet needs to be worn daily for 23 hours. The average age when helmet therapy is introduced is 7.6 months (ranging from 3 to 14 months) [7]. In general, guidelines recommend starting helmet therapy within the first year of the child's life, when skull growth in size is substantial [30]. Helmet therapy is also a good option for patients who had deteriorated despite the corrective steps undertaken [31]. The average length of helmet therapy is 3.7 months [7].

Despite these general recommendations, reaching consensus about the initiation, duration and even application of orthotic management in plagiocephaly remains difficult. In spite of numerous reports on the beneficial effects of custom-made helmets, systematic reviews reveal that most of these studies lack comprehensive methodology, so evidence-based conclusions are compromised [3]. Another problem is that helmet therapy is not free of risks: sores may form and the helmet may be uncomfortable for the child. On the other hand, these complications occur only in 25.4% of the cases, are of a mild degree and resolve spontaneously [27]. The experiences of parents/caregivers are generally positive [4].

There is a strong urge for standardization of orthotic intervention in plagiocephaly, as 5 out of 7 studies prove it to be a more effective way of correcting cranial asymmetry in comparison to positioning alone [20]. In the survey conducted by Lee et al. 58% of all plastic surgeons would recommend helmet therapy for moderate and severe cases of plagiocephaly [32]. (fig. 5).

Effects of helmet therapy (fig. 6, fig. 7, fig. 8)

Surgical intervention

Frontal orbital advancement (FOA) is considered to be the gold standard for coronal suture stenosis and is widely practiced [33]. The development of new technologies

Fig. 5. Vigo Helmet (courtesy of VIGO Ortho Sp. z o.o.).

Ryc. 5 Kask Vigo (udostępnione dzięki uprzejmości VIGO Ortho Sp. z o.o.).

Fig. 7. Six-month therapy (courtesy of VIGO Ortho Sp. z o.o.).

Ryc. 7. Po terapii sześciomiesięcznej (udostępnione dzięki uprzejmości VIGO Ortho Sp. z o.o.).

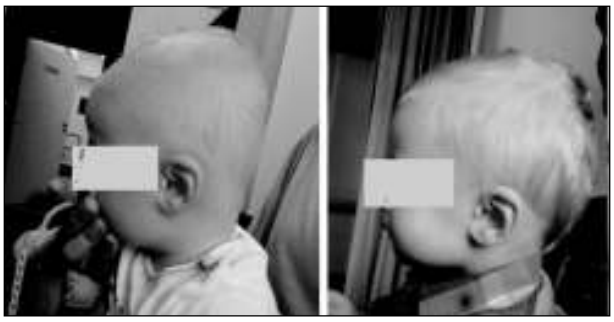

Fig. 6. Six-month therapy (courtesy of VIGO Ortho Sp. z o.o.).

Ryc. 6. Po terapii sześciomiesięcznej (udostępnione dzięki uprzejmości VIGO Ortho Sp. z o.o.).

Fig. 8. Four-month therapy (courtesy of VIGO Ortho Sp. z o.o.).

Ryc. 8. Po terapii czteromiesięcznej (udostępnione dzięki uprzejmości VIGO Ortho Sp. z o.o.).

Fig. 9. Craniostenosis surgery (courtesy of VIGO Ortho Sp. z o.o.).

Ryc. 9. Leczenie operacyjne kraniosynostozy (udostępnione dzięki uprzejmości VIGO Ortho Sp. z o.o.).

brought many modifications to the original approach. The main goal of the procedure is to obtain cranial and facial symmetry and to diminish functional limitations. The procedure requires full anesthesia. In every case the frontal-orbital bar is stabilized with a plate. The temporal bossing may be reshaped and filled with a piece of bone or cut and slid to the front. The disadvantage of this technique is a long incision, the risk of significant blood loss and the long hospitalization period required. Some studies demonstrated a relapse on the follow-up evaluation. The minimum age of the child that qualifies for this kind of surgery is 6 months (fig. 9).

CONCLUSION

The prevalence of plagiocephaly has increased significantly in recent times; still most of the cases are mild to moderate. As the wrong diagnosis and wrong treatment of skull deformities may lead to unnecessary complications, it is important for paediatricians and neonatologists to be trained in diagnosing head deformation. A careful diagnostic process must be applied every time in order to rule out synostosis. There are different suggestions on how to handle the problem of cranial asymmetry. It seems that the most reasonable approach is to implement positioning and physiotherapy. In case of moderate and severe deformations, it is recommended using a custom-made head orthosis as the most effective non-invasive method of forming the skeleton of the skull. Still these approaches need further evaluation as there are contradicting reports about their effectiveness. In the case of synostosis, surgical intervention should be considered.

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