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Sagittal craniosynostosis combined with ossified cephalhematoma—a tricky and demanding puzzle

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Abstract

Introduction Four cases of sagittal synostosis combined with ossified cephalhematoma prompted the authors to present the data and to discuss the implications. Large cephalhematoma of the vertex at birth with subsequent ossification occurred in all with final sizes of 6.5–10 by 4.0–5.5 by 0.8–1.8 cm. At surgery with 2–6 months, the mean skull index was 64.75, sagittal suture completely closed, and a disfiguring bony mass present in all.

Discussion In three of the original cohort of 106 sagittal synostoses, ossified cephalhematoma was removed in one piece together with the suture. In contrast to pathogenesis of common parietal cephalhematomas, cephalhematoma in sagittal synostosis is rather induced by periosteal detachment of the midline by strong shearing forces because molding is hindered in the lateral direction. Frequent and fast complete ossification is possibly directed by the same local factors (e.g., Noggin) which lead to premature fusion of cranial sutures. Some treatment principles of ossified cephalhematoma in sagittal synostosis may be applied to surgery of common types.

Keywords Ossified cephalhematoma · Sagittal synostosis · Pathogenesis cephalhematoma · Ossification cephalhematoma · Treatment

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Introduction

The clinical findings and the early and intermediate natural history of cephalhematoma [22] and of craniosynostoses [17, 26] are well known. The observation of four cases of sagittal synostosis combined with an unusual and nearly identical clinical presentation of an ossified cephalhematoma on the vertex prompted the authors to present their data and to discuss the implications, particularly the following topics: (1) possible factors leading to ossification of cephalhematoma, (2) pathogenesis of cephalhematoma in general and specifically in sagittal synostosis, and (3) surgical treatment of ossified cephalhematoma in sagittal synostosis and its application to those ossified cephalhematomas with classic localization.

Materials and methods

From 1991 to 2007, 106 infants with sagittal synostosis have been operated by a modified extended vertex craniectomy [7] and cranioplasty. Three of them (=2.8%) had a neonatal cephalhematoma which was followed by ossification in any case. A further case of ossified cephalhematoma in sagittal synostosis of a partner university hospital was added to the three cases. A confirmed consent was obtained from the parents to use the data of their children. The clinical and radiological evaluation included the peri- and postnatal history, the local findings and their evolution, data of surgery and histological examination, and follow-up findings. In general, all craniosynostosis patients have prospective follow-ups in regular intervals increasing with age until puberty.

Results

In Table 1, the obstetric history and the condition of the four patients at birth are listed. All are born spontaneously at term by primipara, but in all, instrumental deliveries were needed.

Table 2 shows the precise findings of ongoing ossification on the plain X-ray of the skull at 2 to 2.5 months of age. The area of ossification has always an oblong–oval shape which is lined up in the direction of the closed sagittal suture and distributed symmetrically on both sides. The mean area is 7.3 cm long, 4.8 cm wide, and 1.3 cm deep. Three patients have one osseous prominence (Fig. 1). Case 4 shows two prominences (Fig. 2); in contrast to cases 1 and 2 with a homogeneous osseous structure, in case 3 and 4, one and two central luminescences, respectively, are visible corresponding to two small cavities. All patients had radiological signs of premature closure of the hole sagittal suture.

Table 3 summarizes the natural history of cephalhematoma and the findings of ossified cephalhematoma as well as the head measures at surgery. A distinctly recognizable cephalhematoma has been present in all shortly after birth. It was always localized in the midline over the sagittal suture. The same is true for the disfiguring ossified cephalhematoma with a largest diameter of >5 to 10 cm at surgery which corresponded clinically (Fig. 3) as well as radiologically to a stage 4. Except for case 4, all patients had cavities within the ossified cephalhematoma which were more or less distinctly visible by the naked eye (Fig. 4). In case 4, such cavities were seen histologically (Fig. 5a–c). All patients had a distinct scaphocephaly with a mean skull index of 64.75 at a mean time of 3 months and 1 week.

In Table 4, age at surgery, type of surgery, and follow-up time of the individual patients are quoted. Except for the added case from the partner university hospital, all had a vertex craniectomy in which the ossified cephalhematoma was removed together with the midline bone strip over the sagittal sinus in one piece including the fused sagittal suture (Figs. 6 and 7). For cranioplasty, small pieces of the removed bone parts were used to cover the whole defect like a mosaic except for a 1–2 cm wide zone along the midline (a method which we are using since many years with success). The follow-up time was in every case long enough to prove a regular and complete reossification and permanent correction of the former deformity caused by the sagittal synostosis and the ossified cephalhematoma.

Discussion

In the presented cohort of 106 infants with sagittal synostosis, cephalhematoma has been observed in three during the neonatal period. The percentage of 2.8 cephalhematomas corresponds to some of the figures quoted in the literature in a general population [16, 27], but the fact that ossification occurred in all and was already completely developed to stage 3-4 at the age of 2 to 2.5 months is remarkable. It is different from the common course of ossification which is much less frequent and displays chronologically variable periods of ossification. Besides the possibility of a biased phenomenon, the occurrence of factors leading to frequent and fast complete ossification must be discussed, as for instance, local factors of the adjacent dura and suture cells as Noggin under- and Runx-2 overexpression [8] among other mechanisms [10] which are possibly involved in premature fusion and reossification of the surgically opened sagittal suture. In contrast to the common parietal cephalhematoma which is somewhat apart from the normal sutures, the presented vertex hematomas lie directly over a pathological sagittal suture and by it close to the dura.

In the recent Anglo-American literature, the occurrence of ossified cephalhematoma is quoted as occasional, rare, or unknown and restricted to few case reports [4, 6, 12]. Already in the seventies and much earlier, it has been claimed by some that a persistent osseous prominence is an extremely rare sequel of cephalhematoma because there is even, in case of fully established ossified cephalhematoma, a slow resolution of the mass which merges over months into the general contour of the growing calvaria [22, 31]. At the beginning of the fifties and sixties, Bloch [1] and Vernon [30] quoted studies which, currently took place 10 years ago with an incidence of 3.2% to 10% persistent

Table 1 Obstetric history and condition of the patients at birth (n=4)

Patient	1	2	3	4
Week of gestation at birth	40th	40th	41th	41th
Parity	Primipara	Primipara	Primipara	Primipara
Labor	Spontaneous	Spontaneous	Spontaneous	Spontaneous
Type of delivery	Forceps	Vacuum	Forceps	Vacuum
Body weight (mean 3,440 g)	3,240 g	3,520 g	3,900 g	3,100 g
Body length (mean 49.75 cm)	50 cm	48 cm	52 cm	49 cm
Condition at birth (1 min Apgar score \geq 7)	+	+	+	+

Table 2 Radiological findings of ongoing ossification of cephalhematoma (n=4)

Patient	1	2	3	4
Age at X-ray (months)	2.5	2	2.5	2
Area cephalhematoma	$10.0 \times 5.0 \times 1.0$	$7.5 \times 4.0 \times 1.8^{a}$	$8.0 \times 5.5 \times 1.3$	$6.5 \times 5.0 \times 0.8$
Structure cephalhematoma right-left symmetry	+	+	+	+
Number of prominences	1	1	1	2
Homogeneity	+	+	_b	_b
Signs of sagittal synostosis	+	+	+	+

^aRadiological size of cephalhematoma at age 5 days 7.0×4.75×0.45

^bOne and two luminescences

ossified cephalhematomas, still visible after several years. Figures of the early seventies are 1.6% to 10% [25, 31, 34] although less reliable due to much smaller number of patients.

Some of the cephalhematomas may go clinically and radiologically through different stages of subperiosteal ossification [3, 9, 22, 25, 31]. In stage 1, a ridge is palpable around the edge of cephalhematoma. In stage 2, the formed bone at the edges grows subperiosteally step by step over the dome of the bulging hematoma. A thin pliable layer of bone becomes palpable which has been described as comparable with a rustling parchment [31] or celluloid ball [2]. In stage 3, the bony covering becomes thicker and solid, and in stage 4, a prominent firm mass or a shell-like thickening of the skull is palpable. According to histological



Fig. 1 Anteroposterior X-ray of the skull in case 2 at age 2 months. The ossified cephalhematoma with homogeneous osseous structure lies symmetrically over the middle third of the fused sagittal suture. A distinct impression of the superior sagittal sinus is recognizable without any signs of sagittal suture

examination, all these stages are processes of ossifications and not of calcifications [3] and should be named ossifying cephalhematomas.

Although ossification may start at the end of the first postnatal week and reaches stage 2 already at the end of the second week, permanent ossification only occurs at the end of the sixth week if resorption of cephalhematoma is not completed by this time [31]. The observation that resorption of cephalhematomas may occur by 1 to 6 weeks with a median time of three to four weeks [31] and that different lengths are quoted in the literature for the four stages of ossification [1, 9, 25, 31] points to a large variation of the time course of resorption and ossification of cephalhematoma, and complete resolution is still possible in the early stages of ossification. This conclusion is strengthened by the frequent occurrence of resolution during the early ongoing ossification which can be observed in one third of the patients [31] in comparison to the relatively low incidence of resolution in permanently ossified cephalhematoma.

A relation between the initial size of cephalhematoma and the type of resolution has been rejected by some



Fig. 2 Lateral X-ray of the skull in case 4 at age 2 months. Instead of one, two prominences of ossified cephalhematoma are visible which lies over the middle and posterior third of the fused sagittal suture. In addition, two distinct luminescences are visible inside of the ossified cephalhematoma

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Patients	1	2	3	4
Clinical recognition of cephalhematoma	Day 1–2	Day 1–2	Day 1–2	Day-4
Time of surgery (mean 3 months 1 week)	2.5 months	2.0 months	6.0 months	3.0 months
Site of ossified cephalhematoma	Midline anterior half of sagittal suture	Midline middle third of sagittal suture	Midline middle third of sagittal suture	Midline middle and posterior third
Size of ossified cephalhematoma (largest diameter)	10 cm	>5 cm	>5 cm	>5 cm
Gross and histological morphology	Subtotal ossification	With residual cavity (ies)	With abnormal vascularization	In all patients ^a
Head circumference/skull index	>97%	>97%	>97%	>75%
(mean 64.75)	56	67	68	68

 Table 3 Natural history of cephalhematoma

Findings of ossified cephalhematoma and head measures at surgery (n=4)

^a In case 4, cavity (ies) were not visible by the naked eye but were seen histologically

authors [31]. On the other hand, all case reports including ours with disfiguring permanent ossified cephalhematoma had initially very large cephalhematomas of minimally the same size. According to Chung et al. [3] and Wechselberg and Sanders [31], the individual time course of the two ongoing processes—"resorption" and "ossification"—are crucial to the origin of a permanent ossified cephalhematoma and its possible persistence over years, which means resorption of the clot does not proceed rapidly and is not completed in the early stages of ossification. This hypothesis is confirmed by the observation that in almost all radiological work-ups with computed tomography (CT) or magnetic resonance imaging (MRI) [3, 4, 12] and in all operative specimens, residual cavities with remnants of consolidated hematoma and/or with abnormal vascularization are present as in ours.

The vertical site of cephalhematoma was in all four cases of proven sagittal synostosis identical and unusual in comparison to the typical localization of cephalhematomas. The frequency of neonatal cephalhematomas is reported to be 0.2% to 2.5% of life births [16, 27] or even higher, as for instance following vacuum extraction with a mean incidence of 6% [6], and depends for the most part on the composition of the evaluated women and newborns as well



Fig. 3 Case 1 at age 2.5 months. The disfiguring ossified cephalhematoma is located over the anterior half of the fused sagittal suture and has an oblong–oval shape in the direction of the former suture

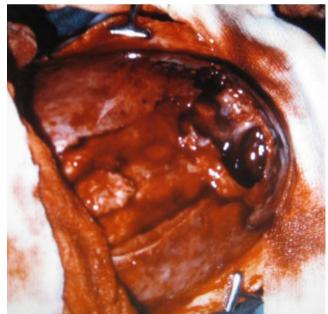


Fig. 4 Operative findings in case 1 at age 2.5 months in whom the sagittal suture (on the *left side*) and the ossified cephalhematoma (on the *right side*) have been removed step by step. A large irregular cavity is visible within the ossified cephalhematoma with a dark-red vascularization

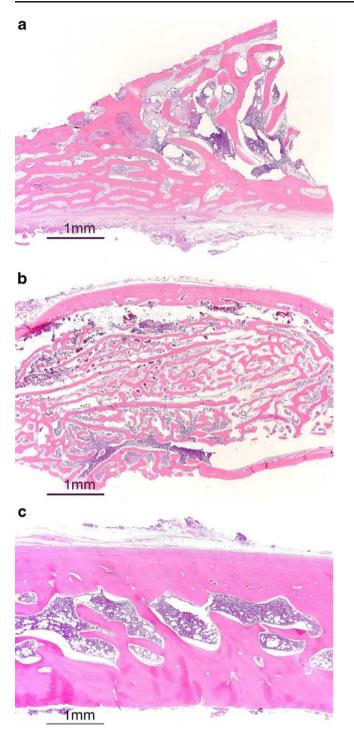


Fig. 5 a-c Histological work-up of ossified cephalhematoma and sagittal suture in case 4 at age 3 months. Hematoxylin and eosin stain. a Site of the transition of the parietal bone (on the *left side*) to the ossified cephalhematoma (on the *right side*). A distinct difference is recognizable between them concerning thickness and structure of the bone. b Central part of ossified cephalhematoma. It consists mostly of trabeculated bone with innumerable vascularized cavities which are different from those in the parietal bone and the former sagittal suture. c Site of the former sagittal suture. There is a normal bone structure without any suture tissue

as on a careful and repeated clinical examination in the newborn period. Independently of frequency, 85% to 91% of cephalhematomas have a unilateral parietal or, less frequently, a bilateral parietal localization [19, 31]. The occipital site is with about 10% the second most frequent site; in addition, rarely frontal and multiple localizations occur. Midline vertical site is quoted only once in the literature [20].

For the most frequent types of vaginal delivery, the preferred localization of cephalhematoma over the parietal bone does not permit a detailed statement about the pathogenesis of cephalhematoma in the individual case because there is no correlation between right or left occipital presentation at birth and the laterality of cephalhematoma [5, 31]. On the other hand, cephalhematoma must develop on the entrance of the head in the osseous pelvis or shortly thereafter because only in this moment equally exists the chance of involvement of the right or left side of the prominent upper posterior angle of the parietal bones. Likewise, cephalhematoma in caesarean section was only present if labor had already started and was interrupted during the first stage [5]. According to Churchill et al. [5], cephalhematomas occur significantly more frequently in occiput posterior and occiput transverse presentations than in the common occiput anterior presentations. The former types of presentations are therefore a risk factor for development of cephalhematoma as primiparity, high-term birth weight, prolonged labor, and instrumental deliveries in general [31, 34]. Although in the latter group, forceps deliveries are prone to development of cephalhematoma specifically in midforceps and forceps rotation [5, 19], vacuum extractions increase the risk of cephalhematoma and certain types of intracranial hemorrhage more than low forceps deliveries [32].

The observation of cephalhematomas by ultrasound during pregnancy [11, 24, 33] on the one hand and after cesarean section [5, 16, 31] and in vertex-born babies on the other hand points to the possibility of different pathophysiological mechanisms which lead to circumscript detachment of periosteum from the underlying bone combined with rupture of transversing blood vessels [16]. Hartley and Burnett [13] suggested a trauma of dragging nature as precipitating factor. Out of the two following mechanisms described in the literature [3, 25]—(1) parallel forces are set up between periosteum and skull which result in a shearing mechanism and (2) sudden or prolonged compression and inward movement of the skull with displacement of the bone from the periosteum-the former one fits better the hypothesis of a movement with effort as shown by the following observations: Smaller parietal cephalhematomas are usually localized in the upper posterior parietal bone and larger types take on an oblong-oval shape. Therefore, in the common vaginal

Patient	1	2	3	4
Age at surgery (months)	2.5	2.0	6.0	3.0
Type of surgery	Stepwise excision cephalhematoma and linear craniectomy	Vertex craniectomy including cephalhematoma and cranioplasty	Vertex craniectomy including cephalhematoma and cranioplasty	Vertex craniectomy including cephalhematoma and cranioplasty
Follow-up time (years)	16.0	15.10	9.6	0.6

Table 4 Time and type of surgery for sagittal synostosis and cephalhematoma and follow-up time (n=4)

delivery, there is shearing of the periosteum in a parallel and longitudinal manner at the level of the promontory and/ or symphysis due to the normal rotation movement of the head. In the already mentioned occiput posterior and transverse presentations, or the in midforceps and forceps rotation deliveries, the same mechanism takes place though to a more severe degree. The above mentioned uniform distribution pattern of cephalhematoma with preference for parietal bone shows that it results from and is due to the anatomical, and physiological characteristics of the birth channel, e.g., impact of the parietal area against the pelvis [13], and its frequency is triggered by the known risk factors.

In case of the observed cephalhematomas in sagittal synostosis, the already mentioned pathogenic mechanisms seem not very likely because the head should be presented in such a way that instead of the parietal bones as in normal delivery, the vertex with the sagittal suture gets into close contact with the promontory and/or symphysis as for instance in increased posterior or anterior parietal presentation (asynclitism). A possible pathogenic mechanism is a bilateral detachment of periosteum including the midline by strong shearing vertical forces because molding of the head is hindered in the lateral direction by premature fusion of the sagittal suture. During surgery of the fused sagittal suture, it can be observed that the site of periosteal insertion to the closed suture and of the latter one to the dura is very



Fig. 6 View to the outer side of the bone strip of 10 cm length in case 3 which includes the site of the former sagittal suture as well as the ossified cephalhematoma. The oblong–oval bony prominence lies in the middle third of the former suture site and is still disfiguring at age 6 months

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small and less adherent in contrast to a normal suture. Therefore, a bilaterally symmetrical cephalhematoma is developing which comes together, takes on an oblong–oval configuration, and occupies a large distance of the midline.

The hypothesis that synostosis of the sagittal suture is secondary to the ossified cephalhematoma claimed by Martinez-Lage et al. [20] in their case report on sagittal synostosis combined with ossified cephalhematoma must be discarded in spite of the cited experimental models of craniosynostosis due to the contradictory statements between the extreme scaphocephaly on X-ray and the clinically seemingly open sagittal suture on both sides of the cephalhematoma. Our cases had already scaphocephaly at birth and clinical and/or radiological signs of premature closure of sagittal suture before ossification of the cephalhematoma.

In our last three cases, the disconfiguring ossified cephalhematoma was removed in one piece together with a bone strip adjacent to the fused sagittal suture. This method is much easier than local excision which might be dangerous and combined with considerable blood loss. The same proceeding could be applied to the common parietal ossified cephalhematoma.

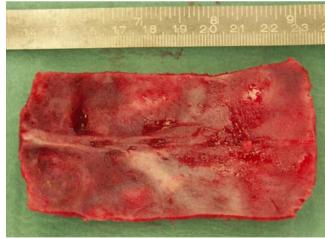


Fig. 7 View to the inner side of bone strip of 9 cm length in case 4 which includes the site of the former sagittal sutures and the ossified cephalhematoma (not visible in the picture). Except for few millimeters of a recognizable suture line adjacent to the coronal suture, the former sagittal suture is replaced by a bony ridge on the *left* and by an impression of the superior sagittal sinus on the *right side*

The operative techniques for the removal of common ossified cephalhematomas have been described by Kaufman et al. [15] and recently by Chung et al., Chung et al., and Gupta et al. [3, 4, 12]. The latter authors propose excision of the outer bulging bone including the remnants of the consolidated hematoma over the inner calvarial bone which is left behind. Remodeling of the operative field with multiple small pieces of the outer bulging bone is only performed if there is a depression of the involved parietal bone or if there is a defect or diminished thickness of it. Excision of the whole ossified cephalhematoma beyond the junctional area of the involved parietal bone as it is proposed above and mentioned in Gupta's case history [12] has several advantages in spite of major surgery. The whole ossified cephalhematoma can be handled outside of the patient, a possible injury of the underlying structures by the osteotome can be avoided, and a depression of the whole parietal bone as it is observed frequently in and after large cephalhematomas can be corrected at the same time. As pointed out by Chung et al. and Gupta et al. [3, 12], CT and/or MRI is not only useful if a cephalhematoma has not resolved within 6 weeks after birth for definition of the stage of ossification but also in every case preoperatively for differential diagnosis and to describe the expansion of the lesion in all three dimensions. Recently, Jang et al. [14] have proposed simple excision and periosteal reattachment in ossified cephalhematoma with good cosmetic appearance instead of additional cranioplasty for the correction of a depressed area.

Evacuation of native cephalhematoma by puncture in the second week of the neonatal period or thereafter is still not yet the usual practice [18, 28] after a controversial debate many years ago [1, 2, 16, 22, 23, 27, 29–31], but there are exceptions of it as suspicion of infected cephalhematoma in which diagnostic puncture is needed [18, 21] or as large cephalhematomas. The latter one may be combined with or lead to depression of the parietal bone which can be temporary if resorption occurs [25] and permanent if ossification develops as shown by the X-rays of several case reports. Depression and/or ossification can be corrected or avoided by proper puncture in time [2, 29], for instance in selected cases with a size of a walnut to an apple [23, 30].

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