CASE REPORT

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Ruptured Intracranial Aneurysm in a 45-day-old Infant

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The incidence of intracranial aneurysms in childhood is rare, especially in infancy. We report a case of a 45-day-old girl who presented with seizure due to a ruptured large saccular aneurysm of the middle cerebral artery(MCA) with subsequent subarachnoid, intracerebral and intraventricular hemorrhage. The baby has enjoyed an excellent clinical outcome after surgical management. The clinical features of the case and review of the literature are presented.

KEY WORDS : Intracranial aneurysm · Infancy · Subarachnoid hemorrhage.

Introduction

Intracranial arterial aneurysms in childhood are rare, comprising 0.5 to 4.6% of all diagnosed aneurysms in large series[4,8,13]. They are particularly uncommon in early childhood less than 1year of age[4], and in the 1st month of life especially they are extremely uncommon[4,11]. However their incidence increases with age during childhood and into adulthood. In children, the location, morphology, and presentation of aneurysms seem to be different from those found in adults.

Approximately 18% of childhood aneurysms occur in infants or less than 1% of all aneurysms[17,20,22]. With the increasing use of magnetic resonance angiography(MRA), cerebral angiography, and computed tomography angiography (CTA), intracranial aneurysms are now being discovered more often, even in neonates with subarachnoid or intracranial hemorrhage[12] and several successful operations on infants have been reported[9,17,20]. In this report a case of aneurysm rupture in a 45-day-old infant is described along with the special features of cerebral aneurysms which occur in children.

Case Report

A 45-day-old girl who delivered to healthy non-consanguineous parents by cesarean section after an uncomplicated pregnancy. She had no history of trauma or infectious disease and her development was unremarkable. She was completely well until 1day prior to admission, when generalized seizure developed. After admission, she had similar two more episodes, each lasting for a few minutes. Upon admission to the hospital, she was conscious, with no posturing or lateralizing signs. The anterior fontanel was not bulging and there was no evidence of retinal hemorrhage or papilloedema on funduscopy. Spontaneous activity and normal motor tone was observed. Magnetic resonance imaging(MRI) disclosed a large hematoma with surrounding edema in the left frontotemporal region, which suggested that the hematoma resulted from rupture of an arteriovenous malformation or an aneurysm (Fig. 1).

Three-dimensional CTA showed a large aneurysm (9×7mm) arising from the left MCA (Fig. 2). Next day, a left pterional craniotomy was performed. After splitting the Sylvian fissure a large aneurysm was identified arising from

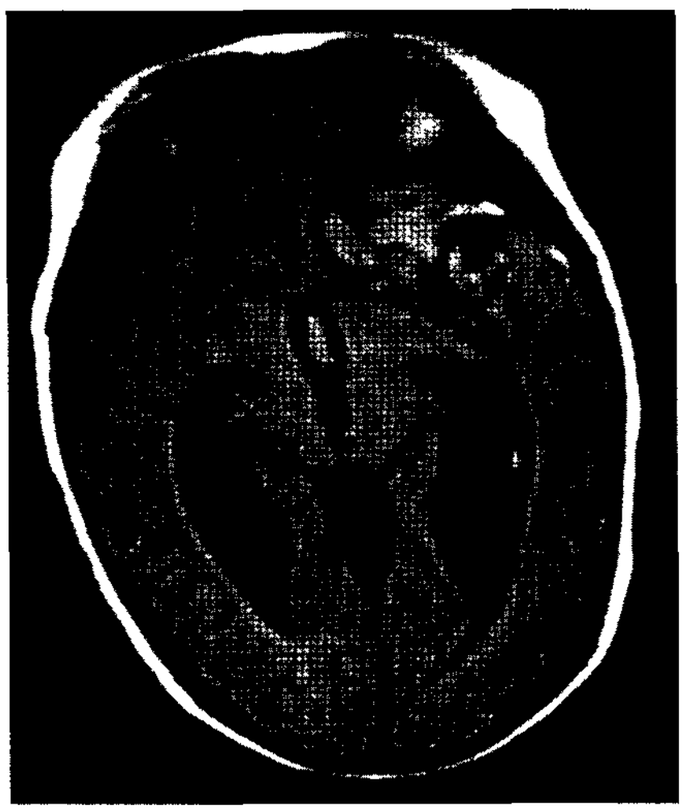

Fig. 1. Preoperative magnetic resonance image (FLAIR) demonstrating a large hematoma in the left frontotemporal region associated with subarachnoid and intraventricular hemorrhage.

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Fig. 2. Three-dimensional computed tomography angiography revealing a cerebral aneurysm(9×7mm) arising from the left middle cerebral artery.

Fig. 3. Intraoperative photograph showing the aneurysm(arrow), the clipped parent artery(arrowhead), the clipped efferent artery(arrowheads) and the M1 portion of left middle cerebral artery(double arrows).

one branch of the middle cerebral artery (M1). The diameter of parent artery and efferent artery were less than 2milimeter, and the aneurysm seemed similar to saccular aneurysm results of dilatation of the artery. Because the diameter of the arteries were less than 2milimeter, and the neck size of aneurysm was 7milimeter, the parent and the efferent artery were clipped respectively with small pediatric curved clips (Sugita's miniclip) (Fig. 3).

The infant was operated successfully without complications. She made a rapid recovery after surgery. One month follow up, computed tomography(CT) scans revealed ventriculomegaly and ventriculoperitoneal shunt was done. At the age of 1year she is developing normally.

Discussion

Differences of intracranial aneurysms in children as compared with adults have been extensively discussed. They are more common in males, tend to be larger and arise more from the MCA and posterior circulation[1,4,10,13,15,18]. The first report of aneurysmal surgery on an infant was by Jones and Shearburn[9] in 1961. Since then, the results of many such operations have been reported[7,21]. Surgical results have generally been reported as better than in adults[2,4,13]. Operative mortality is generally less than 5%[1,2,4,13]. In addition longterm outcome is good[13]. Reasons for better outcome are the greater functional brain capacity, better vascular status and less sensitivity to post-hemorrhagic vasospasm in pediatric patients[4]. This in part may be due to the plasticity of the immature nervous system when neurologic deficits occur.

The unique features of the pediatric aneurysm has led to much speculation regarding etiology. A major controversy concerning etiopathogenesis is whether cerebral aneurysms are congenital or acquired. In 1930, Forbus[5] proposed that aneurysms are acquired lesions, arising from a combination of a congenital medial defect of the arterial wall and degeneration of the internal elastic lamina. He suggested this theory when he observed a medial defect at the apex of cerebral artery bifurcations and found that an aneurysm appeared to arise as a bulging through the medial defect. Since then, much conflicting evidence has been presented. Those proposing acquired mechanisms claim that pediatric saccular aneurysms develop as a result of hemodynamic stresses. Anomolous circulations, such as those occurring with persistent fetal vessels are commonly associated with aneurysms[2]. The high incidence of aneurysms on the MCA may be due to its embryological development. The MCA appears earlier than other vessels, supplies more blood flow to the developing cerebral hemispheres, and is exposed to the hemodynamic stress of direct blood flow for a longer period of time than other vessels. This may be an explanation for the common occurrence of MCA aneurysms in early infancy[14].

Other authors postulate that cerebral aneurysms are congenital in nature. Bremer[3] suggested that aneurysms may be remnants of fetal cerebral plexuses. He thought that during development, the proximal portion of such plexuses enlarges, while the distal segments degenerate and thus produce aneurysmal sacs. Studies examining the arterial walls of vessels giving rise to aneurysms, report quantitative and qualitative deficiencies in the reticular fiber content of these vessels

walls[16]. Type III collagen deficiency has also been demonstrated[15]. Anatomical studies of fetal and neonatal intracranial vasculature have reveald remnants of small vascular trunks stemming from arterial bifurcations[11]. These remnants may develop into saccular aneurysms[11]. Familial incidence of pediatric aneurysms is well documented[2,6,19]. These aneurysms are often associated with multiple congenital anomalies[19] and their heredity is dominant[6]. Most pathologic reports of pediatric aneurysms have shown that they resemble adult aneurysms with absence of both the internal elastic lamina and the muscularis layer of the media[4,11,13].

MRA or CTA is an option and provides a non-invasive method for diagnosis. However, conventional angiography remains the gold standard for diagnosis of a cerebral aneurysm. Since no valid screening parameter is available, diagnosis is often made only after rupture of the aneurysm. This causes problems for emergency management. This case illustrates the entity of pediatric aneurysms well. The patient tolerated surgery well and had an excellent outcome. It is therefore important that the medical community should consider, although it occurs rare, the fact that aneurysms in pediatric population possibly do occur.

Conclusion

Pediatric intracranial aneurysms are rare. However, investigation and treatment of childhood is similar to that in adults. Cerebral arterial aneurysms should be considered in the differential diagnosis of stroke-like symptoms in infant and early childhood and surgical intervention is mandatory.

References

1. Amacher AL, Drake CG : The results of operating upon cerebral aneurysms and angiomas in children and adolescents. Cerebral aneurysms. **Childs Brain 5** : 151-165, 1979
2. Amacher AL, Drake CG, Ferguson GG : Posterior circulation aneurysms in young people. **Neurosurgery 8** : 315-320, 1981
3. Bremer JL : Congenital aneurysms of the cerebral arteries. An embryologic study. **Arch Pathol 35** : 819-831, 1943
4. Ferrante L, Fortuna A, Celli P, Santoro A, Fraioli B : Intracranial arterial aneurysms in early childhood. **Surg Neurol 29** : 39-56, 1988
5. Forbus WD : On the origin of miliary aneurysms of the superficial cerebral arteries. **Bull Hopkins Hosp 47** : 239-284, 1930
6. Fox JL, Ko JP : Familial intracranial aneurysms. Six cases among 13 siblings. **J Neurosurg 52** : 501-503, 1980
7. Grobe ML, Saunders M, Carton CA : Subarachnoid hemorrhage secondary to ruptured aneurysms in infants. Report of two cases. **J Neurosurg 49** : 898-902, 1978
8. Hulsman S, Moskopp D, Wassmann H : Management of a ruptured cerebral aneurysm in infancy. Report of a case of a ten-month-old-boy. **Neurosurg Rev 21** : 161-166, 1998
9. Jones RK, Shearburn EW : Intacranial aneurysm in a four-week-old infant. Diagnosis by angiography and successful operation. **J Neurosurg 18** : 122-124, 1961
10. Kanaan I, Lasjaunias P, Coates R : The spectrum of intracranial aneurysm in pediatrics. **Minim Invasive Neurosurg 38** : 1-9, 1995
11. Lipper S, Morgan D, Krigman MR, Staab EV : Congenital saccular aneurysm in a 19-day-old neonate. Case report and review of the literature. **Surg Neurol 10** : 161-165, 1978
12. Maroun F, Squarey K, Jacob J, Murray G, Cramer B, Barron J, et al : Rupture of middle cerebral artery aneurysm in neonate. Case report and review of the literature. **Surg Neurol 59** : 114-119, 2003
13. Meyer FB, Sundt TM, Fode NC, Morgan MK, Forbes GS, Mellinger JF : Cerebral aneurysms in childhood and adolescence. **J Neurosurg 70** : 420-425, 1989
14. Nishio A, Sakaguchi M, Murata K, Egashira M, Yamada T, Izuo M, et al : Anterior communicating artery aneurysm in early childhood. Report of a case. **Surg Neurol 35** : 224-229, 1991
15. Ostergaard JR : Aetiology of intracranial saccular aneurysms in childhood. **Br J Neurosurg 5** : 575-580, 1991
16. Ostergaard JR : Deficiency of reticular fibers in cerebral arteries. On the etiology of saccular aneurysms in children. **Br J Neurosurg 3** : 113-116, 1989
17. Putty TK, Luerssen TG, Campbell RL, Boaz JC, Edwards MK : Magnetic resonance imaging diagnosis of a cerebral aneurysm in an infant. Case report and review of the literature. **Pediatr Neurosurg 16** : 48-51, 1991
18. Sedzimir CB, Robinson J : Intracranial hemorrhage in children and adolescents. **J Neurosurg 38** : 269-281, 1973
19. Ter-Berg HW, Bijlsma JB, Veiga-Pires JA, Ludwig JW, Heiden C, Tulleken CA, et al : Familial association of intracranial aneurysms and multiple congenital anomalies. **Arch Neurol 43** : 30-33, 1986
20. Thrush AL, Marano GD : Infantile intracranial aneurysm. Report of a case and review of the literature. **AJNR 9** : 903-906, 1988
21. Ventureyra ECG, Choo SH, Benoit BG : Super giant globoid intracranial aneurysm in an infant. Case report. **J Neurosurg 53** : 411-416, 1980
22. Young WF, Pattisapu JV : Ruptured cerebral aneurysm in a 39-day-old infant. Case report. **Clin Neurol Neurosurg 102** : 140-143, 2000