The rare anatomical variant of upper lobe

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Research article

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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 July 2020.

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.

Background

An azygos lobe is a rare congenital variant. It is recognized incidentally on chest X-ray or computed tomography (CT) for the duration of health checkups. Wrisberg firstly described the azygos lobe in 1877 that is a variant of pulmonary anatomy and presenting in 0.1-1% of the people [1]. From an anatomical point of view, unlike the accessory lobes of other anatomical abnormalities in the lungs, the sinusoidal vein refers to the abnormal vein that travels through the tip of the lung [2]. According to the literature, azygos lobe may occur in the left or right lobe of the lungs [3, 4].

Based on a review of the literature on the general electronic database, there are only a few case reports on the study of the azygos veins, and several cases of primary chest disease with azygous veins are described. This

is the first study that included all patients diagnosed with odd veins through surgical findings in English literature. This article mainly summarizes the clinical data, radiological manifestations and treatment strategies of azygos lobe to provide new knowledge for doctors as well as empirical diagnosis and treatment thinking.

Methods

Data sources

We searched the full-text English paper from PubMed, EMBASE and Web of Science from January 1931 to July 2020. Final search criteria included the following keywords that were “azygos lobe” and “surgery”. Furthermore, we also made a second manual exploration of the list of selected references to ensure that the selected articles contained relevant research without duplication. The summary of the findings is listed in Table.

Inclusion and exclusion criteria

Inclusion criteria

(i) azygos lobe was 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) documents without full text were excluded; (iii) Non-English languages papers were not accepted; and (iv) patient without surgery.

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 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 and vomiting. 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 video-assisted thoracic surgery (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

Azygos lobe is a rare abnormality anatomy of the lung, described by Wrisberg [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, most of the patients are young and middle-aged male 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.P.D[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]
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]

Case	Symptoms	Localization	Accompanying disease	Treatment	Author
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	
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

During the embryonic development period, if the main vein in the posterior part of the right or left thoracic cavity is displaced in the embryo at the normal medial side of the apex of the lung, the upper surface of the upper lobe in the developmental stage will be cut by the atypical vein. The lobe medial to azygos vein is developed as the azygos lobe. The upper lobe is separated into two parts by a slanting 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 on 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 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, b and c. The difference between a and b is that the "cut line" of the upper lobe of the lung is close to horizontal or completely vertical. The c-type is the "cut line" that completely cuts the upper lobe of the lung and cuts a small tongue-shaped projection near the inner surface.

Clinical characteristics

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

Using the most common chest X-ray examination, we can see that the typical azygous veins appear as small nodular changes in the upper lobe of the lungs. But such changes may be overlooked by most clinicians [9, 10]. Azygos lobe can be diagnosed by High-resolution chest computed tomography (HRCT). In our case, chest CT scan confirmed that there were azygos lobe in the right upper lobe near the tip of the lung, while a ground glass opacity (GGO) was observed on the outer side of the azygos lobe near the azygous vein whose size was 1.2×1.0 cm (Fig. 1A, B). On HRCT, the position of the azygos arch of the azygos lobe is relatively high compared to the normal azygos vein [11]. From an anatomical point of view, it is understood that the inner pleura is not fused with the top layer of azygos, which ultimately makes the azygos arch appear more plentiful [12, 13]. The elevation of the azygos arch will facilitate the migration of the azygous vein [14]. The azygous vein, based on the azygos arch, is more higher connected to the mediastinum than the normal anatomical path [15–18].

Treatment

In the group of patients with lung cancer, all of them were treated with surgical procedure. Arai et al reported the first case of azygos lobe in a patient with lung cancer and underwent by VATS of right upper lobectomy [10]. According to some researches, tumors may exist directly in the azygos lobe [9, 19, 20]. In Fukuhara et al [20] research, they reported that lesions present in the azygos lobe can be removed by robot-assisted minimally invasive surgery and ultimately confirmed by pathological findings as primary lung adenocarcinoma. Since the azygos lobe are not isolated, but part of the lung tissue separated from the right

upper lobe, if the patient only simply removes the azygos lobe and leave the remaining part of the right upper lobe, it will be considered as a limited operation [20]. 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 [1, 21]. As we showed from the table and the surgical picture, we performed a mediastinal lymph node dissection and a complete resection of the right upper lobe by VATS. During the operation, we can clearly see the azygos lobes located in the upper lobe of the right lung through the display. The area of the pleural cavity near the top of the right chest appears to be divided into two spaces. The parietal pleura and the azygous vein are connected by a dome-like fold (Fig. 2).

According to a small number of studies, in some benign lung diseases, especially in the case of hyperhidrosis or spontaneous pneumothorax, the presence of azygos lobes may be related to these diseases [3, 12, 14]. Azygos lobe has an inhibitory effect in spontaneous pneumothorax possibly. This theoretical basis is mainly derived from three possible mechanisms. Firstly, the reflexed pleura may limit the size of the potential pneumothorax; Secondly, the mechanical stress generated by the azygos lobe at the top of the lung can reduce the pleural effusion of the spontaneous pneumothorax tension in order to avoid the occurrence of pneumothorax; Thirdly, it may directly change the anatomy of the lung lobe to substantially prevent the formation of bullae. 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]. Since the azygous vein is a thin wall, blood flow and a very fragile structure, it must be carefully pushed aside by the surgeon or ligated if necessary during surgery [5–7, 25]. Some scholars believe that it is difficult to achieve hemostasis in the case of damaged of azygous veins [26]. The Azygos vein covers the sympathetic chain between the second and fourth of thoracic ganglia. The third ganglion is the most difficult to identify ganglion in surgery and should be highly valued during surgery [22]. When venous tributaries are not seen in the surgical field, it is useful to create a operating field of exposure to the sympathetic chain for safety reasons [22]. Reminding all thoracic surgeons that at the end of the VATS, it is important to check if the azygos lobe has returned to their original position. If it does not return to its original position, there will be a problem of atelectasis. [2–4, 22].

In the group of patients with esophageal diseases, azygos lobe was also found in surgery [27, 28]. Two cases were babies diagnosed as esophageal atresia. Due to the abnormal anatomical structure of the azygous vein, the azygous lobes are produced, which makes the surgeon difficult to separate the azygous veins during the operation. In some operations, the azygous veins may not be separated successfully [27, 28]. Therefore, literature studies suggest that once the vein is found, the dissection of the azygos vein is safe. Then Koksall et al [29] firstly reported a child with an extralobar pulmonary sequestrations (ELPS). ELPS is a rare congenital lobular abnormality commonly found in the left lobes [30–32]. Under normal circumstances, ELPS receives blood supply from the ascending aorta and right brachiocephalic artery, and through the accompanying venous drainage to the superior vena cava [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 should attach great importance to the azygous lobes caused by the abnormal anatomy of the azygous vein to avoid unnecessary bleeding or other complications during surgery. 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.

Abbreviations

CT—computed tomography—ELPS—extra lobar pulmonary sequestrations—GGO—ground-glass opacity—HRCT—High-resolution chest computed tomography—VATS—video-assisted thoracic surgery.

Declarations

Ethics approval and consent to participate

All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Consent for publish

All the authors consent to publish the paper.

Availability of data and materials

The datasets supporting the conclusion of this article are included within the article.

Competing interests

The authors declare that they have no competing interests.

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Authors' contributions

CS was involved in drafting the manuscript. CS and GC made contributions to the concepts, acquisition and analysis of the data. CS was 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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Table

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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

Figures

Figure 1

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 2

Intraoperative image of the azygos lobe.