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CASE REPORT

Endovascular Treatment of an Aneurysmal Aberrant Systemic Artery Supplying a Pulmonary Sequestrum

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Abstract An aberrant systemic artery originating from the abdominal aorta supplying a pulmonary sequestration is a rare congenital malformation. This causes a left-to-left shunt. Symptoms include recurrent pneumonias, hemoptysis, and, in the long term, heart failure. Aneurysm of the aberrant vessel is rarely seen. Traditionally, treatment of pulmonary sequestrations includes ligation of the feeding vessel and lobectomy. A new promising treatment is an endovascular approach. Only a few cases describe endovascular treatment of pulmonary sequestration. This is the first published case of a giant aneurysmal branch from the abdominal aorta to the normal basal segments of the lung, successfully occluded with an Amplatzer Vascular Plug II (AVP II, St.Jude Medical, MN, USA) alone.

Keywords Pulmonary sequestration · Aberrant artery aneurysm · Endovascular · Amplatzer Vascular Plug II

The work was performed at Odense University Hospital, Denmark.

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Introduction

Pulmonary sequestration is an abnormality in which a bronchopulmonary segment has an anomalous systemic arterial blood supply, normal venous drainage, and non-functioning lung tissue that lacks communication with the normal tracheobronchial tree [1–8]. 50 % of the patients present themselves with chronic cough, expectoration, or recurrent pneumonias before the age of 20 [1]. Treatment includes lobectomy, segmentectomy, anastomosis between the aberrant artery and the pulmonary vein, and ligation of the anomalous artery [1, 3–6, 8]. A rare manifestation of sequestration is abnormal blood supply of the normal basal segments of the lung, a "pseudosequestration" [3–5].

An endovascular approach is a promising alternative to avoid postoperative complications [3, 5, 7]. Few available studies are published regarding successful endovascular closure of the aberrant artery with occluder and/or coils [5, 7, 8]. We describe a rare case of a giant aneurysm of an aberrant artery from the anterior wall of the abdominal aorta supplying a pulmonary sequestrum successfully treated with endovascular Amplatzer Vascular Plug II (AVP II, St.Jude Medical, MN, USA) alone.

Case Report

A 48-year-old man presented with intermittent back pain to the right of the thoracolumbar spine, primarily present with physical activity. The patient, a non-smoker, had a history of five clinically diagnosed pneumonias. He had never experienced haemoptysis. A chest X-ray showed a large mass in the right lower lobe. A contrast-enhanced computed tomography (CT) of the chest and abdomen revealed a giant aneurysmal aberrant branch from the anterior wall

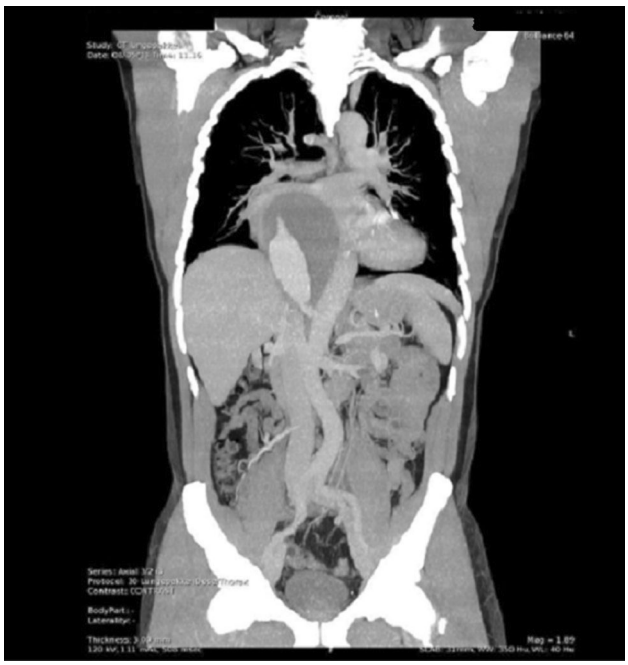

Fig. 1 CT showing the aneurysmatic aberrant branch with contrast-filled lumen and thrombus material supplying the pulmonary sequestrum

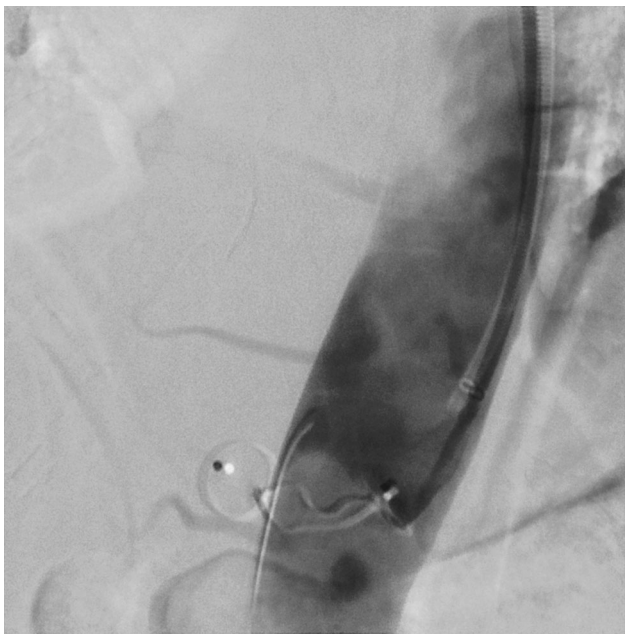

Fig. 2 Close-up view of the abdominal aorta of a well-placed Amplatzer Vascular Plug II in the aberrant artery in anterior–posterior projection. The aberrant artery is in the image anterior to the aorta

of the abdominal aorta proximal of the coeliac trunk to the basal segments of the right lung (Fig. 1). At the branching from aorta, the aberrant artery had a diameter of 1.0 cm distending 10.0 cm cranially and diminishing as it branched further. The total length of the aneurysmal sac measured 16.7 cm with partial mural thrombosis. There was normal pulmonary artery and venous branching in the right lung. A thoracic aortography via the left brachial artery confirmed the CT findings. Classic sequestral arteries were not visualised.

With thoracic and vascular surgeons in back-up, endovascular procedure was performed in local anaesthesia. A 12 French (F) sheath was installed in the common femoral artery in case an aortic occlusion balloon was needed for back-up. An 8 F sheath was installed in the left brachial artery, and from here, a 1.4 cm AVP II was directed into the aberrant aneurysmal artery via the descending aorta central to the aneurysm (Fig. 2). There were no complications during the procedure. An angiogram confirmed suitable device position in the aberrant artery with nothing protruding into the abdominal aorta. Postoperatively, the patient was well and was discharged the following day. Six hours after discharge, the patient returned with fever (39.6 °C) and onset of constant abdominal pain localised in the epigastrium. C-reactive protein was slightly elevated to 21 mmol/L (ref. <10 mmol/L), white blood count in a normal level. A CT scan of chest and abdomen showed an unchanged plug position with total occlusion of the sequestral artery. Aorta and the mesenteric arteries had normal contrast filling (Fig. 3). The symptoms were present for a few hours and successfully treated with analgetics.

After 3 months, a control CT scan of the chest and abdomen showed an unchanged plug position with total occlusion of the aberrant vessel. Clinically, the patient was well and no symptoms had been present since the day after the procedure. The patient experienced light pain in the right hemithorax during exercise. One year after, the patient was feeling well and had no complaints. There was unchanged plug position and the vessel was diminished by 3 cm. We plan to control the patient with CT once a year. Surgery is not scheduled as long as the patient's condition remains stable and potential pain is tolerable.

Discussion

Pulmonary sequestration is a rare congenital malformation [2]. The arterial supply is usually from the descending thoracic aorta (73 %). In 95 % of the cases, the venous drainage is to the left atrium via pulmonary veins which in time can cause a left-to-left shunt leading to ventricular enlargement and heart failure [8]. In our case, it was an aberrant branch from the abdominal aorta right above the coeliac trunk supplying the right lower pulmonary lobe.

Two types of sequestration are recognized, described by Pryce et al. [2]. Intralobar sequestration (75 %) is an

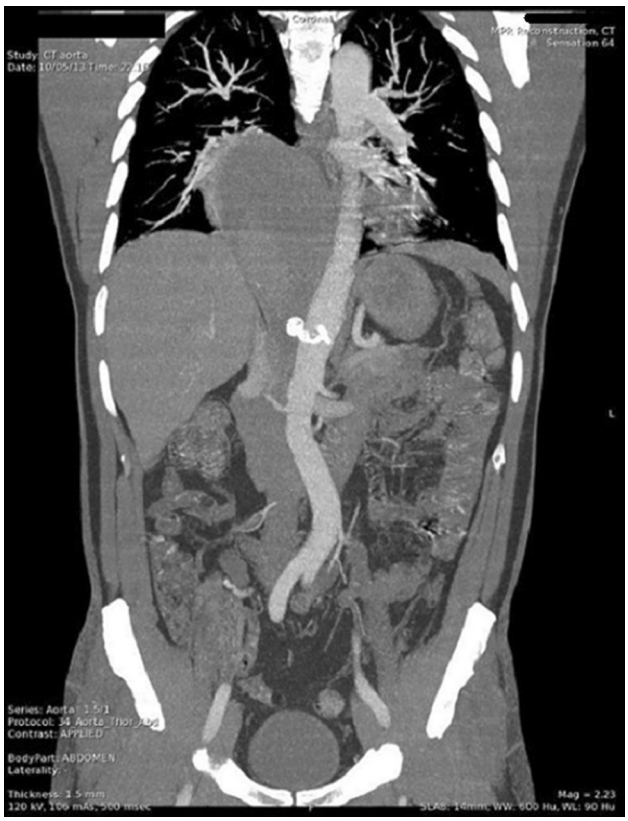

Fig. 3 CT showing the aneurysmatic aberrant branch after embolisation with Amplatzer Vascular Plug II. The aberrant artery is in the image anterior to the aorta

abnormal region within the normal pulmonary visceral pleura. Extralobar sequestration (25 %) is a true accessory pulmonary lobe enveloped in its own separate visceral pleura and can be placed extrathoracic. Pryce et al. first classified the intralobar sequestrations in three types; type one, abnormal artery supply without sequestration; type two, abnormal artery to sequestrated mass and to adjacent normal lung; and type three, abnormal artery confined to the sequestrated segment [2]. Our patient had an intralobar sequestration type one or pseudosequestration with normal bronchial branching, normal pulmonary artery branching in the right lung and normal venous drainage to the left atrium, and no classic sequestral vascularity.

Patients with intralobar sequestration are likely to present themselves with symptoms in early adulthood, but up to 15 % remain asymptomatic at the time of diagnosis [1]. Our patient presented with only one symptom, thoracolumbar back pain, which induced the chest X-ray that leads to his diagnosis.

Only a few cases have been reported of intralobar sequestration or pseudosequestration associated with an aneurysm of an aberrant artery [1, 3–7], and in only two cases, the aberrant artery had a diameter greater than the descending aorta [6, 7], as in our case.

Traditionally, the treatment of pulmonary sequestration is ligation of the feeding vessel and surgical resection of the sequestrated lung parenchyma or lobectomy due to the risk of recurrent infections. We have found one case of an aneurysmatic aortic branch supplying pulmonary sequestration where the aneurysm was embolised prior to surgery to avoid extensive bleeding [7].

We have found only one case of aneurysm of an anomalous systemic artery to the basal segments of the lung endovascular treated with AVP II and coils [5]. In that case, the aneurysm was embolised with coils prior to insertion of the AVP II to prevent retrograde filling of unexpected collaterals. The patient had postembolisation syndrome after the procedure, as did our patient. In our case, the postembolisation symptoms were present for a few hours, and in the following months, the patient had no complaints except light back pain related to exercise as seen prior to the procedure.

In our case, an AVP II was successfully chosen as an approach of permanent occlusion of the aberrant systemic artery. No coils were inserted and no future surgery is planned as long as the patient's condition remains asymptomatic and stable or with tolerable back pain. The advantage in our endovascular treatment of the patient is absence of surgery, and therefore it is less invasive. By surgery, we would have removed lung tissue and thereby reduced the lung volume. We used a plug instead of coils as it is our experience that plugs are more densely woven, and plugs are better when limited space for embolisation is available. This case confirms that endovascular treatment of an aneurysmatic aberrant branch from the aorta with an AVP II alone can be a simple and effective alternative to surgery.

Conflict of interest Katrine Lawaetz Kristensen, Louise Aarup Duus, and Bo Elle have no conflict of interest.

Statement of Informed Consent Informed consent was obtained from the patient included in this case report.

Statement of Human and Animal Rights Does not apply to this article.

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