Abstract

OBJECTIVES: The B^3 downwards-shifting malformation was first proposed by Boyden in 1950. Here, we report 14 cases of this malformation in the right lung and the first case of segmentectomy for this malformation.

METHODS: All patients with pulmonary lesions underwent three-dimensional computed tomography bronchography and angiography (3D-CTBA) between January 2019 and January 2020, prior to surgery. A consecutive 2356 patients were included, and 14 cases of this malformation were identified; bronchovascular patterns were analysed in each patient.

RESULTS: The incidence of this malformation was 0.6%. It was further divided into 3 types: over downwards-shift, partial downwards-shift and normal downwards-shift. The normal downwards-shift type was the most common (8/14), where B^3 shifted downwards completely to merge with B^4+5. In the partial downwards-shift (5/14), only part of the B^3 shifted. In the over downwards-shift type (1/14), both B^3 and B^1b shifted downwards. A bifurcated right upper lobe (RUL) bronchus (B^1 defective) was observed in 3 cases. The incidence of V^1a, V^1b, V^2a, V^2b, V^2c, V^3a, V^3b and V^3c was 100% (14/14). The incidence rates of A^1, A^3a and A^3b were 100% (14/14). The incidence of A^2 rec and A^2 asc was 92.9% (13/14) and 71.4% (10/14), respectively.

Take-home message

Boyden’s triad is a rare anomaly. Exploration of these malformations is important for surgery.

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CONCLUSIONS: The B³ downwards-shifting malformation or 'Boyden's triad' is a rare anomaly. Anatomical exploration of this malformation is important for surgery.

Keywords: Boyden • B3 downward-shifting • Segmentectomy

ABBREVIATIONS

| Abbreviation   | Description                                      |
|----------------|--------------------------------------------------|
| 3D-CTBA        | Three-dimensional computed tomography bronchography and angiography |
| CT             | Computed tomography                              |
| CV             | Central vein                                     |
| RML            | Right middle lobe                                |

INTRODUCTION

The right middle lobe (RML) has the smallest volume of all lung lobes. Therefore, the analysis of its anatomy is limited. Looking back into history, the anatomical analysis of RML can be generally divided into 3 phases. In the 1940s, attention was focused on the vulnerability of the RML bronchus and the possibility of its compression by surrounding lymph nodes. For example, in 1943, Brock identified enlarged nodes and reviewed 60 cases in which obstruction and infection of the middle lobe were attributed to tuberculous lymphadenitis [1]. It is extremely important to know the variations in the anatomy of the RML in order to treat them surgically. In the 1950s and 1960s, the second phase started when segmentectomy became recognized by surgeons. The quest for knowledge on segmentectomy drove surgeons into researching the anatomy of all lung lobes, including the RML [2–4]. However, as the smallest lobe, the benefits of sublobe resection were considered marginal, and studies on the segmental anatomy of the RML have rarely been reported [5]. The interest in research waned because compared with lobectomy, segmentectomy was no better in several aspects, including survival. In the 2000s and 2010s, high-resolution computed tomography (CT) was commonly utilized for healthy examinations; more ground-glass opacities were identified. For the ground-glass opacities, lobectomy means sacrifice of more lung volume and less protection of lung function. These disadvantages fuelled the increasing interest in the thoracoscopic approach to anatomic segmentectomy. Furthermore, advances in CT and volume-rendering reconstruction techniques have allowed for the reconstruction of three-dimensional CT bronchography angiography (3D-CTBA) as a powerful tool for thoracic surgeons to analyse pulmonary anatomy on computer, instead of on cadavers. In 2017, Nagashima et al. [6] first analysed the pulmonary bronchovascular patterns of the RML using 3D-CTBA. Yajima et al. [7] reported the first case series (2 patients) of thoracoscopic RML segmentectomy in 2018.

The anatomy of the RML, although considered simple, can also be accompanied by anomalies, some of which are rare. Boyden reported a gross anomaly involving the right upper and middle lobes, in which the middle lobe bronchus, although correctly placed, gave rise to the anterior segment of the upper lobe bronchus as well as to its own segments [5]. This anomaly was cited in articles about pulmonary anatomy and segmentectomy in the years that followed; however, no one reported a second case.

Here, we report 14 cases of this anomaly with analysis of the bronchovascular variations using 3D-CTBA, providing a more

Figure 1: The partial downwards-shift type of Boyden’s triad. (A) The classic pattern of RUL and right middle lobe bronchus. (B) The partial downward-shift: B³a and B³b shift. (C) The partial downward-shift: B³a shift. (D) The partial downward-shift: B³a and B³bii shift.
Figure 2: The normal downwards-shift type of Boyden's triad. (A) The classic pattern of RUL and right middle lobe bronchus. (B) The normal downward-shift: B3 shift.

Figure 3: The over downwards-shift type of Boyden's triad. (A) The classic pattern of RUL and right middle lobe bronchus. (B) The over downward-shift: B1b and B3 shift.

Figure 4: The branching of veins in Boyden's triad: anterior with central type. (A) Anterior with central type in Boyden's triad. (B) Anterior with central type proposed by Dr. Shimizu [6].
accurate information on this rare anomaly and a comprehensive preoperative evaluation.

**METHODS**

**Patient characters**

The Ethics Committee of the First Affiliated Hospital of Chongqing Medical University approved this study (No. 2020-206), and patients' consent was waived. Between January 2019 and January 2020, all patients with pulmonary lesions underwent 3D-CTBA preoperatively. A consecutive 2356 patients were included, and 14 cases of malformation were identified. The incidence was 0.6%. CT pulmonary angiogram and CTPV by SOMATOM Definition Flash dual-source CT were performed for each patient. 3D-CTBA reconstruction was analysed with the Mimics software (Belgium).

**Variations in bronchovascular pattern and parenchymal division**

The Boyden's triad includes anomalies in 3 aspects: the bronchial anatomy, the vascular anatomy and the parenchymal division. Boyden's triad was therefore defined as: (i) the RML bronchus gave rise to the anterior segment of the RUL bronchus as well as
to its own middle lobe segments, and the RUL bronchus itself has only 2 branches. We named this ‘B3 downwards-shifting’; (ii) the RML artery had at least 3 branches; (iii) the upper and middle lobes are completely fused into ‘one lobe’, with no horizontal fissure. Because downwards-shifting of B3 plays a key role in this anomaly, we define this anomaly as ‘B3 downwards-shifting malformation’ or ‘Boyden’s triad’, in memory of the first author who named it.

RESULTS

The mean age of the 14 patients (9 women and 5 men) was 56 years. This malformation was far more complex than we expect, and it was further divided into 3 types: partial (Fig. 1), normal (Fig. 2) and over (Fig. 3) downwards-shift types. In the partial downwards-shift type (5/14), only a part of B3 shifted (the other part was still located in the RUL). Among the 5 patients with this type, 2 had the B3ai and B3b shifts, 2 had only the B3a shift and 1 had the B3a and B3bii shifts (Fig. 1). The normal downwards-shift type was the most common (8/14), where B3 (both B3a and B3b) completely shifted downwards to merge with B4+5 (Fig. 2). In the over downwards-shift type (1/14), B3 and B3b shifted downwards, leaving only B3a and B3b in RUL (Fig. 3).

The veins were more complex. In the classic nomenclatures proposed by Shimizu in 2015, there are 2 important concepts: the central vein (CV) and the anterior vein. The CV is defined originating from V2a and descends between B2 and B3, finally draining into the superior pulmonary vein from the mediastinal side. The anterior vein is defined originating from V1b and descends down the anterior side of the upper lobe bronchus, finally draining into the superior pulmonary vein from the mediastinal side [8]. Of the ‘anterior’ and ‘central’ RUL veins, all could be classified into 4 types: 2 anterior with central types (Iab and Ib), 1 anterior type and 1 central type. In Boyden’s triad, however, since B3 has shifted downwards, it is impossible to redefine the vein ‘between B2 and B3’ as CV. The traditional branching of the RUL veins (anterior with central, anterior and central types) was therefore not applicable. We modified the concept of ‘central vein’ in the Boyden’s triad and proposed a new simplified model, in which the CV is defined as originating from V2a and descending between B2 and B3. The anterior vein is still defined as originating from V1b as proposed by Shimizu. The branching of veins in Boyden’s triad is thus classified into the anterior with central (Fig. 4) and anterior (Fig. 5) types; the central type has not yet been identified. Details of the vessels are listed in Supplementary Material, Table S1. The incidence of V1a, V1b, V2a, V2b, V2c, V3a, V3b and V3c was 100% (14/14). In 4 patients, there were V1a and V1a. The incidence of V1l, V1t and upper lobe vein posterior to the bronchus intermedius (UVPBI) [9] was 57.1% (9/14), 50% (7/14) and 7.1% (1/14), respectively. There were 2 cases in which V3c drained into V3b (Supplementary Material, Table S1).

For the artery, the incidence of A1 was 100% (14/14), while that of A2 rec and A2 asc was 92.9% (13/14) and 71.4% (10/14), respectively. The incidence of A3a and A3b was 100% (14/14). The details of the artery patterns are listed in Supplementary Material, Table S1.
We recently encountered a patient with a 12 mm nodule in the RUL. He had undergone surgery for colon cancer 3 years ago. To exclude the possibility of metastasis, surgery was performed. The preoperative 3D-CTBA revealed this as a case of Boyden’s triad. ‘Lobectomy’ was obviously not suitable for this patient because in the real sense, it meant sacrificing ‘two lobes’. A wedge resection was also not suitable because the nodule was not located under the pleura. Therefore, we decided to perform segmentectomy. However, segmentectomy for patients with Boyden’s triad has never been reported; therefore, preoperative planning was challenging. Finally, we performed an RS4a subsegmentectomy (Fig. 6). Pathological examination revealed metastasis. The operative time was 65 min and blood loss was 86 ml. The chest tube duration was 2 days. The length of hospital stay was 2 days. There are no postoperative complications.

DISCUSSION

The $B^3$ downwards-shifting malformation was first reported by Boyden in 1950 [5]. In the following 70 years, no other case was reported. The decision to revisit this ‘old’ topic was not sudden. Since high-resolution CT is becoming a common tool for screening patients with lung cancer at a very early stage, segmentectomy is replacing the lobectomy in the surgical treatment of this disease [10]. However, both vascular and bronchial structures vary greatly at segmental level. Because of the anatomical complexity, the segmentectomy is technically more difficult than lobectomy. A thorough preoperative analysis of anatomic features is important for precise surgery. It is difficult to identify all the details from the conventional two-dimensional CT images, especially for those with anatomic anomalies. Fortunately, the 3D-CTBA imaging is useful in assessing bronchovascular anatomy prior to surgery, allowing thoracic surgeons to determine pulmonary anatomy both before and during surgical procedures. The 3D-CTBA is also a powerful tool to analyse and summarise the pulmonary anatomy variation on computer, instead of on cadavers. The wide application of 3D-CTBA in recent years could explain the rapid growth of cases with Boyden’s triad.

The downwards-shift of $B^3_a$ ‘fused’ the margins of the upper and middle lobes, turning the right side into a ‘mirror image’ on the left. This could explain the disappearance of the horizontal fissure, which did not exist in the left lung. It is important to note that this is ‘totally no horizontal fissure’, instead of ‘incomplete horizontal fissure’, which is common in the right lung. Accompanying the downwardly displaced $B^3$, $A^3$ arises from the interlobe artery and moves downwards alongside $B^3$ into the middle lobe. Thus, the RML has at least 3 arteries, including its own.

It is interesting to note that the extent of the $B^3$ downwards-shift varies greatly. Three types were identified as follows: over, partial and normal downwards-shift. However, this has never been reported in literature. The rarest type was the over downwards-shift (Fig. 3). In this type, $B^3$ and $B^1_b$ had shifted. This means a greater ‘mix’ of RUL and RML. For this kind of complex variation, preoperative planning for segmentectomy is more complex and requires more attention. The bifurcated RUL bronchus ($B^1$, defective), which is another anomaly first proposed in 2018 [11], was found in 3 of the 14 patients (Fig. 7).

In 2 cases, we find that the $V^3_c$ drains into $V^1_b$ (Fig. 8). It is theoretically explainable. The $V^3_c$, as the intrasegmental vein of $S^3_b$, is responsible for the drainage of $S^3_b$. In Boyden’s triad, as the $B^3(S^3)$ shifted, the $V^3_c$ always shifted downwardly with the bronchus, draining into the downwardly shifted $V^3_b$. However, in the partial downwards-shift type, there are 2 cases with only $B^3_a$ shifted while the $B^3_b$ is still in original place; accordingly, the $V^3_c$ did not shift downwards but drained into $V^1_b$, leaving $V^1_b$ downwards shifted alone (Supplementary Material, Table S1).

Boydens triad is a rare anomaly. It is far more complex than it seems, and further studies are required to investigate bronchovascular variation in the future.

SUPPLEMENTARY MATERIAL

Supplementary material is available at ICVTS online.

Funding

None.

Conflict of interest: none.
Data Availability Statement

All relevant data are within the manuscript and supplementary files.

Author contributions

Min Zhang: Data curation. Ning Mao: Investigation. Qingchen Wu: Investigation. Hongtao Tie: Data curation. Mingjian Ge: Conceptualization.

Reviewer information

Interactive CardioVascular and Thoracic Surgery thanks Alessia Stanzi, Toyofumi F. Chen-Yoshikawa and the other anonymous reviewers for their contribution to the peer review process of this article.

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