



Mesenteric pseudoaneurysm and cerebral stroke as sequelae of infective endocarditis in an adolescent

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Abstract Infective endocarditis is uncommon in children, and there is a paucity of literature concerning cases that involve unique or resistant organisms. Complications associated with infective endocarditis are distinctly rare and poorly characterized, especially unusual sequelae such as pseudoaneurysms of the abdominal mesentery. Our case involves an adolescent who presented with several weeks of fever and eventual cardiac murmur and was found to have vancomycin-resistant *Enterococcus* growing as a vegetation on a previously undiagnosed bicuspid aortic valve. He had a cerebral stroke presenting as Broca's aphasia before cardiac surgery, as well as a superior mesenteric artery pseudoaneurysm several days postoperatively. The case highlights some of the serious surgical complications that can occur in young persons with infective endocarditis, as well as many of the problems involved in managing a patient with highly resistant pathogens and a surgically challenging location of the aneurysm.

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Sir William Osler [1] first used the term *mycotic aneurysm*, designated specifically for infected aneurysms secondary to bacterial endocarditis. In modern times, infective endocarditis (IE) is very uncommon in the pediatric

population, with an estimated rate of 0.47 to 0.78/1000 pediatric hospital admissions [2,3]. *Enterococcus* is the third most common causative organism of endocarditis, after *Streptococcus viridians* and *Staphylococcus aureus*. However, there are only a few case reports of IE because of highly-resistant *Enterococcus* and little consensus about optimal treatment of these resistant strains [4]. The incidence of complications associated with endocarditis, such as stroke and mycotic aneurysms, are also poorly characterized in the

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pediatric population. In adults, stroke is reported in 5.7% to 21% of patients with IE [5,6], whereas the incidence of infected aneurysms is estimated at 2% to 4% [5].

We report a case of IE because of vancomycin-resistant *Enterococcus* (VRE) in a 14-year-old boy who subsequently developed mycotic aneurysms in the distributions of the proximal superior mesenteric artery (SMA) and middle cerebral artery (MCA). The unusual causative organism, as well as the numerous sequelae he experienced, posed many challenging questions related to the most appropriate management of his disease and the role of surgical intervention.

1. Case report

The patient is a previously healthy 14-year-old male with a distant history of infantile gastroschisis, who presented with a 3-month history of fever. The patient was otherwise asymptomatic during the first month of fevers and was treated only with acetaminophen and ibuprofen. The

following month he developed intermittent symptoms of headache, fatigue, chills, and temperature up to 39.8°C. The patient had corrective orthodontic braces for the last year that received scheduled adjustments. There was no history of any sexual activity or illicit substance use.

After 3 months of fever, the primary care physician noted a new cardiac murmur on examination. An echocardiogram was performed that demonstrated a bicuspid aortic valve, previously undiagnosed, with a large vegetation and regurgitation. He was admitted to a local hospital that day, afebrile with blood pressure of 120/40. He was started on vancomycin and gentamicin, later adjusted to ampicillin and gentamicin when pansensitive α -hemolytic *Streptococcus* was grown. He was transferred to our children's hospital for a planned Ross procedure (replacement of the aortic valve with pulmonary autograft and subsequent placement of pulmonary allograft).

On the day of admission, intermittent headaches led to him receiving a brain magnetic resonance imaging (MRI)/magnetic resonance angiography study, which was suggestive of a small acute infarct within the left MCA region (Fig. 1A). However, on the morning of his cardiac surgery,

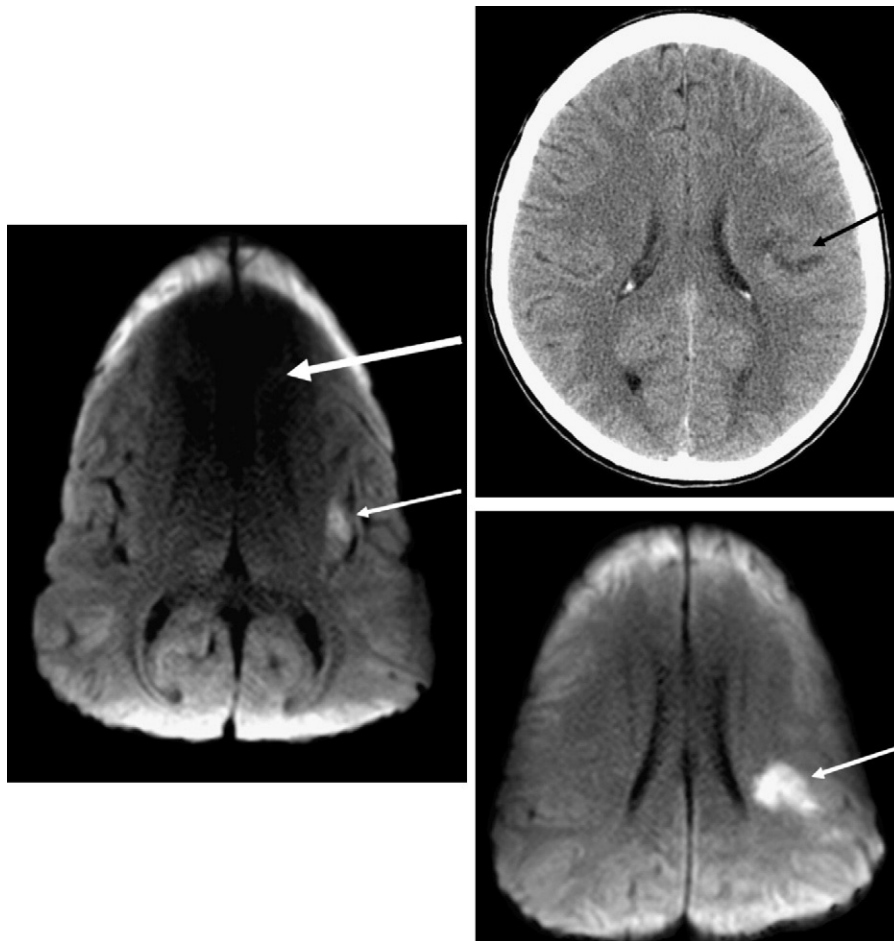


Fig. 1 Magnetic resonance imaging axial diffusion-weighted images and CT head axial image. Left (A, before aphasia), Thick arrow denotes artifact and signal dropout from dental hardware on MRI. Thin arrow denotes focal area of ischemia in left insular cortex. Right (B, day of aphasia onset), Black arrow on CT image and white arrow on MRI image show increase in area of ischemic changes.

the patient began to demonstrate a new onset of persistent Broca's aphasia, characterized by the inability to find basic words previously in his vocabulary. The patient immediately underwent computed tomographic (CT) and MRI scans of the brain, both of which showed increased size of the left MCA distribution infarction, including coverage of Broca's area (Fig. 1B). This appeared as an expansion of the ischemic area noted on MRI 3 days prior, but a second stroke in the same distribution could not be ruled out.

After evaluating the risk of showering further emboli by delaying his operation vs the risk of intraoperative hemorrhage, the patient went to surgery, and the Ross procedure was performed. All visible vegetation was removed and was sent for culture, which eventually grew VRE. The patient recovered well from his operation, with significant improvements in his aphasia and successive head CT scans demonstrating no significant interval changes. On postoperative day (POD) 6, the patient was discharged to home on 6 weeks of intravenous daptomycin to cover VRE, with extensive follow-up planned.

Three days after discharge, the patient presented again to his local emergency department with 24 hours of post-prandial nausea and nonradiating abdominal pain that progressed to extensive nonbilious, nonbloody vomiting that evening. In the emergency department, he was found to be afebrile with normal vital signs. Abdominal CT was performed and was initially noted only for a focal cortical defect within the right kidney thought to represent a small renal infarction (unchanged on later studies). Laboratory

workup revealed mild leukocytosis without overt anemia and essentially normal electrolytes, liver enzymes, amylase, lipase, lactate, and urinalysis. Stool guaiac was also negative. Because of his recent history, the patient was transferred back to our institution the following day. Repeat evaluation of the referring hospital's CT was suspicious for a mycotic aneurysm in a proximal portion of the SMA (Fig. 2). A Doppler sonogram further characterized the lesion to be consistent with a pseudoaneurysm (Fig. 3).

Because of the potential risk that the SMA pseudoaneurysm would lead to intestinal infarction or hemorrhage, the patient underwent immediate exploratory laparotomy. His saphenous veins were exposed in the anticipation that a bypass graft would need to be performed. At surgery, a large 5×3 cm pseudoaneurysm was found at a proximal portion of the main branch of the SMA. Dissection was begun by gaining proximal and distal control of the main SMA vessel and proceeded to follow this main vessel into the mass. Interestingly, because this was a pseudoaneurysm, we were fortunate to isolate the main branch of the SMA that remained intact. Thus, resection of the pseudoaneurysm was performed by retaining this main branch, whereas resecting the aneurysmal branches that extended off of the SMA. The intestine was monitored throughout the dissection, it continued to maintain perfusion, and no bowel resection was necessary. There were no complications, and the patient was taken to the intensive care unit postoperatively and extubated the following morning. Successive echocardiograms and a cardiac MRI study showed no significant



Fig. 2 Sagittal reformatted image and axial source image. Left, White arrow denotes the oval pseudoaneurysm adjacent to superior mesenteric vein (SMV) and SMA branches. Right, Black arrow denotes the pseudoaneurysm. White arrow denotes adjacent SMA and SMV.

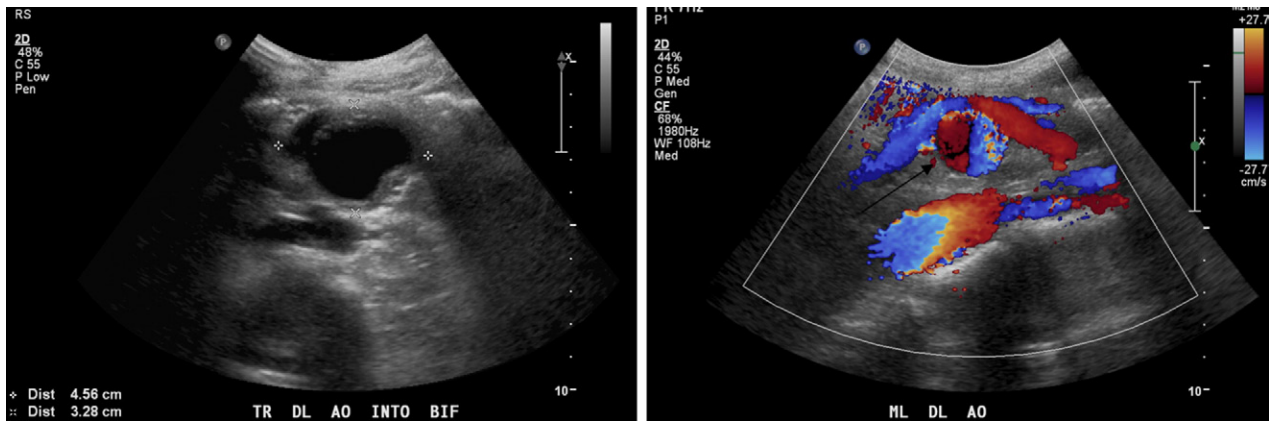


Fig. 3 Ultrasound, 2-dimensional, and color Doppler images. Two-dimensional, complex partially anechoic thick-walled oval lesion. Color Doppler, Characteristic yin-yang pattern of pseudoaneurysm denoted by arrow. Overlying vessel is branch of SMV. Deep to the pseudoaneurysm is aorta.

interval changes, and there was no evidence of new vegetations. Gram stain of the pseudoaneurysm removed during the operation showed gram-positive cocci; however, the cultures did not grow any bacterial or fungal colonies. The CT angiography of the head and neck on POD 4 did demonstrate a small aneurysm measuring 5 mm within the perisylvian branches of the left MCA; because of its small size and location, it was observed without any interventions, and later imaging verified that the aneurysm thrombosed. The patient's previous aphasia continued to improve remarkably with therapy. Despite a period of continued abdominal pain and vomiting that may have been caused by a mild postoperative pancreatitis, he recovered and was discharged on POD 19 with cefotaxime and linezolid for a total course of 6 weeks, with extensive follow-up in multiple clinics and complete recovery of previous functioning. The patient is currently doing well and is without any neurologic, infectious, or gastrointestinal problems 9 months after his initial surgery.

2. Discussion

Infective endocarditis is rarely seen in children and is most commonly associated with those pediatric patients who have underlying valvular abnormalities [7]. This case highlights a highly unusual variant of IE, with a number of associated complications including mycotic aneurysms of the viscera (SMA) and cerebrum (MCA), stroke, and renal infarction.

Several case series have reported that incidence rates of neurologic complications in IE in children are between 15% and 40%, and mortality rates with such complications are as high as 60% [8,9]. Aneurysms in the SMA are very rare, constituting about 6% of all arterial aneurysms and are frequently infectious in etiology [10]. Neurologic manifestations in IE are most frequently associated with infection by

Staphylococcus aureus (21%). Mycotic aneurysms occur most frequently secondary to infections caused by non-hemolytic *Streptococci* (30%). In our case, both α -hemolytic *Streptococcus* and VRE were isolated; the VRE likely colonized secondarily after antibiotics had selected for this resistant and unusual IE pathogen.

Visceral aneurysms associated with IE are extremely rare in children, and few cases have been reported in the literature. In adults, the SMA is the most common artery besides the aorta to have infected aneurysm involvement [10]. Mycotic aneurysms have a high mortality rate because of their early or late potential for rupture; thus, abdominal pain in a patient with a recent history of endocarditis should be evaluated closely. A visceral pseudoaneurysm noted in a febrile patient can be presumed to be a mycotic aneurysm, especially in the absence of etiologies such as trauma or iatrogenic procedures.

Computed tomography is often the first imaging study performed during the evaluation for acute abdominal pain. Pseudoaneurysms present on contrast-enhanced CT as enhancing masses of soft tissue attenuation adjacent to a vessel. In some occasions, the aneurysm can be confused with bowel loops, especially if oral contrast has been administered. In this scenario, the diagnosis can be confirmed with ultrasound. There is often a characteristic "yin-yang" sign on color Doppler imaging where there is swirling pattern of internal flow in the center of the mass (Fig. 3).

The management of a visceral aneurysm should include resection along with appropriate antibiotic therapy for 6 to 8 weeks [11]. In this case, preservation of the entire intestine was possible. However, at the time of surgery, we were prepared to resect a proximal branch of the SMA with a revascularization of the intestine using a saphenous vein graft. The management of cerebral aneurysms should be guided by symptoms and assessment of the risks and benefits of surgical intervention. In our case, the SMA pseudoaneurysm was resected

completely with preservation of the main arterial branch. Should a more proximal portion of the SMA or aorta have been involved, a saphenous vein bypass may have been required and should be anticipated for at the beginning of the surgery. The MCA aneurysm was left untreated with close monitoring by frequent MRI imaging. Appropriate antibiotic coverage is crucial, particularly when a highly resistant organism has been verified. Similar cases warrant attentive management and timely access to any necessary operations.

References

- [1] Osler W. The Gulstonian lectures on malignant endocarditis. *BMJ* 1885;1:467-9.
- [2] Coward K, Tucker N, Darville T. Infective endocarditis in Arkansas children from 1990 through 2002. *Pediatr Infect Dis J* 2003;22:1048-52.
- [3] Van Hare GF, Ben-Shachar G, Liebman J, et al. Infective endocarditis in infants and children during the past 10 years: a decade of change. *Am Heart J* 1984;107:1235-40.
- [4] Stevens MP, Edmond MB. Endocarditis due to vancomycin-resistant *Enterococci*: case report and review of the literature. *Clin Infect Dis* 2005;41:1134-42.
- [5] Pruitt AA, Rubin RH, Karchner AW, et al. Neurologic complications of bacterial endocarditis. *Medicine (Baltimore)* 1978;57:329-43.
- [6] Hart RG, Foster JW, Luther MF, et al. Stroke in infective endocarditis. *Stroke* 1990;21:695-700.
- [7] Ferrieri P, Gewitz MH, Gerber MA, et al. Unique features of infective endocarditis in childhood. *Circulation* 2002;105:2115-7.
- [8] Heiro M, Nikoskelainen J, Engblom E, et al. Neurologic manifestations of infective endocarditis: a 17-year experience in a teaching hospital in Finland. *Arch Intern Med* 2000;160:2781-7.
- [9] Baddour LM, Wilson WE, Bayer AS, et al. Infective endocarditis diagnosis, antimicrobial therapy, and management of complications: a statement for healthcare professionals from the Committee on Rheumatic Fever, Endocarditis, and Kawasaki Disease, Council on Cardiovascular Disease in the Young, and the Councils on Clinical Cardiology, Stroke, and Cardiovascular Surgery and Anesthesia, American Heart Association. *Circulation* 2005;111:e394-e433.
- [10] Lorelli DR, Caimbra RA, Seabrook GR, et al. Diagnosis and management of aneurysms involving the superior mesenteric artery and its branches: a report of four cases. *Vasc Endovasc Surg* 2003;37:59-66.
- [11] Freidman SG, Pogo GJ, Moccio CG. Mycotic aneurysms of the superior mesenteric artery. *J Vasc Surg* 1987;6:87-90.