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Case Report

Delayed subaponeurotic fluid collection on an infant's head: Underreported case and review of the literature

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Received : 22 December 2020
Accepted : 01 April 2021
Published : 17 May 2021

DOI
10.25259/SNI_932_2020

Quick Response Code:

ABSTRACT

Background: Delayed subaponeurotic fluid collection (DSFC) is a relatively uncommon problem, probably under reported soft swelling in the scalp which usually develops in infancy that occurs weeks to months after birth. Although the exact etiology remains unclear, several theories have been postulated such as (i) cerebrospinal fluids (CSF) leak from microfractures of the skull and (ii) disrupted lymphatic drainage, gradually liquefying subaponeurotic bleeding. Here, we reported typical clinical findings of DSFC and analysis of the fluid aspirate from our patient. To the best of our knowledge, this is the first case reported from Asia, particularly from Indonesia.

Case Description: A healthy 2-month-old girl infant presented with 2 weeks history of occipital painless fluctuant scalp mass with no swelling. She was born at term from a nulliparous mother; by emergency cesarean delivery following failed induction of labor. There was no history of scalp injury at birth nor recent head trauma; ultrasonography showed translucent fluid in subaponeurotic or subgaleal space. The fluid collection was noted to be fluctuant, free-flowing across suture lines, without discoloration or bruising; when placed supine, the fluid collected at her midocciput. A diagnostic tap confirmed the presence of serosanguinous CSF led to a diagnosis of DSFC.

Conclusion: Herein, we reported the first DSFC case from Indonesia. With no previous experience of the condition, a definitive diagnosis was possible through a very carefully physical examination and history taking, along with a good communication among the neurosurgeon, pediatrician, and the clinical pathologist.

Keywords: Delayed subaponeurotic fluid collection, Infancy, Underreported case

INTRODUCTION

Acquired scalp swelling in neonates and infants is a common condition. Several diagnostic considerations include subaponeurotic or subgaleal hemorrhage, cephalhematoma, caput succedaneum, and delayed subaponeurotic fluid collections (DSFCs).[3,11] DSFCs are recently acknowledged as one of the causes of idiopathic scalp swelling in infants; the pathophysiology and etiology of DSFCs remain unknown, although several theories have been postulated such as (i) cerebrospinal fluids (CSF) leakage from microfractures of the skull and (ii) disrupted lymphatic drainage, gradually liquefying subaponeurotic bleeding.[3,11] Based on our knowledge, this is the first reported case of DSFCs from Indonesia.

CASE PRESENTATION

A healthy 2-month-old girl infant was referred to our hospital by her anxious parents, presented with 2 weeks history of occipital painless fluctuant scalp mass with no swelling and suspected of having a subaponeurotic or subgaleal hemorrhage. She was initially admitted to the Outpatient Pediatric Clinic, Edelweiss Hospital, Bandung, West Java, Indonesia, before being consulted to the Department of Neurosurgery. She was born at term from a primigravida mother, birth was by emergency cesarean section following failed induction of labor with no history of fetal scalp electrode usage; scalp injury at birth nor recent head trauma. The fluid collection was noted to be fluctuant, free-flowing across the suture lines, without discoloration or bruising skin and when supine, the fluid collected at his midocciput [Figures 1a and b]. Ultrasonography revealed a mobile hypodense anechoic fluid collection beneath the hypoechoic aponeurosis without the presence of cyst wall and crossed sagittal suture lines [Figures 2a and b], internal solid component/active flow signals on color Doppler, and no evidence of fracture. A diagnostic tapping confirmed the presence of serosanguinous fluid [Figure 3]; biochemical results showed glucose 94 μg/dl and albumin 2120 μg/dl with leukocytes 252 cells/μl and Beta$_2$-transferrin positive. Hematology results and coagulation profiles were universally unremarkable. Swelling reappeared after the initial aspiration, but we did not performed re-aspiration, we decided to treat conservatively along with compression-head-bandage. It resolved completely in 3½ weeks and no recurrence during the follow-up period.

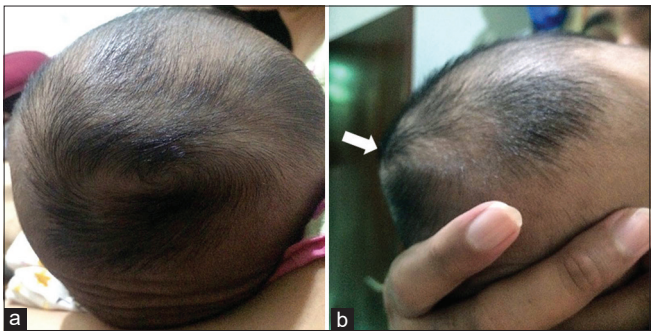

Figure 1: Fluctuant region beneath scalp. Note that there is no discoloration, and the collection does not obey suture lines (a). The arrow indicates the edge of fluid collections, which is migratory depending on patient position (b).

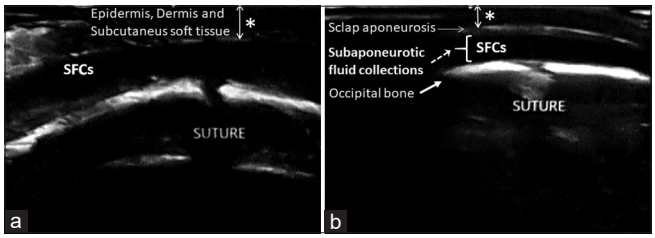

Figure 2: The important diagnosis feature showing subaponeurotic fluid collections crossing above the cranial suture (a). Ultrasonography revealed mobile hypodense anechoic fluid collections beneath the scalp aponeurosis without the presence of cyst wall and crossed sagittal suture lines (b).

DISCUSSION

DSFCs are recently described clinical entity within only 62 case reports in medical literatures up to date (searching PubMed using the search term "DSFCs"). All 16 articles were reviewed by the first author and data were then extracted). There were 15 cases reported from the United States of America,[5,7,9,14] 12 cases from the United Kingdom,[4,6,10,15] 11 cases from Ireland,[12] 9 cases from Canada,[16] 5 cases from Germany,[11] 5 cases from Turkey,[13] 2 cases from India,[2,8] and 1 case each from Australia,[13] the Netherland,[1] and Indonesia (this report) shown in [Table 1]. Overall, mean of age when DSFCs were diagnosed was 8.2 ± 3.7 weeks, ranges from 2 to 18 weeks.[1-16] Mean resolution of scalp swelling was 7.4 ± 5.4 weeks, ranges from 2 to 24 weeks.[1-16] Since its rarity and limited cases reported around the world, many physicians never seen DSFCs before and may not recognize its clinical entity in the first place; Worthen *et al.* published article concerning rare scalp mass that identified through a disease-specific blog, analyzing experiences of 69 families whose infants developed DSFCs.[17]

Hopkins *et al.* reported the first six cases of DSFCs that managed conservatively and one case was managed by a needle aspiration of the collection on two occasions when serosanguinous fluid was aspirated ×2 after diagnosis.[3] Schoberer *et al.*[11] and Munjal and Kumar[8] reported one case managed by 2 times aspiration, since they observed the reaccumulation of the fluid. McAloon and McCabe,[6] Roy

Figure 3: A diagnostic aspirate tap confirmed the presence of serosanguinous subaponeurotic fluid collections.

and Magdum,[10] and Chalipat *et al*.,[2] as in our case, only aspirate once even though we observed reaccumulation of fluid, but decided not to aspirate the fluid for the 2nd time. Further, Hopkins *et al*. showed in one case that the aspirated fluid was sterile on microbiological test and no biochemical analysis was performed. In our case, we did a tap of the fluid; it was beta-trace positive, suggesting that it is CSF as previously described.[2,3,6,8,10,11] The tapping is not always advocated and many authors even advise against tapping these collections.[1,4,5,7,9,12-16] Aspiration itself does not resolve the fluid collection, since its nature as self-resolving lesion; not mention that there is a small risk of introducing infection and bleeding. From our own experience and the literatures, we not recommend needle aspiration for future cases.

All patients have no recent head trauma or illness. Growth, development, and behavior are normal.[3] The patient's physical examination showed a fluctuant fluid collection under the scalp. It is typically located over the superior occiput, crosses suture lines, and is without discoloration; as in our case, clinical examinations consistent with the typical appearance of DSFCs of infancy. Her benign fluid collections occur spontaneously 2 months after birth and probably associated with a difficult labor, which, in our case, was failed of induction. Ultrasonography imaging demonstrates a mobile hypodense anechoic fluid collections beneath the hypoechoic aponeurosis without the presence of cyst wall and crossed sagittal suture lines, internal solid component or active flow signals on color Doppler, and no evidence of skull fracture.[1,3,4,8,9,11-14,16] While the natural history of DSFCs is benign, its presence can be emotionally draining for the parents; as well as for the physicians not familiar with this rare condition.

DSFCs is self-limiting condition that might masquerades a nonaccidental injury and should not prompt work-up in an otherwise clinically well infant.[3,4,7-9,11,14-16] DSFCs can be distinguished from other types of fluid collections (subaponeurotic hemorrhages, cephalohematomas, and caput succedaneum) by physical examination and history taking. DSFCs particularly could be distinguished from subaponeurotic hemorrhages, which occur in the same space, but appear immediately after birth, and could cause intravascular volume loss and sometimes lead to life-threatening conditions.[14,16] Similarly, cephalohematomas and caput succedaneum also present immediately after birth. In contrast, DSFCs appear in between 2 weeks and 4.5 months of life; with mean 2 ± 1 months.[1-16] Cephalohematomas are further distinct since they are limited by suture lines and eventually calcify from the outer edges resulting in a smooth bony lump on the skull. This is in contrast to DSFCs, which gradually reduce in size and resolve within 2 weeks up to 6 months; with mean 2 ± 1.5 months.[1-16] The exact mechanism underlying DSFCs is not completely understood, the diagnosis remains challenging for every physicians to solve their first case. It is important that clinicians are familiar with

Table 1: Summary of the all reporting 62 cases in world literature (including our case, until 2020).

Authors/year/country	No. of cases/gander	Age range/ mean (weeks)	Imaging	Treatment	Resolution of scalp swelling/ mean (weeks)
Hopkins *et al*./2002/UK	6 cases/NR	3–18/8.75	X-ray and US	5 conservative; 1 aspiration×2	2–24/8.8
Vaibhav *et al*./2010/UK	4 cases/3 boys, 1 girl	4–14/7.5	X-ray, US, MRI	Conservative	3–8/4.25
McAloon *et al*./2012/UK	1 case/NR	12	X-ray	Aspiration	NR
Roy *et al*/2014/UK	1 case/boy	10	CT, MRI	Aspiration	NR
Petraglia *et al*./2010/USA	3 cases/NR	5–9/6.3	X-ray, CT	Conservative	5–9/6.7
Medows *et al*./2014/USA	1 case/boy	14	CT	Conservative	5
Lee *et al*./2018/USA	1 case/boy	8	US	Conservative	5
Stephan *et al*./2019/USA	10 cases/5 boys, 5 girls	2–17/9.7	X-ray, US, CT	Conservative	10/NR
Smith *et al*./2016/Ireland	11 cases/NR	3–12/7.1	X-ray, US, CT, MRI	Conservative	1–12/4
Wang *et al*./2016/Canada	9 cases/4 boys, 5 girls	2–11/5.6	X-ray, US, CT, MRI	Conservative	2–20/10.5
Schoberer *et al*./2008/Germany	5 cases/2 boy, 1 girl, NR	7–8/7.8	US, CT, MRI	2 conservative2 aspiration×11 aspiration×2	NR
Ilarslan *et al*./2019/Turkey	5 cases/4 boys, 1 girl	5–14/9.8	X-ray, US, CT, MRI	Conservative	2–12/5.8
Chalipat *et al*./2014/India	1 case/girl	4	MRI	Aspiration	12
Munjal *et al*./2017/India	1 case/boy	13	US	Aspiration×2	7
Aries *et al*./2009/Netherland	1 case/boy	10	US, MRI	Conservative	NR
Smith *et al*./2020/Australia	1 case/girl	8	US, CT,	Conservative	NR
Faried *et al*./2020/Indonesia	1 case/girl	8	US	Aspiration	3.5

UK: United Kingdom, USA: United States of America, NR: Not recorded, US: Ultrasonography, MRI: Magnetic resonance imaging, CT: Computed tomography

this condition as their awareness will facilitate avoidance of anxiety in parents, unnecessary medical and child protection investigations.

CONCLUSION

The diagnosis of DSFCs is primarily clinical examination and history taking, along with a good communication among colleagues; current gold standard treatment remains conservative.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

Financial support and sponsorship

Dr. Ahmad Faried supported by Grants-in-Aids from Universitas Padjadjaran, Bandung, Indonesia.

Conflicts of interest

There are no conflicts of interest.

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How to cite this article: Faried A, Imron A, Aliyannissa A, Indrawati D. Delayed subaponeurotic fluid collection on an infant's head: Underreported case and review of the literature. Surg Neurol Int 2021;12:233.