

## **SERINOMA: REPORT OF A CASE SECONDARY TO MODIFIED SYSTEMIC TO PULMONARY BLALOCK SHUNT**

Jose A. Quibrera-Matienzo MD, Gabriel Camacho-Alva MD, Raul Morales-Cuevas MD

Pediatric Hospital of Sinaloa, Mexico

### **SUMMARY**

The performing of a systemic-to-pulmonary artery shunt (modified Blalock-Taussig) with (polytetrafluoroethylene) tubular grafts is a palliative procedure used frequently in congenital heart disease with obstruction of pulmonary blood flow

The development of serinoma surrounding the graft is an unusual complication of this procedure. We present a nine months old patient that develop right pleural effusion associated to this entity, that decreased spontaneously with persistence of the radiological image of the serinoma at two years follow up.

Key words. Pulmonary atresia; serinoma; Blalock Taussig shunt

### **INTRODUCTION**

The goretex graft (polytetrafluoroethylene, PTFE) has been used to perform the systemic to pulmonary shunt (Blalock Taussig operation). Their use allows better control of the flow, avoid distortion of the pulmonary arteries and preserve the distal circulation toward the subclavian artery<sup>1</sup>. Occasionally important flight of serous liquid can exist through the interstice of the implant, what leads to excessive drainage and pleural effusion<sup>2</sup>, or the development of a serinoma around the implant, defined this as the collection of clear and sterile liquid in a fibrous pseudo membrane<sup>3</sup>.

The incidence of this complication varies from 2 to 18% of the surgeries for modified Blalock shunts, and it occurs at any age<sup>1,2</sup>.

Its clinical presentation usually happens between the second to the 12 postoperative weeks, with dyspnea secondary to the effusion<sup>1,2</sup>.

CORRESPONDENCE: Jose A. Quibrera-Matienzo Donato Guerra y Constitución (without number) Colonia Almada, 80200 Culiacan, Sinaloa, México Phone number: 52 66 77 13 58 07 Fax: 52 66 77 12 89 60 , e-mail: jaquim@cin.megared.net.mx

Its treatment depends on the magnitude of the effusion that has been created; its natural history shows that can be self limited, could needs the use of substances for waterproof the implant or replace the shunt with a new surgery.

## CLINICAL CASE DESCRIPTION

The patient is a 9 months old male with pulmonary atresia and interventricular septal defect without treatment and secondary hipercyanotic spells, and moderate delay in their development. The echocardiogram shows absence of the pulmonary trunk and flow to the confluent pulmonary arteries through a ductus arteriosus. A right systemic to pulmonary shunt was placed with a 5 mm PTFE graft and he was discharged after five days. Two days later he return to hospital with moderate dyspnea and the X ray showed a right pleural effusion affecting the entire lung, and a chest tube was placed. A rounded image was observed in the right superior margin of the heart (Figure 1A), suspecting the development of an aneurism in the place of the shunt or a serinoma; a CT scan and the selective angiography in the right subclavian artery confirmed this last diagnosis (Figures 2 and 3).

The pleural effusion was diminishing gradually and he was discharged after five days. His outcome shows the characteristic of the heart disease awaiting surgical correction and persistence of the radiological image of serinoma (Figure 1B).

## DISCUSSION.

The development of a serinoma in the course of a modified systemic to pulmonar shunt with a PTFE graft (modified Blalock-Taussig operation) is a rare complication whose frequency varies from 2 to 18% with no sex or age preference<sup>1,3</sup>. In its pathogenesis has been postulated the existence of a fibroblastic humoral inhibitory factor with molecular weight of 2000 d that leads to failure of migration and incorporation from fibroblasts in the vascular surface of the implant<sup>4,5</sup>. It has been associated to the trans and postoperative use of



FIGURE 1. Above: Chest X ray after chest tube drainage showing a rounded image in the right superior vascular margin; below: persistence of the image at two years (darts)

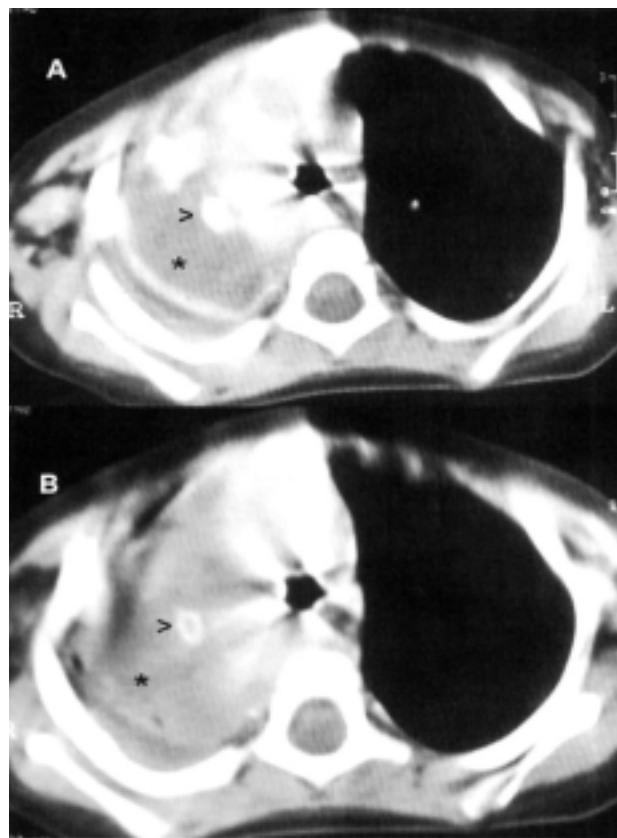


FIGURE 2. Thorax CT scan: the arrow shows the goretex graft; the asterisk points the serinoma.

heparin<sup>6</sup>, as well as hypofibrinogenemia and high levels of antitrombine II, protrombine 1 and 2 and products of fibrine degradation<sup>7</sup>, suggesting an abnormal reaction to the graft that affects the clotting and fibroblast migration.

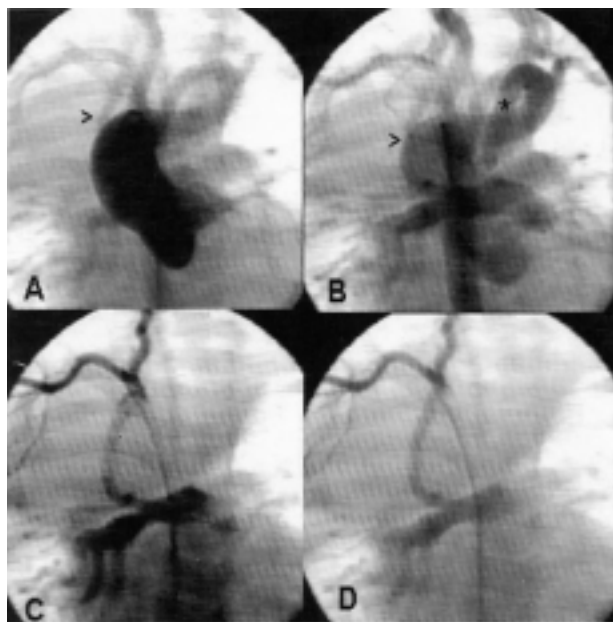


FIGURE 3. Aortic angiography (A and B) and selective injection in right subclavian artery (C and D) on postero-anterior projection: the arrow shows the patent goretex shunt surrounded by the serinoma; the asterisk shows the subclavian to pulmonary conduit

Its clinical presentation occurs between the 2nd and 12th postoperative week, with shortness of breath secondary to pleural effusion<sup>1, 2</sup>. The diagnosis can be done by ultrasonography, tomography or magnetic resonance<sup>3, 8</sup>. The initial management is conservative with chest tube drainage since the flight of the transuded can be self limited; when this doesn't happen it can be necessary to consider some of the surgical options that includes cover the implant with parietal pleura<sup>9</sup>, to instill fibrin products on it<sup>10</sup> or remove it and place another of different material. Success has been reported with the intravenous use of fibrinogen<sup>11</sup>.

The patient developed the serinoma in the 2nd to 3rd postoperative weeks and was managed in a conservative way; its suspicion was given on the initial radiological image and was confirmed by computed tomography and angiography.

At 2 years of follow up he remains with minimum symptoms for his heart disease, with a functional shunt and persistence of the radiological image<sup>12</sup>.

## REFERENCES

1. LeBlanc J, Albus R, Williams WG, Moes CA, Wilson G, Freedom RM, Trusler GA. Serous fluid leakage: a complication following the modified Blalock-Taussig shunt. *J Thorac Cardiovasc Surg* 1984;88:259-62
2. Sahoo M, Sahu M, Kale S, Saxena N. Serous fluid leakage following modified Blalock-Taussig operation using PTFE grafts. *Indian Heart J* 2001;53:328-31
3. Ozkutlu S, Ozbarlas N, Demircin M. Perigraft seroma diagnosed by echocardiography: a complication following Blalock-Taussig shunt. *Int J Cardiol* 1992;36:244-6
4. Ahn SS, Machleder HI, Gupta R, Moore WS. Perigraft seroma: clinical, histologic, and serologic correlates. *Am J Surg* 1987;154:173-8
5. Ahn SS, Williams DE, Thye DA, Cheng KQ, Lee DA. The isolation of a fibroblast growth inhibitor associated with perigraft seroma. *J Vasc Surg* 1994;20:202-8
6. 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;116:286-92
7. Salzer-Muhar U, Pabinger-Fasching I, Zacherl-Wightman S. Is there a possible role for haemostasis in the development of perigraft reaction complicating the modified Blalock Taussig shunt?. *Cardiol Young* 2000;10:261-4
8. van Rijn RR, Berger RM, Lequin MH, Robben SG. Development of a perigraft seroma around modified Blalock-Taussig shunts: imaging evaluation. *AJR Am J Roentgenol* 2002;178: 629-33
9. Ugurlu BS, Sariosmanoglu ON, Metin SK, Hazan E, Oto O. Ann Pleural flap for treating perigraft leak after a modified Blalock-Taussig shunt. *Thorac Surg* 2002;73:1638-40
10. Maitland A, Williams WG, Coles JG, Freedom RM, Trusler GA. A method of treating serous fluid leak from a polytetrafluoroethylene. *J Thorac Cardiovasc Surg* 1985;90:791-3
11. Hiramatsu Y, Atsumi N, Sasaki A, Mitsui T. A successful treatment of serous leakage from a polytetrafluoroethylene Blalock-Taussig shunt with intravenous fibrinogen administration. *J Thorac Cardiovasc Surg* 1999;117:1230-1
12. LeBlanc JG, Vince DJ, Taylor GP. Perigraft seroma: long-term complications. *J Thorac Cardiovasc Surg* 1986;92:451-4