



# Sagittal craniosynostosis associated with midline cephalhematoma or vice versa, case report and a review of the literature

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## Abstract

**Introduction** Sagittal craniosynostosis associated with midline cephalhematoma is a rare finding. Despite the controversy regarding its etiopathogenesis, this condition represents a clear indication for surgery.

**Case report** We present a case of a 10-week-old boy with an ossified midline vertex cephalhematoma and sagittal craniosynostosis. The child underwent a cephalhematoma excision and minimally invasive non-endoscopic narrow vertex craniectomy, with calvarial vault remodeling followed by 2 weeks use of a cranial orthosis. On 5-month follow-up, mesocephaly was achieved.

**Conclusion** Our case is well documented with native CT, 3D CT, intraoperative pictures, and 3D head scan imaging. We described our minimally invasive non-endoscopic technique that led to a rapid cranial vault remodeling with reduction of cranial orthosis need. A review of literature focused on surgical techniques is included.

**Keywords** Scaphocephaly · Cephalhematoma · Minimal invasive technique · Cranial orthosis

## Introduction

Ossified midline vertex cephalhematoma associated with scaphocephaly is a rare finding. It is controversial whether sagittal craniosynostosis develops secondary to the midline cephalhematoma immobilizing the suture or if the vertex cephalhematoma is generated due to loose periosteum above the closed suture [1–6].

Cephalhematomas are subpericranial blood collections limited inferiorly by the skull and latero-laterally by the tight adhesions of the periosteum in sutures. They are predominantly located in the parietal eminences and related to laborious and instrumental delivery. Some of them undergo a fast ossification process [7]. The therapeutical approach to liquid and

ossified lateral cephalhematomas is controversial. However, the following case of vertex cephalhematoma associated with scaphocephaly represents a clear-cut indication for surgery. The case is well documented with native computed tomography (CT), 3D CT, intraoperative pictures, and 3D head scan imaging. Our surgical technique that led to almost immediate skull remodelling and shortened the need for cranial orthosis is described. Furthermore, we provided a review of literature on the topic.

## Case report

Our patient was born to primipara in term. The delivery was prolonged and laborious with abnormal rotation of child's head in deflexion. The boy was eventually delivered vaginally without use of forceps. A midline, reportedly liquid, vertex cephalhematoma was recorded at the birth in a different institution. Six weeks later, an atypical shape of the head with a midline ossified cephalhematoma was noted by the pediatrician. The child was referred to the Department of Neurosurgery, 2nd Faculty of Medicine, Motol University Hospital, Prague. Cephalometry described all markers of premature sagittal suture closure with dolichocephaly, frontal bossing, and parieto-occipital head narrowing (Figs. 2a, b and 3a). CT scan confirmed closure of the sagittal suture, dolichocephalic skull, and a well-developed osseous bridge

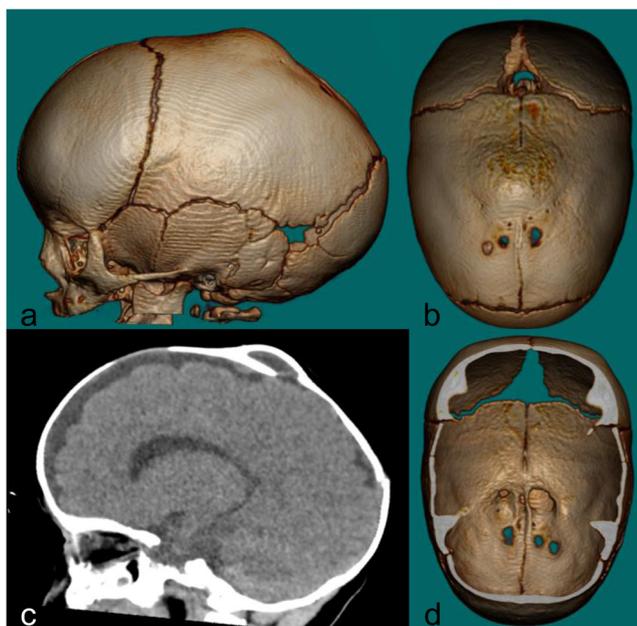
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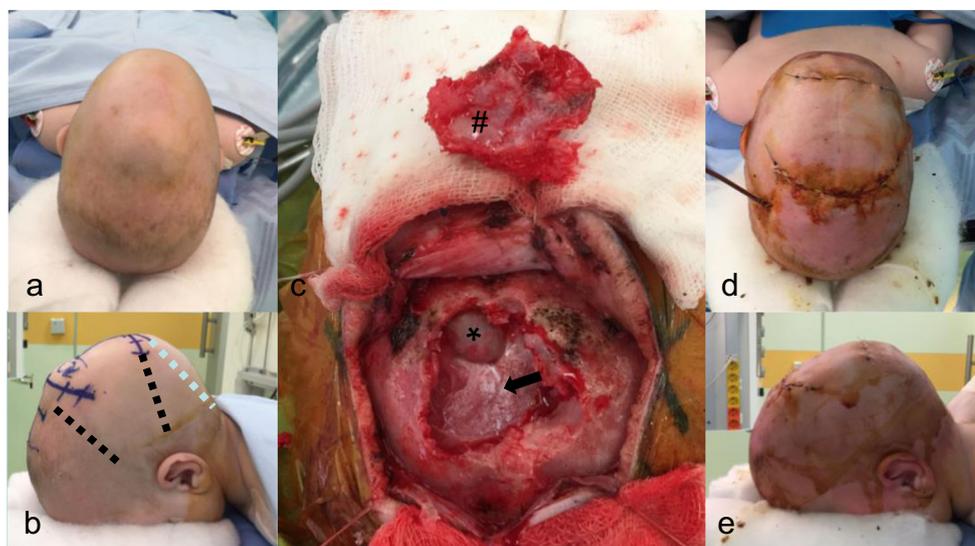
**Fig. 1** Preoperative CT and CT 3D reconstruction in a patient with an ossified midline vertex cephalhematoma associated with sagittal craniosynostosis. **a** A sagittal view of a CT 3D reconstruction demonstrating dolichocephalic skull with ossified vertex cephalhematoma. **b** An axial view, 3D CT reconstruction, showing closed suture along with ossified cephalhematoma and narrow posterior skull. **c** A sagittal view, native CT, depicted defect of the parietal bone beneath the cephalhematoma and benign enlargement of the subarachnoid spaces frontally. **d** An axial view, CT 3D reconstruction of cranial convexity, inner view demonstrating the osseous bridge closing the sagittal suture and bone defects parasagittally bilaterally

connecting both parietal bones, covered by the dome of the ossified cephalhematoma. Furthermore, it showed defects of the skull beneath the cephalhematoma parasagittally and bilaterally (Fig. 1a–d).

At the age of 10 weeks, surgical treatment was undertaken. The cephalhematoma was exposed through a 5-cm-long curved skin incision. Another, 2.5-cm-long skin incision was made above lambda to allow posterior parietal barrel-stave and occipital osteotomies. (Fig. 2b, d). The burr holes around the dome of the cephalhematoma were connected by a craniotome (Fig. 2c). The hemorrhage from the bone was stopped by wax. A 2-cm-wide strip craniectomy was performed along the closed sagittal suture. Barrel-stave osteotomies were performed in parietal bones (Fig. 2b). Due to significant occipital narrowing, we further performed two occipital osteotomies from the posterior skin incision (Fig. 2b). Those in combination with posterior parietal osteotomies had immediate effect on widening of parieto-occipital compartment (Fig. 2d). The postoperative course was uneventful. We left the cranial orthosis for only 2 weeks as the skull was practically remodeled and the child did not tolerate it well. Five-month follow-up cephalometry confirmed mesocephaly with cephalic index 77 units (Fig. 3b). The patient had a good cosmetic result and normal psychomotor development.

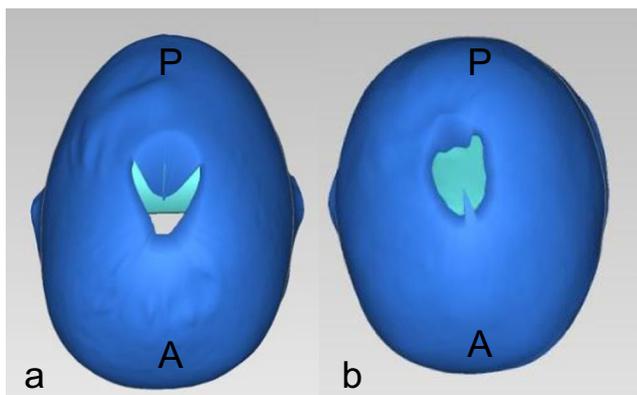
## Discussion

Midline cephalohematomas are a rare finding of not entirely elucidated causes. Martinez-Lage et al. provided the first report and proposed that sagittal craniosynostosis was



**Fig. 2** Perioperative and intraoperative view. **a** Preoperative image of the patient's dolichocephalic head with significant occipital narrowing and vertex cephalhematoma (dotted circle). **b** Preoperative side view showing the vertex cephalhematoma and depicting the scheme of barrel-stave osteotomies (black dotted lines) and additional occipital osteotomies (light blue dotted line). **c** Intraoperative view of resected dome of

cephalhematoma (#), a large defect (\*) uncovering the dura mater is visible along with a closed sagittal suture (arrow). **d** Postoperative picture showing wider occipital region and curved skin incision above the cephalhematoma and in lambda region. **e** Postoperative lateral view demonstrating cephalhematoma resection



**Fig. 3** 3D head scan. **a** Preoperative scan showing dolichocephaly with occipital narrowing and frontal bossing with cephalic index 70 units. **b** Postoperative 3D head scan at 5-month follow-up demonstrated mesocephaly with cephalic index 77 units. (A—anterior, P—posterior)

secondary to a calcified immobilizing cephalohematoma once only portions proximal to its borders were consolidated [1]. Merlob et al. highlighted the continuity of the periosteum across the consolidated suture and went strongly against Martinez-Lage’s theory by stating that a midline cephalhematoma can only occur secondary to a sagittal craniosynostosis and not vice versa [2]. Moreover, the ossified suture has been found to significantly alter birth mechanics determining bilateral detachment of the periosteum that allows for vertex cephalhematoma development [3–5]. In our case, a midline, liquid, vertex cephalhematoma was recorded after laborious birth in a different institution. There was no concomitant record of dolichocephalic-shaped head. This finding was described by pediatrician 6 weeks later and was coupled with sagittal ridge and an already ossified midline cephalhematoma. On CT-3D and intraoperatively, we noted

that the suture closure was pronounced just beneath the cephalhematoma in middle segment of the suture. It is our opinion that Martinez-Lage’s proposal should not be completely dismissed.

Association of ossified midline cephalhematomas with sagittal synostosis has been very scarcely reported. Literature is therefore lacking information regarding the optimal surgical procedure (Table 1). Nguyen et al. documented the first use of the endoscope-assisted technique for a midline cephalhematoma with sagittal craniosynostosis followed by helmeting for 7 months [6]. There is however little evidence of superiority of the endoscopic-assisted craniectomy followed by helmet therapy due to scaphocephaly when compared to minimal invasive variants of non-endoscopic techniques such as the one we used. Mutchnick et al. presented a procedure conducted in this manner [8]. They performed three incisions each of 3 cm in length followed by 3–4 barrel-stave osteotomies oriented by both direct visualization and palpation of the scalp. The authors argument in favor of this more accessible technique for they achieved similar results in surgical time, estimated blood loss, and cosmetic results while disposing of the helmet therapy. Even though this technique has not been used for suturectomy in patients with scaphocephaly and midline cephalhematoma, it closely compares with the procedure we performed. Despite we customarily perform an endoscope-assisted technique for children under 3 months of age, in this case, it was not necessary, because the longer curved incision together with shorter posterior incision gave us sufficient range of view (Fig. 3c, d). Furthermore, we hypothesize that the additional occipital osteotomies contributed to immediate occipital widening and thus reducing orthosis time in our patient.

**Table 1** Summary of the reported cases of sagittal craniosynostosis and ossified vertex cephalhematoma focused on the surgical technique

| References                    | Title  | No. of cases/age                 | Technique used  | Follow-up                     |
|-------------------------------|--|----------------------------------|---|-------------------------------|
| Martinez-Lage et al. 1984 [1] | Craniosynostosis secondary to calcified subperiosteal hematoma: a case report  | 1/5 months                       | Bilateral parasagittal craniectomies limited by length of cephalhematoma that was removed en bloc   | 8 years                       |
| Merlob et al. 1985 [2]        | Crossed sagittal suture cephalhematoma   | 1                                | –   | –                             |
| Currarino 2007 [5]            | Neonatal subperiosteal cephalhematoma crossing a synostosed sagittal suture  | 1/2 months                       | Sagittal synostectomy with cranial reshaping and removal of cephalhematoma  | Few years of follow-up        |
| Kaiser and Oesch 2009 [3]     | Sagittal craniosynostosis combined with ossified cephalhematoma—a tricky and demanding puzzle                            | 4/2.5 months, 2, 6, and 3 months | In one case, excision of the cephalhematoma and linear craniectomy; in the three cases, vertex craniectomy including cephalhematoma and cranioplasty        | 16, 15.10, 9.6, 0.6 years     |
| Piatt 2010 [4]                | Sagittal synostosis and ossified scalp hematoma: cause of consequence?   | 2/4 and 5 months                 | Bicoronal zig-zag incision, removal of outer shell of the hematoma with a power drill, modified pi procedure  | 4 years and 6 months/4 months |
| Nguyen et al. 2014 [6]        | Endoscopic-assisted treatment of sagittal craniosynostosis and calcified cephalhematoma                                  | 1/4 months                       | Endoscopic-assisted wide-vertex craniectomy with barrel-stave osteotomies   | 4 years                       |
| Present case                  | Sagittal craniosynostosis associated with midline cephalhematoma or vice versa, case report and review of the literature | 1/10 weeks                       | Minimally invasive non-endoscopic cephalhematoma excision and narrow vertex craniectomy, with barrel-stave osteotomies and additional occipital osteotomies | 5 months                      |

## Conclusions

We present a case of a 10-week-old boy with an ossified cephalhematoma and sagittal craniosynostosis. The co-occurrence of these two conditions is rare, with only few cases published. Despite the controversy regarding the etiopathogenesis, this condition represents straightforward indication for surgery. The child underwent a cephalhematoma excision and minimally invasive non-endoscopic narrow vertex craniectomy, with calvarial vault remodeling followed by a 2-week use of a cranial orthosis. On 5-month follow-up, mesocephaly was achieved.

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## Compliance with ethical standards

**Informed consent** Informed consent was obtained from the next of kin.

**Conflict of interest** The authors declare that they have no conflict of interest.

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