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CLINICAL CASE REPORT

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A Rare Case of Pulmonary Artery Dissection Associated With Infective Endocarditis

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Abstract: Pulmonary artery dissection (PAD) is a rare condition with high mortality and has not been reported in patient with infective endocarditis (IE). Here, we report the first case of such patient who experienced PDA and survived after surgical intervention.

A 10-year-old female child was diagnosed as IE with a patent ductus arteriosis (PDA) and a vegetation on the left side of pulmonary artery trunk ($10 \times 5\,mm^2$). Following 3-week antibacterial treatment, the body temperature of patient returned to normal, and the size of vegetation reduced ($7 \times 3\,mm^2$). However, the patient had a sudden attack of sustained and crushing right chest pain, orthopnea with increasing respiratory rate (> 60/min), and acute high fever. Echocardiography revealed the detachment of vegetation on the first day and dissection of pulmonary artery on the next day. The patient received immediate surgical intervention. It was found that aneurysm had a size of $28 \times 20\,mm^2$ and its orifice (the dissecting site) located on the opposite side of the PDA opening (right side of the pulmonary artery trunk). The dissected left wall of pulmonary artery trunk was reconstructed followed by the closure of PDA with suture. The patient recovered uneventfully.

From this case, we learned that the surgical intervention should be considered at an early time for IE patients who have a vegetation in pulmonary artery and PDA. After the infection is under control, the earlier surgery may prevent severe complications.

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Abbreviations: CHD = congenital heart disease, IE = infective endocarditis, PAD = pulmonary artery dissection, PDA = patent ductus arteriosis.

INTRODUCTION

Infective endocarditis (IE) is a severe acquired heart disease, which is frequently observed in Kawasaki disease, neonates and infants suffering from severe infections, as well as children with a correction of congenital heart disease (CHD) at a younger age. Unfortunately, IE showed an increasing incidence among children.[1,2] Although antibacterial therapy is the current mainstay for IE management, surgical intervention has been adopted in some patients, especially for those who have a vegetation. Disintegration and/or falling of a vegetation is the most frequently observed severe complications in IE; it may block downstream arteries in pulmonary or systematic circulation based on its location, resulting in paralysis or sudden death.[3] Here, we report a rare IE case with a complication of pulmonary artery dissection (PAD) following the detachment of vegetation, which was successfully treated with surgical intervention. To our best knowledge, this is first case of PAD complication in an IE patient.

CASE REPORT

This study was approved by the Ethics Committee of West China Second University Hospital of Sichuan University. A 10-year-old female child experienced cough and fever (>38°C) for >10 days, with a progressive exertional dyspnea was admitted to our hospital. A continuous murmur was present at the second rib interval with distant heart sounds. The echocardiography revealed a patent ductus arteriosis (PDA, 5 mm shunting duct) and a vegetation on the left side of pulmonary artery trunk ($10 \times 5\,mm^2$), which moved with the heart beating. In addition, pulmonary valvular regurgitation and pericardial effusion have also been identified (Figure 1A). Pulmonary hypertension was not indicated. The bacterial and fungal culture showed negative results, presumably owing to antibacterial treatment before admission. Upon reviewing previous records, there was a positive report for *Staphylococcus aureus* from the blood bacterial culture. In addition, the rheumatoid factor test was positive and CT scan revealed severe pneumonia. According to the Duke criteria, the patient was diagnosed as IE, meeting 1 primary diagnostic criterion (vegetation in pulmonary trunk) and 3 secondary diagnostic criteria (fever, essential heart disease, and positive rheumatoid factor).

Based on the positive result for *S aureus* from the previous blood bacterial culture, vancomycin (1 g/day) and cefoperazone (4 g/day) were administered in combination of prednisone (20 mg/day). Three weeks later, the body temperature of patient returned to normal. Cough and dyspnea were fully corrected. Echocardiography showed the pre-existing PDA, reduction of pericardial effusion, and smaller size of vegetation on pulmonary artery trunk ($7 \times 3\,mm^2$). Chest x-ray displayed amelioration of pneumonia. Taken together, the treatment was proper and effective. Surprisingly, the patient had a sudden attack of sustained and crushing right chest pain, orthopnea with

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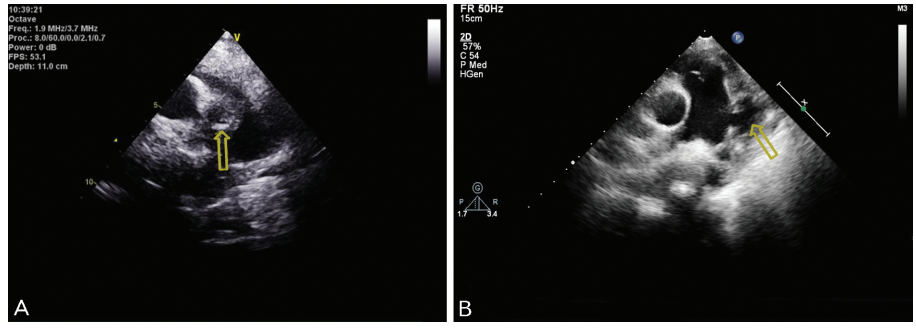

FIGURE 1. Echocardiographic images. (A) The presence of a vegetation in pulmonary artery upon admission to hospital. (B) The dissection of pulmonary artery after the falling of the vegetation.

increasing respiratory rate (> 60/min), and high fever. The emergency echocardiography revealed the detachment of vegetation, which presumably entered the second or third sectional pulmonary arteries. Despite of negative results for blood bacterial culture, the antibacterial regimen was amended as vancomycin (1 g/day) plus meropenem (1.5 g/day) based on our empirical success in managing patient with septic embolism. On the next day, the repeated echocardiography revealed the dissection of pulmonary artery with a newly formed aneurysm with a size of $25 \times 18\,mm^2$ (Figure 1B). Chest x-ray revealed a large prominence of pulmonary artery truck when compared to previous examination (Figure 2A and B). The contrast-enhanced CT scan indicated that the pulmonary artery dissection originated from the site where the former vegetation located (Figure 2C). Considering the risk of aneurysm rupture, the patient was immediately transferred to the operation room for surgical intervention. Because there was no evidence of pulmonary artery hypertension and the size of aneurysm was < 50 mm, the replacement of pulmonary artery and the insertion of endovascular stent-graft were not recommended by surgeons. Instead, the direct repair of aneurysm and closure of PDA with suture were carried out under general anesthesia and standard extracorporeal circulation. Specifically, the operation was performed through a median sternotomy. Following pericardiotomy, the dilated aneurysm on pulmonary artery was exposed. Under mild hypothermia, cardiopulmonary bypass was conducted with aortic and bicaval cannulations. After cardiopulmonary bypass was established, the main pulmonary artery was dissected carefully to expose the lesion site of the PAD. It was found that aneurysm had a size of $28 \times 20\,mm^2$ and its orifice (the dissecting site, on the left side of the pulmonary artery trunk) located on the opposite side of the PDA opening (right side of the pulmonary artery trunk). The dissected left wall of pulmonary artery trunk was reconstructed followed by the closure of PDA with suture. The patient recovered eventfully and was extubated 6 hour postoperatively. Echocardiography on 8th postoperative day revealed normal cardiac function and a smooth pulmonary artery lumen. Patient was discharged on the 10th postoperative day.

DISCUSSION

Pulmonary artery dissection is a rare condition. There are <100 reported cases of PAD in the literature, and most of them were found in autopsies for individuals with a sudden death.[4] Although PAD has a very high mortality rate,[4] no consensus has been reached on how to prevent the formation of PAD. The mainstay of management focuses on primary diseases. The primary pulmonary hypertension or secondary to CHD are considered as the major cause of PAD. Vascular inflammatory disease, aorto-pulmonary fistulas, connective tissue disease, and catheter-induced vessel wall injury are also considered to be potential risk factors.[5–7] To our knowledge, the present study is the first report of a pulmonary artery dissection associated with IE.

Regarding the formation of vegetation and PAD in this patient, we think that it is probably associated with the pre-existing PDA. The abnormal hydrodynamics from the PDA shunting might cause the injury on the wall of pulmonary artery trunk and facilitate bacterial colonization. We speculate that the bacterial growth formed a vegetation and subsequently developed into severe IE. Following the 3-week antibacterial treatment, the vegetation shirked and disintegrated. Unfortunately, the falling of vegetation remainder resulted in the embolization of the sectional pulmonary arteries. Despite the patient survived this severe complication, the vegetation attaching site might be further damaged by the shearing stress from the shunting blood

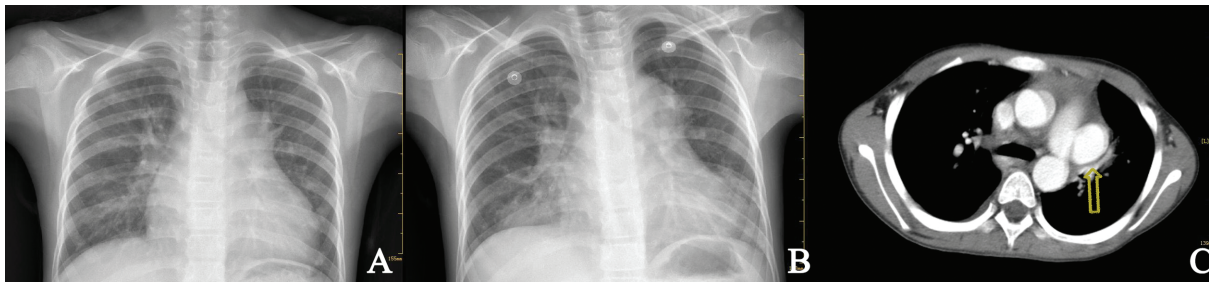

FIGURE 2. The chest x-ray and CT scan. (A) X-ray shows the enlargement of pulmonary artery segment. (B) X-ray shows the significant increase in prominence of pulmonary artery segment. (C) CT demonstrated the dissection of pulmonary artery. CT = computed tomography.

and resulted in a dissection. In summary, PDA might initially facilitate the formation of bacterial vegetation, and the focal lesion from bacteria weakened the pulmonary arty wall. In the meantime, PDA might also contribute to the development of PAD after the falling of vegetation. This case suggested that PAD could occur in the absence of pulmonary hypertension if a PDA is present in IE patient.

In the past decades, extensive studies have been conducted on the treatment of IE, and several guidelines have been established and proved to be effective. According to the standard regimen, the antibacterial therapy should be started as soon as the IE is diagnosed and should be given continuously for at least 4 week. During such long duration of treatment, the most dangerous complication is the falling of vegetation and embolism. Recently, surgical intervention has been considered as the second option for IE patient who have an indication.[8] The surgical removal of vegetation, repair of injured cardiovascular wall, and correction of abnormal structure greatly decrease the risk of complications and improve the prognosis.[9] However, it is still controversial when the best time for such an operation is. Based on our experience with the present case, we propose to conduct the operation at a earlier time for IE patients with a vegetation in pulmonary artery when the following conditions are observed during antibacterial treatment: (1) normal body temperature, (2) negative results for multiple bacterial cultures, (3) reduction of vegetation size, (4) decrease in adhesion and/or increase of vegetation movement, (5) reduction of pericardial effusion, and (6) evidence of pulmonary hypertension. The earlier surgical intervention may be able to prevent the development of severe complication, such as PAD. Of course, in addition to these considerations, the final regimen should be determined by overall surgical risk for specific individuals.

CONCLUSIONS

The present study is the first report of PAD associated with IE and PDA without the presence of pulmonary artery hypertension. This patient received the antibacterial treatment for IE and experienced severe complications. Because of timely surgical intervention, the patient survived PAD and recovered. From this case, we learned that the surgical intervention should be considered at an early time (after the infection is fully controlled) for IE patients who have a vegetation in pulmonary artery and PDA.

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