Acta Neurochirurgica (2019) 161:1095–1098
https://doi.org/10.1007/s00701-019-03924-2

ORIGINAL ARTICLE - PEDIATRIC NEUROSURGERY

Prevalence and severity of positional plagiocephaly in children and adolescents

Federico Di Rocco[1] · Valeria Ble[1] · Pierre-Aurelien Beuriat[1] · Alexandru Szathmari[1] · Laura Nanna Lohkamp[1] · Carmine Mottolese[1]

Received: 11 March 2019 / Accepted: 18 April 2019 / Published online: 30 April 2019

Abstract

Object Though positional posterior plagiocephaly (PPP) is considered common in infants since the pediatric recommendations of "Back to Sleep", several aspects of its natural history still remain unclear. The aim of this study is to understand the actual prevalence and severity of PPP in children and adolescents.

Methods Head CT scans performed for head trauma during the period September 2016–September 2017 were retrospectively analyzed in a total of 165 children ranging from 0 to 18 years of age (101 boys).

Cranial vault asymmetry index (CVAI) was calculated at the level of the superior orbital rim. CVAI values greater 3.5% was considered index of asymmetry.

The results were analyzed according to different age groups: group I: 1 month to 1 year of age (37 children), group II: 2 to 4 years (32 children), group III: 5 to 8 years (36 children), group IV: 9 to 12 years (27 children), and group V: 13 to 18 years (33 children) and the severity of asymmetry according to CVAI values: mild group (CVAI range 3.5–7%), moderate group (CVAI range 7–12%), and severe group (CVAI > 12%).

Result The total prevalence of PPP in the 165 children was 25%. While the prevalence in infants of group I was estimated to be 40.5%, it was 15.6% in group II, 30.5% in group III, 18.5% in group IV, and 12% in group V.

The mean and maximum degrees of deformation were 3.5% and 15.1%, respectively. Most children had a mild asymmetry. One child (group II) presented a severe asymmetry. The degree of the asymmetry varied according to the groups but moderate asymmetry could be found at all ages even in groups IV and V.

Conclusion This study analyzing PPP in an unselected unbiased pediatric population shows that PPP has a high prevalence in adolescents. It confirms that the prevalence of deformational plagiocephaly is more common than usually reported and that PPP may persist at a late age.

Keywords Positional posterior plagiocephaly · Deformational plagiocephaly · Nonsynostotic plagiocephaly · Pediatric population

Introduction

Positional posterior plagiocephaly can be recognized as a unilateral flattening of parieto-occipital region with anterior shift of the ipsilateral ear and bossing of the ipsilateral forehead.

Since 1992, following the American Academy of Pediatrics (AAP) recommendations "Back to Sleep", an increase in the incidence of nonsynostotic plagiocephaly was documented [1]. The AAP recommended that healthy infants should not be placed in the prone position to reduce the risk of sudden infant death syndrome (SIDS). Two years later, Argenta et al. described a dramatic raise in the number of occipital cranial deformities that could not be attributed to lambdoid craniosynostosis [3]. The prolonged use of the supine position was proposed as the main pathophysiological mechanism that first induced a positional occipital deformation and subsequently, a facial deformation. In fact, concomitant to the "Back to Sleep" recommendations, several tools were developed to maintain the child in the supine position not only during sleep but also during the awake phases.

This article is part of the Topical Collection on Pediatric Neurosurgery

✉ Federico Di Rocco
federico.dirocco@chu-lyon.fr

[1] Department of Pediatric Neurosurgery, French Referral Center for Craniosynostosis, Hôpital Femme Mère Enfant, Hospices Civils de Lyon, Université de Lyon, 59 Boulevard Pinel, 69677 Lyon, France

Argenta et al. proposed a conservative treatment, through repositioning by the parents, aimed at reducing the compression on the affected side, while in the most severe forms, they suggested the use of a helmet [2]. In the same year, another study underlined the temporal coincidence between AAP recommendation and the increase in plagiocephaly without synostosis [6]. Since then, several studies have tried to identify numerous risk factors (such as premature birth, restrictive intrauterine environment, birth trauma, and torticollis) and to comprehend the physiopathology with the aim to understand the correct management [9]. The natural history is actually unknown. Some authors have suggested that PPP tends to spontaneously solve with time on the long term implying the absence of need of treatment in infants, such as an orthotic device (helmet). The aim of this study is to assess the prevalence and the degree of severity of PPP in the children and adolescents in an unselected population.

Methods

For this retrospective analysis, we enrolled a total of 300 patients, ranging from 0 to 18 years of age, submitted to a skull CT scan at Hôpital Mère Enfant of Lyon (reference center) for head trauma during the period September 2016–September 2017. All of them had undergone a CT scan to exclude post-traumatic focal and/or diffuse lesions. We excluded 135 patients, because CT scan showed movement artifacts or malpositioning of the head that could not allow a correct evaluation of the craniometric measurements. Therefore, 165 healthy children were included, negative for skull fracture, without history of previous craniofacial interventions. Cranial vault asymmetry index (CVAI) was calculated according to the literature at the level of the superior orbital rim [7]. CVAI values greater than 3.5% were considered pathological [8]. Loveday et al. defined the "Diagonal DA" as the smaller diagonal and the "Diagonal DB" as the larger, but using this formula, the result would always be negative, so we prefer using the formula modified by Dörhage [4], and we divided the difference of the diagonals (DA−DB) by the shorter DB (Fig. 1). The results were analyzed according to different age groups: group I: 1 month to 12 months of age (37 children), group II: 2 to 4 years (32 children), group III: 5 to 8 years (36 children), group IV: 9 to 12 years (27 children), and group V: 13 to 18 years (33 children) and the severity of asymmetry according to CVAI values: mild group (CVAI range 3.5–7%), moderate group (CVAI range 7–12%), and severe group (CVAI > 12%).

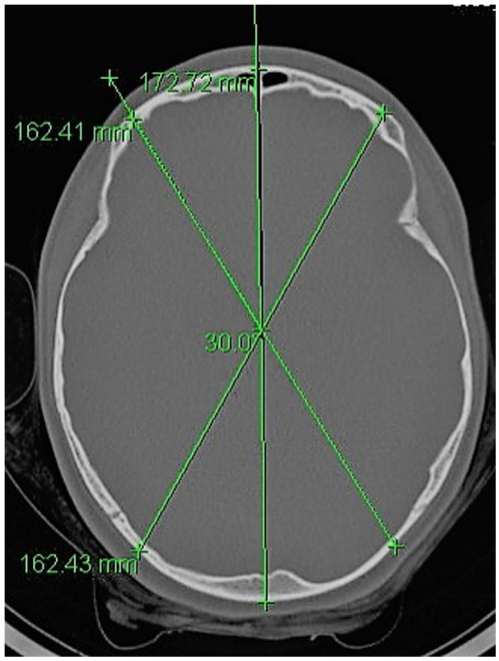

Fig. 1 Cranial vault asymmetry index (CVAI) was calculated in axial views at lower cranial level (the superior orbital rim level)

Results

The overall prevalence of PPP in the 165 children was 25%. As expected, it varied according to age: in group I, it was 40.5%; in group II, 15.6%; group III, 30.5%; group IV, 18.5%; and finally, group V, 12%.(Figs. 2 and 3). The population cohort is summarized in Table 1. All the patients in the first group with PPP ($n = 15$), 10 of which are boys, showed mild CVAI values with an average value of $4.9 \pm 0.98\%$. In the second group, there were 7 patients with plagiocephaly, 6 of which with mild CVAI and 1 patient with a value of 15.09%; in the third group, there were 11 patients with PPP of which 7 with values between 3.88 and 5.85% and 3 patients with values above 7.14%. In the group IV, the 5 patients with PPP, all boys, had mild values except for a patient with a value of 8.54%. Finally, the last group had 2 patients with mild values of the CVAI and 2 with CVAI of 7.98 and 8.25%. If

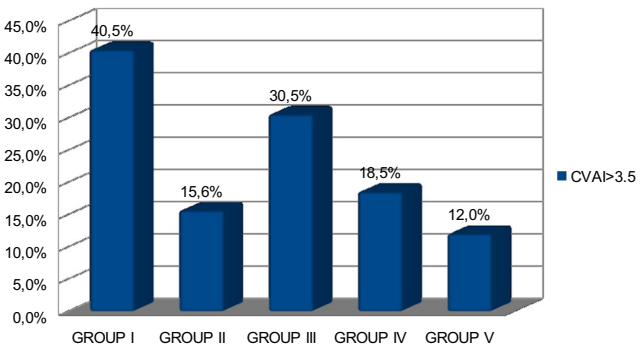

Fig. 2 PPP distribution according to age groups

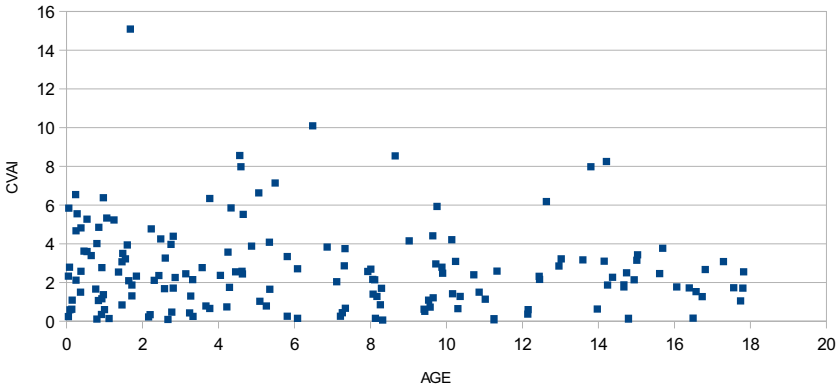

Fig. 3 CVAI distribution according to age

we consider the sex distribution, we have a total of 101 males, which represent 61%, against 39% of girls ($n = 64$). In the group of plagiocephaly ($n = 42$), we found 33 boys (79%) compared with 21% girls ($n = 9$).

Discussion

The prevalence of PPP is age dependent. Hutchison et al. have reported in a prospective study cohort that the overall prevalence rates for the cohort were 16% at 6 weeks, 19.7% at 4 months, 9.2% at 8 months, 6.8% at 12 months, and reduced spontaneously to 3.3% at 24 months [5]. Van Vlimmeren et al. found in a prospective cohort that the prevalence increased from 6.1 at birth to 22.1% at 7 weeks [12], and in another study in the same patients, the PPP at 5 years of age within the mild range was found in 19% and within the moderate/severe range in 1% [11]. The only work evaluating the prevalence of PPP in a cohort of patients born after 1992 but before 1998, the year in which the FDA approved the use of helmet, found a prevalence of 1% positional plagiocephaly accompanied in 38% by facial deformity [10].

This is the first study aimed at examining the presence of PPP in a group of children born after 1998, ranging between 0 and 18 years and shows different results about the prevalence of PPP compared with the result of the literature. As expected in the first group, the prevalence was extremely high: 40.54% with values of the CVAI between 3.5 and 7. However, unexpectedly, we found the persistence of moderate forms, ranging between 7 and 12 CVAI, in groups IV and V of 18.5% and 12% respectively. We found that values of CVAI above 3.5 are present not only in the first group as we expected but also in all the groups although with lower prevalence.

Therefore, in contrast with the literature, we can hypothesize that PPP does not correct spontaneously in all children. Nevertheless, the prevalence is superior in the first years of life and it reduces after the first year. Thus, there is however a possible correction. The more severe form of group V could be explained by an incorrect management of PPP due to poor knowledge at the time. In fact, these patients, of the last group, have an age between 13 and 18 years; therefore, they are born after the recommendations of the AAP of 1992, but probably the attention towards PPP among families, pediatricians, and neurosurgeons were lower compared today. The limitation of this study is that it was based only on radiological analysis; no clinical evaluation was possible. However, we consider that the radiological study allows a proper analysis without any bias.

Because this study is purely CT based, we cannot conclude on the efficacy of potential treatments that the children might have or not undergone. Though these data might not reflect the natural history of PPP, they show that a number of children do present a significant deformation in the late childhood. We can thus confirm that PPP might not completely disappear.

Unfortunately, there is no cohort that followed up for several decades to assess the evolution and impact of treated and untreated PPP. It would be interesting to evaluate the adolescents presenting a PPP with anthropomorphological facial measures and neuropsychology tests in an attempt to identify any eventual impact of PPP. Secondly, as for any traumatic pediatric population, boys are overrepresented, which is also the case in the population with plagiocephaly.

Our study is the first that measure the prevalence of PPP in the pediatric population ranging from 0 to 18 years and shows unexpected results with high prevalence and persistency of important asymmetry in a late age. Our findings show that the prevalence of PPP is still high in adolescence and thus,

Table 1 Cohort description (n 165)

Variable		n (%)
Gender	Male	101 (61%)
	Female	64 (39%)
Age	0–1 years	37
	2–4 years	32
	5–8 years	36
	9–12 years	27
	13–18 years	33

no spontaneous complete improvement maybe achieved, in contrast to the idea that the cranial deformation spontaneously regresses within the second year of life. These data underline the importance of prevention and early management when present of PPP.

Compliance with ethical standards

Conflict of interest All authors certify that they have no affiliations with or involvement in any organization or entity with any financial interest (such as honoraria; educational grants; participation in speakers' bureaus; membership, employment, consultancies, stock ownership, or other equity interest; and expert testimony or patent-licensing arrangements), or non-financial interest (such as personal or professional relationships, affiliations, or beliefs) in the subject matter or materials discussed in this manuscript.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional committee of HFME of Lyon and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. For this type of study formal consent is not required.

References

1. American Academy of Pediatrics (1992) Positioning and SIDS pediatrics 89:1120–1126
2. Argenta L, David L, Thompson J (2004) Clinical classification of positional plagiocephaly. J Craniofac Surg 15:368–372
3. Argenta LC, David LR, Wilson JA, Bell WO (1996) An increase in infant cranial deformity with supine sleeping position. J Craniofac Surg 7:5–11
4. Dörhage K (2010) Ursache und diagnostik der lagebedingten plagiozephalie. Man Med 48:125–134
5. Hutchison BL, Hutchison LA, Thompson JM, Mitchell E (2004) Plagiocephaly and brachycephaly in the first two years of life: a prospective cohort study. Pediatrics 114:970–980
6. Kane AA, Mitchell LE, Craven KP, Marsh JL (1996) Observations on a recent increase in plagiocephaly without synostosis. Pediatric 97:877–885
7. Lee MC, Hwang J, Kim YO, Shim KW, Park EK, Lew DH (2015) Three-dimensional analysis of cranial and facial asymmetry after helmet therapy for positional plagiocephaly. Childs Nerv Syst 31: 1113–1120
8. Loveday BP, de Chalain TB (2001) Active counterpositioning or orthotic device to treat positional plagiocephaly? J Craniofac Surg 12:308–313
9. Mottolese C, Szathmari A, Ricci AC, Ginguene C, Simon E, Paulus C (2006) Positional plagiocephaly: the place of cranial orthotics. Neurochirurgie 52:184–194
10. Roby BB, Finkelstein M, Tibesar RJ, Sidman JD (2012) Prevalence of positional plagiocephaly in teens born after the "Back to Sleep" campaign. Otolaryngol Head Neck Surg 146:823–828
11. van Vlimmeren LA, Engelber RH, Pelsma M, Groenewoud HM, Boere-Boonekamp MM, der Sanden MW (2017) The course of skull deformation from birth to 5 years of age: a prospective cohort study. Eur J Pediatr 176:11–21
12. van Vlimmeren LA, van der Graaf Y, Boere-Boonekamp MM, L'Hoir MP, Helders PJ, Engelbert RH (2007) Risk factors for deformational plagiocephaly at birth and at seven weeks of age. A prospective cohort study. Pediatrics 119:e408–e408