

## CASE REPORT

## Unusual Radiological Sign in Bronchial Atresia

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## Abstract

Bronchial atresia is usually diagnosed by incidentally detecting opacity at hilar region and hyperinflation around this opacity on chest X-ray. It may rarely be detected as air sac like atresic bronchus. The breath sounds in the right hemithorax were heard less when compared to the left hemithorax in the auscultation of a 16-year-old male patient with allergic rhinitis. The patient had no pulmonary complaints, and this finding was not recorded in his previous follow-up. In order to determine the etiology of hyperinflation seen on chest X-ray, computed tomography was performed. Hyperinflation was identified in the lower lobe superior segment of the right lung, which could be secondary to bronchial atresia. It was confirmed that in the evaluation of computed tomography with three-dimensional reconstruction, lower lobe superior segment bronchus of the right lung was atresic and contrary to expected mucus opacity in the distal of atresia, dilated bronchus was filled with air. This case was especially presented to lay emphasis on careful auscultation and share its unusual radiological presentation which had been reported twice before.

**KEYWORDS:** Bronchial atresia, bronchial diseases, congenital, hyperinflation, radiology**Received:** 19.06.2015**Accepted:** 31.08.2015

### INTRODUCTION

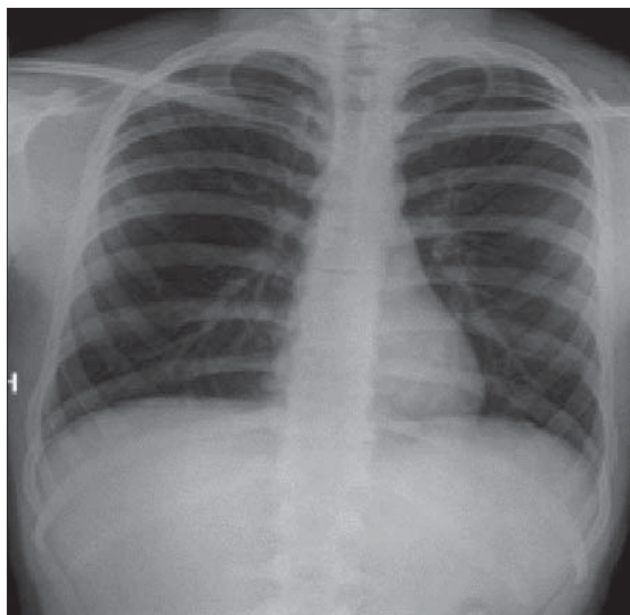
Congenital lung malformations are mostly diagnosed prenatally or in early childhood. However, it can be detected in later periods and even coincidentally, with unrelated clinical symptoms [1]. Bronchial atresia, which is one of the congenital lung malformations that receive late diagnosis, develops as a result of a cut in the continuity of lobar, segmental or subsegmental bronchus. Bronchial atresia is characterized by an increase in inflation in the lung region inflated by the atresic bronchus and accumulation of mucus in the distal of the atresic bronchus [2]. Generally the diagnosis is made by lung graphy with a coincidental detection of opacity in the hilar region and increase in the inflation around it [3]. Rarely, it can be diagnosed by seeing air-fluid level in the distal of the atresic bronchus [4]. The air sac shape of the atresic bronchus is very rare and has been reported twice in the literature.

This study presented the case of a patient with allergic rhinitis, who was diagnosed with atypical bronchial atresia as a result of the radiological examinations performed upon hearing less breath sounds in lung auscultation.

### CASE PRESENTATION

A 16-year-old male patient had been followed up in our polyclinic with a diagnosis of seasonal allergic rhinitis for four years when it was detected during his routine physical examination that the breath sounds in the right hemithorax were heard less when compared to the left hemithorax. There was no peculiarity in patient history and laboratory findings, and pulmonary function test was within normal limits. Upon detecting hyperinflation in the right lung on lung graphy (Figure 1), lung tomography was performed to have an etiologic evaluation. On lung tomography, hyperinflation was identified in the lower lobe superior segment of the right lung, which was thought to be secondary to bronchial atresia or bronchus obstruction. Flexible fiber optic bronchoscopy was conducted in an attempt to investigate the reasons of bronchus obstruction, particularly foreign body aspiration. Trachea and major bronchi were normal on bronchoscopy; however double entry variation anomaly was detected coincidentally. It was detected in the evaluation of the lung tomography with a three-dimensional reconstruction that there was not truncus exit point in the lower lobe superior segment of the right lung and that there was a hyperlucent volume in low vascularity that displaced other lung segments due to hyperinflation in the localization of right lung lower lobe superior segment. A bronchial tree wider than usual and showing branching to the periphery was present within the identified hyperlucent volume. This bronchial tree could draw close to bronchial structures in the hilus at most 6.8 mm. The air-filled bronchiectasis structure, unrelated





**Figure 1.** Hyperinflation in the right lung on the posteroanterior lung graphy.

to the bronchial structures in the hilus, ended with a sharp point proximally (Figure 2).

Since congenital cardiovascular anomalies can sometimes accompany bronchial atresia, cardiologic evaluation was performed, revealing no pathologies. Although the case was asymptomatic, since there was hyperinflation in a very wide region of the lung, the case was closely followed up in terms of probable infection and progression.

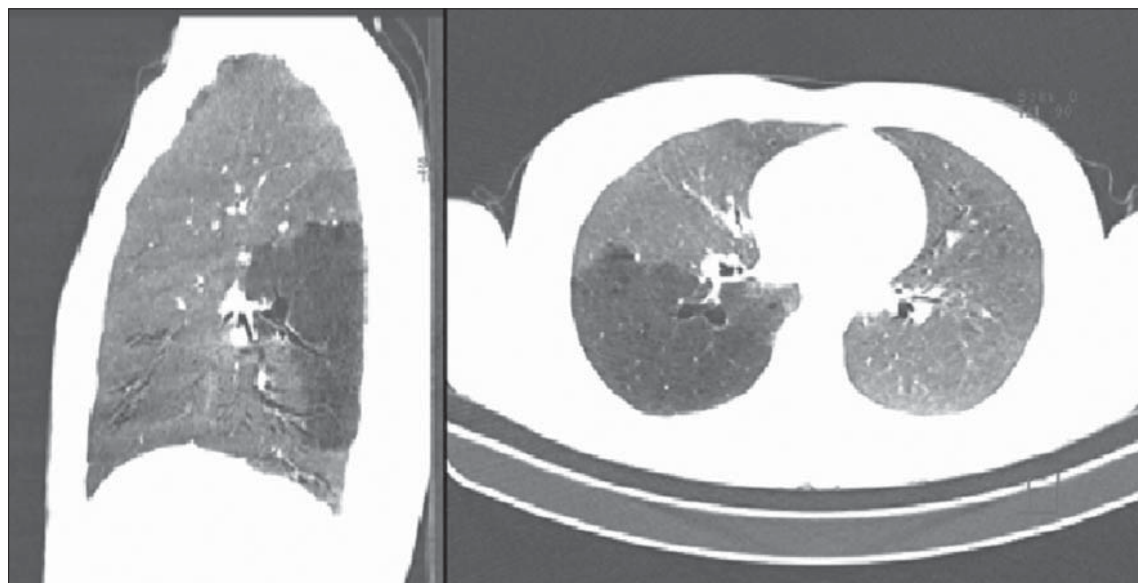
### DISCUSSION

The first case of bronchial atresia was reported by Ramsay and Byron in 1953 [5]. The main reason of bronchial atresia is unknown. The localization of atresia is linked to the time intrauterine is affected. If fetal development is affected on the

5<sup>th</sup>, 6<sup>th</sup> and 16<sup>th</sup> weeks, lobar, segmental and subsegmental bronchial atresia are seen respectively [6]. Cases have been reported, where bronchial atresia is mixed or coexists with other congenital lobar emphysema or congenital cystic malformation. In a recent study, it has been put forward that bronchial atresia is a component of a series of congenital lung anomalies. The formation time of bronchial atresia in fetal period and its response level are associated with the etiology causing this anomaly [2].

Air access to the lung segment ventilated by the atresic bronchus happens with Kohnpores and Lambert channels. As these allow for more air flow during inspiration in proportion to expiration, hyperinflation develops in the lung segment ventilated by the atresic bronchus. Generally bronchial mucocele occurs as a result of mucus accumulation in the distal of the the atresic bronchus [7]. Bronchial atresia is usually asymptomatic, just as it was in our case; however, it may cause repetitive lung infection, cough, wheezing, and dyspnea in some cases [8,9]. It has been reported to cause spontaneous pneumothorax very rarely [10].

Generally, lung tomography is sufficient for the diagnosis of congenital bronchial atresia. Characteristics finding of bronchial atresia on lung tomography is the image of air and mucus-filled, widened bronchus [11-13]. The surrounding area of the mucus-filled, widened bronchus is seen hyperlucent with focal parenchymal oligemia and air trapping that occur as a result of hypoxic vasoconstriction and intrapulmonary vascular compression [13]. Bronchoscopy can be used in terms of the diagnosis of proximal atresia and differential diagnosis [8,9]. Mucocele, which is a typical finding of bronchial atresia on lung graphy, can be seen as nodule close to the hilar region, ovoid, bronchus structure or tubular [14]. Until now, the dilated bronchus has been shown in two other cases to be filled with air instead of mucus, just as it was in our case [4,15]. Congenital diseases or diseases causing acquired bronchus obstruction should be considered in differential diagnosis.



**Figure 2.** The atresic image of the lower lobe superior segment bronchus of the right lung on the reconstruction of thoracic computed tomography and it is seen that the dilated bronchus is filled with air contrary to the expected mucus opacity in the distal of the atresia.

The diagnosis of bronchial atresia is made late since it does not usually cause symptoms [16]. It is diagnosed at about 17 years of age. Two thirds of the reported cases are patients that have not had pulmonary complaints until that day and that have received coincidental diagnosis as a result of the performed lung graphy, just as it was in our case. Treatment is not recommended in asymptomatic patients. Surgical excision is needed if complications, primarily infection, secondary to atresic bronchus develop [17].

Even though classic radiological finding is defined as the mucus opacity in the distal of the atresia and rarely as the opacity that gives air-fluid level in bronchial atresia, the dilated bronchus can be seen as fully filled with air, just as it was in our case. This case was presented in order to emphasize the importance of careful auscultation during physical examination and share the atypical radiologic presentation that has only been reported twice in the literature.

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