Case Report

Spontaneous Subgaleal Hemorrhage in a Girl with Impaired Adrenaline-Induced Platelet Aggregation

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Introduction: *Subgaleal hematomas usually develop followed a birth trauma in neonates. This entity is extremely rare in older children and may be associated with coagulation disorders or subaponeurotic vascular malformations.*
Case Report: *The authors report a spontaneous subgaleal hematoma in a 9-year-old girl without scalp injury. Only platelet aggregation test was identified as an impaired responsiveness of platelets to adrenaline with normal coagulogram. This patient underwent both computerized tomography and complete magnetic resonance imaging including angiography and venography of the head. Of these, extensive bilateral frontotemperoparietal subgaleal hematoma was observed without vascular malformation.*
Conclusion: *Spontaneous subgaleal hematoma can be associated with platelet function defect.*

Keywords: *Spontaneous subgaleal hematoma, Impaired adrenaline-induced platelet aggregation*

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Case Report

A 9-year-old girl with subgaleal hematoma presented with diffuse swelling of the scalp above supraorbital line 8 days before visiting the hospital. She had no previous history of trauma or hair-pulling. This swelling area was mildly tender and her head circumference gradually increased from 54 to 62 cm over 3 days of admission. She also complained of a severe generalized headache. There was no history of an underlying bleeding disorder except for easy bruising at both legs and prolonged blood oozing after a dental extraction in the past two years.

On physical examination, she was febrile at 38.5°C and mildly tachycardic. She had a soft, mildly tender, non-pitting, fluctuating, and swelling of scalp over supraorbital ridge. She looked pale but not icteric. No audible pulsatile bruit was detected over an affected area. Neurological examination was normal. Initial laboratory tests revealed a hemoglobin of 8.4 g/dL, hematocrit of 25.7%, platelet count of 690 x 10^9/L, mean corpuscular volume (MCV) of 66.5 fL, mean corpuscular hemoglobin (MCH) of 21.7 pg, red cell distribution width (RDW) of 16.0% and red cell morphology showed moderately hypochromic and microcytic erythrocytes. The white blood cell count and differential were normal. A non-contrasted computed tomographic scan of the head described a large bilateral frontotemperoparietal extracranial subgaleal collection with blood radiodensity. Magnetic resonance imaging (MRI), angiography (MRA) and venography (MRV) of the head showed no evidence of aneurysm and vascular malformation.

A bleeding time by modified Ivy test and full coagulation profiles were reported within the normal range (Table 1). A platelet aggregation study with collagen (10 μg/mL), adenosine 5'-diphosphate or ADP (5 and 10 μM), ristocetin (1.2 μg/mL) and adrenaline (5 and 10 μM) was performed in platelet rich plasma (PRP) using PACK-4. Aggregation was measured by light scattering on this platelet aggregation chromogenic kinetics system. In the presented case, only abnormal low platelet aggregation response to adrenaline 5 and 10 μM of 4.1% and 9.5% were identified, respectively. Furthermore, platelet aggregation study of her younger sister showed a similar abnormality which suggested hereditary

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impaired platelet responsiveness to adrenaline in this family (Table 2). Unfortunately, no platelet aggregation test in both parents was performed because they worked in a long distance away.

Needle aspiration of a total 150 mL blood was performed for headache relief and aspiration culture which yielded no organism growth. Two days after the aspiration, she became afebrile and hematoma gradually resolved. The pressure dressing was applied over the head for one week to prevent blood and serous reaccumulation. Without an evidence of re-bleeding, platelet transfusion was not given after the scalp aspiration. The patient was discharged on oral cloxacillin and iron supplement. Four weeks after hospitalization, head circumference returned to baseline and repeated platelet aggregation study revealed persistent abnormal platelet aggregation similar to the previous study.

Table 1. Results of coagulation studies in this patient

Measurements	Findings	Normal range
APTT (sec)	35.0	30.0-38.0
PT (sec)	15.0	11.5-15.5
TT (sec)	11.0	9.0-12.0
Bleeding time (min)	3	<8
FV (%)	99.0	40.0-150.0
FVII (%)	71.0	50.0-50.0
FVIII (%)	84.0	40.0-150.0
FIX (%)	61.0	50.0-150.0
FX (%)	76.0	60.0-150.0
FXI (%)	69.0	65.0-150.0
FXII (%)	70.0	60.0-150.0
FXIII screening	Normal	Normal
VWF Ag (%)	140.0	50.0-150.0
RisCoF (%)	72.0	50.0-150.0
Fibrinogen (mg/dL)	257.0	184.0-402.0

APTT = activated partial thromboplastin time; PT = pro-thrombin time, TT = thrombin time; FV = factor V clotting activity; FVII = factor VII clotting activity; FVIII = factor VIII clotting activity; FIX = factor IX clotting activity; FX = factor X clotting activity; FXI = factor XI clotting activity; FXII = factor XII clotting activity; FXIII screening = factor XIII screening; VWF Ag = von Willebrand factor antigen; RisCoF = ristocetin cofactor activity

Table 2. Results of platelet aggregation study of the patient and younger sister

Agonists		Findings (%)		Normal control (%)
		Patient	Sister	
Collagen	5 μM	109.1	67.7	97.3
ADP	5 μM	60.9	62.3	69.1
ADP	10 μM	92.7	73.6	88.0
Ristocetin	1.2 μg	115.5	76.4	78.6
Adrenaline	5 μM	4.1	10.5	81.4
Adrenaline	10 μM	9.5	9.7	87.3

ADP = adenosine 5'-diphosphate

Discussion

Subgaleal hemorrhage usually develops between aponeurosis and periosteum of the cranium. Infants are more susceptible to subgaleal bleeding in this area due to multiple small and large vascular supplies. Subgaleal hemorrhage can lead to the collection of a significant volume of blood[1]. Not only post-traumatic in etiology but also several reports had described spontaneous subgaleal hematoma from the following conditions included periosteal varix which is an anomalous periosteal venous structure on the outer table of skull, bilateral arteriovenous shunting lesion involving the diploic veins and aggressive osteoblastoma of the skull[2-4]. Coagulopathies including vitamin K deficiency, congenital factor VII deficiency, hemophilia A and B, and platelet dysfunction have been reported as bleeding causes of subgaleal hemorrhage[5-8].

From a previously reported patient with abnormal platelet function of Kirkpatrick and et al[9], prolonged bleeding time over 20 minutes and defective platelet aggregation response to collagen, ADP and epinephrine were evaluated. After 6 units of platelet transfusion, platelet aggregation was normalized and scalp hematoma was tapped to release a loculation of blood even the repeated bleeding time was persistently prolonged. However, no magnetic resonance imaging or angiography was performed to identify a vascular malformation or to ascertain the site of bleeding in this report.

The present report is the second case of spontaneous subgaleal hemorrhage in older children with the absence of trauma. The presented patient revealed remarkably abnormal low platelet aggregation response only to adrenaline along with a previous history of easy bruising and delayed blood oozing after dental procedure. Although with normal bleeding time, the possibility of platelet dysfunction could not be entirely excluded. From literatures review, the role of bleeding time to predict the bleeding risk is poorly

defined in a patient with unknown history of abnormal hemorrhage[10-13]. Adrenaline receptor defect is the most likely etiology explaining this bleeding manifestation due to impaired responsiveness of platelet aggregation to adrenaline is the merely solid abnormal finding in this patient and her sister. Besides no reported case of spontaneous subgaleal hematoma in Thai children was previously described in non-neonatal age group and only one patient from India was stated as having subgaleal hemorrhage after hair-pulling without identifiable cause of bleeding abnormality[14].

Subgaleal hematomas often resolve spontaneously[15]. From these reasons, needle aspiration and surgical drainage should be omitted to avoid scalp infection[16]. Most cases had been closely observed only. However, subgaleal tapping was indicated in some cases in order to shorten the time required for blood resorption and to reduce the risk of calcification and reaccumulation of blood[17]. However, the time taken for hematoma to resolve could not be altered by an aspiration[18]. On the other hand, in patients with coagulation disorder, urgent replacement of specific clotting factor, desmopressin or platelets therapy are more efficacious than other modalities to manage subgaleal blood loss.

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ภาวะเลือดออกใต้เยื่อหุ้มกะโหลกศีรษะในเด็กโตจากความผิดปกติในการเกาะกลุ่มของเกล็ดเลือดซึ่งไม่ตอบสนองต่อสารอะดรีนาลิน

สะการะ หัศภาดล, อำไพวรรณ จวนสัมฤทธิ์, อนุชา สร้อยสำโรง

รายงานผู้ป่วยเด็กหญิงไทยอายุ 9 ปี ซึ่งหนังศีรษะบวมขึ้นในระยะเวลา 8 วันโดยปฏิเสธการกระทบกระแทกบริเวณศีรษะ จากการตรวจร่างกายพบเลือดออกใต้เยื่อหุ้มกะโหลกศีรษะ ระหว่างที่นอนพักในโรงพยาบาลพบว่าเส้นรอบศีรษะมีขนาดเพิ่มขึ้นจาก 54 เป็น 62 เซนติเมตร ผลการตรวจทางห้องปฏิบัติการเบื้องต้น ได้แก่ การ ตรวจนับเม็ดเลือด การวัดค่าการแข็งตัวของเลือด และ bleeding time ไม่พบสาเหตุที่อธิบายภาวะเลือดออกง่าย รวมไปถึงการตรวจเพิ่มเติมด้วยภาพถ่ายด้วยคลื่นแม่เหล็กไฟฟ้า (magnetic resonance angiography and venography) การวัดระดับ สารที่ช่วยในการแข็งตัวของเลือดต่างๆ ก็ล้วนเป็นปกติ มีเพียงความผิดปกติจากการเกาะกลุ่มของเกล็ดเลือด ซึ่งไม่ตอบสนองต่อสารอะดรีนาลินซึ่งมีค่าต่ำกว่าค่าควบคุมปกติอย่างมาก นอกจากนี้ยังพบความผิดปกติของเกล็ดเลือดดังกล่าวในน้องสาวของผู้ป่วยอีกด้วย จึงแนะนำให้ส่งตรวจการเกาะกลุ่มของเกล็ดเลือดเพื่อวิเคราะห์หาสาเหตุของเลือดออกใต้เยื่อหุ้มกะโหลกศีรษะในผู้ป่วยเด็กโต เพื่อให้การรักษาที่เหมาะสมต่อไป