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Case Report

Subaponeurotic Fluid Collection of Infancy with Associated Skull Bone Thinning and Protrusion

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Abstract Subaponeurotic (Subgaleal) fluid collection of infancy (SFC) is a rare entity characterised by its delayed presentation with a highly fluctuant non-tender scalp swelling which crosses suture in a stable and neurologically intact infant. SFC typically resolves with conservative management and rarely recurs. To our best knowledge, this is the first reported case with associated localised skull bone thinning and protrusion directly underneath the fluid collection, which lends support to the hypothesis that the fluid collection may result from self-limiting cerebrospinal fluid leakage from skull micro-fracture or fistula.

Key words Collection; Protrusion; Scalp; Subaponeurotic; Skull; Thinning

Introduction

Infant scalp swelling is a well observed phenomenon at birth, ranging from caput succedaneum, cephalohaematoma to life threatening causes such as subaponeurotic haematoma. Clinical examination in general is sufficient in differentiate the cause of scalp swelling. We hereby present a case of delayed scalp swelling of infancy which cannot be attributed to the aforementioned diagnosis.

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Case Report

A 31-day old well baby boy was admitted with a 4-day history of scalp swelling. He was born full term at 37 weeks and 6 days via vacuum extraction due to fetal distress. Birth weight was 2.99 kg. Fetal scalp electrode was placed before delivery. Right parietal caput succedaneum was noted after birth which spontaneously subsided. However, baby developed progressive scalp swelling in the same region at 4 weeks of age. There was no history of head trauma.

Physical examination noted a large fluctuant, mobile, compressible, non-tender fluid collection with fluid thrill at the right parietal region of scalp. The collection was situated above the skull which crossed both coronal and sagittal sutures. The overlying skin was non erythematous. No crepitus was noted on palpation (Figure 1) (Video image). Baby was otherwise well with normal growth parameters, vital signs and feeding. No neurological deficit was identified.

Scalp ultrasound revealed a 1 cm thick anechoic collection over the right parietal region of the scalp in the subaponeurotic region. Computed tomography (CT) scan of the brain showed a hypodense 8 mm thick collection in the aforementioned region with a radiodensity of +6 Hounsfield unit. Of special note an asymmetrical localised

focal thinning of skull was also seen underneath the swelling. The focal calvarial thinning was not associated with any periosteal reaction. Continuity of the skull on imaging was preserved (Figure 2). Haemoglobin, platelet and clotting profile were all within normal range. No clinical evidence of skeletal dysplasia or dysmorphism were present.

Baby was managed conservatively with frequent follow up for monitoring. Aspiration was not performed in our patient as it offered no diagnostic or therapeutic benefit. Clinically the swelling remained fluctuant all along but gradually decreased in size. Repeat ultrasound of the scalp at 5 weeks after presentation showed a reduced thickness of 0.5 cm. The swelling completely resolved on follow up at 12 weeks after initial presentation.

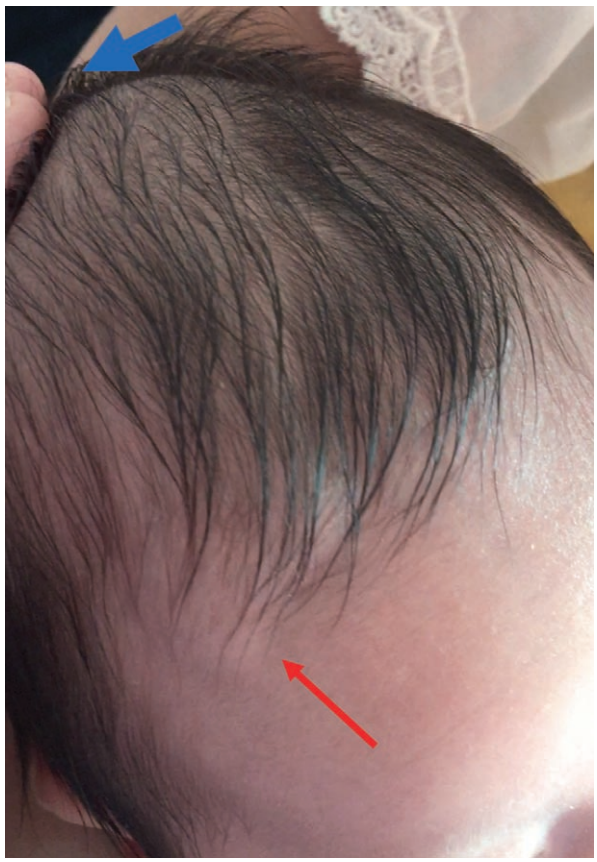

Figure 1 Photo of baby with blue thick arrow highlighting the compressibility of the scalp swelling and the red thin arrow showing the swelling crossed the coronal suture to the upper forehead.

Discussion

Scalp swellings, such as caput succedaneum, cephalohaematoma and subaponeurotic haemorrhage are common and well documented in neonates.[1] Caput succedaneum is usually a result of difficult labour due to compression of the neonate's skull against the maternal pelvis. It usually presents at birth with scalp oedema at the vertex that crosses suture lines. The oedema typically resolves within days. Cephalohaematoma is caused by ruptured blood vessels underneath the periosteum. It typically presents after birth but resolves over the course

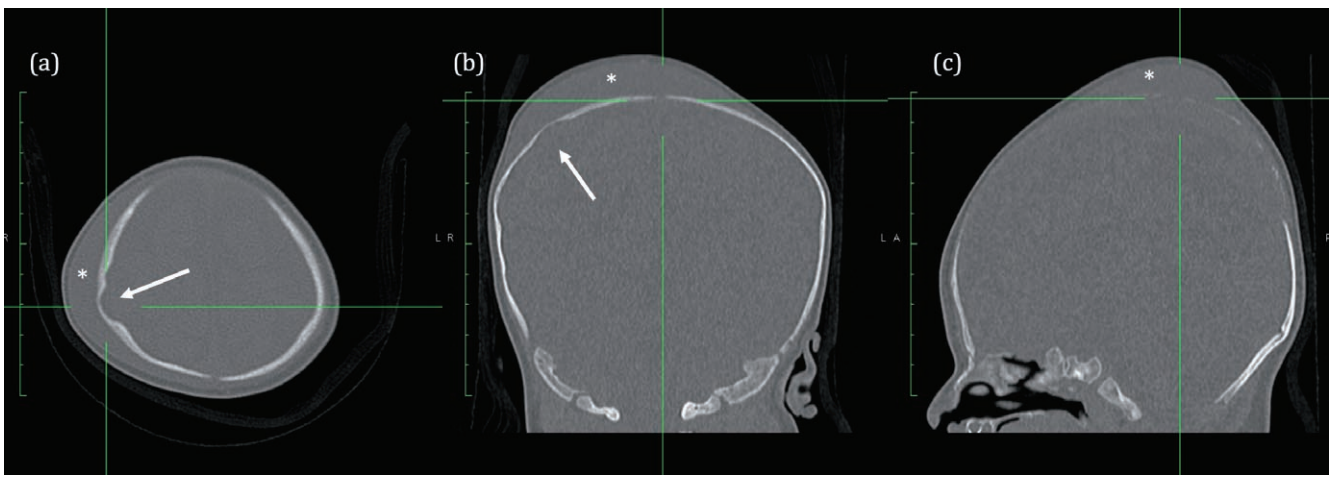

Figure 2 CT scan with (a) axial, (b) coronal and (c) sagittal reconstruction of the skull. The subaponeurotic fluid collection (*) is well appreciated. The associated localised skull thinning and mild protrusion (white arrow) can be seen underneath the swelling. Continuity is preserved with no periosteal reaction is seen.

of weeks. The cardinal feature of cephalohaematoma is its confinement by suture lines. Subaponeurotic haemorrhage is a medical emergency due to bleeding between the aponeurosis and periosteum. It presents within hours of delivery and the neonate may present with hypovolaemic shock and fluctuant scalp swelling that crosses suture.

Subaponeurotic fluid collection of infancy on the other hand is a clinically distinct and rare entity.[2,3] Our experience suggests that its delayed presentation with a highly fluctuant non-tender scalp swelling which crosses suture in a neurologically intact and stable infant are its cardinal features. The disease was first coined by Hopkins[4] in 2002 based on the disease's natural history and radiological findings from six infants. A recent literature review by Wang[5] identified a total of 21 infants in 7 literatures published from 2002 to 2014. Based on the review the swelling presented in infants at the age of 2 to 18 weeks. Complicated delivery as per our case was strongly associated, with 10 out of 21 infants having a history of vacuum extraction, 4 out of 21 infants having a history of forcep delivery and 4 out of 21 infants having a history of fetal scalp electrode monitoring. The swelling invariably resolved over weeks to months, with longest documented duration being 24 weeks.

The exact etiology and pathogenesis remains to be proven. Hopkins[4] initially hypothesised that the swelling may be due to haemorrhage acquired at birth which gradually liquified and increased in size due to subsequent gradual exudation. Schoberer[6] offered an alternative hypothesis in that the swelling was due to cerebrospinal fluid (CSF) leakage. Aspirates were performed on three infants and the content all had serosanguineous appearance. Biochemical analysis of the aspirates showed presence of β2-transferrin and high concentrations of β-trace protein, suggestive of CSF content. As major fractures are not present, Schoberer speculated that the leakage of CSF may be due to microfractures or self-limiting fistula[2] undetectable by neuroimaging. Petraglia's[7] case series of three infants suggests a strong association with fetal scalp electrode monitoring before birth. The author, aligning with Schoberer's hypothesis, suggests that the use of fetal scalp monitoring may result in a small puncture wound leading to gradual leakage and delayed CSF accumulation on the scalp.

The uniqueness of this case lies on the CT scan finding of a localised asymmetrical thinning of the skull bone with mild protrusion just underneath the swelling, which as per our best understanding was not previously reported. Close review by radiologist suggested that skull continuity was preserved and no periosteal reaction was seen, thus speaking against a major fracture. Maternal labour documentation revealed the attachment location for the suction cup used during vacuum extraction to be near the location of the skull defect. Documentation of the fetal scalp electrode insertion location by obstetrician was neither routine nor feasible due to the tiny size of the electrode. We speculate, albeit without documented proof, that the localised skull defect may be induced by birth trauma during fetal scalp electrode monitoring and/or vacuum extraction. There remains a possibility that it represents a congenital skull defect.

In addition, our case lends further support to the hypotheses suggested by recent authors in that the swelling was likely CSF. Clinically the high degree of fluctuance of the swelling was maintained throughout the gradual resolution of the swelling, thus making the content more likely to be CSF than liquified blood. The low radiodensity of the swelling represented by +6 Hounsfield unit in CT scan of the brain is consistent with fluid density as compared to blood. The location relationship between the swelling and skull thinning is likely not coincidental. We speculate that CSF was leaked via the focal skull defect through a self-limiting microfracture or a small fistula leading to a delayed accumulation of swelling. Nevertheless, the swelling responded with conservative management with time, consistent with various international experiences.

Conclusion

Subaponeurotic fluid collection of infancy is a rare entity of infant scalp swelling. Its cardinal features include the delayed presentation of a highly fluctuant non-tender scalp swelling which crosses suture in a neurologically intact and stable infant. Strong association exist between SFC and complicated delivery procedures such as vacuum extraction and fetal scalp electrode placement. We speculate that these procedures prior to birth in our case may have caused the localised asymmetrical skull bone thinning and protrusion just underneath the swelling, therefore leading to a delayed leak of CSF through a microfracture or small fistula.

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Declaration of Interest

The authors declare that there is no conflict of interest.

Video Image

Additional video images may be found in the online version of this article.

http://hkjpaed.org/video/v26n01_video1.mp4

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