

# The rare anatomical variant of upper lobe: clinical features of 46 cases with surgical procedure

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## Research

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# Abstract

## Background

Primary disease in thorax associated with an azygos lobe is extremely rare. It is usually identified incidentally on chest X-ray or CT during health checkups with an incidence of up to 0.2%. This is the first study involving 46 of patients found with azygos lobe in surgery of English literature from January 1931 to October 2018.

## Methods

PubMed, EMBASE and the Web of Science databases were searched for full-text literatures met out inclusion criteria. We summarized the clinical data, radiological manifestation, accompanying disease and treatment strategy of all patients.

## Results

18 eligible studies involving 46 patients were selected for this research. The mean age was 36.5 years old. There were 26 male patients and 20 female patients and the male to female ratio nearly to 1.3:1. There were many different primary diseases with azygos lobe including lung cancer (n = 8), spontaneous pneumothorax (n = 5), esophageal cancer (n = 1), pulmonary sequestration (n = 1), esophageal atresia (n = 2), hyperhidrosis (n = 29). The azygos lobe (azygos lobe in Figs. 1 and 2) is an uncommon anomaly that is found in 1% of anatomic specimens, on about 0.4% of chest radiographs and 1.2% of high resolution CT. The azygos lobe is a developmental anomaly but not a true accessory lobe. Azygos lobe of all patients was diagnosed during the operation.

## Conclusions

Azygos lobe occurs in 0.2% of the population and can make clinical diagnosis difficult. The detection of this anomaly and clarification of its precise anatomical features are important to alert the surgeon to potential problems during surgery.

## Introduction

An azygos lobe is a rare congenital variant. It is recognized incidentally on chest X-ray or CT for the duration of healthcheckups. Wrisberg firstly described the azygos lobe in 1877 that is a variant of pulmonary anatomy and presenting in 0.1-1% of the population[1]. The different from other accessory lobes of anomalies of the lung is that the abnormal azygos vein crosses the apex of the lung[2]. The azygos lobe has been recognized in both the right and left lungs from the studies [3, 4].

According to the review of the literatures from the universal electronic databases, there are only some cases reports of azygos lobe with primary thorax disease have been reported. This is the first study containing all the series of patients with azygos lobe diagnosed in surgery in English literature. The purpose of this study was to study clinical data, radiological manifestation, and treatment strategy of patients.

## Methods

### Data sources

Three universal electronic databases, PubMed, EMBASE and Web of Science, were selected to identify the full-text English literature published from 1 January 1931 to 31 October 2018. Final search criteria included the following keywords that were "azygos lobe" and "surgery". Furthermore, we also manually explored the reference lists of relevant papers to detect any one included study with no duplication. The summary of the findings is listed in Table.

### Inclusion and exclusion criteria

#### Inclusion criteria

(i) azygos lobe were not only found in X-rays or CT, but also diagnosed in surgery; (ii) azygos lobe was independently investigated in original literature; (iii) manuscripts were accessible in the full-text literature; and (iv) only English language manuscripts were considered for the study.

#### Exclusion criteria

(i) Letters and conference abstracts were excluded; (ii) manuscripts without full-text were excluded; (iii) manuscripts in non-English languages were not accepted; and (iv) patient without surgery in report of the literature.

### Statistical analysis

The statistical analysis was performed using IBM SPSS Statistics, version 16.0 (IBM Corporation, Armonk, NY, USA).

## Results

In our Table, there were 26 male patients and 20 female patients and the male to female ratio nearly to 1.3:1. The mean age was 36.5 years old. All the azygos lobes were located in the right upper lobes. The presenting symptoms were dyspnea, excessive sweating, head injury, murmur in the mesocardiac area, hemoptysis, hoarseness, vomiting according to the Table. There were lots of different primary diseases with azygos lobe including lung cancer ( $n = 8$ ), spontaneous pneumothorax ( $n = 5$ ), esophageal cancer ( $n = 1$ ), pulmonary sequestration ( $n = 1$ ), esophageal atresia ( $n = 2$ ) and hyperhidrosis ( $n = 29$ ). Most of them

were treated with VATS. One case was operated by Robot-assisted azygos lobectomy for adenocarcinoma (Case 3). The patients with esophageal diseases were treated with thoracotomy (Case 15, 19 and 20).

## Discussion

## Epidemiology

An azygos lobe is a well-known normal variant of the lung that was described by Wisberg[4]. The literature has pronounced its incidence from 0.4% on chest radiographs to 1.2% on chest CT[5–7]. It arises at any age, which varied from 0.9 to 76 years (Table 1). As in our analysis, the male to female ratio was 1.3:1 and the mean age was 36.5 years.

Table 1  
Characteristics of the patients

Case	Symptoms	Localization	Accompanying disease	Treatment	Author
1	none	right lung	Adenocarcinoma	VATS	Samancilar.O[33]
2	dyspnoea	right lung	Adenocarcinoma	VATS	Shakir.H.A[19]
3	cough	right lung	Adenocarcinoma	RATS	Fukuhara.S[20]
4	none	right lung	Adenocarcinoma	VATS	Arai.H[10]
5	dyspnoea	right lung	Spontaneous pneumothorax	VATS	Azoury.F.M[15]
6	excessive sweating	right lung	Hyperhidrosis	VATS	Kauffman.P[22]
7	excessive sweating	right lung	Hyperhidrosis	VATS	
8	excessive sweating	right lung	Hyperhidrosis	VATS	
9	excessive sweating	right lung	Hyperhidrosis	VATS	
10	excessive sweating	right lung	Hyperhidrosis	VATS	
11	excessive sweating	right lung	Hyperhidrosis	VATS	
12	excessive sweating	right lung	Hyperhidrosis	VATS	
13	none	right lung	Spontaneous pneumothorax	Thoracic closed drainage	Betschart.T[16]
14	head injury	right lung	Spontaneous pneumothorax	Thoracic closed drainage	
15	none	right lung	Esophageal cancer	Esophagectomy	Maldjian.PD[27]
16	murmur in the mesocardiac area	right lung	Pulmonary sequestrations	Thoracotomy	Koksal.Y[29]
17	hemoptysis and hoarseness	right lung	Adenocarcinoma	VATS	Delalieux.S[9]
18	dyspnoea	right lung	Spontaneous pneumothorax	VATS	Internullo.E[17]

<b>Case</b>	<b>Symptoms</b>	<b>Localization</b>	<b>Accompanying disease</b>	<b>Treatment</b>	<b>Author</b>
19	vomiting	right lung	Esophageal atresia	Thoracotomy	Eradi.B[28]
20	vomiting	right lung	Esophageal atresia	Thoracotomy	
21	none	right lung	SCLC	VATS	Sen.S[34]
22	excessive sweating	right lung	Hyperhidrosis	VATS	Gill.A.J[24]
23	none	right lung	NSCLC	VATS	Grismer.J.T[35]
24	excessive sweating	right lung	Hyperhidrosis	Thoracotomy	Sieunarine.K[26]
25	dyspnoea	right lung	Spontaneous pneumothorax	VATS	Sadikot.R.T[18]
26	excessive sweating	right lung	Hyperhidrosis	VATS	Reisfeld.R[23]
27	excessive sweating	right lung	Hyperhidrosis	VATS	
28	excessive sweating	right lung	Hyperhidrosis	VATS	
29	excessive sweating	right lung	Hyperhidrosis	VATS	
30	excessive sweating	right lung	Hyperhidrosis	VATS	
31	excessive sweating	right lung	Hyperhidrosis	VATS	
32	excessive sweating	right lung	Hyperhidrosis	VATS	
33	excessive sweating	right lung	Hyperhidrosis	VATS	
34	excessive sweating	right lung	Hyperhidrosis	VATS	
35	excessive sweating	right lung	Hyperhidrosis	VATS	
36	excessive sweating	right lung	Hyperhidrosis	VATS	
37	excessive sweating	right lung	Hyperhidrosis	VATS	

Case	Symptoms	Localization	Accompanying disease	Treatment	Author
38	excessive sweating	right lung	Hyperhidrosis	VATS	
39	excessive sweating	right lung	Hyperhidrosis	VATS	
40	excessive sweating	right lung	Hyperhidrosis	VATS	
41	excessive sweating	right lung	Hyperhidrosis	VATS	
42	excessive sweating	right lung	Hyperhidrosis	VATS	
43	excessive sweating	right lung	Hyperhidrosis	VATS	
44	excessive sweating	right lung	Hyperhidrosis	VATS	
45	excessive sweating	right lung	Hyperhidrosis	VATS	
46	none	right lung	Adenocarcinoma	VATS	Our patient

## Anatomy

The superior surface of the developing lung will be sliced by the azygos vein if the normal medial migration of the right posterior cardinal vein over the apex of the lung fails in the embryo. The lobe medial to the azygos vein is developed as the azygos lobe. The upper lobe is separated into two parts by an oblique fissure. This abnormal fissure closely looks like a normal lung fissure, ranging from the lung substance to within. It is closed by apposition of the surfaces bounding it and is oval in section. The additional tongue-shaped lobe isolated by the fissure and the material of it is free from macroscopic pathological change with normal lung.

We can see the interior of the right pleural sac after removal of the right lung from picture 1 (quote from Stibbe et al [8]). The upper part of the pleural cavity is realized to be divided into two sections by a dome-like fold seen in picture 1. The fold is a duplication of the parietal pleura. Its bowed margin is attached along a line on the thoracic wall; the attachment that commences posteriorly at the fifth thoracic vertebra in right thorax passes simply upwards across the posterior parts of the intercostal spaces to the middle of the second rib [8]. After that it changes downwards and forwards to the first costal cartilage. The azygos vein is contained between two layers in the fold and the pleural fold and azygos vein are related to one another [8]. The azygos vein lies behind the esophagus and on the right of the midline till it touches the level of the sixth thoracic vertebra [8]. It dips into the material of the upper lobe and pulls down with the pleural fold.

Summarizing from the literatures, the azygos lobe is divided into three types[8]:

### **Type a**

More or less horizontal and cutting the outer (lateral) surface of the lung at some point between the apex and a point two inches below the apex.

### **Type b**

More nearly vertical and dividing the apex of the lung into lateral halves.

### **Type c**

Vertical and cutting off a small tongue-shaped lobe from the inner surface, the pedicle being attached to the upper margin of the root of the lung.

## **Clinical characteristics**

It is very rare to find an isolated case of azygos lobe without any associated anomaly. From our Table, there are many primary thorax diseases with azygos lobe, such as lung cancer (n = 8), spontaneous pneumothorax (n = 5), esophageal cancer (n = 1), pulmonary sequestration (n = 1), esophageal atresia (n = 2), hyperhidrosis (n = 29). The azygos lobe is typically asymptomatic. It tends to be incidentally discovered during radiological investigation of symptoms related to primary thorax disease. In the eight patients with lung cancer, half of them are asymptomatic, even in our case. In Delalieux et al.[9] report, however, the patient presented with hemoptysis and hoarseness. In the research of patients with spontaneous pneumothorax, most of them presented with dyspnea. 29 cases diagnosed as hyperhidrosis presented with excessive sweating typically.

## **Imaging characteristics**

Chest radiographs are the most generally performed imaging learning to evaluate the mass in the thorax, but it may not be possible to distinguish azygos lobe from others. Typical chest X-ray shows a fine, curved line suggesting the meso-azygos and a small nodule shaped like a tear drop telling the azygos vein[9,10]. Azygos lobe can be dependably diagnosed by High-resolution chest computed tomography (HRCT). In our case, HRCT scans confirmed presence of an azygos lobe and a GGO measuring 1.2 × 1.0 cm in the anterior segment of the right upper lobe adjacent to the arch of the azygos vein (Figs. 2A,B).

On HDCT, the azygos vein is seen as a thicker structure following the same path as the fissure. The position of the azygos arch is higher than normal one [11]. The visceral and parietal layers of pleura forming the mesoazygos are not fused, as is shown by the common occurrence of pleural effusion extending into the azygos fissure [12,13]. This state favors mobility of the azygos vein and enables it to jump from its usual position in the fissure and migrate to the mediastinum[14]. Because the repositioned azygos vein is joined to a structure whose location is higher than the normal anatomic path of the intramediastinal azygos vein [15–18].

# Treatment

In the group of patients with lung cancer, 8 cases were treated with surgical procedure. The first report of a right upper lobectomy by video-assisted thoracic surgery (VATS) in a patient with an azygos lobe was published by Arai et al [10]. Some tumors may originate directly from the azygos lobe reported in several research [9,19,20]. In Fukuhara et al [20] research, they firstly reported the case with operative demonstration of a primary adenocarcinoma arising from an azygos lobe, which was treated with robot-assisted azygos lobectomy. As the azygos lobe is a portion of the right upper lobe isolated by the azygos vein and not a developmentally separate lobe, lobectomy in the patient with azygos lobe without concurrent resection of the right upper lobe is considered to be a limited resection [20]. However, an azygos lobectomy with mediastinal lymph node dissection may be an acceptable healing alternative for elderly individuals with poor pulmonary function and this method is considered to be a better way for preserving of postoperative pulmonary function and reducing morbidity and mortality [1, 21]. Some other cardiopulmonary pathology might be existing in patients with azygos lobe so that it is important to keep this in mind when examining such patients. As showed in our case from the Table, we approached the neoplasm with mediastinal lymph node dissection and then it was removed by VATS. The azygos lobe was visible during the operation. The upper part of the pleural cavity was seen to be divided into two compartments by a dome-like fold and occupied the fissure. The fold is a duplication of the parietal pleura (Fig. 3).

The presence of an azygos lobe is considered a complicating issue, especially in cases with hyperhidrosis or spontaneous pneumothorax. An azygos lobe might have a protective effect against the improvement of spontaneous pneumothorax reported from some studies [3,12,14]. Three mechanisms were offered: the reflected pleura might be limiting the size of a potential pneumothorax; the mechanical stress transferred to the apex of the lung will be lessening with the meso-azygos; or the changed anatomy may essentially shield against bullae formation. As it is relatively under-inflated, there is decreased perfusion and ventilation of the azygos lobe physiologically. The anatomical explanation for the decreased ventilation is distortion of the bronchi caused by the azygos fissure. On the other hand, the similar bronchial anatomy could dispose the azygos lobe to air trapping and develop into emphysema, bronchiectasis, or atelectasis. VATS is used for the management of a spontaneous pneumothorax proposing its superiority to open thoracotomy [15,17,18].

Several researches also reported the surgical difficulty in patient with azygos lobe [22–24]. As the azygos vein is a thin wall, blood flow and very breakable structure, it has to be pushed aside or ligated with extreme carefulness [5–7, 25]. The first case was reported by Sieunarine et al [26] in 1997. It was considered that difficulty would have been experienced in achieving haemostasis in the event of injury [26]. Azygos covered the sympathetic chain between the second and fourth thoracic ganglia. The third ganglion was the most difficult one to identify during the surgery [22]. When there were no venous tributaries in the curtain, it was useful to create a window to expose the sympathetic chain [22]. At the end of VATS, it is important to check whether the azygos lobe has gone back to its original location or there is a possibility of atelectasis [2–4, 22].

In patients with esophageal diseases, azygos lobe was also found in surgery [27,28]. Two cases were babies diagnosed as esophageal atresia. The overarching of the azygos vein in the extrapleural plane compromises the preferred extrapleural approach to the posterior mediastinum. Its apparent passage through the upper lobe may be disconcerting, and doubt about the anatomy depresses the surgeon from simply dividing it [27,28]. They suggested that once the vessel is recognized, ligation and division are safe and permit dissection to continue. Then Koksal et al [29] first reported a child with an extralobar pulmonary sequestrations (ELPS) located in the upper posterior mediastinum associated with the azygos lobe. ELPS is a rare congenital anomaly that commonly occurs on the left side [30–32]. In their case, ELPS tissue was receiving blood supply from the ascending aorta and right brachiocephalic artery, and draining to the superior vena cava by an accompanying vein [29].

## Conclusions

An azygos lobe is a rare anomaly of the lung. Anatomic variations may misperceive routine operations while the surgeon performs a thoracotomy or VATS on the right side. No matter whether we preserve the vein or not during the surgery, the surgeon must be informed about the disparities in vascular drainage to the azygos vein. At the same time, we should pay more attention to where the azygos vein truly drains to so that the operation will be proceeding smoothly. Preoperative contrast CT of the chest is useful to assess the anatomic variation of the vein.

## Declarations

### Ethics approval and consent to participate

All the data supporting our findings in this paper were freely downloaded from the PubMed, EMBASE, the Web of Science and China National Knowledge Infrastructure websites. No ethical approval or written informed consent for participation was required.

### Consent for publish

Not applicable.

### Availability of data and materials

All data for this study are publicly available and are ready for the public to download at no cost from the official websites of the PubMed, EMBASE, the Web of Science and China National Knowledge Infrastructure. There is no need to have the formal permission to use data for this study. The sources and data robustness have been described in the section of “Methods”.

### Competing interests

The authors declare that they have no competing interests.

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## Authors' contributions

CS were involved in drafting the manuscript. CS made contributions to the concepts, acquisition and analysis of the data. CS and GC were involved in acquisition of data and preparing the figures. GC designed and revised the manuscript. All authors have read and approved the final manuscript.

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## Figures

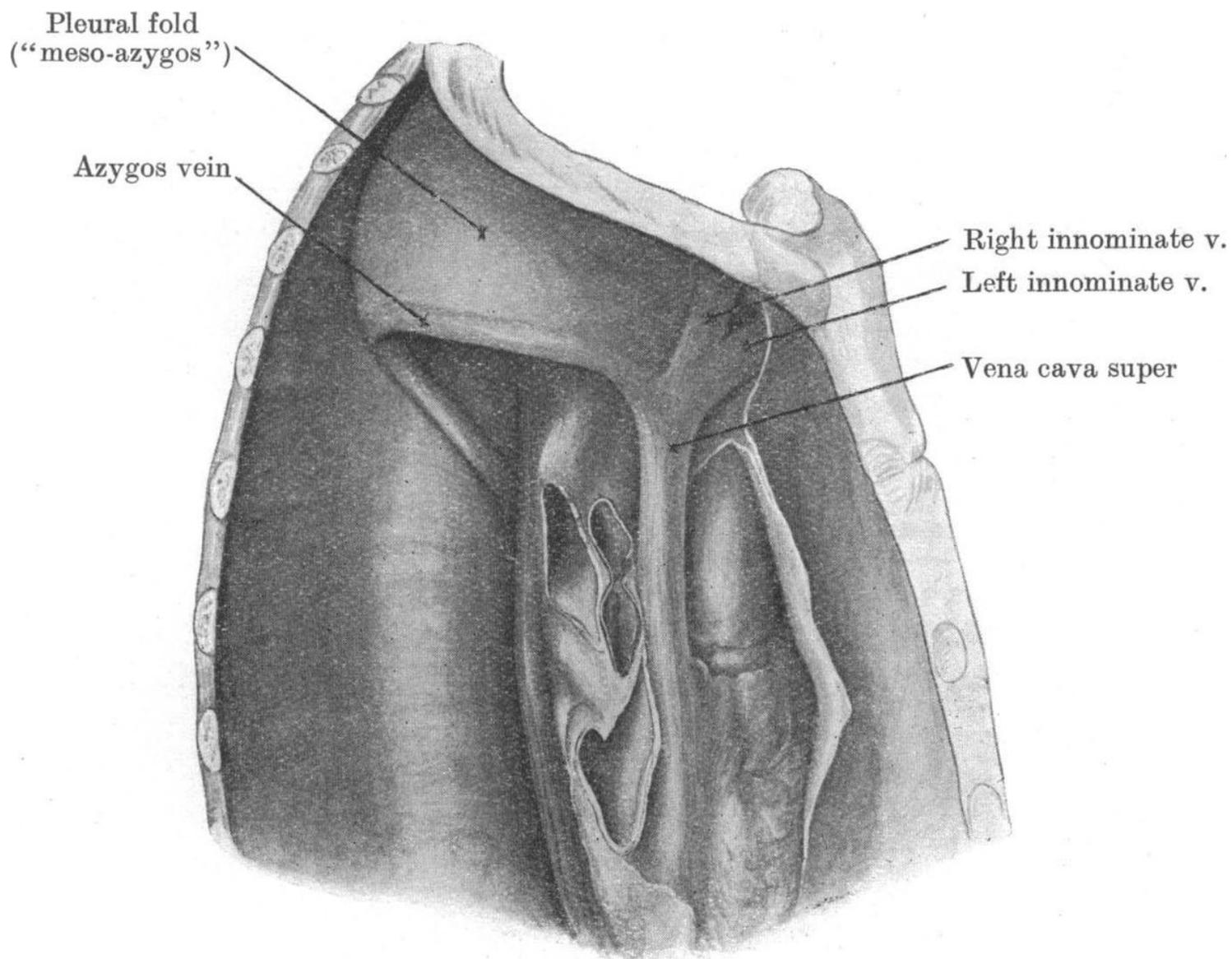
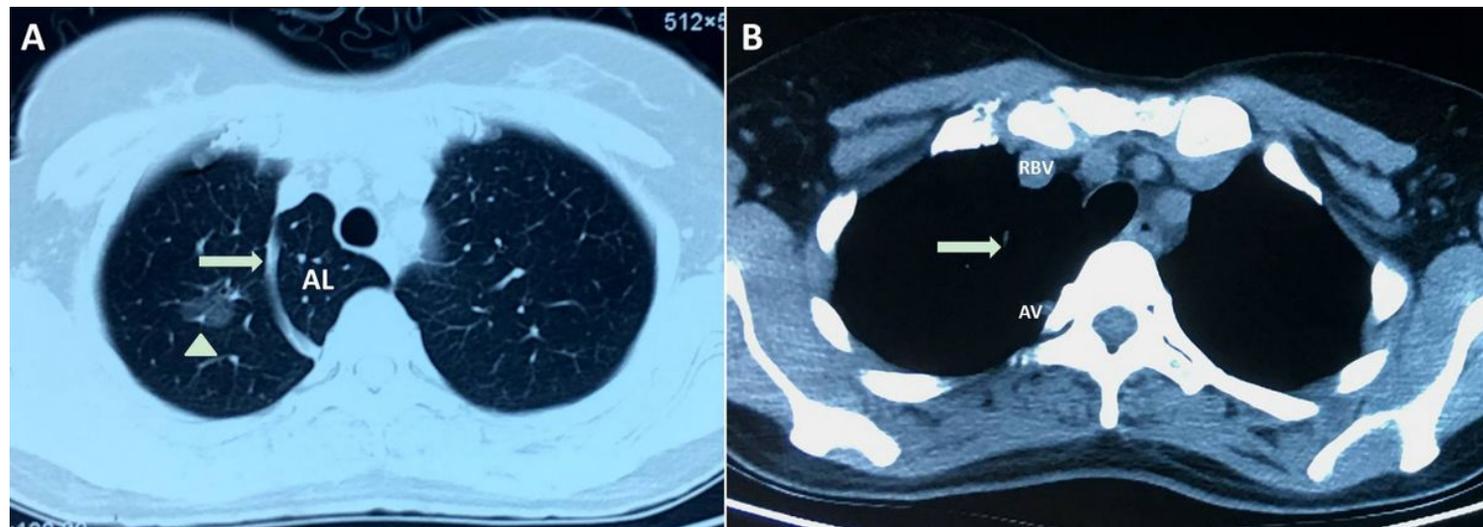


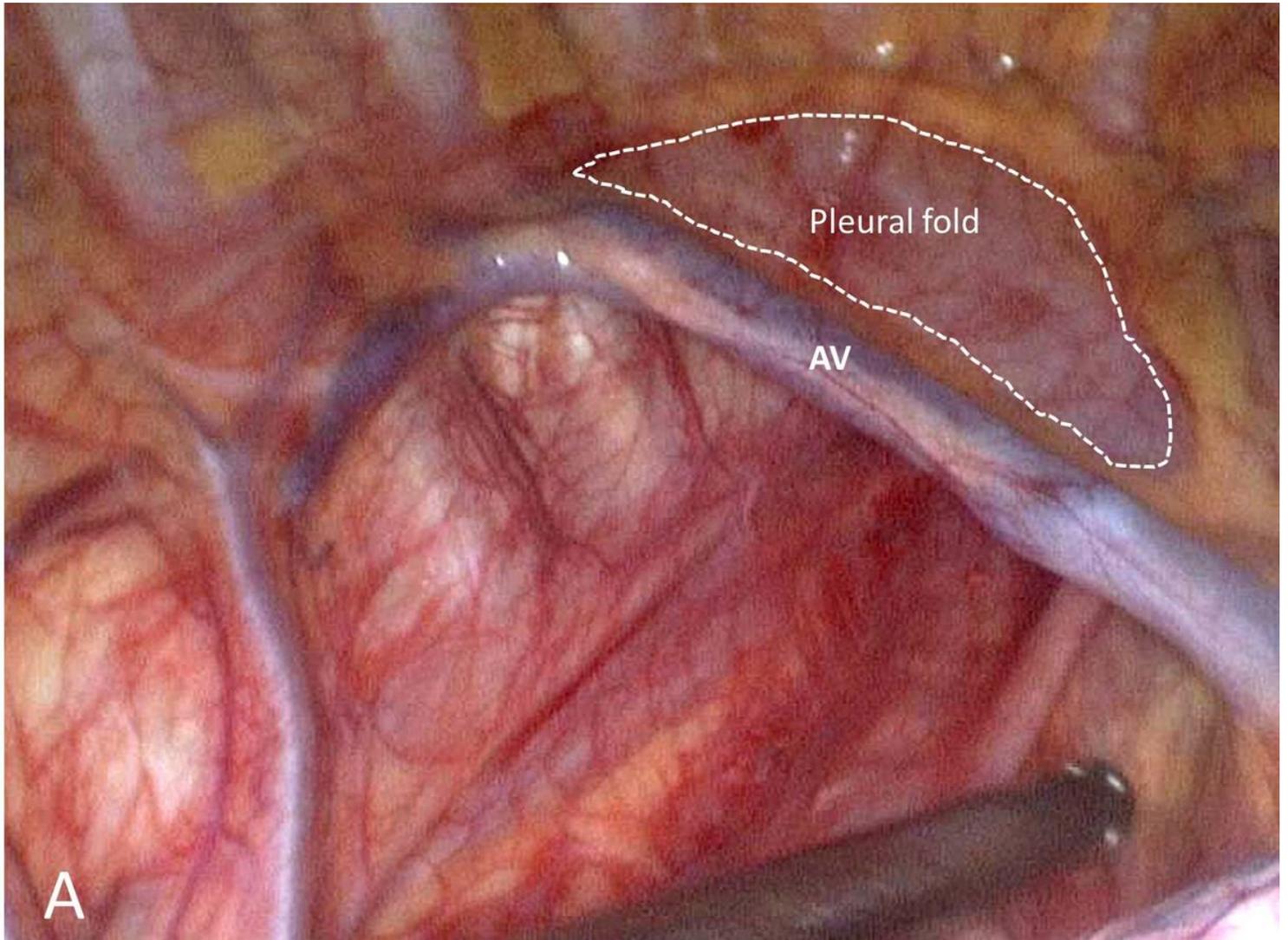
Figure 1

A view of the interior of the right pleural sac after removal of the right lung.



## Figure 2

Computed tomography features of the case. A: Chest HRCT showing azygos lobe (AL) and azygos vein (arrow); GGO in right upper lobe adjacent to azygos lobe (arrowhead). B: Chest HRCT also showing the arch of the azygos vein (arrow) between the azygos vein (AV) and right brachial vein (RBV)



## Figure 3

Intraoperative image of the azygos lobe.