Case Report

Modified Blalock-Taussig Shunt and Giant Perigraft Reaction

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Abstract

This is a case of a modified Blalock-Taussig shunt, which was complicated by perigraft transudative, fibrinous fluid accumulation and recurrence after surgical intervention. Follow-up and expectant management of the patient was successful. Our experience regarding this complication is presented.

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Introduction

Over the past several years, the indications for and timings of palliative procedures have changed. Shunt surgery is one of these palliative procedures intended to increase the pulmonary blood flow when indicated. Usually a prosthetic material is used for the construction of a systemic pulmonary shunt.[1] There have been reports of successful allograft saphenous vein grafts for shunt surgery.[2] Shunt surgery is usually performed when there is an inadequate pulmonary flow. Examples of indications for increasing the pulmonary blood flow and shunt surgery are as follows: The tetralogy of Fallot when the patient is less than 3 months old and is, therefore, too small for an adequate reconstruction of the right ventricular outflow tract, pulmonary atresia with or without ventricular septal defect, tricuspid atresia with pulmonary stenosis, single ventricle with pulmonary stenosis, and transposition of the great arteries with ventricular septal defect and pulmonary stenosis.

In summary, aortopulmonary shunts will benefit any patient with pulmonary obstruction, which usually presents as cyanosis, dyspnea, and/or failure to thrive.1 Modified Blalock-Taussig shunts are easy to construct and, by comparison with other types of shunts, cause less PA distortion and contribute to a better growth of the pulmonary arteries. Be that as it may, like other types of shunts, they may be complicated by shunt stenosis or thrombosis. What is also deserving of note is that Blalock-Taussig shunts are less durable than classic Blalock-Taussig shunts.[3-5] Persistent serum leakage through the prosthetic graft causing perigraft seroma is a rare but devastating complication resulting in an increased duration of tube drainage or reinsertion of chest tubes.[6,7] Confined seroma, which has also been reported,[8-13] may be diagnosed by chest X-ray, CT scan, MRI, sonography, or echocardiography.[8,10,13,14] Some authors believe that the homeostatic milieu of the patient can be effective in seroma

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leakage or seroma formation[15] and suggest that while heparin can increase the risk, such measures as using fibrinogen to prevent serous fluid leakage through polytetrafluoroethylene tubular graft can decrease the risk.[16-17] There are reports of infection in the existing literature.[18] Seroma formation may give rise to graft occlusion;[19] this complication can be difficult to treat and may even recur, sometimes necessitating innovative procedures like wrapping the polytetrafluoroethylene shunt with the parietal pleura flap harvested from the adjacent chest wall.[20]

The case presented here is one of a modified Blalock-Taussig shunt, which ran into a rare complication.

Case Report

A 3-month-old girl was referred to our hospital with a one-month history of cyanosis and spells of attacks. On physical examination, a well-developed and well-nourished baby with moderate-to-severe cyanosis without clubbing was found; and on auscultation, a systolic murmur grade 3/5 in the left sternal border was heard.

Initial arterial blood gas (ABG) revealed: PH=7.44, PO_2=20mmHg, O_2S=34%, PCO_2=25mmHg, HCO_3=16meq/L, and BE=_3meq/L. Other lab tests were normal.

Echocardiography showed: pulmonary atresia with small and confluent pulmonary artery branches, left pulmonary artery (LPA)=0.32cm, right pulmonary artery (RPA)=0.34cm, small patent ductus arteriosus (PDA) and ejection fraction (EF)=88%.

With the diagnosis of pulmonary atresia, ventricular septal defect, and patent ductus arteriosus, the patient became a candidate for a systemic-to-pulmonary (modified Blalock Taussig) shunt. The operation was carried out using a Gore-Tex conduit (6mm). Postoperative control echocardiography showed a normal functioning shunt.

Four days after discharge and on the 8th postoperative day, however, the patient was re-admitted due to fever, diarrhea, and vomiting. Physical examination showed no new findings, but chest radiography revealed an opacity with round borders and sharp margins at the apex of the left hemithorax (Figure 1).

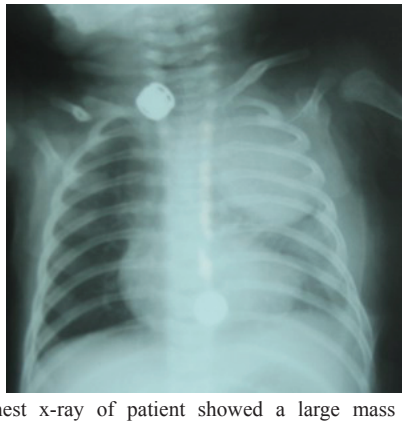

Figure 1. Chest x-ray of patient showed a large mass in upper left hemithorax.

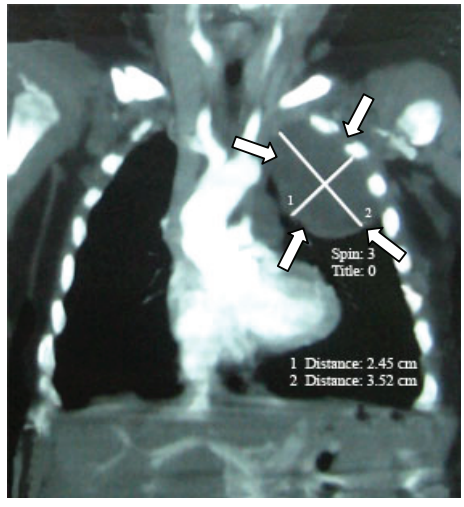

Figure 2. CT scan, showed an opacity with fluid density, (about 4×3cm) in the upper left hemithorax with slightly compressed apical lung parenchyma

CT scan (Figure 2) revealed an ovaloid opacity with fluid attenuation and without enhancement measuring about 4×3 cm in the left hemithorax, which slightly compressed the adjacent lung parenchyma and was suggestive of loculated pleural fluid or hemothorax. The

other possibility was empyema in that setting. The shunt was patent in this evaluation.

Because the patient's O_2 saturation was diminished, she underwent redo surgery, during which we found a round, well-bordered

white cystic mass located in the apical area of the left hemithorax with moderate adhesion to the adjacent structures. The mass was excised; it was encapsulates and contained gelatinous material, which was excised completely and sent for pathologic examinations.

She was re-explored a few hours later because of excessive chest tube drainage, begotten by blood oozing from the surrounding tissues. In pathologic evaluations, sections showed extensive fibrin exudation and amorphous hyaline material with scattered inflammatory cells (fibrinous material). A few days later, chest X-ray showed the re-development of the mass at the same location. The fluid was, thereafter, aspirated percutaneously for analysis, which was indicative of a transudative fluid.

The patient's good general condition prompted us to discharge her, and a one-year follow-up shows that the mass has decreased in size and the patient is well.

Discussion

The appearance of a localized mass on the chest film surrounding the prosthesis requires the exclusion of hematoma, aneurysm, or inflammation. In the case presented here, we are inclined to believe that the diagnosis could

have been confined seroma or, in other words, a perigraft transudative, fibrinous fluid accumulation. A retrospective review of the case through correlating the clinical and pathologic findings is indicative of the fact that the mass was a giant perigraft seroma and or transudative, fibrinous fluid accumulation around a Gore-Tex prosthetic shunt. Local variations in porosity and structure of the implanted prosthesis may play a critical role in the pathogenesis of perigraft-reaction,[21] as is indicated by our case. In the event of an unsuccessful observation of a localized perigraft seroma or of unsuccessful chest drainage of a massive pleural serous effusion, graft replacement and placement of a different graft material are reported to have the best results. [21]

With respect to our own patient, the fluid was identical with serum; consequently, its pathogenesis is likely to have been plasmaphresis. Excision of the mass is associated with a high rate of recurrence, and we recommend a conservative management of such cases when the patient is in good clinical condition.

In retrospect, it seems that the surgical excision of the mass was not necessary and that the patient could have derived greater benefit from a policy of conservative management.

Conclusion

We presented a case of a giant perigraft transudative, fibrinous fluid accumulation in a modified Blalock-Taussig shunt. We recommend a conservative approach to such complications, if possible.

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