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CASE REPORT

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Bronchial atresia as an incidental chest CT finding: a case report

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ABSTRACT

Bronchial atresia corresponds to a congenital anomaly of the airways, in which a bronchial segment presents a focal obstruction. As a result, there are mucoid impactions with or without bronchocele, resulting in air trapping and distention of the lung parenchyma. In the present study, we report the case of a patient incidentally diagnosed with bronchial atresia when undergoing chest computed tomography after performing thoracic sympathectomy as a treatment for palmoplantar hyperhidrosis. Classically, the diagnosis is made incidentally with an asymptomatic patient, but cough, shortness of breath, wheezing, hemoptysis, chest pain, or pneumothorax may be present. There is still no established consensus on the treatment of this pathology, but in asymptomatic patients, the usual approach is conservative, with outpatient follow-up.

KEY WORDS: Bronchial Atresia, congenital anomaly, incidental discoveries, pulmonology, thoracic surgery.

INTRODUCTION

Bronchial atresia is characterized by focal obstruction of a bronchial segment, proximal or distal, being more frequent in the left upper lobe [1]. A deficiency in vascularization during the period of development of the bronchial tree that results in the formation of a bronchus with a blind bottom and distension of the adjacent lung parenchyma is the pathophysiological mechanism of the condition, a process responsible for the classification of the aforementioned atresia in the group of congenital airway anomalies [2].

As a consequence of this anomaly, there is an accumulation of mucus in the affected parenchymal region, forming mucoid impactions associated or not with bronchocele, resulting in areas of air trapping and distension of the lung parenchyma [3]. Therefore, the clinical picture of bronchial atresia varies from the absence of symptoms to the presence of recurrent pulmonary infections or spontaneous pneumothorax [4]. Since most cases are asymptomatic, the findings on physical examination are scarce, with the possibility of localized reduction of the vesicular murmur in the affected area, and the diagnosis is more commonly made by imaging exams, of which chest tomography stands out as the most accurate [5,6]. The characteristic tomographic pattern of bronchial atresia consists of segmental or adjacent lobe hyperinflation by collateral duct ventilation, with the possibility of visualizing reduced vascularization in the affected segment or lobe and mucoid impaction in adjacent areas [6]. The management of such a condition, however, remains challenging, as there is a disagreement in the literature between the need to address asymptomatic bronchial atresia [7]. This discussion results from the paradox between the indolent behavior of the anomaly and the greater tendency to develop complications such as recurrent pneumonias, often with colonization by atypical microorganisms, and spontaneous pneumothorax, which can be fatal if inadequately manager [8]. Therefore, the diagnosis of this health condition reduces the chance of such complications in the long term [7,8].

In this sense, we report the case of an asymptomatic female patient whose diagnosis of bronchial atresia was made occasionally in the postoperative period of a thoracic sympathectomy for the treatment of palmo-plantar hyperhidrosis, by means of a chest tomography requested because of the risks of postprocedural pneumothorax and hemothorax [9].

CASE REPORT

PATIENT INFORMATION: The patient is a 21-year-old student, coming from a family with no reported comorbidities, livin in a Brazilian metropolis.

CLINICAL FINDINGS: A 21-year-old female patient sought thoracic surgery service complaining of excessive sweating in the palmar region of her hands and the plantar region of her feet, referring to decreased quality of life as a result of such symptoms. At the time, she denied other complaints and other comorbidities, so complementary exams were requested in order to perform a thoracic sympathectomy.

TIMELINE: In April 2022, the patient sought the thoracic surgery service with complaints of excessive palmar and plantar sweating refractory to drug treatment, and tests were requested at the time for thoracic sympathectomy to correct hyperhidrosis. At the end of the month, the procedure was performed, and the patient was discharged uneventfully one day after surgery. In mid-May, the patient returned for follow-up, with a chest CT showing bronchocele related to bronchial atresia, generating areas of air retention and mucoid impaction in the adjacent parenchyma. In this sense, the diagnosis of bronchial atresia was confirmed, with the option for outpatient segment at 6 months due to the asymptomatic nature of the condition.

DIAGNOSTIC ASSESSMENT: After the procedure, the patient evolved well and had no complaints. However, the chest computed tomography revealed an area of hyper transparency in the upper lobe of the right lung, with a tight hilar oval image with apparent continuity with the segmental bronchus, which, in turn, shows wall thickening and mucus accumulation inside (figure 1). This tomography was requested because of the risk of pneumothorax and post-procedure hemothorax. Thus, a diagnosis of bronchocele related to bronchial atresia was made, generating areas of air retention and mucoid impaction in the adjacent parenchyma.

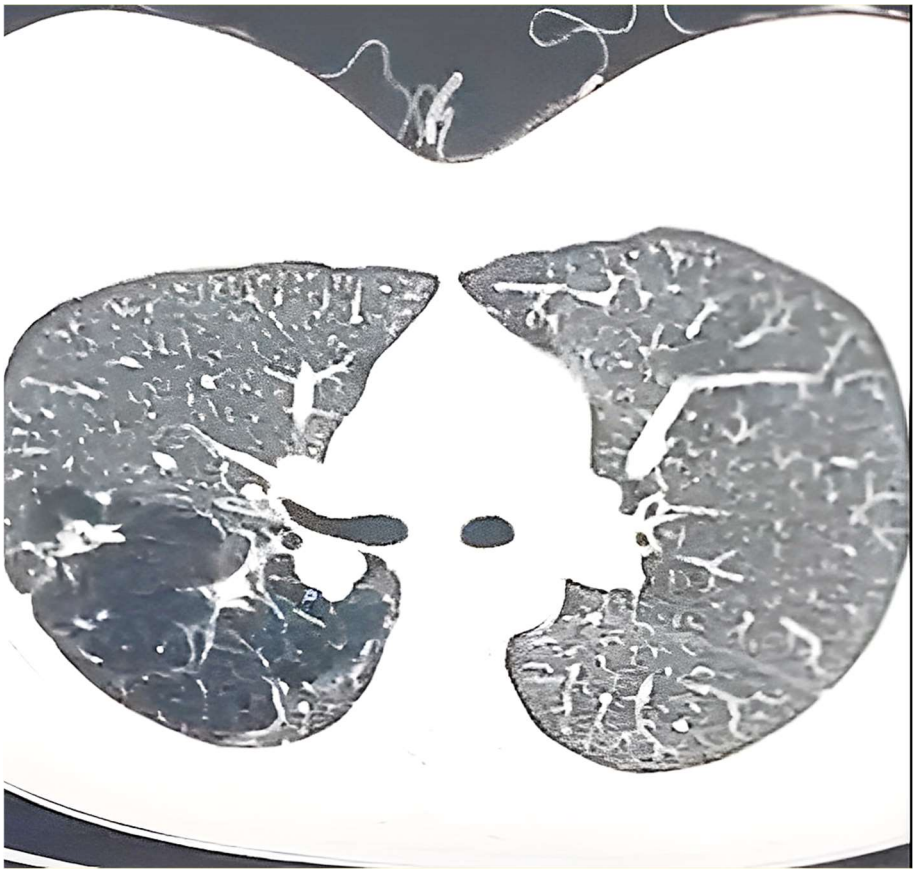

Figure 1. Chest CT sagittal section showing areas of air trapping in the middle and posterior regions of the right upper lobe with the presence of a juxta hilar oval image continuous with segmental bronchus, whose wall is thickened and with signs of mucoid impaction.

THERAPEUTIC INTERVENTION: Due to the asymptomatic nature of the condition, we opted for a conservative treatment with follow-up of the patient every 6 months.

FOLLOW UP AND OUTCOMES: In this context, the patient experienced remission of the main symptoms, with evident objective and subjective improvement in quality of life regarding hyperhidrosis and remained asymptomatic regarding bronchial atresia.

DISCUSSION

Bronchial atresia was first reported in 1953 by Ramsay et al. but was described in 1987 by Jederlinic et al. after findings in 86 cases [10-12]. Still, the condition remains rare with an incidence of 1.2 cases per 100,000 population [13]. The pathophysiology of the malformation is not yet completely understood, but two mechanisms are theorized. The first hypothesis is that the proliferating cells lose their connection to the developing respiratory sprout during normal lung maturation. An alternative mechanism is that a repetitive vascular insult to the lung parenchyma during early fetal development leads to obliteration of an already complete bronchus [1,12,15].

The reported patient was asymptomatic, and was diagnosed as an incidental finding on chest CT scan. Approximately two-thirds of patients are diagnosed in this way, the remaining third report symptoms such as cough, shortness of breath, wheezing, hemoptysis, chest pain, or pneumothorax [1,16,17]. Figure 1 show the classic finding of bronchial atresia on computed tomography: an oval opacity radiating from the hilum with a "*finger in a glove*" appearance due to the formation of mucoid impaction of the distal bronchus, i.e., a mucocele. However, there are cases reported in which there is no mucocele [14,17]. The patient in this case had atresia in the right upper lobe, a site little reported in the literature. The most common sites are the upper left pulmonary lobe (64%), followed by the lower left lobe (14%) and the upper right lobe (12%) [12].

There is still no clear consensus for the management of this malformation. In general, asymptomatic patients are not approached surgically; however, some studies recommend resection of the involved segment to prevent secondary complications, such as infection or involvement of the adjacent lung parenchyma [1,18]. In the case reported here, we opted for a conservative management of the patient's follow-up.

CONCLUSIONS

Bronchial atresia is a rare congenital airway malformation characterized by obstruction of a bronchial segment, the etiology of which remains to be clarified. In the present study, we report the case of an asymptomatic patient with an incidental diagnosis upon chest CT scan. Although there are still no guidelines on the management of the pathology, asymptomatic surgical intervention is not routinely performed, although some studies recommend early resection of the lesion area. In this case, the patient is being followed up with a conservative approach.

SUPPLEMENTARY INFORMATION

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Institutional Review Statement: *The study was conducted according to the guidelines of the Declaration of Helsinki.*
Informed Consent Statement: *Informed consent was obtained from all subjects involved in the study.*
Data Availability Statement: *The datasets generated and analyzed during the current study are available from the corresponding author on reasonable request.*
Conflicts of Interest: *The authors declare no conflicts of interest.*

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