AJNR Am J Neuroradiol 21:308–309, February 2000

Case Report

Intraosseous Hematoma in a Newborn with Factor VIII Deficiency

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Summary: We present an unusual case of an intraosseous hematoma in a newborn with a known bleeding disorder. This cephalohematoma was diagnosed shortly after birth, was entirely within the bony skull, and was in fact determined to be an intraosseous hematoma. The initial CT scans showed the unusual appearance and location of the lesion; later scans showed a significant amount of remodeling, with resolution of the hematoma. Although the coagulopathic diagnosis was independent of this finding, a bleeding disorder might be considered in other patients with similar CT findings.

Cephalohematomas in the newborn are well described and are most often related to vaginal birth trauma. Hematomas may occur in the subcutaneous, subaponeurotic, or subperiosteal spaces (1). We present a case of an intraosseous hematoma as a presenting sign of factor VIII deficiency in a newborn. This case is unique because of the unusual appearance and location of the hematoma and the remarkable amount of subsequent bone remodeling. We thought that the bone remodeling was related to the expansile nature of this lesion, which suggested repetitive in utero hemorrhage.

Case Report

A 2-month-old white male patient was initially seen in a general pediatric clinic for evaluation of bruised and swollen hands. The child was born by uncomplicated spontaneous vaginal delivery after an uneventful pregnancy. At the time of delivery, the infant had a right parietal skull mass that was presumed to be a cephalohematoma. The infant was also circumcised at birth and had a significant amount of bleeding from the incision, requiring a return trip to the emergency department.

Blood coagulation parameters drawn at the initial outpatient evaluation showed a prolonged activated partial thromboplastin time, which corrected when mixed 1:1 with normal plasma. This test suggested the presence of a factor deficiency, and a subsequent check of the factor VIII level revealed activity of less than 1%. This placed the infant in the ''severe'' range for hemophilia A (2).

A physical examination performed during the 2-month postpartum visit again showed a palpable lesion in the right parietal area. This mass gradually decreased in size such that by 6 months it was barely palpable. The results of the neurologic examination were normal for the patient's age, and the patient never had any symptoms referable to the parietal mass.

Plain films of the skull obtained 4 weeks after birth showed a lucency over the right parietal area. Subsequent CT performed at the 2-month postpartum evaluation showed an unusual lytic lesion of the high parietal bone on the right, with expansion and scalloping of the bony margins (Fig 1A), consistent with an intraosseous hematoma. MR imaging performed at that time showed an extra-axial hematoma along the right occipital convexity that exerted mild mass effect, indenting the skull, without intra-axial extension. T1-weighted images revealed bright signal, characteristic for methemoglobin, confirming that the mass was from hemorrhage rather than intraosseous tumor (Fig 1B).

Follow-up CT performed approximately 15 weeks after the initial CT showed an interval decrease in the right intraosseous hematoma and substantial bone remodeling (Fig 1C).

Discussion

Cephalohematomas are not uncommonly identified in normal infants at birth. Nonetheless, our case of this unusual appearance and location of an intraosseous hematoma, related to the known factor VIII deficiency, seems to be unique for two reasons. First, retrospectively, this was the first sign of the child's coagulation pathologic abnormality as it was present at the time of birth and likely indicated in utero bleeding. Cephalohematomas related solely to vaginal birth trauma in normal infants usually do not occur until several hours after birth (3). Second, the appearance is different from that of a typical cephalohematoma. This lesion was very expansile in nature, involving both the outer and inner tables of the skull. Subsequently, there was extensive bone remodeling of both tables as well. Again, this pattern was suggestive of repetitive in utero bleeding events.

Several reports of interosseous hematomas have been described, although none that occurred in a pediatric patient with coagulopathy. Yuasa et al (4) described a case of an interosseous hematoma that developed after a remote head injury. Similarly, Palatinsky et al (5) also described a case of chronic subperiosteal hematoma in a patient who had sustained a head injury 40 years previously. Both of these cases showed calcification radiographically and required surgical intervention for resolution of the hematoma.

Generally, risk factors such as hemophilia or other coagulation disorders, such as thrombopathies or anticoagulant therapy, will place a patient

Received July 6, 1999; accepted after revision August 30.

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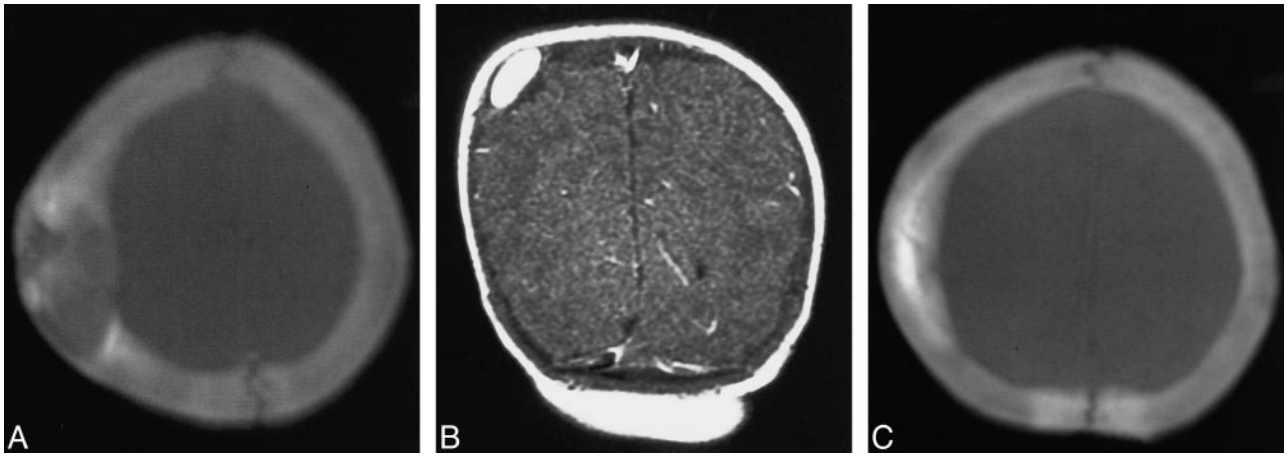

FIG 1. Images from the case of a 2-month-old white male patient who was initially seen in a general pediatric clinic for evaluation of bruised and swollen hands.

A, CT scan of the skull shows a large lytic lesion of the right parietal bone, with expansion and scalloping of the bony margins. The patient had a known factor VIII deficiency, and a cephalohematoma was present at birth.

B, T1-weighted MR image of the lytic lesion present on the CT scan shows a bright signal, characteristic for methemoglobin. This indicated that the mass lesion in the skull was secondary to hemorrhage rather than an intraosseous tumor.

C, CT scan obtained 15 weeks after the initial CT scans shows significant bone remodeling and near resolution of the intraosseous hematoma.

at risk for chronic hematomas, particularly subdural hematomas (6). Also, patients with severe hemophilia, as seen in our patient, will often experience small muscular hematomas (as evidenced by hand bruising in our patient) and may have hematuria or intracranial bleeding as well (2). Not described, though, is isolated, intraosseous bleeding within the skull present at parturition. Therefore, we suggest that the diagnosis of a bleeding disorder might be considered in a newborn infant with such an unusual skull lesion that is present at birth.

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