Bronchocele, a common but underrecognized condition: a systematic review

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Abstract

Bronchocele is an abnormal accumulation of mucus often with associated bronchial dilatation. It can be due to either increased production or impaired drainage of mucus in the airways. Diseases like chronic bronchitis, bronchial asthma, bronchiectasis are characterized by high mucus production and other atypical conditions are bronchorrhea and plastic bronchitis with different physical characteristics and compositions of mucus. Improper drainage can lead to bronchocele formation due to underlying benign, malignant tumours or bronchial stenosis. Allergic bronchopulmonary aspergillosis (ABPA) has a peculiar appearance with high attenuated mucus (HAM) in imaging. Careful evaluation of bronchocele is needed as it can be associated with bronchial obstruction or rare causes like plastic bronchitis. Proper identification, evaluation for the underlying cause is key for not missing the underlying diagnosis and accurate treatment.

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Key words: Bronchocele; bronchorrhea; plastic bronchitis; high attenuation mucus.

Contributions: The Author designed the study, acquired the data, analyzed, interpreted, drafted, revised for important intellectual content and finally approved the publication of this article.

Conflict of interest: The author declares no competing interests and confirm accuracy.

Ethics Approval: not applicable.

Received for publication: 21 October 2021.
Accepted for publication: 13 April 2022.

Monaldi Archives for Chest Disease 2023; 93:2133
doi: 10.4081/monaldi.2022.2133

Introduction

Bronchocele is defined as an abnormal accumulation of mucus with or without underlying bronchial dilatation according to the Fleischner Society Glossary. It can present as an incidental finding or in association with other conditions like bronchial asthma, cystic fibrosis, airway stenosis, tumours and congenital abnormalities of the airways. Excessive production of normal mucus, abnormal mucus or associated anomalies of airways which can lead to impairment of drainage needs evaluation and proper specific therapy since the aetiologies vary. Because of a diagnostic challenge, it can pose and the need for complete evaluation once it's faced, we aimed to review this entity and closely related conditions like bronchorrhea and plastic bronchitis.

Methods

We searched PubMed using the word 'Bronchocele', English and humans as filters with no time limit.

Results

We retrieved 25 articles and analysed all articles along with relevant literature. There were 12 case reports, 11 original articles and cases series, and 5 reviews, comments. The relevant data has been analysed and included in the discussion. Relevant information also was extracted from other articles and included in the discussion part as needed. The main focus is on bronchocele, although other relevant entities like bronchorrhea and plastic bronchitis are discussed.

Discussion

Bronchocele is an abnormal accumulation of mucus with or without underlying bronchial dilatation. According to the Fleischner society of glossary, a bronchocele is a bronchial dilation due to retained secretions (mucus impaction) usually caused by proximal obstruction, either congenital or acquired [1]. Bronchocele word is derived from "bronchos" meaning airway, "cele" meaning hernia or swelling. Historically it was described as one of the eight types of hernias by Celsus in the 1st century mentioning it as one type of thy-

roid swelling rather than the present usage which defines it as mucus accumulation [2]. Several other descriptions also mentioned thyroid as bronchocele, some studies have mentioned any swelling from the neck as bronchocele [3-6]. It was also described as "blocked bronchiectasis" in the literature [7].

Mucus is a gel-like substance produced by secretory cells and submucosal glands. It has 97% water and 3% solids consisting of mucin and non-mucin proteins, lipids, cell debris [7]. Excessive and abnormal production of mucus and impaired drainage are two main mechanisms leading to the accumulation of mucus. Excessive bronchial secretions are seen in asthma, chronic obstructive pulmonary disease (COPD), bronchorrhea as in bronchoalveolar carcinoma, and lymphatic abnormalities. Goblet cell hyperplasia is the characteristic feature in conditions like COPD and pathways activated are mitogen-activated kinase/extracellular signal-regulated kinase, calcium-activated chloride channels [8]. Impaired drainage can be seen due to obstruction due to tumours- benign and malignant, foreign body, post-infective sequelae leading to bronchostenosis, ciliary abnormalities like primary ciliary dyskinesia (PCD) and mucus abnormalities as in cystic fibrosis (CF) [9].

Congenital bronchial atresia is an anomaly with completely atretic bronchial, segmental or subsegmental bronchus with an accumulation of the mucus in the distal airway [10,11]. The shape of the accumulation can be circular or branching type depending on the airways involved. [12]. It can be detected in utero and postnatal life. Presentation is common in childhood although adult manifestation is not unusual. Magnetic resonance imaging (MRI) has been compared with computerized tomography (CT) scan to detect various congenital lung anomalies including bronchocele in postnatal life and there was complete agreement between MRI and CT suggesting MRI is a good alternative imaging modality with avoidance of radiation in the paediatric age group [13]. Congenital segmental emphysema a lesion, between the spectrum of congenital cystic adenomatoid malformation (CCAM) and sequestration can manifest bronchoceles [14]. In a study prenatal diagnosis of oesophageal bronchus was done with the orientation of bronchocele towards the gastroesophageal junction and towards hilum in bronchial atresia cases [15]. Calcification is also sometimes a feature of bronchocele in congenital bronchial atresia [16]. Bronchocele with distal lung hyperinflation is a universal finding in bronchial atresia, more than half will be branching and few cases can have air within them [17]. As bronchial atresia can be associated with sequestration, a search for associated other anomalies should be done [18].

CF is a genetic disorder caused by a defect in the chloride transporter, which results in mucus buildup in the airways, repeated infections, respiratory failure, and a shortened life span. It is very common in Caucasians. Bronchocele is a common finding in cystic fibrosis, accounting for roughly two-thirds of cases [18]. This group is predisposed to allergic bronchopulmonary aspergillosis (ABPA), a fungal sensitization that leads to increased mucus secretion and damage to the underlying airways [19]. ABPA is characterized by high density (70 HU) mucus accumulation hyperattenuated mucus (HAM) in some cases. ABPA is usually a feature of asthmatics although it has been described in non-asthmatics also [20]. High serum levels of aspergillus specific IgE, positive precipitin antibody, blood eosinophilia in the appropriate clinical context point towards a diagnosis of ABPA. High bronchocele density as mentioned above, can suggest the presence of ABPA in these patients [19]. The high density of HAM has been attributed to calcium oxalate deposition. Other contents like iron, manganese, calcium phosphate, calcium sulphate are also proposed as causative factors of high attenuation as seen in allergic fungal sinusitis and airway disease, sinobronchial allergic mycosis (SAM) [21-23]. In the author's opinion ABPA is a common but underrecognized cause of bronchocele and if it's present in asthmatics, a workup is needed accordingly with IgE levels, Aspergillus serology and imaging since management differs in both conditions.

Bronchial stenosis can lead to the accumulation of mucus distal to the obstruction. Infections like tuberculosis especially endobronchial form is a common cause of bronchostenosis in countries like India and can lead to bronchocele formation distal to the stenosis [24,25]. The collection of mucus itself can mimic malignancy and bronchoscopy is an investigation of choice to confirm stenosis [26]. In our experience, this mass-like appearance of bronchocele is a common presentation of bronchial stenosis. Since it may need interventions like bronchial dilatation or surgical resection depending upon the severity of presentation proper workup is essential (Figure 1). Other causes of obstruction like benign or malignant tumours [18], foreign body [27] and broncholiths will be suggested by imaging and confirmed by bronchoscopy. Carcinoid tumour [28], bronchogenic carcinoma [29] and occasionally metastasis [30,31] can present similarly. Rare case reports available in the literature are with oat cell carcinoma [29], mucinous cystadenocarcinoma [32] can be present in this manner. Among airway tumours, neuroendocrine tumours (NET) have a significant association with bronchoceles when compared with salivary type malignancies [33].

Bronchorrhea is a condition characterized by watery sputum exceeding 100 ml per day [34]. Diverse conditions are associated with bronchorrhea like bronchial asthma [35], bronchoalveolar cell carcinoma [36,37], organophosphorus poisoning [38], infections like tuberculosis [39]. An interesting study by Smyrnis showed that copious sputum can be originated from the upper respiratory tract as seen in postnasal drip syndrome and term bronchorrhea should be used carefully for specific diagnoses in which sputum origin is from the lower respiratory tract [40]. Various mechanisms have been proposed like increased secretion by active ion transport by epithelial secretory cells, increased epithelial permeability leading to transudation of fluid, negative pressure from airway lumen and increased serous secretion from submucosal glands [35]. Chemically bronchorrhea sputum is characterized by low PH and high histamine concentration than mucoid sputum. Sputum of acute severe asthma with mucus plugging is characterized by highly mucin enriched [9]. However, these secretions are usually expectorated leaving little room to get impacted but can lead to bronchocele if conditions like dehydration predispose.

Another rare but grossly under-recognized condition that can present with excessive mucus is plastic bronchitis (PB). It's characterized by expectoration of bronchial casts. Consistency, cohesiveness, formation of bronchial casts differentiate this entity from other mucus pluggings. [41]. Usually seen in the paediatric population who underwent cardiac surgeries like Fontan's operation. Postoperative chylothorax, chest drainage duration at stage 2, postoperative chylothorax are considered as risk factors for developing PB. Elevated systemic venous pressures, lymphatic anomalies and inflammation have debated their role in PB [42]. Other aetiologies include viral infections, asthma, ABPA, cystic fibrosis, bronchiectasis, post coronary artery bypass graft (CABG), lung transplantation, silicosis etc. in adults. Pulmonary lymphatic abnormalities have been proposed to be the main abnormality in adults in contrast to the paediatric age group [43,44]. The content of casts is composed of fibrin, mucus, lymphatic fluid or inflammatory cells like eosinophils. This can be classified into two types based on the content of casts [45]:

1. Mucus with inflammatory cell content.
2. Pure mucin without inflammatory cells.

Plastic bronchitis is a clinical diagnosis. The patient can present with recurrent lobar or segmental collapse, respiratory distress. It can be fatal unless recognized. 'Lymphatic plastic bronchitis' has been proposed as a new terminology in adults as the majority were associated with pulmonary lymphatic flow abnormalities [46].

Classification of bronchoceles can be based on whether obstruction is present or not, mucus nature and whether bronchial dilatation is present or not.

Mechanical [47]:

1. Obstructive: congenital bronchial atresia, post stenotic due to tuberculosis, foreign body;
2. Non obstructive: asthma, cystic fibrosis.

Based on mucus characteristics on imaging:

1. Highly attenuated mucus: ABPA ;
2. Normal mucus density.

Based on mucus content:

1. Normal mucin;
2. Fibrin, mucin along with lymphocytes and macrophages as seen in lymphatic abnormalities;
3. Mucin /degenerated eosinophils as in ABPA [41].

Investigations

Hemogram, serum IgE levels, spirometry, bronchoprovocation testing, CFTR analysis may be needed according to the underlying causes of asthma, cystic fibrosis.

Imaging

Chest radiograph is an insensitive investigation although branching opacities can be appreciated as in ABPA. Indirect signs on chest radiographs include nodular density, branching density representing airways accumulated with mucus, the collapse of lobe or lung. CT-chest is the investigation of choice. Low-density nodules (Figures 1 and 2), masses, branching opacities, finger in glove appearance are classical findings [48]. In a study of cystic fibrosis, the density of bronchocele in the non-HAM group was 28HU [19]. Detailed characters of bronchi like wall thickening, bronchostenosis, bronchiectasis pattern and the most important characteristic-density of mucus itself-HAM (Figure 3) as defined by the density of more than paraspinal muscle density i.e. approximately 70HU [49]. Other conditions which can present with high attenuation densities are fresh alveolar haemorrhage and aspiration of contrast material. Hyperinflated lung distal to mucus is seen in congenital bronchial atresia. Double artery sign suggests mucus-filled bronchi and pulmonary vessel appearing as two vessels adjacently in lung window [50]. The radiological appearance of bronchocele can be deceiving sometimes since mucinous tumours with high mucous content and low cellularity and false-negative FDG PET scan can give a false assurance of benignity and can be easily missed out indicating the high index of suspicion is again necessary to rule out malignancy. Unexpected associations of prostatic and colorectal carcinoma metastases are available in the literature [30,31]. Calcification of a bronchocele is also not an abnormal finding [16,51]. As pulmonary arterial venous malformation(PAVM) also has a similar presentation in the form of branching opacity, it has to be considered as another differential diagnosis [48,52].

Malignancy is always a concern to be ruled out in bronchocele patients although malignant lesion itself has presented like bronchocele [31]. Bronchoscopy with analysis of bronchial washings

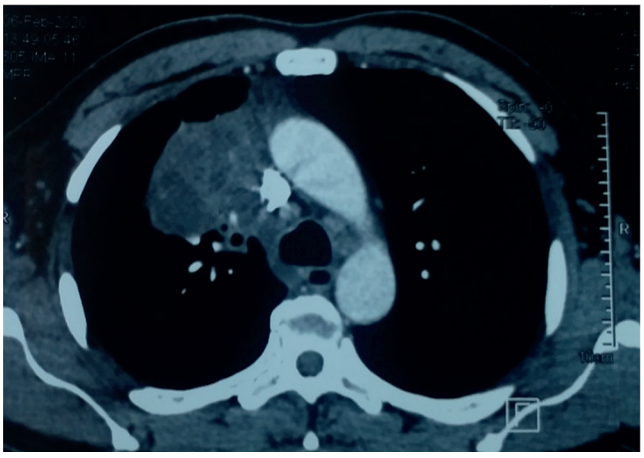

Figure 1. Right upper lobe lesion initially diagnosed as malignancy, later proved to be a bronchocele.

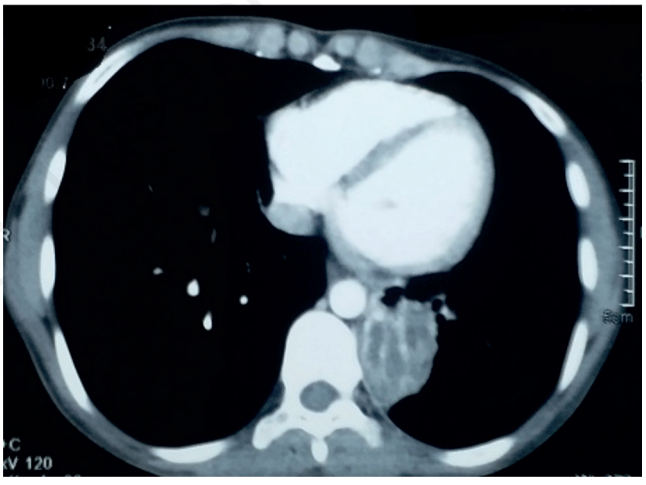

Figure 2. Left lower lobe mucocele.

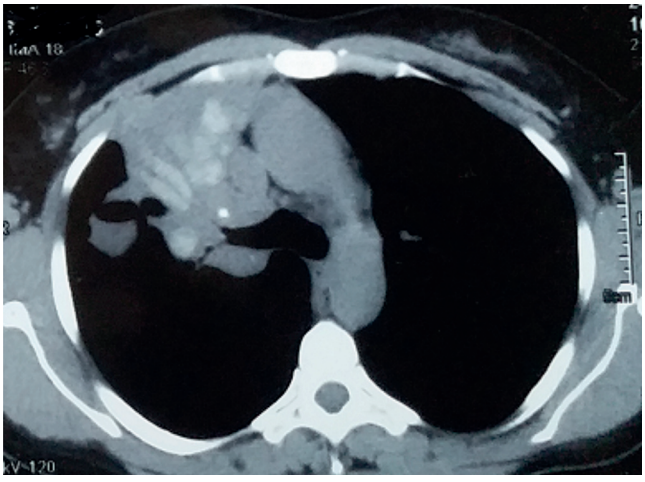

Figure 3. High attenuation mucus in the right upper lobe in a patient with ABPA.

and analysis of cellular material is essential to rule out malignancy, infections like tuberculosis. Calcified metastatic adenocarcinoma has been reported in one case report [18]. Plastic bronchitis is characterized by CT findings of centrilobular nodules, consolidation, atelectasis [41]. There may not be associated bronchiectasis in patients who underwent Fontan's operation [53]. Dynamic contrast-enhanced magnetic resonance (MR) lymphangiography (DCMRL) is in good agreement with intranodal lymphangiography in evaluating plastic bronchitis in adult patients [46]. MRI role is increasing in recent literature for prenatal diagnosis of CBA or other anomalies. No radiation with MRI is the advantage when compared to CT scans. MRI with a high T2 signal can differentiate from the tumour [54].

Bronchoscopy

Diagnostic bronchoscopy may show unexpected findings like foreign bodies [27]. Bronchostenosis can be confirmed by flexible bronchoscopy (Figure 4) and may provide material to rule out infections like tuberculosis. Rigid bronchoscopy may be needed to remove the foreign body, debulking of tumours and bronchoplasty [27].

Treatment

Depending on the underlying condition- inhaled corticosteroids, bronchodilators in asthma, oral steroids, antifungals in ABPA, aggressive chest physiotherapy along other medications are used for CF. Infections like endobronchial tuberculosis require antituberculous medication, along with steroids to prevent bronchial stenosis, or balloon bronchoscoplasty may be needed if the distal lung is functional and surgical resection may be needed if the parenchyma is dysfunctional. Underlying benign tumours may require bronchoscopic or surgical removal. Malignant neoplastic lesions may require chemo, radiotherapies and surgery.

Therapeutic bronchoscopy with cryo may be needed for the removal of casts in plastic bronchitis [41]. Thoracic duct embolization, ligation [46], pulmonary lymphatic intervention (PCL), alpha chymotrypsin [55] tissue plasminogen activator have been used in the treatment [56]. Bronchorrhea may respond to treatment of underlying conditions like malignancy and asthma. Various other treatment modalities like inhaled indomethacin [57,58], corticosteroids [37], octreotide [59], tyrosine kinase inhibitors [60], erythromycin [61], anticholinergics have been tried in bronchorrhea [62]. The antifungal agent [63] even radiation therapy also has been used as reported in one case report [64].

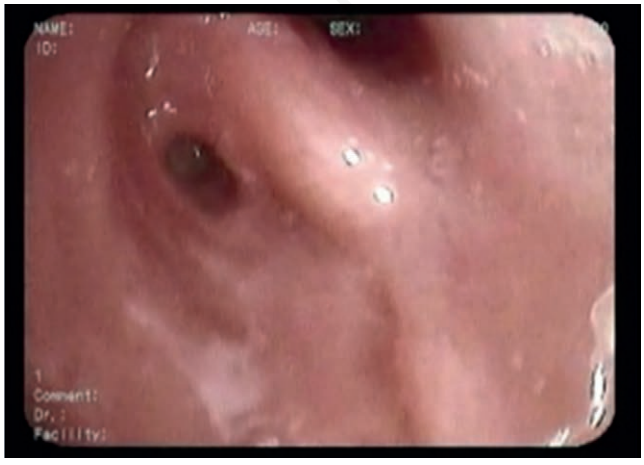

Figure 4. Bronchoscopy showing right upper lobe anterior segment stenosis.

Conclusions

Bronchocele can present as a result of abnormalities of production or drainage of mucus of the airways. Special types and situations like plastic bronchitis and bronchorrhea needs attention. The main concern is to rule out underlying neoplastic lesions or conditions predisposing to it. Underlying abnormality dictates management and prognosis.

Take-home messages

- Bronchocele is associated with excessive, abnormal mucus accumulation or impaired drainage caused by ciliary, airway or lymphatic abnormalities.
- Workup for associated lesions in bronchocele is necessary.
- Bronchocele itself can represent malignancy.
- Recognition of bronchocele may reduce the unnecessary investigations and apprehension to the patient.

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