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Case report

Bronchial to subclavian shunt in a CF patient. A potential pitfall for embolization

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Abstract

Bronchial artery embolization is a well accepted and widely used technique in the management of massive haemoptysis in cystic fibrosis (CF). It can be a complex procedure requiring a deep knowledge of the bronchial artery anatomy including the possible bronchial anastomoses. We report a case of complex vascular anatomy of the left bronchial artery with multiple anastomoses with the ipsilateral subclavian artery as cause of non-attempted embolization.

Keywords: Bronchial artery; Subclavian artery; Embolization

1. Introduction

Bronchial artery embolization (BAE) is an accepted and effective method of controlling massive haemoptysis in patients with cystic fibrosis (CF) when conservative treatment fails [1,2]. Usually the bleeding site arise from a bronchial artery, but many reports have also emphasized aberrant origin of the hemorrhage from non-bronchial collateral vessels or from anastomoses between bronchial and non-bronchial circulation [3,4]. These entire occurrences increase the technical difficulties of the procedure and the complication rate [4]. In this paper we describe a case of complex vascular anatomy with multiple anastomoses between the left bronchial artery and ipsilateral subclavian artery as cause of non-attempted embolization in an 18-year-old female CF patient with massive haemoptysis.

2. Case report

Eighteen-year-old female with CF diagnosed from the age of 8 weeks for rectal prolepses. The diagnosis was confirmed with sweat test (Gibson and Cooke method), chloride 101 mEq/l. The gene mutations were ΔF508/ΔF508. The patient developed pancreatic insufficient and liver cirrhosis from the age of 12. Sputum culture was positive for persistent Pseudomonas Aeruginosa infection from 1996 and Burkholderia Cepacia from 1999. FEV_1 maintained a constant value (80% of predicted) in the last 3 years since she received oral and aerosolized continued antibiotic treatment and every 3–4 months for 12–14 days, intravenous therapy with 2 different antibiotics. She was referred for minor haemoptysis from the age of 12 treated conservatively; at the age of 17 for massive haemoptysis and angiographic study was performed but no bronchial embolization was accomplished due to inconclusive findings. 9 months later, the patient showed frequently minor haemoptysis episodes, almost daily producing 30 ml of bloody sputum for over 2 weeks, treated conservatively with tranexanic acid and aggressive antimicrobial therapy. Every time she reported upper thoracic gurgle preceding each incoming episode. The haemoptysis rate increased after the first 2 weeks with an average of 70–80 ml, per day; so second bronchial angiography was per-

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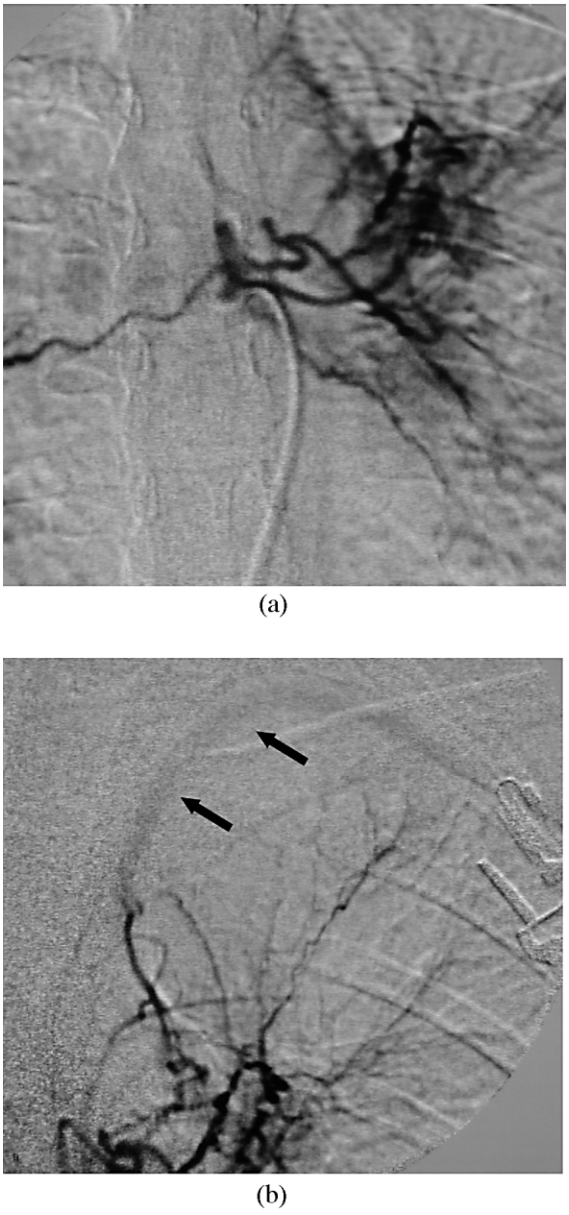

Fig. 1. (a–b) Selective catheterization of the bronchial trunk: (a) The early phase of the selective angiogram shows enlarged arteries and extensive collateral flow into the upper lung. (b) In the late phase a subclavian artery filling is visible (arrows) via collaterals vessels, probably an aberrant hypertrophied mediastinal artery arising from the middle aspect of the lung.

formed. The common bronchial trunk angiogram showed on the left side a markedly dilated and tortuous artery with multiple shunts and opacification of the ipsilateral subclavian and vertebral artery (Fig. 1a–b) sustained from some collateral arising from the middle aspect of the lung, probably an aberrant mediastinal artery. Despite the use of a coaxial catheter system, the embolization for the high flow going in the shunt, was considered a too risk procedure for non-targeted embolization and then non-attempted.

3. Discussion

It has been estimated that approximately 7% of adult CF patients may develop significant haemoptysis, which is usually a self-limiting episode. But a small number of patients may develop severe heamoptysis that require a more aggressive treatment to control massive bleeding [2]. BAE has proven to be an effective treatment to control massive haemoptysis [1].

Generally in most healthy individuals, the bronchial arteries arise from the proximal descending aorta at the level of T4–T6. However, anomalous origin of the bronchial arteries from the subclavian, internal mammary, brachiocephalic trunk has been described [3]. In addition to bronchial arteries, the bleed may originate from non-bronchial arteries via trans-pleural vessels [4].

In fact organizing pulmonary disease, associated with chronic infection leads to the hypertrophy of systemic vessels supplying the lungs and may open multiple shunts in the pulmonary and systemic circulation [4].

Usually intercostal arteries are commonly involved in chronic lung disease, but also the internal mammary, branches of the thyro-cervical and costo-cervical trunks, oesophageal, phrenic and left gastric arteries may be involved. In case of apical lung disease, it is possible to see numerous thoracic branches arising from the axillary and subclavian arteries supplying abnormal lung tissue, but these shunts were not previously reported in CF patients [4–7].

These extensive anastomotic network that exist between the bronchial arteries an other systemic arteries of the mediastinum can became hypertrophic in the region of interest and create enlarged collateral channels to the bronchial arteries or vice versa [5,6]. Shunts should be searched whenever an embolization procedure is attempted, because they increase the risk on non-targeted embolization as in our case or failure of the procedure [5–7].

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