Totally endoscopic lobectomy and segmentectomy for congenital bronchial atresia

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Received 30 November 2008; received in revised form 24 February 2009; accepted 25 February 2009; Available online 15 April 2009

Abstract

Congenital bronchial atresia is a congenital obliteration of a segmental or lobar bronchus resulting in an inflation of the correspondent parenchyma. It may lead to infectious complications and in the long-term to alteration of the adjacent lung parenchyma. As it usually occurs in young and healthy patients with normal lungs, this disorder is particularly suitable for a full endoscopic pulmonary resection. We report our recent experience of two lobectomies and one segmentectomy in three patients.

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Keywords: Congenital bronchial atresia; Lobectomy; Segmentectomy; Thoracoscopy; VATS

1. Introduction

Bronchial atresia (BA) is a rare congenital obliteration of a segmental or lobar bronchus resulting in an inflation of the correspondent parenchyma. As it usually occurs in young and healthy patients with normal lungs, this disorder is particularly suitable for a full endoscopic pulmonary resection.

2. Case reports

2.1. Patient 1

A 13-year-old adolescent presented with chest pain, fever and cough. Chest roentgenograms revealed a left upper pneumonia. He was successfully treated by amoxicillin but recurred one year later. Chest computed tomography (CT) revealed a hypoperfusion of the lingula and a 2 cm diameter cystic process. Bronchoscopy was performed by his personal pulmonologist and was considered normal without obvious bronchial obliteration. The patient was operated on via a totally endoscopic approach under split ventilation, according to a recently described technique, using a 100% video display and pure endoscopic dissection without utility incision [1]. We first considered performing a lingulectomy but the remaining culmen was of poor quality and we found it safer to perform an upper lobectomy. The interlobar fissure was almost complete. All vessels were controlled using thermofusion (Ligasure™, Valleylab) and the lobar bronchus was stapled. Pathological examination showed a whole abnormal lobar tree with multiple irregular fibrotic extrinsic stenosis and a bronchiectasy of the lingular bronchus without connection to the bronchial tree. The patient had an uneventful postoperative course and was discharged on the 3rd postoperative day.

2.2. Patient 2

A 24-year-old woman had a severe pneumonia with sudden thoracic pain and high fever. Chest roentgenograms detected pneumonia of the right upper lobe. She was successfully treated by ceftriaxone and levofloxacin. Pectus excavatum was noticed on physical examination.

Assessment by chest roentgenograms showed radioluency in the right upper pulmonary field. Chest CT revealed infectious alveolar opacities of the posterior segment with a heterogeneous multicystic anterior segment while the middle lobe was squeezed (Fig. 1a).

At bronchoscopy, the anterior segmental bronchus of the right upper lobe was absent. Thoracoscopy showed a distended and emphysematous anterior segment of the upper lobe (Fig. 1b). The apical and posterior segments of the upper lobe were small and there was a clear separation between the apical and anterior segment, as seen on the CT.
performed a right upper lobectomy under totally endoscopic control. Pathological examination confirmed atresia of the anterior bronchus and lack of communication with the rest of the bronchial tree. Because of a prolonged air leak, the patient was discharged only on the 10th postoperative day.

2.3. Patient 3

An asymptomatic 25-year-old woman had a previous history of pulmonary embolism related to prothrombin gene mutation. The chest CT done at that time not only confirmed the pulmonary embolism but incidentally demonstrated hypoperfusion of the left basilar segments and excavated opacity of the malformative bronchus (Fig. 2a). At thoracoscopy, there was a clear difference between the normal superior segment and the basilar segments that looked emphysematous and contained an abscess (Fig. 2b). A basilar segmentectomy was performed (Fig. 2c and d). The presence of an unusual fissure between the superior and the basilar segments was helpful. Pathological examination confirmed atresia of the basilar segments with bronchial ectasia and dystrophic parenchyma. Patient left hospital on the 5th postoperative day after a simple postoperative course.

3. Discussion

Bronchial atresia is a rare congenital malformation resulting in an obliteration of a segmental or lobar bronchus, and a hyperinflation of the affected segment or lobe. This results in an emphysematous aspect of the parenchyma. The affected segment remains ventilated from collateral canals, causing hyperinflation by air trapping. Although BA most often occurs on the segmental level and rarely on the lobar level [2], most recent articles have reported lobectomy as the treatment of choice, since the adjacent parenchyma is usually compressed and affected by the consequences of mucus accumulation and air trapping [3,4]. In the series of 29 BA in children reported by Morikawa et al. only three patients underwent segmentectomy [5]. Some congenital abnormalities may be associated to BA, such as pectus excavatum, as noticed in one of our patient [2,6]. From a pathological standpoint, congenital BA is defined as a blind-ending bronchus associated with distal mucous-filled bronchocele surrounded by hyperinflated lung parenchyma. A bronchus connection is sometimes preserved but with an occluded lumen [2]. So normal bronchoscopy does not rule out BA but CT is sufficient to suggest it. Video-assisted (VATS) lobectomies in infants and children for various congenital anomalies have recently been reported. In children, a thoracoscopic approach not only results in decreased postoperative pain and better cosmetic result, but also reduces the consequences of thoracotomy, i.e. chest wall deformity and scoliosis [7]. VATS lobectomy for benign disease has also been reported in adults [8,9], but its conversion rate is high because of intraoperative difficulties related with fused and/or inflammatory fissures, or inflammatory lymph nodes [8]. Patients presenting with BA are young and have a healthy parenchyma. The marked limit between affected segment and normal parenchyma facilitates a thoracoscopic segmentectomy, usually a challenging operation because of the difficulty showing up the intersegmental plane [5]. However, in two of our three patients, the adjacent segments were so much compressed that lobectomy was more advisable. If BA is incidentally discovered like the third case, resection is justified by the fear of long-term consequences, i.e. compression and damage to the intact adjacent lung parenchyma [2]. Performing a segmentectomy or lobectomy with minimal cosmetic and functional input is appreciated in these young and healthy patients.

References