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Endovascular Treatment of Primary Infectious Aneurysm in Childhood: A Case Report

Çocukluk Dönemi Primer İnfeksiyöz Anevrizma Endovasküler Tedavisi: Olgu Sunumu

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ABSTRACT

Infectious aneurysms constitute 4% of all intracranial aneurysms. The microorganisms responsible are most commonly streptococcus viridans, staphylococcus aureus and combined bacterial infections. Nonetheless, cases with no reproduction in their cultures are rather frequent. A 6-year-old patient admitted with complaints of sudden headache, nausea, vomiting and high temperature. Intracerebral hematoma and saccular aneurysm located at the distal posterior cerebral artery were diagnosed as a result of the laboratory investigations and neuroradiological examinations. Infectious aneurysm was considered due to the clinical findings, morphology and location of the aneurysm. Although the causative microorganism was detected in blood culture, no focus could be detected. The aneurysm was hindered by endovascular intervention. In this manuscript, we discuss the infrequently seen childhood infectious aneurysm in the light of the pertinent literature.

KEY WORDS: Childhood, Endovascular treatment, Infectious aneurysm

ÖZ

İnfeksiyöz anevrizmalar tüm intrakranial anevrizmaların %4'ünü oluştururlar. En sık sorumlu mikroorganizmalar streptococcus viridans, staphylococcus aureus ve karma bakteri enfeksiyonlarıdır. Bununla birlikte kültürlerinde üreme olmayan olgu sayısıda oldukça fazladır. 6 yaşındaki çocuk hasta; ani başlayan baş ağrısı, bulantı, kusma ve yüksek ateş şikayetiyle başvurdu. Yapılan laboratuvar ve nöroradyolojik incelemeler sonucu intraserebral hematom ve distal posterior serebral arter yerleşimli sakküler anevrizma saptandı. Klinik bulgular, anevrizma morfolojisi ve yerleşimi itibariyle infeksiyöz anevrizma düşünüldü. Kan kültürlerinde etken mikroorganizma tespit edilmesine karşın herhangi bir odak saptanamadı. Endovasküler girişimle anevrizma devre dışı bırakıldı. Bu makalede; nadir görülen çocukluk çağı infeksiyöz anevrizmaları ve tedavi seçenekleri literatür eşliğinde tartışılmıştır.

ANAHTAR SÖZCÜKLER: Çocukluk dönemi, Endovasküler tedavi, İnfeksiyöz anevrizma

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INTRODUCTION

Infectious aneurysms (IA) constitute 4% of all intracranial aneurysms. They are more common at young ages and 80% of the cases are younger than 40 years of age (7, 14). They are also observed in immune suppressed patients and 2-10% of patients with subacute bacterial endocarditis (SBE). Infectious aneurysms develop secondary to the invasion of the muscular layer of the tunica media by an infectious pathogen, most frequently via the blood stream and, occasionally, dissemination through the arterial wall of the inflamed tissues around the arteries, causing necrosis (27). Infectious aneurysms are generally located in the distal parts of the cerebral arteries and can be multiple in number (17). The most frequent location is the distal branches of the middle cerebral artery. Hemorrhage occurs in the form of intra-cerebral hematoma (ICH) and subarachnoidal hemorrhage (SAH). Culture results may sometimes be negative although a microorganism can be detected (7). Clinical findings are variable and the condition may deteriorate and result in fatality. Mortality rates between 50% and 80% have been reported in different series (2,23). In addition to antimicrobial treatment in hemorrhagic mycotic aneurysms, aneurysm clipping and endovascular treatment comprise other therapeutic modalities for selected cases. In this paper, rgw clinical manifestation and laboratory findings in a pediatric patient with mycotic aneurysm are presented and treatment alternatives are discussed in the light of the relevant literature.

CASE REPORT

A 6-year-old patient was admitted with complaints of sudden headache, nausea, vomiting and high temperature. Neurological examination revealed a Glascow coma scale of 11/15, pupillary anisocoria (left>right) and neck stiffness. Fever (39ºC) and tachycardia (110/min) were found during systemic examination. Although moderate rises were observed in the patient's sedimentation rate, CRP level and leukocyte count, staphylococcus cohnii was isolated in blood culture while the focus of primary infection could not be found. An acute intra-cerebral hematoma 4x4 cm in size and opening to the ventricular system at the left parieto-occipital region was visualized by computed tomography (CT) (Figure 1). A minuscule saccular aneurysm on the P4 segment of the left posterior cerebral artery was observed in cerebral angiography (Figure 2).

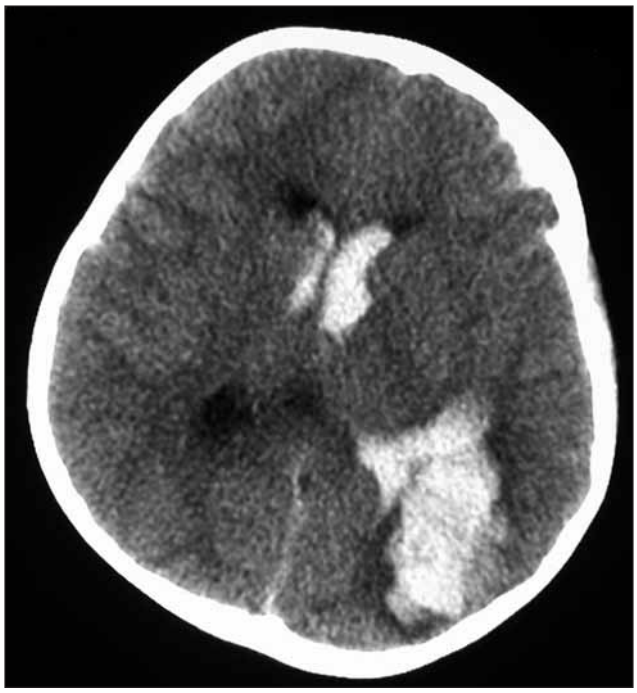

Figure 1: *Computed tomography demonstrating an acute intra-cerebral hematoma measuring 4x4 cm and opening to the ventricular system at the left parieto-occipital region.*

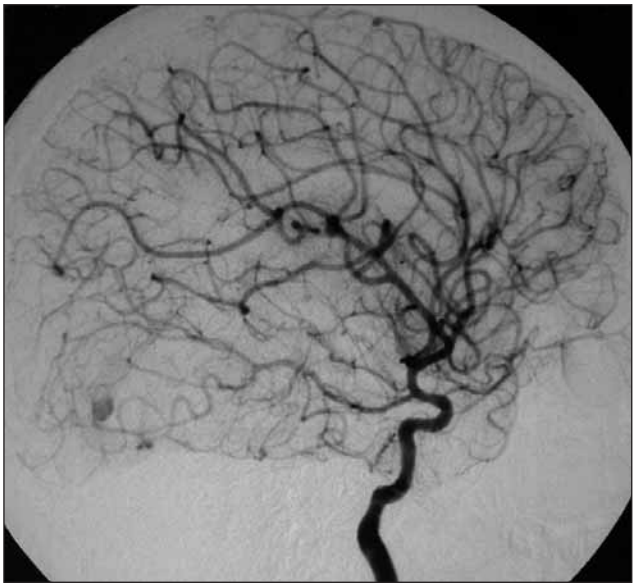

Figure 2: *Cerebral angiography demonstrating a saccular aneurysm on the P4 segment of the left posterior cerebral artery.*

The morphology and location of aneurysm, together with the patient's clinical behavior suggested infectious aneurysm. The aneurysm was embolized by endovascular intervention using 25% glue-lipiodol mixture and the aneurysm disappeared in the follow-up cerebral angiography (Figure 3). Other predisposing factors related to infectious aneurysm

(SBE, vasculitis, meningitis, immune deficiency etc.) were excluded after the embolization and the case was considered a primary infectious aneurysm. Appropriate antimicrobial treatment was administered for 4 weeks. Definite regression in the dimensions of the ICH was observed during the control CT scan. The general condition of the patient gradually improved and the patient was discharged from the hospital with total cure.

Figure 3: *The aneurysm was embolized by endovascular intervention using 25% glue-lipiodol mixture, resulting in disappearance of the aneurysm in the control cerebral angiography.*

DISCUSSION

The term "bacterial aneurysm" was introduced to replace "mycotic aneurysm", which was first used by Osler in 1885, subsequent to high frequency of endocarditis in its etiology. At present, "infectious aneurysm" is preferred for all infection-related aneurysms. The most frequently isolated microorganisms are staphylococcus aureus and streptococcus viridans. Pneumococci, enterococci, corynebacteria and haemophilus species are less frequently encountered. Fungal pathogens generally seen in patients with a poor immune system are aspergillus and candida species (5,16,29). A recent study reported a 53% "no growth in culture" while it was reported to be 12% previously (21,29).

Infectious aneurysms may appear as a result of invasion of the arterial wall by bacteria originating from a close or distant focus, septic or aseptic embolization of vasa vasorum and immune complex storage resulting in damage in the artery wall (5). The source of septic emboli is frequently infective endocarditis and infectious aneurysm develops in 2-10% of patients with SBE. In addition to septic embolism, it may also develop secondary to dissemination of an infection such as meningitis, osteomyelitis, sinusitis and cavernous sinus thrombophlebitis (3,18,29). Frequency of cases with primary or cryptogenic IA but no focus of infection anywhere in the body was reported between 10 and 12.5% (10).

Although IAs are usually fusiform in shape, they may also be saccular in shape, as in the present case, and their dimensions vary between 1 and 10 mm. Necrosis, hemorrhage, abscess formation and colonization of bacteria may also be observed while acute and chronic inflammation is often observed (5).

The clinical course of infectious aneurysms varies and they frequently progress asymptomatically. A rapid loss of consciousness together with a sudden headache is observed in patients with IA who develop ICH or SAH (2,10,16,23). A study carried out in a large series reported that complaints at the time of admittance were related to SAH in 32.5%, ICH in 25.5%, embolic stroke in 16% while 11% of the cases experienced symptoms such as headache, epileptic crisis and high fever (11). If a thorough investigation is not carried out in cases with IA, the diagnosis may be delayed or incorrect in the presence of insidious neurological symptoms (10,16).

Since laboratory findings in IA cases are not specific, there is no consensus on which diagnostic tests need to be carried out (4,6). Although the presence of hemorrhage in an atypical location detected on CT was an indirect evidence of IA in our patient, cerebral angiography is a reliable test to diagnose IA with a sensitivity of 90%. However, it is not possible to determine with current diagnostic techniques which IAs will rupture and which will regress. Some infectious aneurysms may also rupture before they exhibit dilation angiographically (5,6,18,28).

No universal agreement exists on optimal treatment plan for infectious aneurysms and treatment varies based on clinical and/or radiological findings. From long-term antibiotic treatment to surgical clipping and resection, wide

range treatment modalities can be employed (10). Antibiotic treatment should be initiated promptly for unruptured aneurysms. The duration of antibiotic treatment was between 4 and 6 weeks in most patients, similar to our case. Even if regression of the aneurysm is observed in cerebral angiographies of the patients under antibiotic treatment, the aneurysm may re-grow or occur somewhere else (18). The greatest danger of medical treatment is the development of rupture during treatment. However, the frequencies of real hemorrhage and recurrent hemorrhage are not known in cases with IA (8,9,24,25). The mortality rate may be as high as 90% when the aneurysm is ruptured (11).

The aneurysm may be clipped if and when no regression is evident in the control cerebral angiographies or if it causes hemorrhage (9,25). Due to the small diameter of the vessel and fragile wall of the aneurysm, it may be difficult and risky to clip IAs with a fusiform shape. Surgery should be carried out as soon as possible if the risk of excising or clipping the lesion is low (7,13,15).

Current modalities such as stereotactically-guided determination of surgical location, image-guided surgery and endovascular interventions in inoperable cases, as was the case in our patient, facilitate IA treatment considerably (10,12,19,22,26). It is generally not possible to spare the main vessel during the surgery. Endovascular intervention is the most popular method since the outcome of its alternative, aneurysm clipping, is risky. The outcome of aneurysm clipping was risky for our patient as well. Moreover, endovascular treatment should also be preferred in patients with SBE who use anticoagulant drugs due to heart valve replacement (7,15).

Thanks to the advances in imaging techniques and the development of the effective antimicrobial and antimycotic drugs that result in early diagnosis, the prognosis of intracranial IAs has improved significantly. Aneurysm rupture is the most important factor affecting the prognosis and it increases the mortality rate by a factor of 3. The mortality rate is between 43% and 63% in cases emergency surgery is performed while that is between 0% and 18% when elective surgery is performed (3,20,29). The mortality rate of endovascular treatment, an alternative treatment protocol, derived from large series is not available in the literature (10,19,22).

CONCLUSION

IA is one of the disorders to be kept in mind in pediatric patients with intra-cerebral hemorrhage, immune deficiency and unexplained high fever (1). It carries high morbidity and mortality rates. Although surgical treatment is recommended in elective conditions, a combination of endovascular treatment together with antibiotic treatment should be preferred in selected cases when surgical treatment is difficult and risky (6,7,21).

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