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CASE REPORT

A rare cause of scalp swelling in infancy: delayed subaponeurotic fluid collections in five cases

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Abstract

Subaponeurotic (subgaleal) fluid collection (DSFC) is a rare clinical entity of unknown etiology. We aimed to present our series of infants who were diagnosed with DSCF at Ankara University Children's Hospital. We retrospectively reviewed clinical findings, imaging studies, laboratory tests, management, and clinical courses of infants diagnosed with DSCF between June 2014 and June 2018. Five infants (4 males, 1 female), aged 5–14 weeks, were identified during the study period. All deliveries were non-progressive (3 normal deliveries, 2 cesarean sections) while instrumentation (vacuum extraction or forceps) was used in 2. History of recent trauma, concern for child abuse, and family or personal history of coagulopathy were negative for all patients. Conservative management with the spontaneous resolution was observed in 2 to 12 weeks in all infants. Although rare, DSFC should always be kept in mind in the differential diagnosis of scalp swelling in young infants. The diagnosis is primarily clinical, and current treatment is conservative.

Keywords Scalp swelling in infancy · Subgaleal · Subaponeurotic fluid collection · Traumatic delivery

Abbreviations

DSFC	Delayed subaponeurotic (subgaleal) fluid collection
CSF	Cerebrospinal fluid
MRI	Magnetic resonance imaging
US	Ultrasonography
CS	Cesarean section
CT	Computed tomography

Introduction

Scalp swelling is a frequent clinical condition in young infants. It may be caused by cephalohematoma, caput succadaneum, subgaleal hemorrhage, and delayed subaponeurotic (subgaleal) fluid collection (DSFC). DSFC, defined as fluid accumulation between the scalp aponeurosis and the periosteum, is a rare clinical entity manifesting beyond the neonatal period [1]. Few case reports exist in the current literature, and studies suggest inadequate awareness of health care providers considering DSCF [2].

Although its etiology is unclear, birth trauma leading to disruption of venous drainage, lymphatic drainage, and cerebrospinal fluid (CSF) leakage has been implicated as the main mechanism [1, 3, 4]. It is described as a soft, mobile, compressible, non-tender swelling which crosses the suture lines. It usually emerges between postnatal 4 and 18 weeks in otherwise healthy infants with no history of recent trauma. The diagnosis is clinical and management is conservative.

We aimed to call attention to DSFC, a clinical condition which represents as a diagnostic challenge for most physicians because of its rareness. Herein, we present our experience at Ankara University Children's Hospital.

Case report

We have investigated 5 infants, aged between 5 and 14 weeks (4 males, 1 female), who were diagnosed with DSCF between June 2014 and June 2018 (Table 1). We have reviewed clinical

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Table 1 Summary of cases with delayed subaponeurotic fluid collections in infancy

Patient	Mode of delivery	Age at onset (weeks)	Description of the lesion	Imaging	Treatment	Resolution (weeks)
1	Vaginal, vacuum	9	Left parieto-occipital, 8 × 5 cm	CT	Conservative	4
2	CS (non-progressive delivery)	8	Right parieto-occipital, 6 × 5 cm	Skull radiography, US, MRI	Conservative	2
3	Vaginal, vacuum, and forceps	13	Occipital, 2 × 2 cm	Skull radiography, US	Conservative	4
4	CS (non-progressive delivery)	14	Occipital, 10 × 10 cm	Skull radiography, US	Conservative	7
5	Vaginal, prolonged	5	Right parieto-occipital 15 × 15 cm	US	Conservative	12

CS cesarean section, *US* ultrasonography, *CT* computed tomography, *MRI* magnetic resonance imaging

findings, imaging studies, laboratory tests, management, and clinical courses. The study protocol was approved by our university's ethical committee (study approval number: 11-730-18, June 2018). Informed consent was obtained from all included individual participants' families.

All deliveries were non-progressive (3 normal deliveries, 2 cesarean sections (CS)). Instrumentation (vacuum, forceps) was implemented in two normal deliveries. None of the infants received fetal scalp electrodes. Two infants displayed cephalohematoma after birth which resolved in 7–10 days. Age of onset of scalp swelling was postnatal 5–14 weeks. There was no history of recent trauma, concern for child abuse, family/personal history of coagulopathy, swelling, hematoma, or bleeding in other areas of the body in any infant. Scalp swelling occurred spontaneously and increased in size in a few days. Swelling, located at the occipital or parieto-occipital regions, was soft, non-tender, and fluctuant and crossed the suture lines (Fig. 1). There was no bruising of the surrounding skin. Vital signs, general appearance, and neurological examination were normal.

Skull X-ray showed large soft tissue swelling and no skull fracture (Fig. 2). Ultrasonography (US) demonstrated compressible, hypoechoic subgaleal fluid collection which crossed the suture lines. Case 1 had brain computed tomography (CT) before admission, and a homogenous, hypodense subgaleal collection with normal brain parenchyma was noted. Brain magnetic resonance imaging (MRI) of case 2 revealed subgaleal fluid collection overlying the right parieto-occipital region (Fig. 3). Whole blood count and coagulation profiles were normal. All patients were managed conservatively with spontaneous resolution in 2–12 weeks.

Discussion

Although extremely rare, DSFC may easily be diagnosed clinically and managed conservatively. We have presented the

Fig. 1 Image of case 5 shows a soft, compressible scalp swelling (magnification: 0.68 × 0.80)

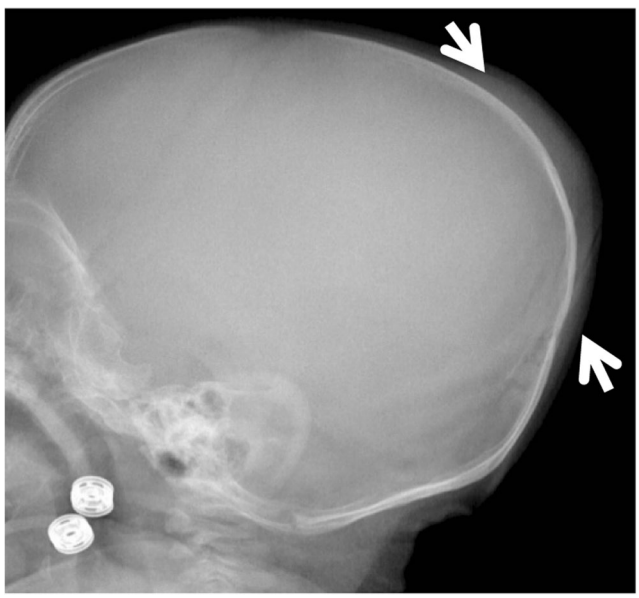

Fig. 2 Skull X-ray of case 2 shows soft tissue swelling in parieto-occipital region (arrows)

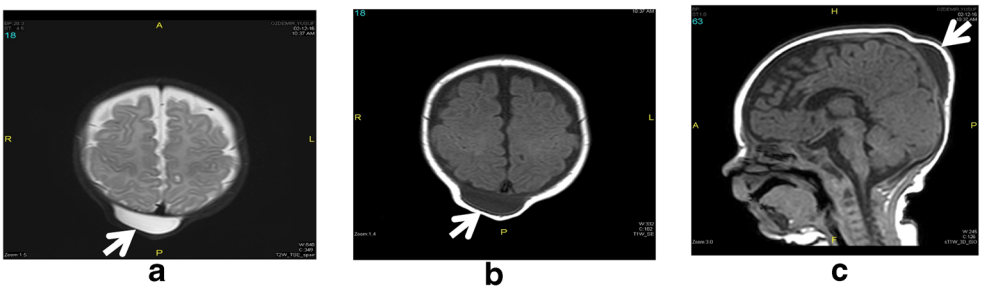

Fig. 3 Brain magnetic resonance imaging of case 2 showing subaponeurotic fluid collection located at the right parieto-occipital region and crossing the sagittal suture. **a** Axial T2-weighted image. **b** Axial T1-weighted image. **c** Sagittal T1-weighted image

first cases diagnosed with DSFC in infancy from Turkey to raise awareness of this entity.

Worthen et al. published an article concerning a blog, analyzing experiences of families whose infants developed DSFC [2]. The authors realized family concerns regarding lack of provider qualification, the performance of nonessential procedures, and suspicion of child abuse. We believe that increasing emphasis on DSFC will provide physicians better understanding of this condition.

Although the pathophysiology of DSFC is not fully elucidated, most reported infants share a similar history of prolonged or instrumented delivery [5, 6]. Smith et al. reported 11 infants diagnosed with DSCF, all of whom underwent instrumented delivery or emergent CS [5]. In our series, two normal deliveries required instrumentation while CS was preferred because of non-progressive delivery in two cases. Yet, it is uncertain why swelling represents later. Hopkins et al. proposed that hematoma formed at birth may liquefy over time while further exudation causes further enlargement [1]. They also postulated that birth trauma may lead to disruption of lymphatic drainage or CSF leak, resulting in a slow collection of fluid. Petraglia et al. reported 3 infants with DSFC who were monitored by fetal scalp electrodes during delivery and speculated that this procedure may have caused CSF leak [4]. Another case report presented an infant who developed DSFC following lumbar puncture, raising concern for the potential effect of CSF dynamic changes [7].

As compatible with literature, our patients showed typical clinical characteristics. The age of onset of scalp swelling also indicated similarity with published reports [1–10].

The differential diagnosis for DSFC consists of accidental/non-accidental head injury leading to subgaleal (subaponeurotic) hemorrhage, caput succadaneum, and cephalohematoma resulting from traumatic delivery [8]. Clinical assessment can distinguish DSFC from these reasons. Subaponeurotic hemorrhage resulting from traumatic delivery appears right after birth and may be life-threatening because of massive intravascular volume depletion. It may also occur in shaken baby syndrome, but most cases are unstable [8]. Cephalohematoma and caput succadaneum also occur immediately after birth. Cephalohematoma does not cross the suture lines. Inversely, DSCF of infancy appears weeks after birth and crosses the suture lines.

Routine radiological evaluation is not indicated for the diagnosis of DSCF. Petraglia et al. recommended MRI to evaluate for a focus of CSF leak in persistent cases [4]. Medows et al. proposed further investigation if trauma or child abuse is of concern [7]. Vaibhav et al. suggested skull X-ray to rule out fractures in case of suspected abuse [8]. Although we performed more extensive radiological investigations initially, we were more confident with US and/or X-ray as we became familiar with this specific diagnosis. Even though most authors do not recommend routine laboratory evaluation, we have tested our patients for coagulation defects and observed normal results. Similarly, Wang et al. also reviewed coagulation profiles in their series [6].

Current management is conservative for DSCF. Although few researchers have attempted to aspirate the fluid, reacculumation was observed in all [3, 10]. Spontaneous resolution was achieved in other case reports, as in our cases, within weeks to months without recurrence [4–8].

Conclusion

Although rare, DSFC should always be kept in mind in the differential diagnosis of scalp swelling in young infants. The diagnosis is primarily clinical. Further investigations should only be undertaken in unclear diagnosis. Current management is conservative.

Author contributions EI and NECI designed the study. NECI had the primary responsibility in the formation of the initial draft. All authors provided substantial contributions to the design of the work and the acquisition or the interpretation of the data, revised the initial draft, approved the final manuscript, and agreed to be accountable for all aspects of the work.

Compliance with ethical standards

Conflict of interest The authors declare no conflict of interest.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

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