Case report

Systemic arterial supply to the left basal segment without the pulmonary artery: four consecutive cases

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Abstract

Systemic arterial supply from the descending thoracic aorta to the basal segment of the left lower lobe without a pulmonary artery supply is a rare congenital anomaly within the spectrum of pulmonary sequestration cases. We encountered four consecutive cases, which were treated successfully by three basalectomies and one lower lobectomy to preserve lung function.

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1. Introduction

Systemic arterial supply to the basal segment of the left lower lobe without a pulmonary artery supply is a rare congenital anomaly, requiring surgery because of hemoptysis caused by localized pulmonary hypertension. In this case report, the anomaly was encountered in four consecutive cases, and treatments consisted of three basal segmentectomies and one lower lobectomy to preserve lung function.

2. Preoperative diagnosis and clinical summary

All patients were referred to us with suspicion of pulmonary sequestration. We performed several careful examinations to confirm the existence and type of lesions. Computed tomography (CT) was used to scan for the presence of a sequestrated lobe and an abnormal blood supply. Examination with a bronchofiberscope or bronchovideoscope was used to detect abnormal branches of bronchial trees. Angiography for the pulmonary artery and the descending thoracic aorta depicted the distribution of the pulmonary artery, the pulmonary vein and the systemic artery around the descending thoracic aorta. These cases are summarized in Table 1 and Fig. 1A, B show representative preoperative angiographies corresponding to Case 1.

3. Operation and pathological findings

At thoracotomy, the basal segment of the left lower lobe with textiform capillaries on the surface was poorly developed, whereas the superior segment of the left lower lobe with a normal surface was well developed (Fig. 1C). We could successfully perform the basal segmentectomy at the mark of the capillary dilatations in the first three cases. It is not so difficult to distinguish the basal segment with textiform capillaries from the superior segment, which is without them, at the pulmonary vein of V6b and c. We found no major air leaks on the surface of the abscission. The anomalous systemic artery arose from the descending thoracic aorta and could be ligated by double ligation at the merge region of the aberrant artery (Fig. 1D). However, in the last case, the segmentectomy was not completed, but instead, a lower lobectomy was performed, since a drainage vein of the superior segment unexpectedly merged into a vein of the basal segment at the periphery.

Microscopic examination of the aberrant systemic artery disclosed an elastic artery with atherosclerosis, intimal thickening and with fibrosis and intra-alveolar hemorrhage in some part of parenchyma. The pulmonary arteries or their

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Table 1
Characteristics of patients with systemic arterial supply to the left basal segment instead of to the pulmonary artery

<table>
<thead>
<tr>
<th></th>
<th>Case 1</th>
<th>Case 2</th>
<th>Case 3</th>
<th>Case 4</th>
</tr>
</thead>
<tbody>
<tr>
<td>Sex</td>
<td>Female</td>
<td>Male</td>
<td>Female</td>
<td>Male</td>
</tr>
<tr>
<td>Age (years)</td>
<td>50</td>
<td>17</td>
<td>43</td>
<td>20</td>
</tr>
<tr>
<td>Gestation and delivery</td>
<td>Normal</td>
<td>Normal</td>
<td>Normal</td>
<td>Normal</td>
</tr>
<tr>
<td>Symptom</td>
<td>Bloody sputum in childhood</td>
<td>Hemothysis (150 ml)</td>
<td>Hemothysis (200 ml)</td>
<td>(−)</td>
</tr>
<tr>
<td>Accompanying disease</td>
<td>Nephrotic syndrome</td>
<td>(−)</td>
<td>Breast cancer</td>
<td>(−)</td>
</tr>
<tr>
<td>Heart murmur</td>
<td>(−)</td>
<td>(−)</td>
<td>(−)</td>
<td>Inspiratory phase on his back</td>
</tr>
<tr>
<td>Laterality of the lesion</td>
<td>Left</td>
<td>Left</td>
<td>Left</td>
<td>Left</td>
</tr>
<tr>
<td>Abnormal branching of bronchial trees</td>
<td>(−)</td>
<td>(−)</td>
<td>(−)</td>
<td>(−)</td>
</tr>
<tr>
<td>Sequestrated lung</td>
<td>A8, A9, A10^a</td>
<td>A8, A9</td>
<td>A8, A9, A10</td>
<td>A8, A9, A10</td>
</tr>
<tr>
<td>Defects of the pulmonary artery</td>
<td>10 mm diameter from DTA^b</td>
<td>10 mm diameter from DTA</td>
<td>8 mm diameter from DTA</td>
<td>8 mm diameter from DTA</td>
</tr>
<tr>
<td>Aberrant artery</td>
<td>LIPV^c</td>
<td>LIPV</td>
<td>LIPV</td>
<td>LIPV</td>
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<tr>
<td>Drainage vein</td>
<td>Basal segmentectomy</td>
<td>Basal segmentectomy</td>
<td>Basal segmentectomy</td>
<td>Lower lobectomy</td>
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<tr>
<td>Surgical procedure</td>
<td></td>
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</tbody>
</table>

^a Fig. 1C.
^b Fig. 1D; DTA, descending thoracic aorta.
^c LIPV, left inferior pulmonary vein.

Fig. 1. (A) A preoperative digital subtraction angiography for the left pulmonary artery is shown in lateral view. A4, A5 and A6 could be identified, although the pulmonary arteries for the basal segments were completely deleted. (B) A preoperative digital subtraction angiograph for the descending thoracic aorta is shown in posteroanterior view. The aberrant artery distributed into the basal segments, where the pulmonary artery for the basal segments should be spread. (C) The left lower lobe at thoracotomy. The basal segment (+) with textiform capillaries on its surface was poorly developed, whereas the superior segment (*) with a normal surface was well developed. The volume of the underdeveloped basal segment appeared almost the same as that of the superior segment of the left lower lobe. (D) The aberrant artery at thoracotomy. The aberrant artery is shown with a red tape, the inferior vein with a blue tape and the lower lobe bronchus with a yellow tape.
remnants could not be identified in the basal segment, which did not present any chronic inflammation, sequestration, consolidation, or dysplastic change in any of the resected specimens. The superior segment in the fourth case and the posterior basal segment in the second case presented normal lung parenchyma and vessels except for the existence of some larger vessels in the subpleural peripheral region.

4. Discussion

Regardless of pulmonary artery supply, the systemic arterial supply to the basal segment of the lower lobe without intralobar sequestration has been categorized as type I according to Pryce’s nomenclature [1]. However, in some rare cases, there is no pulmonary artery supply to the basal segment, as in the four cases presented here. We conclude that the four cases presented here are consistent with the systemic arterial supply to the basal segment of the lower lobe without intralobar sequestration. This conclusion was reached after examining preoperative findings of bronchoscopy, CT, angiography for the pulmonary artery and the aberrant artery and microscopic findings for the resected specimens. The common characteristics in our patients were as follows: the anomaly was on the left side and there was no sequestrated lung, no basal pulmonary artery and no abnormal bronchial branches. An aberrant systemic artery extended from the descending thoracic aorta to the pulmonary arterial defective segments.

The etiology of the systemic arterial supply to the basal segment of the left lower lobe without a pulmonary artery supply has been contentious [2,3]. A similar acquired lesion of ‘pseudosequestration’ [4] has been described as the combination of systemic arterial supply to the lung with normal bronchial connections, but with coexistent recurrent pulmonary infection. The systemic arterial supplies of the pseudosequestration are plural, due to hypervascularization of systemic arteries supplying the lung and chest wall. This condition results from recurrent pulmonary infection. Thus, we believe that the lesions in our patients must be congenital, because of their common characteristics and because there were no episodes of repetitive inflammation at the lesions.

Surgery has been adopted as the appropriate treatment for lesions of the left basal segment because of hemoptysis that follows pulmonary hypertension. Left lower lobectomies have been performed in most reported cases, rather than a basal segmentectomy and an anastomosis of the abnormal systemic artery to the pulmonary artery [2,5]. We believe that a basal segmentectomy is more appropriate than a lower lobectomy to remove the lesion, in order to preserve postoperative lung function, because the volume of the underdeveloped basal segment appeared almost the same as that of the superior segment of the left lower lobe. Three patients under our care have followed a completely successful clinical course, as indicated by postoperative CT and blood gas examination. Yamanaka et al. reported that the anastomosis procedure would likely be ineffective to preserve normal circulation of the lung via a perfusion scanning method [2]. Moreover, it is unclear whether the aberrant systemic artery could exchange blood gas in the alveolar-capillary region, because of thickening and sclerosing of the arterial wall following the application of systemic arterial pressure.

The systemic arterial supply to the basal segment without a pulmonary artery supply, which presented common features, as mentioned earlier, belongs within the spectrum of pulmonary sequestration cases [6]. We should be keen to select basal segmentectomy to the atrophic basal region as the best treatment to preserve lung function, instead of a lower lobectomy or anastomosis of the abnormal systemic artery to the pulmonary artery.

References