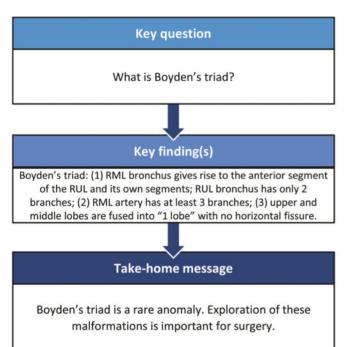
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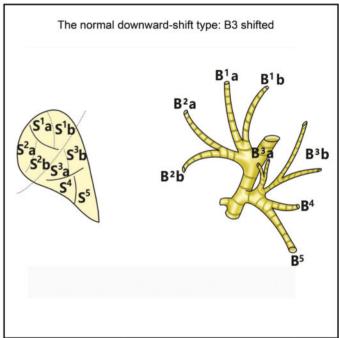
Boyden's triad: the past, present and future

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Abstract

OBJECTIVES: The B³ 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^1 b shifted downwards. A bifurcated right upper lobe (RUL) bronchus (B^1 defective) was observed in 3 cases. The incidence of V^1 a, V^1 b, V^2 a, V^2 b, V^2 c, V^3 a, V^3 b and V^3 c was 100% (14/14). The incidence rates of V^3 0, V^3 1, V^3 2 as was 92.9% (13/14) and 71.4% (10/14), respectively.

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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

ABBREVIAITONS

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, highresolution 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

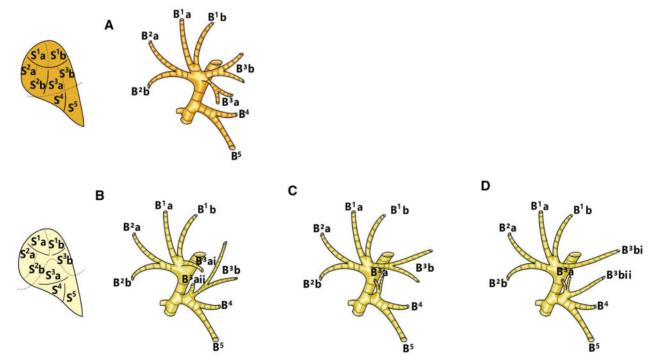


Figure 1: The partial downwards-shift type of Boyden's triad. (A) The classic pattern of RUL and right middle lobe bronchus. (B) The partial downward-shift: B³aii and B³b shift. (C) The partial downward-shift: B³a shift. (D) The partial downward-shift: B³a and B³bii shift.

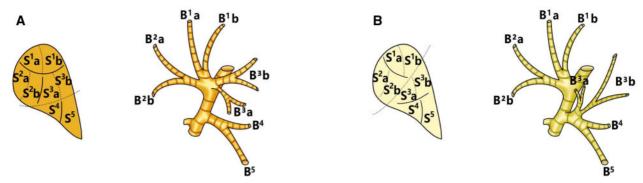


Figure 2: The normal downwards-shift type of Boyden's triad. (A) The classic pattern of RUL and right middle lobe bronchus. (B) The normal downward-shift: B³ shift.

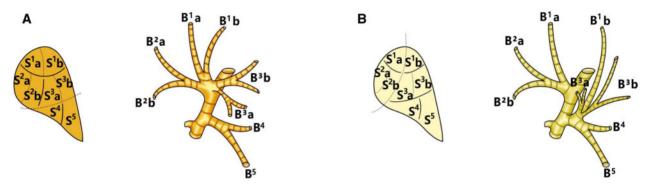


Figure 3: The over downwards-shift type of Boyden's triad. (A) The classic pattern of RUL and right middle lobe bronchus. (B) The over downward-shift: B¹b and B³ shift.

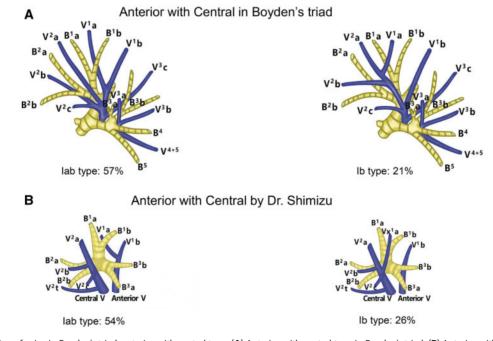


Figure 4: The branching of veins in Boyden's triad: anterior with central type. (A) Anterior with central type in Boyden's triad. (B) Anterior with central type proposed by Dr Shimizu [6].

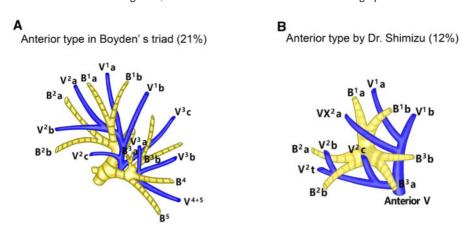


Figure 5: The branching of veins in Boyden's triad: anterior type. (A) Anterior type in Boyden's triad. (B) Anterior type proposed by Dr Shimizu [6].

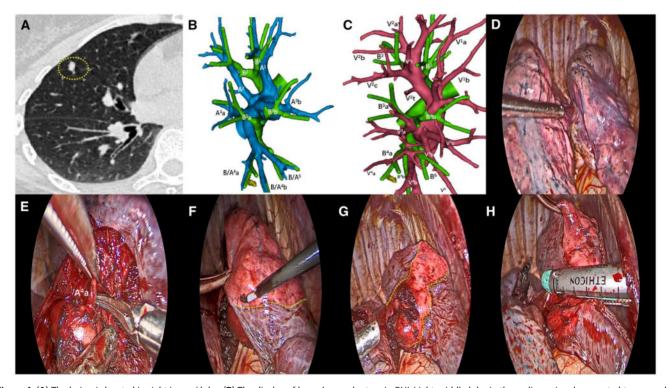


Figure 6: (A) The lesion is located in right 'upper' lobe. (B) The display of bronchus and artery in RUL/right middle lobe in three-dimensional computed tomography bronchography and angiography. (C) The display of bronchus and vein in RUL/right middle lobe in three-dimensional computed tomography bronchography and angiography. (D) The RUL and right middle lobe are completely fused into 'one lobe', there is no horizontal fissure. (E) The A⁴a is dissected and resected. We ventilate the lung with 100% oxygen. About 10 min later, the intersegmental plane is visualized. The inflation-deflation method was used for the visualization of intersegmental plane. (F) The intersegmental plane is marked by electrocautery from interlobar surface. (H) The resection of RS⁴a by stapler.

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

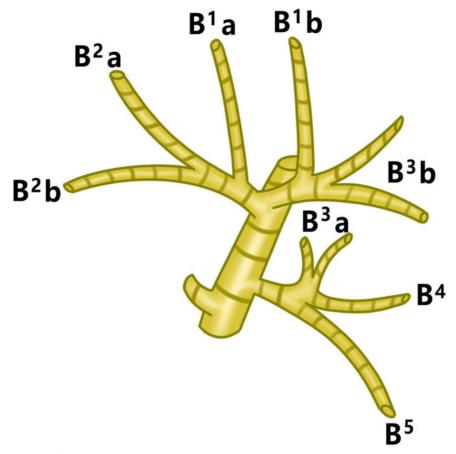


Figure 7: The bifurcated RUL bronchus (B¹ defective) in Boyden's triad.

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 B³aii 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 B¹b shifted downwards, leaving only B¹a and B² 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 V^2 a and descends between B^2 and B^3 , 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 (lab 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 B¹ and B². 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 $V^{1}a'$. The incidence of $V^{1}I$, $V^{2}t$ 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 V¹b (Supplementary Material, Table S1).

For the artery, the incidence of A^1 was 100% (14/14), while that of A^2 rec and A^2 asc was 92.9% (13/14) and 71.4% (10/14), respectively. The incidence of A^3 and A^3 b was 100% (14/14). The details of the artery patterns are listed in Supplementary Material, Table S1.

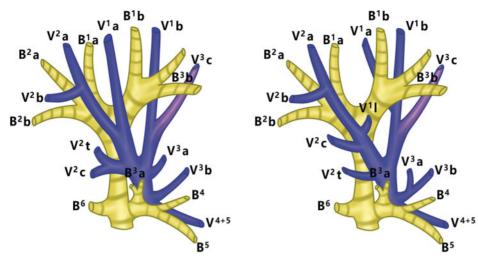


Figure 8: V³c drains into V¹b in Boyden's triad (V³c is highlighted in different colour).

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 B³ 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 summarize 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 B³a '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 B³, A³ arises from the interlobe artery and moves downwards alongside B³ 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 B³ 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, B³ and B¹b 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 (B¹ 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^3c drains into V^1b (Fig. 8). It is theoretically explainable. The V^3c , as the intrasegmental vein of S^3b , is responsible for the drainage of S^3b . In Boyden's triad, as the $B^3(S^3)$ shifted, the V^3c always shifted downwardly with the bronchus, draining into the downwardly shifted V^3b . However, in the partial downwards-shift type, there are 2 cases with only B^3a shifted while the B^3b is still in original place; accordingly, the V^3c did not shifted downwards but drained into V^1b , leaving V^3b downwards shifted alone (Supplementary Material, Table S1).

Boyden's 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

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