Spontaneous Coronary Sinus Infective Endocarditis in a 9-year-old: a rare case report with a favorable outcome

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Abstract

Coronary sinus infective endocarditis is extremely rare, more so in children. We report a case of coronary sinus infective endocarditis diagnosed by echocardiography in a 9-year-old boy with no pre-existing heart disease or any of the usual risk factors known to predispose to infective endocarditis. He was successfully managed non-surgically.

Introduction

Infective Endocarditis (IE) is an infection of the inner lining of the heart (the endocardium) and its related structures, especially the valves or any prosthetic valve inside the heart. It is a rare condition, with reported incidences of 2-12 cases per 100,000 people.[1,2] However, it is among the leading causes of cardiac-related deaths and among the four infectious causes of death worldwide.[3] Right-sided IEs are rarer compared to the left-sided types. Coronary Sinus (CS) IE is perhaps the rarest, even among the right-sided IEs. Most of the reported cases were in adults.[4-14] Like other forms of IEs, reported cases of CSIE, were mostly in those with underlining cardiac conditions like atrial septal defect, coronary artery fistula, tricuspid regurgitation, *etc*.[5,15,16] Other identifiable risk factors for CSIE include central venous catheterization (*e.g*. among patient on hemodialysis),[6] heart surgeries,[7] Intravenous Drug Abusers (IDAs),[4,8] or prolonged used of intravenous catheter, and conditions associated with tricuspid regurgitation.[6,14] However, in some instances, there was no identifiable risk factor.[9,10]

We report a rare case of coronary sinus IE in a child, a 9-year-old boy who presented with prolonged fever and a soft systolic murmur. He had an echocardiography, which showed a large vegetation in the CS, extending into the Right Atrium (RA) in an apparently normal heart. The patient was commenced on antibiotics, among other management. He fully recovered, was discharged home, and has remained stable for more than six months after discharge.

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Case Report

A 9-year-old boy presented to our facility with a complaint of fever of two weeks duration. Fever was described as high grade, initially intermittent, but it became persistent a week prior to presentation. There were associated chills and rigors. He also had anorexia, weight loss, and abdominal pain. A few days prior to presentation, he became progressively weak but had no history suggestive of paroxysmal nocturnal dyspnea or orthopnea. He had no chest pain and no history of recent dental procedures. With the onset of the fever, he had oral and parenteral (both intramuscular and intravenous) antimalarials and antibiotics (details not remembered by caregivers). These medications were procured and administered at the Primary Health Care Centre (PHC), and the injections were at a local patent medicine store. With no improvement, he presented at a secondary health facility where he was referred for an echo on account of a cardiac murmur. The echo showed a CS vegetation, on account of which the patient was referred to our facility. He was fully vaccinated. Other aspects of his history were not adversely remarkable.

At presentation, he was ill-looking, febrile (axillary temperature of 40.2), pale, mildly icteric, acyanosed, and had no edema. He had no 'peripheral stigmata' of IE. He was severely wasted. He was dyspneic and tachypneic with a respiratory rate of 40 breaths/min. The chest was clear. He was tachycardic with PR of 136 bpm, normal BP (96/58mmHg), and apex beat was not displaced. There

was a grade 2/6 systolic murmur that was loudest at the left lower sternal area. His oral exam was unremarkable. He had a mild generalized abdominal tenderness with hepatomegaly, 4 cm below the right costal margin (liver span of 12 cm). The spleen was not palpably enlarged. He had a repeat echo done in our facility, which confirmed the presence of vegetation attached to the posterior wall of the CS. It measured 18 mm with a thickness of about 1 mm. It was freely mobile, extending into the RA. There was turbulence in the CS and mild tricuspid regurgitation. No other structural anomaly was seen in the heart (Figure 1). The electrocardiogram showed sinus tachycardia. The full blood count showed elevated white cells (White Blood Cell count, WBC, 13.5x10^9/L) and neutrophil (10.2x10^9/L) counts; moderate anemia (Hematocrit Test, HCT, 19.1%), normal platelet counts of 390x10^9/l and markedly elevated Erythrocyte Sedimentation Rate (ESR) of 130 mm/ hr. He also had microscopic hematuria. His electrolytes, urea, and creatinine were normal. The retroviral screening was non-reactive. Blood samples were taken for culture, and he was commenced on intravenous ampicillin-flucloxacillin with amikacin. However, on day 3 of admission, he had 2 episodes of generalized, tonic-clonic convulsions, two hours apart. He had no differential body weakness. He was also noticed to have developed a tender, reddish swelling on the mid sternum. He had a lumbar puncture done, the result of which was unremarkable; no organism was isolated, and Cerebrospinal Fluid (CSF) biochemistry was normal: glucose and protein were 3.3 mmol/L and 17 mg/dL (normal ranges 2.5 mmol/L and 15-40mg/dL) respectively. The fever had persisted. The blood cultures also yielded no growth. Antibiotics were changed to IV ceftriaxone and IV clindamycin on day 4. The fever resolved within 48 hours of the commencement of the antibiotics. He also had packed red cell transfusion on account of the anemia. He continued to show remarkable improvement with progressive resolution of the vegetation from serial echo monitoring and normalization of Full Blood Cout (FBC), ESR, and other investigation results. By the second week, the vegetation had completely resolved (Figure 2). He no longer has the sternal swelling or murmur. He was discharged home after four weeks of intravenous antibiotics. He was continued on oral cefpodoxime and clindamycin for another two weeks. More than six months after discharge, he has remained stable and is doing well.

Discussion

IE is generally a rare infection, but with high mortality, especially if diagnosis/treatment is delayed.[3] The index case has none of the usual risk factors.[17] The clinical features, though unspecific, were very much in keeping. These include prolonged fever, weight loss, convulsions, features of nephritis, and sternal swelling, which could be due to septic embolus, markedly raised ESR, anemia, and microscopic hematuria, among other features. The absence of immunologic features like splinter hemorrhages, Janeway lesions, Roth spots, Osler nodes, *etc*., is, however, not surprising because they are rare in children.[17,18] The site of the vegetation (the coronary sinus) is very unusual. But the echocardiographic features were also very similar to those of CSIEs earlier reported: large, highly mobile vegetation, usually extending into the right atrium, a dilated CS with turbulent flow, absence of vegetation on the valves or elsewhere, *etc*.[5,6,10]Using the modified Duke criteria, the index patient has definite endocarditis; 1 major criterion (echo proven cardiac vegetation) and at least three minor criteria (fever >38 , elevated ESR, immune complex phenomenon like nephritis evidence by hematuria, serologic evidence of infection like the elevated white cell count, and vascular phenomena such as the sternal swelling and the aseptic meningitis).[19,20] The prior use of intravenous and intramuscular injections may be a predisposing risk factor, but we do not think it can be given the same significance as those of IDAs. The unique feature of this case is that it is perhaps the first detailed reported case of CSIE in a child, more so with no background cardiac disease or any known usual risk factor.[19,20] We came across two related cases of IE in children. One was CS IE, which was mentioned in a retrospective review of children with IE.[21] The mean age of the subjects was seven years. No other details of that particular patient were available. A related case was that of IE and pseudoaneurysm of the sinus of Valsalva in 6 a 6-year-old girl with a bicuspid aortic valve.[22]

Although the blood cultures yielded no growth, the index patient responded to the combination of ceftriaxone and clindamycin. The negative culture may not be unconnected with the prior antibiotic given to this child.[17] While ceftriaxone is recommended for non-valvular endocarditis,[23] the choice of clindamycin was informed by our experience with its effectiveness in the management of staphylococcal infections. *Staphylococcus aureus* is the major cause of IE in a normal heart, and most of the reported culture-positive CS IE was due to staphylococcus aureus.[6,17]

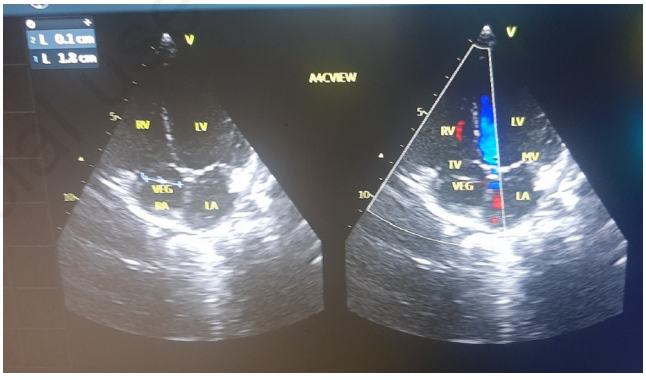

Figure 1. Modified apical four-chamber (coronary sinus) view transthoracic echocardiogram showing a vegetation (VEG) attached to the posterior wall of the Coronary Sinus (CS), extending into the Right Atrium (RA); it measured 18 mm by 1 mm.

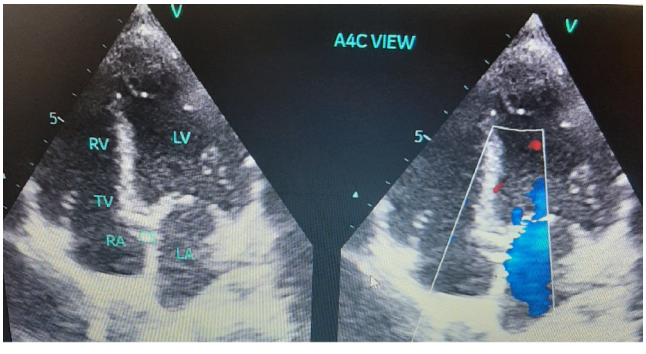

Figure 2. Modified apical four-chamber view showing coronary sinus free of vegetation (echo done after resolution of Coronary Sinus InfectiveEndocarditis, CSIE). RV right ventricle; LV, left ventricle; LA, right atrium; VEG, vegetation; MV, mitral valve; TR, tricuspid valve.

Conclusions

This case highlights the need for a high index of suspicion and the need for prompt performance of echocardiography in any child suspected of IE. It also brings to the fore the need for a thorough check of the 'unusual sites' like the CS during such echocardiography.

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