Perigraft seroma complicating the modified Blalock-Taussig shunts: two consecutive instances with review of literature

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Systemic-to-pulmonary artery shunts using polytetrafluoroethylene (PTFE) (modified Blalock-Taussig shunt) are being used successfully in palliation of cyanotic congenital heart diseases. Graft thrombosis is by far the most common complication of the procedure. Persistant serum leakage through PTFE graft causing perigraft seroma is a rare but devastating complication resulting in increased duration of tube drainage or reinsertion of chest tubes, prolonged hospital stay, and multiple operations. Two consecutive modified Blalock-Taussig shunts complicated by perigraft seroma formation are presented here with review of the literature.

Key words: perigraft seroma, polytetrafluoroethylene, modified Blalock-Taussig shunt.

In patients with diminished pulmonary blood flow, systemic-pulmonary artery shunts remain important for palliation. Since the classic Blalock-Taussig (B-T) shunt operation was first introduced, it has been considered the best procedure whenever possible[1]. Direct aortopulmonary anastamoses like Waterston (ascending aorta to right pulmonary artery) and Potts (descending aorta to main pulmonary artery) shunts are now rarely employed because of difficulties in controlling shunt flow, distortion of the right pulmonary artery, and difficulties in interrupting the shunt during intracardiac repair. Presentation of the modification of the classic B-T shunt by interposing a polytetrafluoroethylene (PTFE) vascular graft between the sides of the subclavian and pulmonary arteries provided a pressure- and volume-controlled pulmonary perfusion with preservation of distal blood flow in the subclavian artery[2,3].

Graft thrombosis, infection, and pseudoaneurysm formation are commonly reported complications related to modified B-T shunt operations. Perigraft seroma is unusual and infrequent, but nevertheless a devastating complication. Serous fluid leakage causing seroma formation around a PTFE graft was first recognized as a complication in arteriovenous fistulas[4]. Subsequently, seroma formation following peripheral arterial reconstructions and systemic-pulmonary arterial shunts has been reported[5-7].

Two consecutive modified B-T shunts which were complicated with perigraft seromas diagnosed in the early postoperative period are presented in this paper.

Case Reports

Case 1

A one-year-old girl was admitted with the diagnosis of tetralogy of Fallot and pulmonary atresia with confluent pulmonary artery branches. A palliative modified B-T shunt was planned because pulmonary vasculature was hypoplastic. A 5 mm PTFE expanded GoreTex graft (W.L. Gore & Associates, Inc., Elkton, MD) was interposed between the right subclavian and right pulmonary arteries through a right posterolateral thoracotomy. Ten hours after the operation, a consolidation was observed in the right upper zone on chest X-ray (Fig. 1). This image persisted and became larger despite controlled ventilation. The patient underwent rethoracotomy on the same day because of a probable hematoma formation around the anastamoses. A right superior mediastinal mass of 6 to 8 cm in size surrounding the functional graft was explored. The clear, gelatinous material

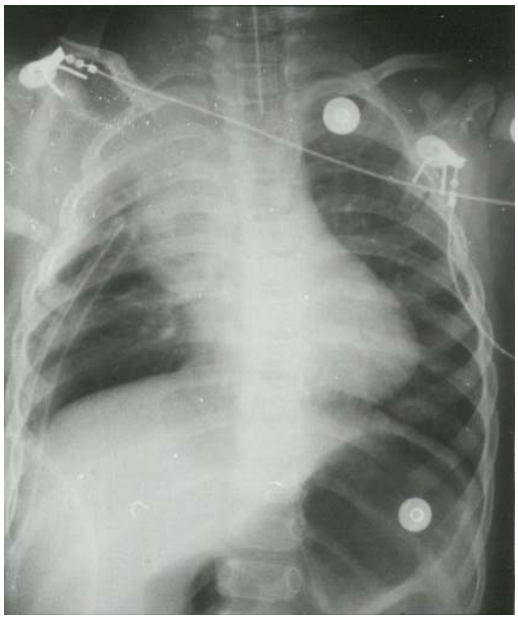

Fig. 1. Early postoperative chest X-ray demonstrating consolidation of right upper zone indicating seroma formation.

with a fine capsule was aspirated and removed but the graft continued weeping colorless serum. A thin sheath of Surgicell® (ETHICON Ltd. Bankhead Ave. Edinburgh EH11 4HE IRL-Tallagh, Dublin 24) was wrapped around the graft. Two silicone drainage tubes were placed in the right pleural cavity. The amount of drainage decreased slowly and ceased on the third postoperative day. The patient had an uneventful recovery and was discharged on the seventh postoperative day. Eventual follow-up chest X-ray showed no evidence of fluid collection (Fig. 2).

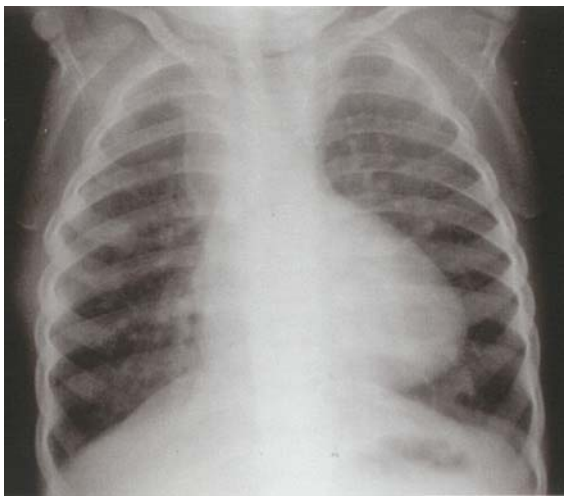

Fig. 2. Follow-up chest X-ray showed no evidence of fluid collection.

Case 2

A single stage surgical repair was planned for a 10-month-old boy with tertalogy of Fallot. A well developed conal branch of the right coronary artery which was crossing the right ventricular outflow tract was observed through a median split sternotomy, and impeded a total correction. A left modified B-T shunt was then performed with a 5 mm thin walled PTFE graft. Early postoperative period was uneventful, and the drainage tube was removed on the second postoperative day. The patient developed respiratory distress on the fifth day. Chest X-ray revealed left superior mediastinal mass. Thoracal computerized tomography (CT) demonstrated perigraft seroma (Fig. 3). Rethoracotomy was performed and the clear and gelatinous seroma material was removed and silicone drainage tube was replaced. The lung fully re-expanded soon after surgery and the patient was kept under controlled ventilation for one day. The composition of the drainage changed to chylous in nature on the third postoperative day and continued for seven more days (200 cc/day). Dietary control and continuous intravenous somatostatin infusion (5 μg/kg/hr) controlled the drainage.

Fig. 3. Computerized tomography of thorax showing polytetra fluorethylene graft surrounded by the seroma.

Discussion

Serous leakage causing seroma formation around a PTFE graft is a rare but challenging complication of modified B-T shunt operations. Perigraft seroma is a collection of clear, sterile fluid confined in a fibrous pseudomembrane around a graft. It was first described in arteriovenous

fistulas[4]. Blumenberg and associates[5] reported a large series of perigraft seroma following peripheral arterial interventions, in which data was gathered by a North American questionnaire. Perigraft seromas following systemic-pulmonary artery shunts when PTFE grafts were used were reported subsequently[6,7]. The incidence of perigraft seroma following the modified B-T shunt is 9.4%[6] to 12%[8]. However, very few patients (2-3%) require a reoperation for persistent leakage[9]. Ahn and associates[10] reported a perigraft seroma incidence of 1.2% for peripheral vascular interventions. Since most cases cannot be detected, the actual incidence of perigraft seroma is unknown.

The most frequently complicated graft with serous leakage is knitted Dacron, followed by PTFE; autogenous vein grafts seem to be least affected by this complication[5]. The cause of perigraft seroma is unknown, but various theories have been postulated to explain etiologic mechanisms of seroma formation. The hydrophobic surface of PTFE becomes hydrophilic when in contact with blood and interstitial fluid, called wetting. In peripheral arterial reconstructions, grafts placed in subcutaneous tissue are more prone to perigraft seroma formation because of liquefied fat, thus proper tunneling becomes important. Contact of graft material with organic solvents, povidone iodine, excessive manipulation, and forced irrigation through the graft wall can lead to serum leakage through the interstices of the graft material[6].

As a result of imbalance between wetting and sealing of the graft surface, surrounding connective tissue fails to incorporate the graft[5]. Interfibrillar distance and intraluminal pressure are theoretically related to leakage through the graft wall[11]. Graft handling like stretching, bending, clamping, and resterilization would influence seroma formation. Resterilized grafts were used in both our cases.

Histopathologic examination of failed grafts reveals that the graft is surrounded by clear proteinaceous material devoid of cellular structures and inflammatory infiltrate, circumscribed by a fibrous pseudomembrane and scarce fibroblasts lining the graft surface which actually play a role in the poor incorporation of the surrounding connective tissue[5,6,9,12]. The content of the fibrous pseudomembrane and inner secretory membrane is biochemically similar to serum, which led to definition of the seroma as an ultrafiltrate of the blood[12].

Sladen and associates[13] proposed a hypothesis and explained the mechanism of seroma formation by fibroblast inhibition. They tried to isolate a humoral fibroblast inhibitor in sera of patients with seromas. Ahn and associates[10], in a similar study, also showed presence of humoral factors in sera of patients with persistent perigraft seroma which caused in vitro arrest of fibroblast growth. They also demonstrated loss of the inhibitory effect of the sera after spontaneous resolution of the seromas.

Age and graft diameter seemed to influence seroma formation in the LeBlanc series[6], that is, older patients with higher mean arterial pressure and with greater graft diameter tended to develop perigraft seromas. However, Berger and associates[8] have reported that the effect of age and graft size diminished after a multiple logistic regression analysis.

The relationship between seroma formation and postoperative heparin administration is controversial. LeBlanc and associates[6] reported a nonsignificant relationship between the incidence of perigraft seroma and heparin use. In multivariate analysis of the Berger series, heparin was the only independent factor influencing seroma incidence[8]. However, in that series, heparin was mostly administered to patients in whom unifocalization was performed.

Perigraft seromas are presented as a growing mediastinal mass in chest X-ray, in most cases appearing three days following the operation. Hydrophobic PTFE grafts commonly become hydrophilic two to four days following implantation[9]. In our first case, we experienced an acute development of a perigraft seroma clearly defined in the same postoperative night, which is quite uncommon. The consequence of repeated thoracotomy, removal of the seroma, and wrapping of the graft with surgicell was satisfactory; drainage ceased in three days. Sustained PTFE graft leakage can resemble traumatic chylothorax. In our second case, pleural drainage following removal of the seroma became chylous, making the case more complicated. Perigraft seroma must be differentiated from resolving hematoma, atelectasis, pseudoaneurysm, and graft infection. The differentiation of seroma and hematoma is sometimes difficult because calcifications can be present in both instances, and can be distinguished by demonstration of

hemosiderin in a hematoma. In most instances, the diagnosis is made in the operation theater[14]. Echocardiography is a simple, safe, and noninvasive method for evaluation and follow-up of a complicated B-T shunt[15]. CT and magnetic resonance imaging (MRI) can demonstrate graft and perigraft seroma with high accuracy in almost every case. Graft wall enhancement in CT following intravenous contrast injection may provide evidence of abnormal graft permeability[16].

The natural history of perigraft seromas, which are complications of PTFE grafts, is unknown, but nearly 65% of seromas are reported to resolve spontaneously[5], although resolution may be slow or incomplete[6]. Nonetheless, there is no evidence to suggest that graft compression by perigraft seroma and compression of the lung may deteriorate pulmonary functions[12]. The important complication of a perigraft seroma is infection, observed in one of 22 patients followed without any intervention. On the other hand, the highest incidence of infection was observed in patients who were treated only by removal of the cyst but not replacement of the graft[5]. Persistence and recurrence of seroma can be as high as 40% when patients are treated with aspiration or drainage; however, satisfactory results were obtained when failed grafts were replaced, especially with a different material[5,12]. conservative approach is justified in most instances when high incidence of spontaneous resolution is considered.

In symptomatic cases surgical interventions have been employed, including aspiration of the cyst, enhancement of the graft by wrapping with absorbable collagen hemostat or collagen soaked in fibrin glue, topical application of acrylic tissue adhesives, and intraluminal injection of cryoprecipitate (containing aprotinin, thrombin, and calcium)[9,17].

We concluded that perigraft seroma complicating a modified Blalock-Taussig shunt is still a challenging problem despite all measures to protect PTFE grafts. Perigraft seromas increase the hospital stay significantly and may necessitate repeat thoracotomy, rendering B-T shunts, technically a simple operation, troublesome. In this paper we presented two consecutive modified B-T shunts which were both complicated with perigraft seroma formation. We point at the acute onset of seroma formation in the former case and development of chylothorax following rethoracotomy in the latter, which further complicated the cases.

REFERENCES

1. Blalock A, Taussig HB. The surgical treatment of malformations of the heart in which there is pulmonary atresia. JAMA 1945; 128: 189.
2. de Leval MR, McKay R, Jones M, et al. Modified Blalock-Taussig shunt. J Thorac Cardiovasc Surg 1981; 81: 112.
3. McKay R, de Leval MR, Rees P, et al. Postoperative angiograph assessment of modified Blalock-Taussig shunts using expanded polytetrafluoroethylene (GoreTex). Ann Thorac Surg 1980; 30. 137.
4. Bolton W, Cannon JA. Seroma formation associated with PTFE vascular grafts used as arteriovenous fistulae. Dial Transplant 1981; 10: 60.
5. Bulumenberg RM Gelfand ML, Dale WA. Perigraft seromas complicating aterial grafts. Surgery 1985; 97: 194-204.
6. LeBlanc J, Albus R, Williams WG, et al. Serous fluid leakage: a complication following the modified Blalock-Taussig shunt. J Thorac Cardiovasc Surg 1984; 88: 259-262.
7. Damus PS. Seroma formation after implantation of Gore-Tex vascular grafts in cyanotic children. J Thorac Cardiovasc Surg 1984; 88: 310-311.
8. Berger RM, Bol-Raap G, Hop WJ, Bogers AJ, Hess J. Heparin as a risk factor for perigraft seroma complicating the modified Blalock-Taussig shunt. J Thorac Cardiovasc Surg 1998; 116: 286-293.
9. Maitland A, Wiliams WG, Coles JG, Freedom RM, Trusler GA. A method of treating serous fluid leak from a polytetrafluoroethylene Blalock-Taussig shunt. J Thorac Cardiovasc Surg 1985; 90: 791-793.
10. Ahn SS, Machleder HI, Gupta R, Moore WS. Perigraft seroma: clinical, histological, and serologic correlates. Am J Surg 1987; 154: 173-178.
11. Tabata R, Kobayashi T, Mori A, et al. A computer simulation of the plasma leakage through a vascular prosthesis made of expanded polytetrafluoroethylene. J Thorac Cardiovasc Surg 1993; 105: 598-604.
12. Lewis P, Wolfe JH. Lymphatic fistula and perigraft seroma. Br J Surg 1993; 80: 410-411.
13. Sladen JG, Mandl MA, Grossman L, Denegri JF. Fibroblast inhibition: a new and treatable cause of prosthetic graft failure. Am J Surg 1985; 149: 587-590.
14. Leblanc JG, Vince DJ, Taylor GP. Perigraft seroma: long-term complications. J Thorac Cardiovasc Surg 1986; 92: 451-454.
15. Özkutlu S, Özbarlas N, Demircin M. Perigraft seroma diagnosed by echocardiography: a complication following Blalock-Taussig shunt. Int J Cardiol 1992; 36: 244-246.
16. Fink AM, Ditchfield MR. Wall enhancement of leaking polytetrafluoroethylene grafts: a new sign. Pediatr Radiol 1997; 27: 327-329.
17. Sim EK, Wong ML, Tan CT, Lee CN. Histoacryl tissue adhesive: an alternative means of stopping polytetrafluoroethylene graft sweating. J Thorac Cardiovasc Surg 1993; 106: 1227-1228.