Case report

Fistulae of the internal thoracic vessels: report of two cases

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Abstract

Arteriovenous fistulae involving the internal thoracic vessels are extremely rare. The multiple causes, variable clinical presentation, and inconsistent delay of onset of clinical symptoms in addition to their rarity make their diagnosis difficult. However, the complications of untreated internal thoracic fistulae are potentially fatal, emphasizing the need to make a prompt and correct diagnosis. Once the diagnosis is made and the investigational workup complete, treatment consists of either surgical ligation and excision or percutaneous transcatheter embolization. We report two cases of patients with internal thoracic fistulae: one arising as a complication of a tube thoracostomy and the other following an anterior thoracotomy. The second case report describes a rupture of the internal thoracic fistulae, a complication not reported earlier.

Keywords: Fistula; Internal thoracic vessels; Angiography; Ligation; Embolization

1. Introduction

Internal thoracic fistulae are extremely rare. A complete review of the literature was published by Senno et al. [1] which reported a case series of 19 patients. The causes of internal thoracic fistulae are multiple. However, the majority are either traumatic or iatrogenic in origin. Diagnosis is made difficult by their relative scarcity and their variable and often subclinical presentation. Internal thoracic fistulae can be associated with serious complications including congestive heart failure [1,2], bacterial endocarditis [3], rupture, and proximal arterial degeneration. This emphasizes the importance of an accurate and timely diagnosis. We present two case reports of patients with internal thoracic fistulae, one arising as a complication of a tube thoracostomy and the other as a late complication of an anterior thoracotomy.

2. Case 1

A 23 year-old male presented in 1985 with a large spontaneous pneumothorax. This was treated with a left-sided tube thoracostomy. One week later, there remained a persistent air leak. The patient was taken to the operating room and via an anterior thoracotomy, a left apical resection and pleurectomy was performed. During the post-operative course, the left lung did not completely re-expand because of a persisting pleural effusion. A left-sided thoracentesis was performed via a posterior approach following which the patient improved and was discharged home a total of 13 days after the operation. Over the next seven years, the patient noted increasing dyspnea and vague pain, tenderness, and easy bruising of the left anterior chest wall. Despite these complaints, he did not seek medical attention again until 1992. At this time, he was noted to have hyperesthesia of the left anterior chest wall and a loud, continuous ‘machinery-type’ murmur, maximal in intensity over the medial aspect of the left anterior thoracotomy scar. Palpation over the scar revealed an obvious thrill. A tentative diagnosis of an acquired chest wall arteriovenous fistula was made. Selective angiography confirmed a fistula between the left internal thoracic artery and the left internal thoracic vein (Fig. 1). Management proceeded with percutaneous transcatheter embolization of the left internal thoracic artery, which resulted in resolution of the murmur and angiographic resolution of the arteriovenous communication and accompanying collateral vessels.

Two months later, the symptoms of hyperesthesia and left anterior chest wall pain and tenderness recurred. Repeat angiography confirmed recurrence of the arteriovenous communication between the left internal thoracic vessels.
Three days after discharge (eight days after the injury), he developed a sudden increase in the severity of right-sided chest pain accompanied by increasing dyspnea. He presented to the regional hospital where clinical examination revealed decreased air entry to the entire right lung field. A new, loud and continuous machinery-type murmur was noted on auscultation, maximal in intensity precisely at the site of the previous tube thoracostomy. A chest X-ray revealed a new, large right pleural effusion. The effusion was drained via thoracentesis and was found to be sanguinous, thus confirming a hemothorax. A doppler echocardiogram showed no evidence of an intracardiac shunt or any valvular abnormality. The patient was transferred again to our institution for further evaluation.

A tentative diagnosis of a right internal thoracic fistula was made and then confirmed by selective angiogram with DSA imaging. After an unsuccessful attempt at percutaneous transcatheter embolization, the patient underwent a right posterolateral thoracotomy. A large hemothorax was evacuated and a pseudoaneurysm with a fistula of the right internal thoracic artery and vein was identified. The vein and the artery were individually clipped proximal and distal to the aneurysm. The patient’s symptoms and clinical findings subsequently resolved. Three years after surgery, the patient is well without any recurrence of a murmur.

4. Discussion

Fistulae of the internal thoracic vessels are extremely rare. Since the exhaustive review of literature by Senno et al. [1], only individual case reports have been published describing a wide range of clinical presentations and possible etiologies associated with internal thoracic fistulae. The cases presented herein involved injury to the internal thoracic vessels which occurred at the time of anterior thoracotomy and tube thoracostomy and resulted in eventual fistula formation.

One of the significant but rare complications of internal thoracic fistulae is congestive heart failure and is likely related to the duration and the size of the fistula. Other complications include bacterial endocarditis and proximal arterial degeneration. In our second case report, the initial presentation of a hemothorax was likely secondary to the rupture of the internal thoracic fistula. This complication is being reported in the literature for the first time.

Diagnosis of internal thoracic fistulae is suspected on the basis of a characteristic continuous, machinery-type murmur. Intra- or extra-cardiac pathology must be ruled out prior to making the final diagnosis. While computerized tomography (CT) scans and magnetic resonance imaging (MRI) scans may be used to help locate the sites of fistula formation, the gold standard for diagnosis of internal thoracic fistulae is selective angiography.

Once the diagnosis has been made, successful surgical ligation or percutaneous embolization [3–6] of the internal thoracic arteriovenous fistula results in resolution of clinical signs and symptoms. New treatment options may include minimally invasive attempts at surgical ligation either through video-assisted thoracoscopic surgery (VATS) or with the aid of robotic technology in the future.
While internal thoracic fistulae remain rare, their complications can be potentially fatal. Unlike many rare conditions, the workup and treatment of internal thoracic fistulae are both established and effective. By reporting these two cases, we hope that we have reminded clinicians to consider internal thoracic fistulae as part of their differential diagnosis when examining a patient with a machinery-type murmur and vague complaints of dyspnea.

References


