

Research Article – Human Anatomy Case Report

Bathrocephaly: a case report of a head shape associated with a persistent mendosal suture

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Abstract

Bathrocephaly is a posterior cranial deformity associated with failure of fusion of the mendosal suture. Infants are born with a prominent occipital bone. Here we discuss a case report of bathrocephaly in a newborn discussing management, imaging finding, differential diagnosis and relevant bony anatomy and development.

Key words

Bathrocephaly, mendosal suture.

Introduction

Bathrocephaly is posterior cranial deformity in which the mendosal suture fails to fuse. It has been described as a step-like deformity of the occipital bone. Due to the failure of the suture to fuse appropriately, the patients are often found to have a significant bulge of the interparietal portion of the occipital bone (Mulliken and Le, 2008). The purpose of this paper is to discuss a recent case of bathrocephaly seen at our institution. We will also discuss the relevant bony development, differential diagnosis, and management options.

Case report

The patient is a product of a 39 weeks gestation birth through Caesarean section secondary to breech positioning. He was discharged home and seen in our clinic five days after birth due to a prominence in the occipital region noted by the pediatrician. X-rays were obtained by the primary physician to workup craniosynostosis or skull fracture. At the time of presentation, he was noted to have a head circumference of 37 cm, which placed him at 25th-30th percentile. Both his anterior and posterior fontanelles were noted to be open. The sagittal and bilateral coronal sutures were noted to be open as well as the left lambdoid suture and the superior portion of the right lambdoid suture. He was noted to have no neurologic deficits on examination. His x-ray showed open sagittal and bilateral coronal sutures (Figure 1). The left

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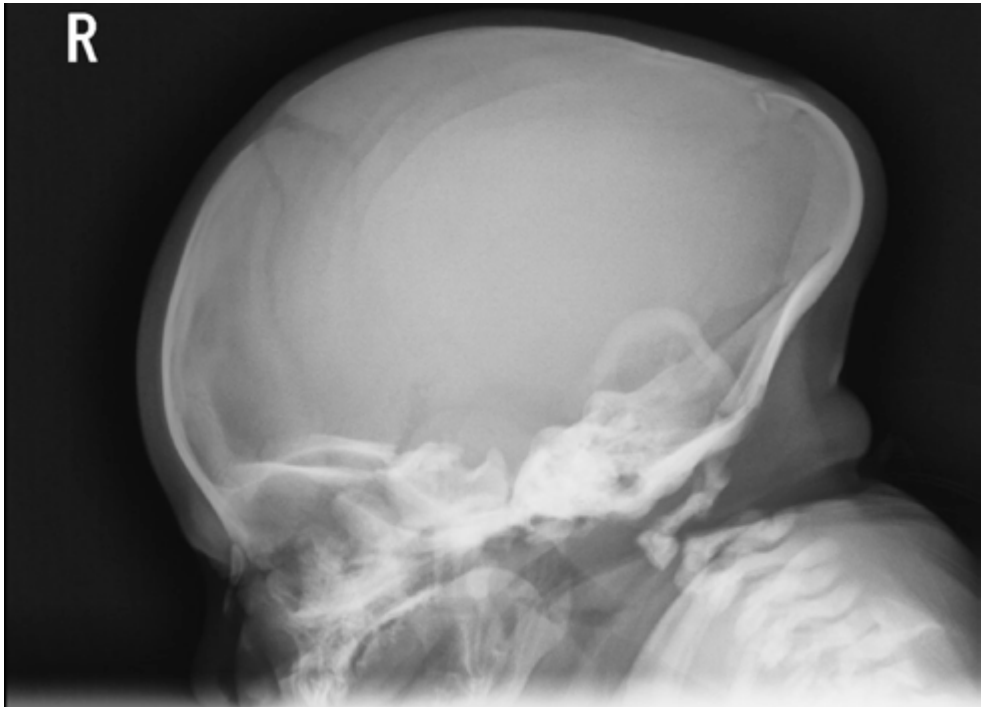


Figure 1 – Lateral skull XRay showing prominent occipital boss.

lambdoid suture was also open, but the right was diminished. In addition, a significant occipital boss was seen. After obtaining appropriate digital imaging (Figure 2), the plan was to have the patient return to clinic in approximately 1 month with the working diagnosis of bathrocephaly. Upon returning to the clinic, his head circumference was 38 cm, placing him at 30th percentile. He remained without any neurological deficits. At that time the decision was made to obtain a low dose 3D CT scan with image reconstruction to complete the evaluation. In his return clinic appointment the skull shape has significantly improved from the previous visit (Figure 3). The CT scan showed no evidence of synostosis (Figure 4). It was determined that no surgical intervention would be necessary at the time.

Discussion

Occipital bone development

The development of the occipital bone occurs in two portions. The skull base and inferior most portion of the bone are formed by endochondral ossification, while the interparietal portion of the bone is formed by membranous ossification (Mowbray, 2005; Gallagher et al., 2013) These two regions are separated by the mendosal suture



Figure 2 – Digital image at the first craniofacial follow up visit.



Figure 3 – Digital image obtained 10 weeks after initial evaluation.

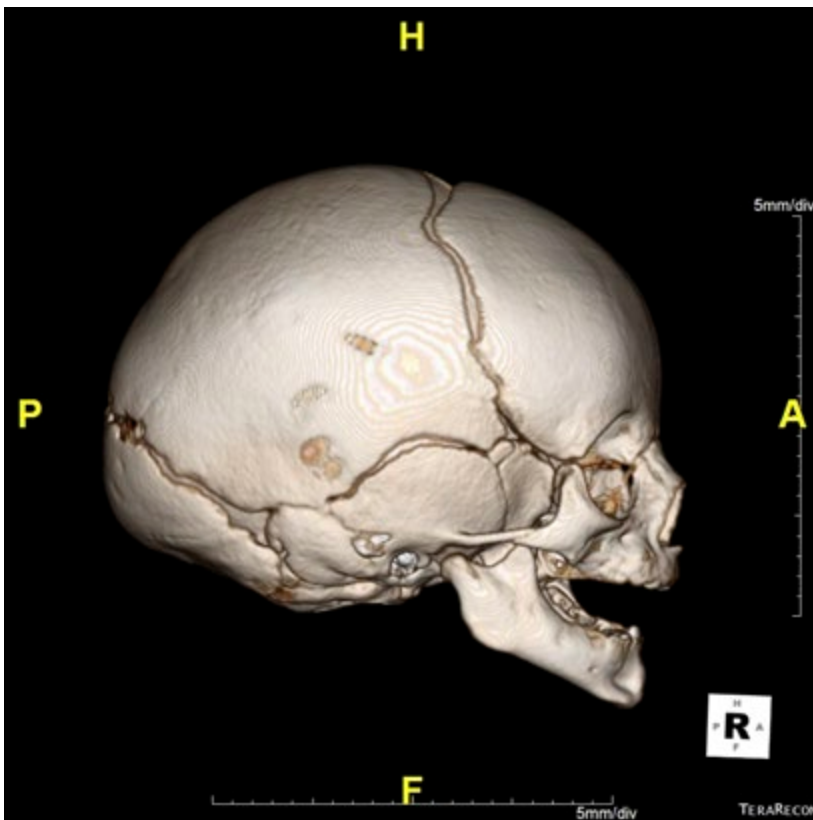


Figure 4 – CT scan with 3D reconstruction 10 weeks since initial evaluation.

(Wu et al., 2011). The skull base and inferior most portions are formed by two ossification sites. This portion of the bone is most typically ossified by 8-9 weeks gestational age (Matsumura et al., 1993; Wu et al., 2011). The interparietal portion undergoes a very different developmental process. It typically has three separate pairs of ossification sites, all of which ossify at different periods of time. The first pair of ossification centers, also known as the primary pair, also ossifies at a gestational age of approximately 8-9 weeks. The secondary pair, which appears just superiorly to the primary pair, undergoes ossification sites at approximately 9-12 weeks gestational age. Finally, a third pair of ossification appears at the uppermost angle of the interparietal portion. Ossification of these centers occurs no earlier than 16-20 weeks of gestational age. However, in most cases, the ossification of both the inferior portion and the interparietal portion is completed by the 20th week of gestation.

Mendosal Suture

The mendosal suture is located at the junction of the interparietal portion and the inferior portion of the occipital bone. The suture line is noted to travel from the asterion on one side of the skull, through the inion, and to the asterion on the other side of the skull (Mulliken and Le, 2008; Gallagher et al., 2013). This suture corresponds with the highest nuchal line (Srivastava, 1992; Wu et al., 2011). It is said by many that the fusion of this suture begins to occur prior to birth. In fact, some postulate that this is the first of all the sutures to fuse (Mulliken and Le, 2008). Shapiro and Robinson (1980) state that midline fusion begins at approximately 12 weeks. However, the lateral portions of the mendosal suture remain open at that time. In fact, it was found that up to 80% of infants younger than 1 year of age had the lateral portions of the mendosal present on skull radiograph. The prevalence of these lateral openings decreases with age, but can still be present in up to 6% of 4-5 year olds. It is important to note this mainly because these lateral openings can be confused radiographically with skull fractures in young children (Miller et al., 2010).

Bathrocephaly

As stated, bathrocephaly is an occipital bone deformity which results from failure of mendosal suture fusion. This is often noted to be an isolated finding, with no associated underlying cranial pathology. The true incidence of this disorder is unknown, with few case reports in the current literature. Gallagher et al. (2013) reported the largest series of 17 patients who were found to have bathrocephaly.

While most frequently bathrocephaly is noted to be an isolated, benign disorder, there are a few reported instances in which other pathology has been noted including synostosis in the lambdoid, coronal, and metopic sutures. It has been postulated that the mendosal suture remains open in these instances to allow for growth in the occipital region, although this has not been definitively proven (Sze et al., 2005; Wu et al., 2011).

It is important to consider other diagnoses when evaluating a posterior cranial deformity. The differential includes ossified cephalohematoma, deformational plagiocephaly, encephaloceles, caput succedaneum, non-ossified cephalohematomas and subgaleal hematomas.

With regards to treatment of bathrocephaly, there is currently no evidence for surgical management of the disorder. It appears that the natural history of the disorder involves resolution without surgical management. As stated prior, this disorder is the result of incomplete fusion of the mendosal suture which leads to bulging of the interparietal portion of the occipital bone. It appears that, even if incompletely fused at birth, the mendosal suture eventually fuses in a delayed fashion. When this occurs, the bathrocephaly deformity resolves.

In conclusion, it appears that bathrocephaly is a relatively rare posterior skull deformity that is benign in nature. There is currently no evidence for a surgical role in the management of this disorder. It is in our opinion that the appropriate management of this disorder involves non-operative treatment and follow up to ensure appropriate development and correction of the deformity over time.

References

- Gallagher E.R., Evans K.N., Hing A.V., Cunningham M.L. (2013) Bathrocephaly: a head shape associated with a persistent mendosal suture. *Cleft Palate Craniofac. J.* 50: 104-108.
- Matsumura G., Uchiumi T., Kida K., Ichikawa R., Kodama G. (1993) Developmental studies on the interparietal part of the human occipital squama. *J. Anat.* 182: 197-204.
- Miller A.J., Kim U., Carrasco E. (2010) Differentiating a mendosal suture from a skull fracture. *J. Pediatr.* 157: 691.
- Mowbray K. (2005) Surface bone histology of the occipital bone in humans and chimpanzees. *Anat. Rec. B New Anat.* 283: 14-22.
- Mulliken J.B., Le M.N. (2008) A craniofacial glossary. *J. Craniofac. Surg.* 19: 705-712.
- Shapiro R., Robinson F. (1980) *The Embryogenesis of the Human Skull: An Anatomic and Radiographic Atlas.* Cambridge, Mass., Harvard University Press.
- Srivastava H.C. (1992) Ossification of the membranous portion of the squamous part of the occipital bone in man. *J. Anat.* 180: 219-224.
- Sze R.W., Hopper R.A., Ghioni V., Gruss J.S., Ellenbogen R.G., King D., Hing A.V., Cunningham M.L. (2005) MDCT diagnosis of the child with posterior plagiocephaly. *AJR Am. J. Roentgenol.* 185: 1342-1346.
- Wu J.K., Goodrich J.T., Amadi C.C., Miller T., Mulliken J.B., Shanske A.L. (2011) Interparietal bone (Os Incae) in craniosynostosis. *Am. J. Med. Genet. A* 155A: 287-294.