Childs Nerv Syst (2006) 22: 295–299
DOI 10.1007/s00381-005-1198-1

CASE REPORT

Hyeong-Joong Yi
Kwang-Myung Kim
Yong Ko
Young-Soo Kim
Suck-Jun Oh
Seong-Hoon Oh

A spontaneous giant pseudoaneurysm presenting with chronic headache in adolescent

Received: 4 March 2005
Published online: 14 June 2005

H.-J. Yi (✉) · K.-M. Kim · Y. Ko · Y.-S. Kim · S.-J. Oh · S.-H. Oh
Department of Neurosurgery,
Hanyang University Medical Center,
Haengdang-dong, Sungdong-gu,
Seoul, 133-792, South Korea
e-mail: hjyi8499@hanyang.ac.kr
Tel.: +82-2-22908499
Fax: +82-2-22810954

Abstract *Background:* Intracranial pseudoaneurysms are rare vascular lesions, particularly in children and adolescents, and are characterized by the presence of organizing hematoma and fibrosis outside the true lumen, instead of true vascular elements. Most pseudoaneurysms result from an eminent insult, such as major trauma or grave infectious illness. Development of pseudoaneurysm without antecedent incident is rare. Furthermore, clinical manifestation with only a headache is also unusual in this age group. *Case history:* We now describe one patient who had a giant pseudoaneurysm arising at the distal middle cerebral artery. A 17-year-old boy complained of headache that had become apparent 3 years ago and intractable to medicine 3 months ago. Brain computed tomographic scan and lumbar cerebrospinal spinal fluid study revealed no trace of recent hemorrhage. However, digital subtraction angiography revealed a huge aneurysmal dilatation along the right M2 segment with the features of delayed filling and emptying of contrast agent. Surgical obliteration of the corresponding aneurysm with tandem clipping and aneurysmectomy rendered him free of headache thereafter. *Conclusions:* The actual cause and mechanisms of this case are not certain; nonetheless, we suggest that traumatic cause produced such a lesion, and minor repeated bleeding also elicited headache, albeit no evidence of recent major injury.

Keywords Intracranial aneurysm · Pseudoaneurysm · Spontaneous · Headache · Adolescent

Introduction

Intracranial pseudoaneurysms (false aneurysms) are terms based on histopathological aspects and are unexceptionally associated with the apparent preceding major trauma or serious infectious disease [1, 3, 4, 9, 16]. Therefore, individualization of their subtypes in clinical implication is still confusing and somewhat overlapping in itself. Although there has not been any single report of a large cohort regarding the pseudoaneurysms proper, they seem to occur rarely in the general population. Traumatic and infectious aneurysms, on the other hand, are more prevalent in childhood and adolescence than in adults [10, 12]. The etiology of most childhood aneurysms is unexplained and remains controversial. However, the mechanism of pseudoaneurysm formation strongly suggests traumatic cause, particular in this vulnerable age group. Even if they are histologically proved, the underlying etiology and pathophysiological process cannot always be elucidated unless there are definite historical and physical evidences. In this sense, the term "spontaneous" should be selected very cautiously.

In this report, we present a case of a 17-year-old boy who harbored giant pseudoaneurysm arising at the nonbranching site of the middle cerebral artery (MCA) and presented with nothing but a chronic headache. He sustained no previous major trauma or severe infection. After careful review of the present case as well as relevant literature, we concluded possible traumatic etiology.

Case report

A 17-year-old boy presented with intractable headache of 3-month duration. The patient had no familial history of specific inherent illness, previous history of major head injury, or grave infectious disease. He was born at full-term pregnancy; however, delivery with forceps was necessary due to the footling breech presentation. Despite this troublesome delivery, he had been well without any developmental or acquired anomalies until he entered puberty. Since he was 14 years old, he had been suffering headache once or twice a month, which lasted for several hours. The headache originated in the right temple region, occasionally propagated toward the high parietal area, and was usually preceded by nausea instead of aura. It was pin pricking and pulsating in nature and was not related to the patient's position or diurnal progression. His parents assumed it as a simple stress-related phenomenon and occasionally gave him only migraine medication instead of visiting the neurosurgical clinic, until the patient complained of sustained intractable headache. On presentation, he was alert, oriented, and showed a full range of higher cortical functions. Physical and neurological examination, including systemic review, revealed no specific abnormal deficits. All the laboratory blood tests yielded normal results, and cerebrospinal fluid examination disclosed no evidence of recent hemorrhage.

Plain skull radiographs disclosed no evidence of fracture. Nonenhanced computed tomography (CT) of the brain revealed only a suspicious high-density mass at the right distal sylvian fissure with no evidence of hemorrhage or vascular malformation, and enhanced CT disclosed a well-demarcated round mass with mixed high density, measuring 13×13 mm in size, at the same site (Fig. 1).

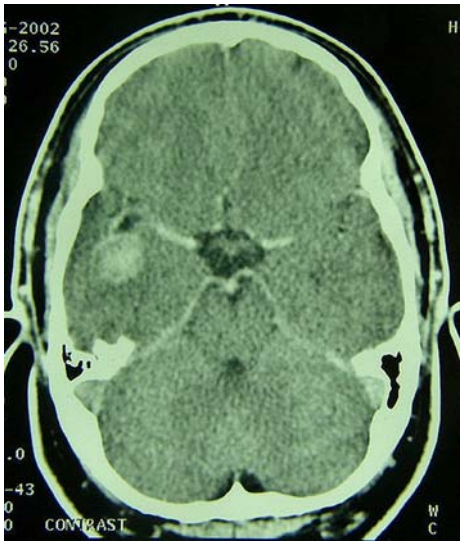

Fig. 1 Preoperative enhanced axial computed tomographic scan reveals a round mixed high-density mass at the right distal sylvian fissure

Magnetic resonance (MR) imaging, on the other hand, was not done because of his claustrophobia. Digital subtraction angiography (DSA) obtained on the next day after admission showed an aneurysmal dilatation at the midway between the right M2 and M3 branches, measuring 30×25×30 mm in size, and unusually elongated M1 segment. DSA also revealed delayed contrast filling and emptying, particularly at the medial and lower portion of the aneurysm, along the inferior M2 branch but without definite aneurysmal neck (Fig. 2).

A right pterional craniotomy was performed for obliteration of the aneurysm from the cerebral circulation. There was no evidence of subarachnoid hemorrhage or cortical

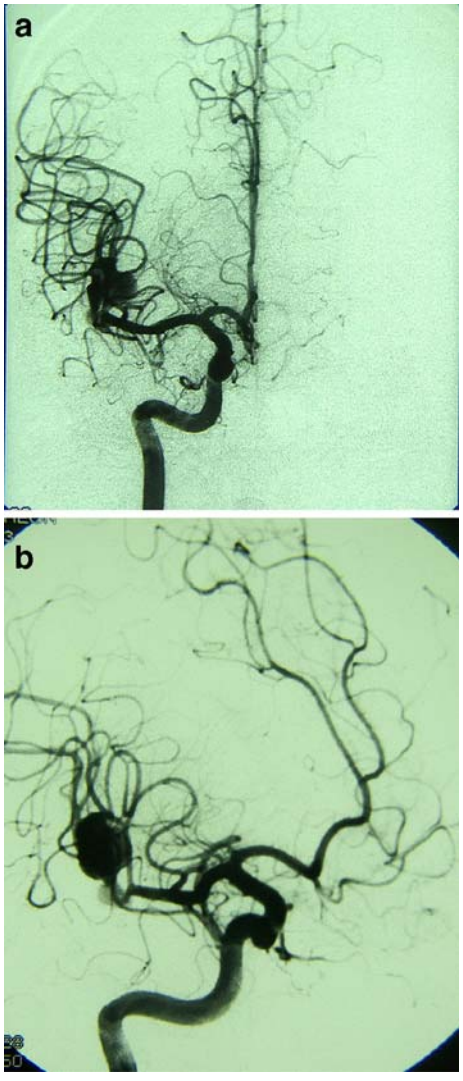

Fig. 2 Digital subtraction angiography of the right internal carotid artery. **a** Anteroposterior views and **b** oblique views show aneurysmal dilatation at the midway between the right M2 and M3 branches, measuring 30×25×30 mm in size. The aneurysm revealed a poorly defined neck, delayed contrast filling (figure not shown), particularly at its medial and lower portion. Also, note disproportionately long M1 segment

contusion. After splitting the whole sylvian fissure and tracing distally along the entire course of an extraordinarily long M1 segment, proximal portion of the inferior M2 segment was found to embed into a giant unruptured aneurysm. It compressed and splayed out the inferior M2 segment toward the superficial direction. Fortunately, temporal branches and the lateral lenticulostriate arteries were some distance away from the peripheral extent of the aneurysm. The aneurysm wall was composed of thick solid fibrous tissue and appeared dark purplish. Decompression was achieved by removal of impacted thrombi and dense fibrous wall altogether in piecemeal fashion by scalpel and pituitary forceps. During this procedure, the M1 segment distal to the lateral lenticulostriate arteries was temporarily clipped. After partial relief of mass effect, a clip was applied to seal off the incision site on the aneurysm surface. Throughout the operative procedure, a portable transcranial Doppler microprobe (Multidop X4/TCD8, DWL Electronische System GmbH, Sipplingen, Germany) and self-manufactured mirror were used to prevent inadvertent sacrifice of perforators. Finally, a huge aneurysmal sac was totally peeled off from the surrounding major MCA branches and excised by microscissors. The origin of the aneurysm was behind the surface of the inferior M2 segment and was clipped by four Sugita cobalt alloy clips in tandem fashion (Fig. 3).

Histopathological examination revealed a pesudoaneurysm. The dense fibrous vascular wall constituted multiple foci of old hemorrhage and chronic inflammatory cells,

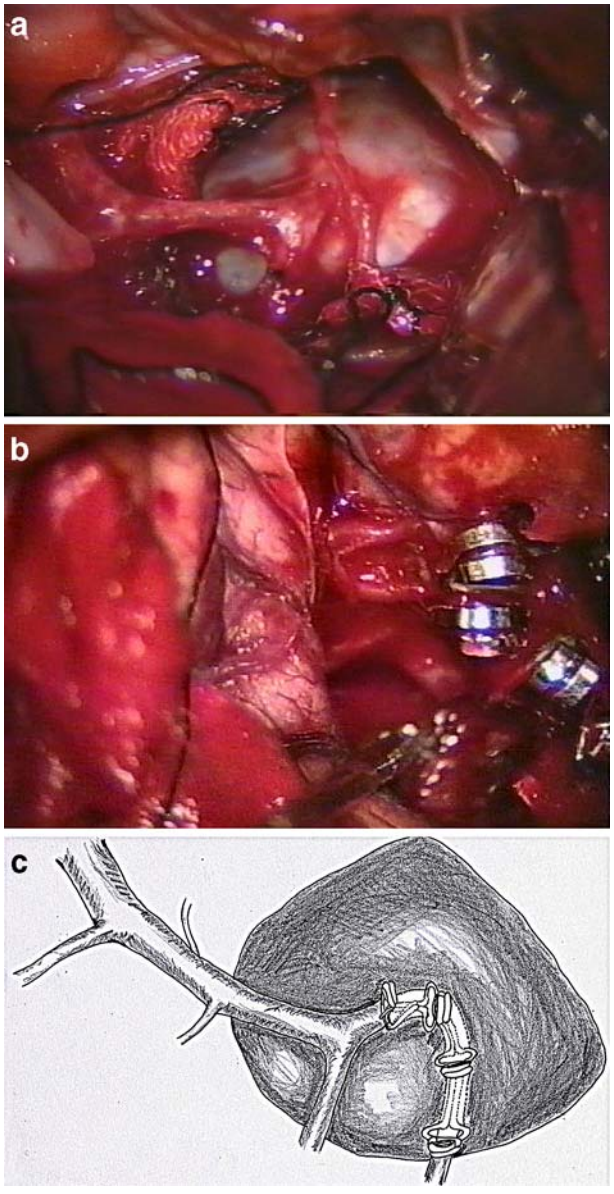

Fig. 3 Intraoperative photographs and schematic diagram in right transsylvian approach. **a** Note huge aneurysmal sac compressing and splaying the inferior M2 segment. **b** More superior and counterclockwise tilted view than **a**, after withdrawal of the frontal self-retractor, shows four Sugita clips arranged in tandem fashion along the inferior segment of M2. **c** Schematic diagram shows configuration of aneurysmal sac with regard to the underlying inferior M2 segment and final placement of the clips

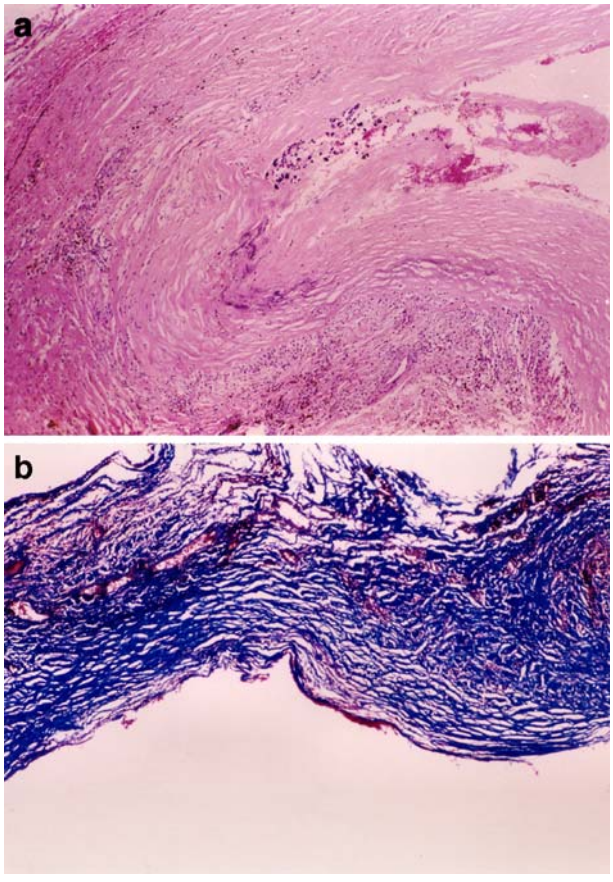

Fig. 4 Photomicrograms of surgical specimen show dense fibrous vascular wall with foci of old hemorrhage and chronic inflammatory cellular infiltration. Luminal surface also demonstrates thrombus but no endothelial lining cells. (**a** H & E, original magnifications×40, **b** Masson's trichrome, original magnification×100)

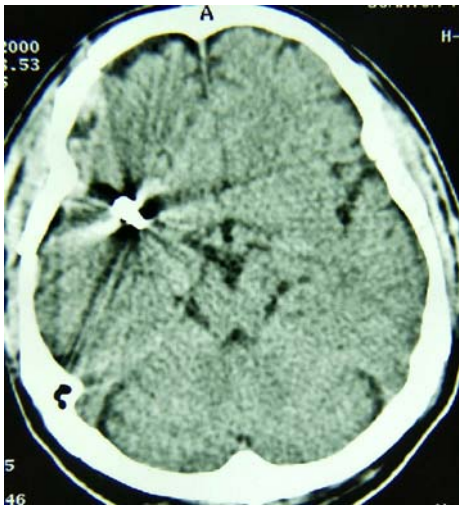

Fig. 5 Postoperative nonenhanced axial CT scan obtained at the seventh postoperative day shows metallic artifact caused by the implanted clips. Evidence of intraparenchymal hemorrhage or infarction is not seen

instead of endothelial linings, internal elastic lamina, or muscle coat (Fig. 4). Interestingly enough, he showed a complete postoperative resolution of headache and made a full postoperative recovery. Neither seizure nor hemiparesis was observed. CT of the brain taken at the seventh postoperative day did not reveal any presence of infarction or hemorrhage (Fig. 5).

Discussion

Intracranial aneurysms are uncommon in childhood and adolescence, and their overall incidence is rated between 0.17 to 4.6% [5, 8–11, 13, 17]. Traumatic intracranial aneurysms (TICAs) are even more rare, constituting less than 1% of all the intracranial aneurysms [1, 3, 7, 9, 15, 16]. Up to one third of TICAs occur in the first two decades of life; therefore, TICAs are relatively more common in children than adults [1, 14]. The clinical presentation of all aneurysms as well as TICAs in this age group is usually symptomatic and typically presents with subarachnoid or, less commonly, intracerebral hemorrhage [2, 4, 5, 7, 13, 16]. Besides hemorrhage, depending on their specific location [9], there may be unexplained neurological deterioration, massive epistaxis, and cranial nerve palsy. All the above descriptions clearly indicate that aneurysm formation is rare, whereas the risk of rupture is relatively high compared with adults, irrespective of their subtypes [11]. In addition, seizures and symptoms from mass effect due to large or giant unruptured aneurysms are more frequent in younger children than in adults [10]. Clinical manifestation of the mass effect in most cases represents hemispheric symptoms: seizure, hemimotor or sensory change, and any other ischemic symptoms. Headache, on the other hand, is not necessarily translated into the hemispheric involvement, and another mechanism might be involved, as shown in the present case.

As for the pathophysiology involved, it is generally accepted that injury to the internal elastic lamina by hemodynamic stress at the previously altered vascular wall is the initial pathophysiological change of aneurysm formation, almost identical to that of the adults, although there are some reports stressing congenital basis for aneurysm formation, such as the composite of quite a few reticular fibers seen in the tunica media [8, 11–13]. TICAs are frequently found in the MCA and pericallosal arteries in addition to the supraclinoid carotid artery [7, 15]. Among them, cortical MCA TICAs may develop beneath an overlying linear or depressed skull fracture, whereas supraclinoid carotid and pericallosal TICAs are not [9]. Nevertheless, most intracranial aneurysms in this younger age group are unexplained and the etiology remains controversial [12]. Several characteristic features specific to the aneurysms in this age group have already been reported: males are more affected than females, higher incidence at the posterior circulation or internal carotid artery, tendency to form a larger size aneurysm (30–45%), and higher incidence of infectious and traumatic origin [6, 8, 10, 13, 17]. As stated before, TICAs comprise larger parts in all childhood aneurysms, and most of the above specific features appear to represent the traumatic cause [10, 12]. Angiographically, TICAs involve more commonly the arterial wall along the course of the secondary branch, typically do not have a neck, are more irregular in their dome contour, and are frequently observed to be subject to delayed filling and emptying. Overall their sizes range from 2 to 15 mm; therefore, large or giant form is much rarer [2, 7].

Reports on intracranial pseudoaneurysms almost exclusively dealt with traumatic or infectious etiology; therefore, pseudoaneurysms that developed spontaneously or are not followed by major head injury are seldom documented up to the present. Pseudoaneurysms that result from rupture of the entire arterial wall comprise a larger percentage of TICAs, particularly those at the skull base [9, 16]. Histologically, the pseudoaneurysm is composed of extraluminal hematoma that is contained by the surrounding connective tissues and occurs after complete vessel wall injury. The hematoma may recanalize or remain in communication with the vascular lumen, and the wall of this structure is formed through hematoma organization and fibrosis. Therefore, as its name implies, none of the normal vascular structures are present [1, 3, 14]. Because of the above reasons, almost all (94%) of the pseudoaneurysms seem to have concurrent intracranial pathology, such as fracture or hematoma [3]. The reasons we believe the present case to be traumatic etiology are based on histologically proven pseudoaneurysm and solidifying aneurysm wall. Hematoma and false lumen designate any kind of traumatic involvement, regardless of how minute the cause can be. In contrast to the "friable" wall typically seen

in the infectious aneurysm, densely hardened aneurysm wall implies traumatic process more aptly, although it progresses very slowly and indolently. Although optimal operative technique must be individualized for each patient, surgical obliteration by trapping, excision, or clipping is still believed to be the most effective [2, 14]. In the current case, we were able to exclude the aneurysm neck by using three different clips with tandem maneuver as well as aneurysmectomy, without the aid of bypassing surgery. TICAs rarely regress and generally have a high incidence of rupture [3]; therefore, isolation of the aneurysm from the cerebral circulation, the sooner the better, is mandatory.

Classification of TICAs only on the basis of histology presents several problems in clinical decision making. Furthermore, radiographic criteria to differentiate presumed TICAs from other aneurysms may not correlate with the histological diagnosis, and histopathology does not necessarily correlate with the mechanism of injury in every case [16]. In the present case, we uncovered neither history of recent major head injury nor history of infectious illness, except remote obstetric history of forceps delivery. Although there has been "one" case report of a traumatic pericallosal aneurysm in a patient with no major, albeit mild (roller-coaster rides), injury [15], there is no such a report in the literature of a "traumatic" aneurysm arising in a patient without any history or physical evidence of trauma at presentation, except our present case. Therefore, our case seems to be unique. We failed to uncover the exact cause of such a giant pseudoaneurysm, and remote past history of forceps delivery seemed to be very conjectural. As proven at the operative procedure, the headache was most likely elicited by small repeated bleedings within the thrombosed extramural wall. This distended wall seemed to have triggered the pain-sensitive structures within and/or adjacent to the vessels and the dura in a certain, but not fully understood manner, the clipping of the aneurysm resolved the headache. However, a question still remains on his clinical manifestation, and lack of hemispheric symptoms, including seizure and hemiparesis, cannot be fully explained by this assumption.

Acknowledgements We are grateful to Dr. Woon-Ki Paik for editing this manuscript and Dr. Seung-Sam Paik for reviewing histopathologic findings.

References

1. Buckingham MJ, Crone KR, Ball WS, Tomsick TA, Berger TS, Tew JM Jr (1988) Traumatic intracranial aneurysms in childhood: two cases and review of the literature. Neurosurgery 22:398–408
2. Chedid MK, Vender JR, Harrison SJ, McDonnell DE (2001) Delayed appearance of a traumatic intracranial aneurysm. Case report and review of the literature. J Neurosurg 94:637–641
3. Dowd GC, Awasthi D (2002) Management of infectious and traumatic intracranial aneurysms. In: Batjer HH, Loftus CM (eds) Textbook of neurological surgery, vol 3. Lippincott Williams & Wilkins, Philadelphia, pp 2473–2480
4. Gutierrez FA, Bailes J, McLone DG (1987) Intracranial aneurysm and pseudo-aneurysm occurring during infancy and childhood: diagnosis and surgical results. Concepts Pediatr Neurosurg 7:153–168
5. Heiskanen O (1989) Ruptured intracranial arterial aneurysms of children and adolescents. Surgical and total management results. Childs Nerv Syst 5:66–70
6. Humphreys RP, Hendrick EB, Hoffman HJ, Storrs BB (1985) Childhood aneurysms—atypical features, atypical management. Concepts Pediatr Neurosurg 6:213–229
7. Lath R, Vaniprasad A, Kat E, Brophy BP (2002) Traumatic aneurysm of the callosomarginal artery. J Clin Neurosurg 9:466–468
8. Meyer FB, Sundt TM Jr, Fode NC, Morgan MK, Forbes GS, Mellinger JF (1989) Cerebral aneurysms in childhood and adolescence. J Neurosurg 70:420–425
9. Mustafa U, Murat C, Sait SM, Murat H, Naci K, Cetin SA, Civan I (1998) Traumatic intracranial carotid tree aneurysms. Clinical study. Neurosurgery 43:1314–1322
10. Norris JS, Wallace MC (1998) Pediatric intracranial aneurysms. Neurosurg Clin North Am 9:557–563
11. Østergaard JR (1991) Aetiology of intracranial saccular aneurysms in childhood. Br J Neurosurg 5:575–580
12. Piatt JH Jr, Cluine DA (1992) Intracranial arterial aneurysm due to birth trauma. Case report. J Neurosurg 77:799–803
13. Proust F, Toussaint P, Garnieri J, Hannequin D, Legars D, Houtteville JP, Freger P (2001) Pediatric cerebral aneurysms. J Neurosurg 94:733–739
14. Raju BSV, Purohit AK, Murthy SR, Sundaram C, Sanjay T (2001) Traumatic distal anterior cerebral artery aneurysm in a child: a case report. Neurol India 49:295–298
15. Senegor M (1991) Traumatic perocallosal aneurysm in a patient with no major trauma. Case report. J Neurosurg 75:475–477
16. Ventureyra ECG, Higgins MJ (1994) Traumatic intracranial aneurysms in childhood and adolescence. Childs Nerv Syst 10:361–379
17. Wojtacha M, Bazowski P, Mandera M, Krawczyk I, Rudnik A (2001) Cerebral aneurysms in childhood. Childs Nerv Syst 17:37–41