

High-dose octreotide treatment for persistent pleural effusion after the extracardiac Fontan procedure

Ekstrakardiyak Fontan işleminde gelişen persistan pleural efüzyonun tedavisinde yüksek doz oktreotid

Ergin Koçyıldırım, Yavuz Yörükoğlu, Enver Ekici, Coşkun İkizler*

From Departments of Cardiovascular Surgery and *Pediatric Cardiology, Medical School, Ufuk University, Ankara, Turkey

Introduction

The Fontan procedure and its several modifications have been successfully used in palliation of functionally univentricular hearts. Although results of the procedures are excellent, morbidity is still a problem. Persistent pleural effusions are one of the serious and considerable causes of morbidity. At present, there is no specific cause or completely effective treatment. Postoperative medical treatment after Fontan procedure shows a wide variability between institutions. Cava et al. (1) indicated that a standardized therapy after the Fontan procedure plays an important role in minimizing the morbidity. Octreotide, an analogue of somatostatin has been reported to be effective in the treatment of chylothorax. de Leval et al. (2) suggested the use of octreotide in patients who had persistent pleural effusions after the Fontan procedure.

We report here a successful treatment of persistent pleural effusion after the extracardiac Fontan procedure with high dose octreotide.

Case report

A 10-year-old patient with tricuspid atresia, ventricular septal defect and severe pulmonary stenosis had underwent a left modified Blalock-Taussig shunt on the 7th day of life. Bidirectional cavo-pulmonary anastomosis with closure of the shunt was performed at age of 3 years. Completion of the total cavo-pulmonary connection was performed using an 18 mm extracardiac polytetrafluoroethylene (PTFE) (Gore-Tex®, W. L. Gore & Associates, Inc. Medical Products Division, Arizona, USA) graft with a 4 mm fenestration. Hemodynamic function was satisfactory. Postoperative echocardiogram showed good ventricular function with an unobstructed blood flow throughout the tube graft. The patient was extubated 5 hours postoperatively with a central venous pressure (CVP) of 10-11 mmHg. On the postoperative first day, the mediastinal drain was removed. However, right pleural serous drainage persisted and the average daily drainage was about 500 ml. The low serum protein level was replaced by human albumin. The patient was on enteral feedings enhanced by oral high protein formula (Protifar, Nutricia, the Netherlands). On postoperative day 6, intravenous octreotide at an initial dose of 3.5 µg/kg/hr was started. The amount of drainage did not change, subsequently the dose was increased to 7 µg/kg/hr. Over the next 5 days, the pleural drainage diminished to 200 ml daily. On day 12, the drainage peaked to 400

ml/day. The octreotide dosage was increased to 12 µg/kg/hr and over the next 7 days the pleural drainage decreased to 10 ml/day and the drain was removed on postoperative day 20. Patient was discharged to home on postoperative day 22. None of the main side effects such as nausea, vomiting, melena and abdominal pain was experienced. Chest X-ray showed no pleural effusion during a routine check two months after the patient's discharge.

Discussion

Somatostatin was first discovered as a hypothalamic hormone, which inhibits growth hormone (GH) secretion. Besides its GH-inhibiting activity, in both children and adults, somatostatin has been used to treat refractory diarrhea, enterocutaneous fistulas, and bleeding from esophageal varices. Octreotide is an octapeptide analogue of somatostatin with a longer half-life but can be used intravenously in the same doses as somatostatin. Octreotide has been used to treat persistent neonatal hyperinsulinism, the severe diarrhea and other symptoms that occur with certain intestinal tumors. In addition, chylothorax as one of the most serious complications of cardiothoracic surgery has been successfully treated with both somatostatin and octreotide. Recently, Goto et al. (3) described a chylothorax in a preterm infant with severe lung disease and pulmonary interstitial emphysema successfully treated with a very low dose somatostatin.

Our current standard octreotide treatment was based on the results of several recent studies, which had used a continuous infusion of octreotide with an initial dose of 3.5 µg/kg per hour with a doubling of this dose at 24 hours if there was no improvement in the patient's condition (4, 5). In our case the initial dose of 3.5 µg/kg per hour was increased to 7 µg/kg per hour and finally to 12 µg/kg per hour. Buettiker et al. (6) increased the dose to 12.5 µg/kg per hour in one patient but did not note any therapeutic advantage. No side effects of the therapy had been noted. Although liver function abnormalities, hypoglycemia and hyperglycemia, nausea, vomiting, melena and abdominal pain have been described, we did not observe any of them in our patient.

Fenestration is a modification of the extracardiac Fontan procedure, which allows right to left shunting and is closely related with low morbidity and mortality (7, 8). The possible reasons of persistent pleural effusions are absence of fenestration, high CVP, injury of thoracic duct, significant aortopulmonary collaterals. In our patient, none of these possible contributors of persistent pleural drainage was present.

Conclusion

Our case report shows a successful treatment of a post Fontan persistent thoracic drainage by the help of high dose octreotide. Although the reason of persistent pleural drainage was not chyle, high dose octreotide diminished the amount of pleural effusion. Further investigations are needed to understand the effect of octreotide in both chylothorax and post Fontan persistent thoracic drainage.

References

1. Cava JR, Bevandic SM, Steltzer MM, Tweddell JS. A medical strategy to reduce persistent chest tube drainage after the Fontan operation. *Am J Cardiol* 2005; 96: 130-3.
2. Walther T, Theune P, Sullivan I, de Leval MR. Successful medical treatment of persistent pleural drainage after the Fontan operation. *Interact CardioVasc Thorac Surg* 2003; 2: 348-49.
3. Goto M, Kitano M, Watanabe K, Watanabe K, Chiba Y. et al. Treatment of chylothorax in a premature infant using somatostatin. *J Perinatol* 2003; 23: 563-4.
4. Pratap U, Slavik Z, Ofoe VD, Onuzo O, Franklin RC. Octreotide to treat postoperative chylothorax after cardiac operations in children. *Ann Thorac Surg* 2001; 72: 1740-2.
5. Rosti L, Bini RM, Chessa M, Butera G, Drago M, Carminati M. The effectiveness of octreotide in the treatment of post-operative chylothorax. *Eur J Pediatr* 2002; 161: 149-50.
6. Buettiker V, Hug MI, Burger R, Baenziger O. Somatostatin: a new therapeutic option for the treatment of chylothorax. *Intens Care Med* 2001; 27: 1083-6.
7. Jacobs ML, Norwood WI Jr. Fontan operation: influence of modifications on morbidity and mortality. *Ann Thorac Surg* 1994; 58: 945-51.
8. Lemler MS, Scott WA, Leonard SR, Stromberg D, Ramaciotti C. Fenestration improves clinical outcome of the Fontan procedure: a prospective, randomized study. *Circulation* 2002; 105: 207-12.

Recanalization of occluded modified Blalock-Taussig shunt with balloon angioplasty and intraarterial streptokinase

Balon anjiyoplasti ve intraarteriyel streptokinaz ile rekanalize olan tromboze modifiye Blalock-Taussig şant olgusu

*Cemşit Karakurt, Gülendam Koçak, Ünsal Özgen**

From Departments of Pediatric Cardiology and *Pediatric Haematology, Faculty of Medicine, İnönü University, Malatya, Turkey

Introduction

Progressive stenosis and acute thrombosis, months to years after surgical creation, are the most known complications of the modified Blalock-Taussig shunt (1, 2). Children who developed acute Blalock-Taussig shunt occlusion usually require some form of intervention which includes thrombolytic therapy. Tissue plasminogen activator and streptokinase have been used successfully in some children (3, 4). Balloon angioplasty, stent implantation and surgery are the other therapeutic options (5-7).

We report an 11 months old patient with acute modified Blalock-Taussig shunt occlusion. The patient was treated successfully with balloon angioplasty and intraarterial streptokinase.

Case Report

An 11 months old girl with situs inversus totalis, double outlet right ventricle, malposition of great arteries, complete atrioventricular septal defect, pulmonary stenosis, hypoplasia of pulmonary arteries and right-sided modified Blalock-Taussig shunt was admitted to our hospital due to severe cyanosis and respiratory distress. Arterial blood gas parameters were consistent with hypoxia, and oxygen saturation was 35%. Detailed history revealed that she had undergone a right-sided

modified Blalock-Taussig shunt with 4 mm Gore-tex graft three months ago. She was discharged after one week of uneventful follow-up and oral anticoagulation was started after surgery with aspirin. Her condition had deteriorated suddenly, approximately 12 hours before admission.

On admission physical examination showed severe cyanosis and respiratory distress, but the lungs were clear on auscultation. There was no continuous murmur audible at the right sternal border. The typical high velocity, continuous flow profile of the shunt could not be identified by color Doppler echocardiography. Her oxygen saturation was 35%. She was taken up for urgent cardiac catheterization within 4 hours of hospitalization to confirm the diagnosis and perform transcatheter recanalization. Using a percutaneous left femoral artery approach, 50 U/kg heparin was given intravenously and 5F multipurpose catheter was advanced into the right subclavian artery. Right subclavian artery angiogram confirmed complete occlusion of right-sided modified Blalock-Taussig shunt (Fig. 1, 2). A 0.014 inch floppy guidewire was advanced to the left pulmonary artery via B-T shunt. The multipurpose catheter was exchanged over the guidewire with a 4X20 mm percutaneous transluminal coronary angioplasty (PTCA) catheter. After placement the PTCA catheter within the shunt, balloon angioplasty was performed twice (Fig. 3, 4). An irregular contour on the pulmonary side of the shunt was observed in control angiograms performed after balloon angioplasty (Fig. 5). Fibrinolytic treatment with streptokinase was