Case report - Pulmonary

Endovascular treatment of pulmonary sequestration in adults using Amplatzer® vascular plugs

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Abstract

Two adult patients were diagnosed with extralobar and intralobar pulmonary sequestration. One patient presented with haemoptysis. Both patients suffered from recurrent episodes of severe pulmonary infections. Both patients were treated by means of endovascular embolization using Amplatzer® vascular plugs (AVPs). They were discharged from hospital after 48 and 24 h and then followed up for 24 and six months, respectively. No recurrence of symptoms was observed. Computed tomography scans were obtained every six months. Persistent occlusion of vascular supply and moderate regression of the sequestered lung tissue are evident after 24 and six months in both patients. Just one case of an adult patient affected by pulmonary sequestration and treated by endovascular embolization has been reported to date. The present report is the first on the use of the AVPs in adults for this condition. The potential advantages and drawbacks of this treatment modality in adults are discussed, as well the specific benefit represented by the AVPs.

Keywords: Lung; Congenital lesions; Adult; Endovascular procedures; Device

1. Introduction

Pulmonary sequestration is a congenital malformation representing an area of non-functional pulmonary tissue, not communicating with the bronchial tree and supplied by one or more systemic arteries [1]. It is typically classified into intralobar and extralobar sequestration. According to Pryce, there are three types of pulmonary ‘sequestration’ (type I: the systemic artery supplies a segment of otherwise normal lung, without lung sequestration; type II: the systemic artery supplies the area of sequestered lung together with an area of adjacent normal parenchyma; type III: the systemic artery supplies only the sequestered lung tissue) [2].

Two adult patients were diagnosed with pulmonary extralobar and intralobar sequestration (Pryce type III and II, respectively). Both patients were treated successfully by means of embolization using Amplatzer® vascular plugs (AVPs) (AGA Medical Corporation, Golden Valley, MN, USA). Only one case of endovascular treatment of such malformation in adults has been reported to date [3]. To our knowledge, the present is the first report on the use of AVPs in adults for this purpose.

2. Patients and methods

2.1. Case 1

A 29-year-old male presented with haemoptysis (approx. 100 ml). He had a history of recurrent respiratory infections. Physical findings were unremarkable. Chest radiography showed a consolidation in the left lower lobe. Contrast-enhanced multi detector computed tomography angiography (MDCTA) showed an area of consolidation and cystic components (Fig. 1a). Two feeding arteries were seen arising from the aorta with venous drainage via the azygos vein system (Fig. 1b). Bronchoscopy showed a normal bronchial tree with traces of fresh blood arising from the left lower bronchus. The patient underwent digital subtraction angiography (DSA). An AVP 1 of 10 mm (20% of oversize) was advanced into the proximal portion of the largest artery. The device was then released after confirmation of its position. An AVP 1 of 8 mm (30% of oversize) was used for the second smaller artery (Fig. 1c).

2.2. Case 2

A 52-year-old male reported a history of recurrent respiratory infections and one episode of haemoptysis. Physical findings were unremarkable. A small consolidation in the left lower lobe was evident on a standard computed tomography (CT)-scan. MDCTA scan showed one artery arising from the thoraco-abdominal aorta and supplying the area of consolidated lung, with the venous drainage via the
Fig. 1. Case 1 (a) MDCTA, axial view. Area of consolidation and cystic components in the left lower lobe; (b) MDCTA, maximum intensity projection (MIP) reconstruction. Radiological findings consistent with extralobar sequestration. Arrows: feeding arteries. Asterisk: venous drainage; (c) DSA. Two AVP 1 (asterisks) deployed at the origin of the two feeding arteries; (d) final DSA confirmed the occlusion of the two vessels (arrows) within 7 min. MDCTA, multi detector computed tomography angiography; DSA digital subtraction angiography; AVP, Amplatzer® vascular plugs.

inferior pulmonary vein (Fig. 2a). Bronchoscopy showed a normal bronchial tree. DSA confirmed the presence of the feeding artery (Fig. 2b). An AVP 4 of 8 mm (30% of oversize) was used to occlude the proximal portion of the artery.

3. Results

Final aortograms were obtained at the end of the procedures (Fig. 1d). Patients were discharged from hospital after 48 and 24 h. Both patients are completely free of symptoms after 24 and six months. They both report a clear improvement of their quality of life. The first patient obtained a diving licence. Control CT scans were performed every six months (Fig. 2c and d).

4. Discussion

Embolization has been reported previously as a safe alternative to surgery in pediatric patients [1, 4, 5]. For adults, embolization has been reported exceptionally for the treatment of severe haemoptysis in Pryce type I sequestration [2, 6–8], while surgery remains the gold standard for treatment for Pryce type II and III sequestrations [9, 10].

We report the cases of two adult patients affected by typical pulmonary sequestration. Previously reported experience with embolization is limited to one case in this setting [3]. Major concerns include possible incomplete occlusion of vascular supply, subsequent evolution of the sequestered tissue and possible recurrence of symptoms. Both patients were eligible for traditional surgical treatment. Nonetheless, based on results with embolization in pediatric patients, we made the assumption that the same modality of treatment could be offered to adults. Both patients were informed that a less invasive method than surgery was available in order to minimize complications and to shorten the hospital stay and that surgical treatment should have been considered in case of complete or partial failure of the endovascular treatment.

Incomplete embolization and incomplete or null regression of the sequestered tissue are reported in 25–47% of pediatric cases [1, 3–5]. Incomplete embolization may result from distal occlusion of the feeding artery with possible subsequent opening of collateral supply. After incomplete embolization, recurrence of symptoms and infection may occur [1, 4]. Complications are also reported, including distant migration of embolization material causing embolization of non-targeted arteries [1, 4].

Metallic coils represent the preferred embolization material in most of the previously reported cases [1–3, 7, 8]. The AVP family (divided into four types of devices) is particularly suited for embolization of large, short and high-flow vessels where coil migration is possible or multiple coils may be needed [5]. AVPs seemed to us the optimal tool in order to achieve a complete proximal occlusion of the vessel minimizing the risk of regurgitation and distant embolization.

Size reduction and, sometimes, complete involution of the sequestration after embolization has been observed in infants and pediatric patients [1, 4, 5]. This process is far from being proved in adults. Only one case has been reported to date. The authors report the patient to be asymptomatic after one year but they do not mention if regression of the sequestration was observed [3].

Fig. 2. (a) Case 2. MDCTA MIP reconstruction. Systemic artery (arrow) supplying the sequestration (asterisk) and areas of normal lung (Pryce type II sequestration); (b) Case 2. Selective angiography of the systemic artery. Multiple collaterals are evident (asterisks). Proximal occlusion must be accomplished in order to obtain a complete embolization; (c) Case 1. MDCTA, axial view. The sequestered cystic area is moderately reduced in size after 24 months; (d) Case 1. MDCTA MIP reconstruction. Persistent occlusion of the feeding arteries (arrows) and reduction in size of the venous drainage (asterisk) after 24 months. MDCTA, multi detector computed tomography angiography; MIP, maximum intensity projection.
Our patients are totally asymptomatic since embolization was performed two years and six months previously, respectively, and their quality of life drastically improved. Nonetheless, the sequestered areas were just moderately reduced in size (Fig. 2d). A higher number of cases and longer follow-up is needed to answer the question if regression may occur and, above all, if this has clinical significance. In conclusion, pulmonary sequestration in adults may be successfully managed by means of percutaneous endovascular treatment, resulting in satisfactory long-term clinical results. AVPs should be used, in our opinion, in order to minimize complications. The endovascular procedure can be used in patients who are not suitable for surgery and/or refuse the operation. Finally, wider experience and lasting follow-up are needed to propose the embolization as an alternative to surgery in the treatment of pulmonary sequestration in adults.

References


