Management of broncholithiasis

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Abstract: Broncholithiasis is a condition in which calcified material has entered the tracheobronchial tree, at times causing airway obstruction and inflammation. Broncholiths generally originate as calcified material in mediastinal lymph nodes that subsequently erode into adjacent airways, often as a result of prior granulomatous infection. Disease manifestations range from asymptomatic stones in the airway to life-threatening complications, including massive hemoptysis and post-obstructive pneumonia. Radiographic imaging, particularly computed tomography scanning of the chest, is integral in the evaluation of suspected broncholithiasis and can be helpful to assess involvement of adjacent structures, including vasculature, prior to any planned intervention. Management strategies largely depend on the severity of disease. Observation is warranted in asymptomatic cases, while therapeutic bronchoscopy and surgical interventions may be necessary for cases involving complications. Bronchoscopic extraction is often feasible in cases in which the broncholith is freely mobile within the airway, whereas partially-embedded broncholiths represent additional challenges. Surgical intervention is indicated for advanced cases deemed not amenable to endoscopic management. Complex cases involving complications such as massive hemoptysis and/or bronchomediastinal fistula formation are best managed with a multidisciplinary approach, utilizing expertise from fields such as pulmonology, radiology, and thoracic surgery.

Keywords: Surgical management; bronchoscopy; broncholithiasis

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Overview

Broncholithiasis refers to the presence of calcified material that erodes into the lumen of the tracheobronchial tree or lung parenchyma, potentially causing inflammation and obstruction (1). Consequences from broncholithiasis range widely from being asymptomatic to life threatening such as recurrent pneumonias, hemoptysis, and bronchoesophageal fistula. Management varies depending upon several factors including patient symptoms, associated lung disease, and mobility of the broncholith within the airway, but typically involves either observation or intervention via bronchoscopy or thoracic surgery. This paper reviews the etiology, clinical presentation, radiologic appearance, and management of broncholithiasis.

Definition and etiology

The term broncholithiasis is the condition that arises when a broncholith causes airway irritation, obstruction, or erosion into adjacent structures (2). The vast majority of broncholiths are formed by the partial or complete erosion

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Publication Patients Etiology Clinical presentation Radiographic findings Bronchiectasis (n=28, 100%) Kennedy 28 Primary ciliary Lithoptysis (n=5, 18%) 2007 (8) dyskinesia Tsubochi 2 Fungal infection Hemoptysis (n=2, 100%) Consolidation (n=2, 100%), bronchiectasis (n=1, 2007 (9) (aspergillosis and 50%) actinomycosis) Ford 9 Not specified Cough after drinking (n=8, 89%), Esophageal radiocontrast studies showed fistula 2005 (10) dysphagia (n=1, 11%), lithoptysis (n=6, (n=9) 66%), recurrent pneumonia (n=6, 66%) Kim 9 Fungal infection Cough (n=8, 89%), sputum production Distal obstruction (n=8, 89%) atelectasis (n=2, 22%), 2005 (11) (actinomycosis) (n=8, 89%), hemoptysis (n=5, 55%) central necrosis within consolidation (n=4, 44%), cavitary lesion (n=3, 33%), airway narrowing (n=2, 22%), airway obstruction (n=7, 78%), bronchiectasis (n=4, 44%), pleural thickening (n=2, 22%)Conces 15 Not specified Cough (n=13, 87%), hemoptysis (n=6, Atelectasis (n=11, 73%), distal obstruction (n=4, 1991 (12) 40%), chest pain (n=3, 20%), sputum 27%), bronchiectasis (n=4, 27%), air trapping (n=1, production (n=3, 20%), fever (n=2, 7%) 13%), lithoptysis (n=2, 13%), halitosis (n=1, 7%), wheezing (n=1, 7%)

Table 1 Summar	v of publication	s of clinical p	presentations and	radiographic	findings in bror	ncholithiasis

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of an adjacent calcified peribronchial lymph node into the airway lumen. Erosion of a broncholith into the airway is thought to result from the repeated physical contact of a calcified lymph node with an adjacent bronchus during normal respiratory motion (1). Nodal calcification in the thorax most often occurs from long-standing lymphadenitis related to fungal or mycobacterial infections, with Histoplasma capsulatum representing the most common causative organism in the United States. Less common infectious etiologies include tuberculosis, actinomycosis, coccidioidomycosis, and cryptococcosis (3). Due to airway anatomy and normal lymph node distribution, the most commonly affected bronchi are the proximal right middle lobe bronchus, the bronchus intermedius, and the origin of the anterior segment right upper lobe bronchus, in order of frequency (1,3-5).

Other etiologies of broncholithiasis are less common. Broncholiths can arise as extrinsic calculi from aspirated foreign material (4), such as aspirated fragments of bone or foreign debris which calcifies *in situ* within the airway over time. Calcified cartilage rings in the airway wall can also extrude and become sequestered inside the lumen of the bronchus forming broncholiths. Calcified material from remote locations within the thorax, such as fragments of calcified pleural plaques, can migrate over time and eventually erode into a bronchus (6). Extremely rare cases of calcified endobronchial hamartomas and carcinoid tumors, primary ciliary dyskinesia-associated broncholiths, and silicosis-associated egg shell lymph node broncholiths have also been reported (6-8).

Clinical presentation, symptoms, and radiographic findings

The severity and type of patients' symptoms are typically related to the degree of bronchial inflammation, obstruction, or involvement of adjacent structures (*Tables 1-3*). Given that the most common symptom is chronic cough, most patients present in the outpatient setting (3). Cough associated with broncholithiasis can be dry or productive of mucous, purulence, or "gravel". The latter symptom, termed lithoptysis, occurs when fragments of the broncholith are expectorated. Symptoms or conditions that may necessitate inpatient care include pneumonia, hemoptysis, and the development of fistulae between the bronchi and mediastinal structures (1).

While routine chest radiographs can reveal findings of hilar calcifications or parenchymal infiltrates (3), they lack the resolution to identify the causative broncholith (*Figure 1A*). On occasion, a broncholith cannot be identified even on diagnostic bronchoscopy due to partial erosion of the node into the airway, with a reported incidence

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Publication	Patients	Procedures	Type of bronchoscopy	Outcomes	Complications
Cerfolio 2008 (1)	50	38	Rigid	29 attempted, 29 removed (100%)	None reported
Menivale 2005 (13)	4	4	Flexible	2 attempted, 2 removed (100%)	None reported
Yi 2005 (14)	2	2	Flexible	2 attempted, 2 removed	None reported
Olson 1999 (15)	95	95 (127 broncholiths identified)	Flexible and Rigid	71 attempted, 46 removed (65%) overall; 67% with rigid, 30% with flexible	Hemorrhage (n=1, 1.4%) broncholith dislodgement (n=1, 1.4%)
Cole 1986 (16)	42	40	Flexible and Rigid	8 (19%) removed; attempts not reported; 75% with rigid	None reported
Dixon 1984 (3)	19	18	Flexible and Rigid	8 attempted, 4 removed (50%); (75%) with rigid	None reported
Trastek 1985 (17)	52	12	Unavailable	12 attempted, 8 removed; additional 40 patients treated surgically	Unavailable
Moersch 1959 (18)	87	33	Unavailable	33 attempted; success rate unknown	Unavailable

Table 2 Maio	r series of	bronchosconic	management of	broncholithiasis
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Table 3 Major series of surgical management of broncholithiasis

Publication	Patients	Etiology	Type of surgery	Complications
Hammoud 2009 (19)	13	Fixed broncholiths, recurrent pneumonia and/or hemoptysis from histoplasmosis	Lobectomy (n=7, 54%); Bilobectomy (n=5, 38%); transbronchial excision of LN (n=1; 8%)	Air leak (n=1, 7.7%); empyema (n=1, 7.7%)
Cerfolio 2008 (1)	5	BE fistula (n=2, 40%); fixed broncholith (n=3, 60%)	Thoracotomy with LN curettage and/or removal (n=5, 100%)	None reported
Potaris 2000 (20)	47	Hemoptysis (n=21, 45%); recurrent pneumonia (n=11, 23%); suspected BE fistula (n=5, 11%); suspected malignancy (n=6, 13%); failed bronchoscopy (n=4, 9%)	Lobectomy (n=16, 34%); bilobectomy (n=4, 8.5%); segmentectomy (n=7, 15%); wedge resection (n=2, 4%); pneumonectomy (n=1, 2%); broncholithectomy with pulmonary resection (n=30, 64%); broncholithectomy only (n=9, 19%); broncholithectomy with bronchoplasty (n=7; 15%); sleeve resection of left mainstem (n=1, 2%)	Intraop (n=6, 12.8%): PA laceration (n=4, 8.5%); esophageal laceration (n=1, 2.1%); mainstem bronchus laceration (n=1, 2.1%); postop (n=16, 34%): hemothorax (n=2); empyema (n=2); wound dehiscence (n=1); PA thrombosis (n=1); lingua necrosis (n=1); respiratory insufficiency (n=3); confusion (n=2); prolonged air leak (n=2); pneumothorax (n=1); air atrial fibrillation (n=1);
Cole 1986 (16)	25	Fixed broncholiths, symptoms after bronchoscopy, hemoptysis from histoplasmosis, aspergillosis, and tuberculosis	Thoracotomy (n=26, 100%); segmental resection (n=11, 42%), lobectomy (n=11, 42%), bronchotomy with broncholithectomy (n=3, 12%), tracheoesophageal fistula repair (n=1, 4%)	None reported

BE, bronchoesophageal; LN, lymph node; PA, pulmonary artery.

ranging from 28–56% of cases (3,12,17,21). Computed tomography (CT) therefore plays a crucial role in the initial diagnosis, accurately identifying the specific location of

the broncholith, the degree of endobronchial obstruction, and the presence of associated findings such as atelectasis, consolidation, bronchiectasis, air trapping, mucoid impaction

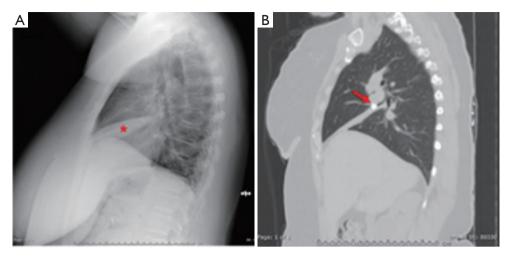


Figure 1 Comparison of plain radiograph to computed tomography (CT) scan in the same patient with broncholithiasis. (A) Lateral chest radiograph demonstrates right middle lobe atelectasis (asterisk); (B) CT scan in sagittal reformat reveals the causative broncholith (arrow) obstructing the proximal right middle lobe bronchus.

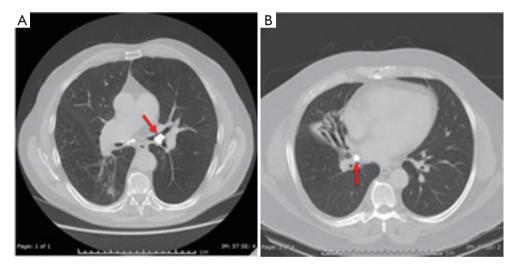


Figure 2 Common CT imaging findings in patients with broncholithiasis. (A) Axial chest CT image demonstrates a broncholith (arrow) in the left mainstem bronchus without parenchymal involvement; (B) axial chest CT image of a different patient demonstrates a broncholith (arrow) at the origin of the right middle lobe bronchus with associated bronchiectasis and parenchymal atrophy.

of a bronchus, or frank lung destruction (*Figure 1B* and *Figure 2*) CT imaging may also show other sequelae of long standing lymphadenitis or granulomatous disease, including mediastinal calcifications, nodules, and scarring.

Management

Conservative

Management of broncholithiasis depends on several factors

including patient symptoms, the size of the broncholith, its relationship with the airway wall, and its proximity to or involvement of adjacent thoracic structures. Indications for bronchoscopic or surgical intervention include symptoms related to intractable cough, recurrent pulmonary infections, symptomatic bronchiectasis, hemoptysis, and fistula formation. In general, conservative management in the form of observation is warranted in asymptomatic patients and those with minimal or non-recurring, non-life-threatening

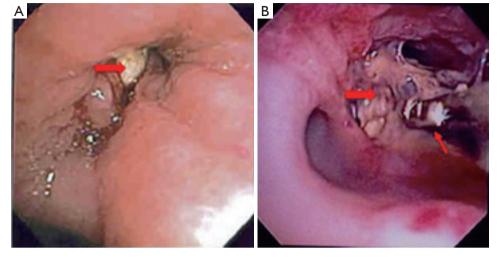


Figure 3 Bronchoscopic visualization of broncholiths. (A) Bronchoscopic visualization of broncholith in right mainstem bronchus (thick red arrow) with associated mucosal edema; (B) forceps extraction (thin red arrow) of broncholith (thick red arrow) during bronchoscopic intervention.

symptoms. Also, patients with significant comorbidities or limited life span who may not tolerate invasive therapy are candidates for conservative therapy. Conservative measures to consider include cough suppression, bronchodilators, and empiric antimicrobial therapy. If sputum gram stain and culture or other laboratory testing identifies a specific infection, then antimicrobial therapy can be tailored accordingly.

Bronchoscopy

Bronchoscopy has been described for both diagnostic and potentially therapeutic purposes in the symptomatic patient (22). When broncholithiasis is suspected based on clinical and radiographic findings, flexible bronchoscopy is often employed to confirm the diagnosis by direct visualization of a calcified stone in the airway (3,15). The typical broncholith appears as a yellow or white concretion, sometimes with areas of dark discoloration (*Figure 3A*). When tapped with forceps through the working channel of the bronchoscope, it has a hard texture and may be either mobile or fixed. The adjacent airway mucosa is often inflamed and friable with blood oozing when touched. Granulation tissue may also be present. When the broncholith cannot be identified, evidence of airway distortion or inflammatory obstruction is typically observed.

In addition to confirming the diagnosis, bronchoscopy can aspirate secretions for therapeutic purposes and obtain cultures which may guide antibiotic therapy. During bronchoscopy, the degree of broncholith mobility should be assessed, as this may dictate further management. It is, however, occasionally difficult to assess the degree of mobility as well as any involvement of important structures beyond the bronchial wall, such as pulmonary vasculature. This underscores the need for CT imaging with intravenous contrast prior to any attempt at bronchoscopic removal with attention directed at identifying major vascular structures in the vicinity of the broncholith. Saline irrigation and forceps manipulation during bronchoscopy have been described as methods to assess the degree of attachment (13,16) (*Figure 3B*). Use of topical lidocaine and epinephrine have been described to minimize bronchial wall bleeding during broncholith manipulation (23).

The pathophysiology of broncholithiasis represents a distinct entity when compared to other forms of foreign bodies in the airways (19). As such, bronchoscopic management may differ from traditional forms of foreign body removal where extraction using flexible or rigid bronchoscopy is often feasible (17). Historically, bronchoscopic broncholithectomy was not recommended due to concern of complications including bleeding, displacement of the broncholith causing obstruction, or iatrogenic creation of a fistula. The role of therapeutic bronchoscopy in the context of broncholithiasis has evolved however to include select cases (2). Olson *et al.* reported on 48 patients who underwent bronchoscopic removal with only a 4% complication rate (15). The authors determined that mobile broncholiths could be safely removed, as well

as some partially-eroding broncholiths. In this series, there was a high risk of complications, particularly hemorrhage, when broncholiths were partially embedded in the bronchial wall (15). Cerfolio and colleagues reported their experience in 50 patients with broncholithiasis (1). The authors reached a similar conclusion that broncholiths which are mobile can be safely removed with bronchoscopy, whereas surgery is typically indicated for symptomatic patients not amenable to bronchoscopic treatment. In summary, retrospective studies support an initial attempt at bronchoscopic removal in cases where broncholiths are mobile or loosely attached to the bronchial wall.

During bronchoscopy, if forceps extraction is not readily feasible, more investigational options are available to facilitate removal. The use of a balloon catheter has been described for removal of a partially eroded broncholith (14). With this technique, the balloon is inserted distal to the stone. The balloon is then inflated and pulled back in an attempt to dislodge the stone proximally. This technique mimics that where a closed forcep is passed beyond the broncholith, opened, and then retracted. Stone fragmentation which facilitates extraction using holmiumvttrium aluminum garnet (Ho:YAG) (24) and neodymiumdoped yttrium aluminum garnet (Nd:YAG) (15,25) lasers has been reported. Bronchoscopic lithotripsy similar to that used in nephrolithiasis has also been reported (26). More recently, the use of a cryotherapy probe for broncholithectomy was described. Some have advocated the use of rigid bronchoscopy with larger working channels (1) to facilitate stone extraction, however, this modality is generally now reserved for very select cases in which flexible bronchoscopy has failed (15). Use of a pediatric bronchoscope has been described to reach endobronchial lesions in the distal airways (27) Lastly, the use of a covered self-expandable metallic stent has been described in a case in which the broncholith was associated with a bronchoesophageal fistula (28). In a separate but similar case, endoscopic placement of an esophageal stent was described for a poor surgical candidate with bronchoesophageal fistula (29).

In the patient presenting with hemoptysis as a complication of broncholithiasis, diagnostic bronchoscopy should be pursued to identify the location of bleeding and etiology of hemoptysis, if possible. The decision to proceed with observation versus broncholith removal, however, should be based on the frequency and quantity of hemoptysis coupled with the predicted ease of broncholith removal. There are published reports of massive hemoptysis caused by broncholithiasis (30); however, management of bleeding due to bronchoscopic removal of broncholiths has scarcely been described. Techniques that have been described for other etiologies of hemoptysis may be useful in these cases. Placement in the lateral decubitus position, with the affected side down, has been proposed, as has the use of selective mainstem bronchus intubation (31). Likewise, insertion of a balloon catheter adjacent to the endotracheal tube to tamponade the site of bleeding has been described (31). These should be regarded as temporizing measures while definitive therapy is planned. Flexible or rigid bronchoscopy with various methods of cautery may be of some use. The use of endobronchial hemostatic devices placed via bronchoscopy has been described but has yet to achieve widespread adoption (32). Bronchial artery embolization or thoracic surgery is required for definitive therapy (33,34).

In addition to hemorrhage, complications of bronchoscopic broncholithectomy include airway obstruction due to stone dislodgement, fistula creation, and damage to the bronchial wall (15,35). On occasion, attempts at stone extraction can result in inadvertent advancement of the broncholith with distal airway occlusion, necessitating further intervention (23). Cases involving embedded broncholiths may lead to formation of bronchomediastinal or bronchoesophageal fistulae when the stone is removed (28,36). Table 2 summarizes published series on bronchoscopic management of broncholithiasis. The highest reported success rates approach 100% for freely mobile broncholiths. The success rate of intervening on partially eroded or fixed broncholiths ranges from 0-48% (1,15). Bronchoscopic interventions for adherent broncholiths should only be attempted by experienced operators at specialized centers with appropriate interventional radiology and thoracic surgery expertise in case of serious complications. Moreover, in an otherwise healthy patient with significant distal lung destruction such as bronchiectasis or fistulization into an adjacent structure, high-risk bronchoscopic procedures should be avoided in deference of surgical intervention.

Surgery

Surgical intervention is indicated for symptomatic patients without precluding comorbidities in whom therapeutic bronchoscopy is unable to relieve the airway obstruction. These patients often suffer from complications related to recurrent pneumonia, hemoptysis, substantial distal bronchiectasis or parenchymal destruction, or fistulae between the airway and mediastinal structures (*Figure 4*) (13). While there is current enthusiasm to

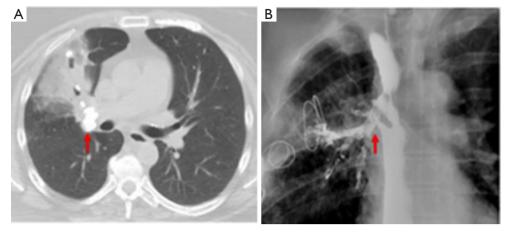


Figure 4 Advanced complications of broncholithiasis that required surgery. (A) Axial CT image of an otherwise healthy patient with several calcified lymph nodes surrounding the distal bronchus intermedius (arrow) with complete obstruction of the right middle lobe airway and secondary bronchiectasis and pneumonitis that was treated with thoracotomy and bilobectomy; (B) a separate case demonstrating a bronchoesophageal fistula (arrow) seen on gastrografin esophagram that developed after surgical removal of a broncholith in the bronchus intermedius. This case required a second thoracotomy with esophageal repair.

perform pulmonary surgery using minimally invasive approaches such as thoracoscopy or robotic technology, calcific fibrosis surrounding airway and pulmonary vasculature typically requires an open approach for the majority of these cases. Occasionally broncholiths can be removed through a small bronchotomy, followed by repair of the airway and takedown of any fistula if present, with interposition of pedicled soft tissue between the two structures, sparing normal pulmonary parenchyma (1).

An anatomic pulmonary resection is usually required for broncholiths causing hemoptysis or chronic obstruction with resultant end stage parenchymal damage (Figure 2B and Figure 4) Inflammation and dense fibrosis are frequently encountered during lobar hilar dissection, which makes branch pulmonary artery isolation and division risky. In these cases, initial placement of an umbilical tape proximally around the main pulmonary artery prior to lobar hilar dissection will allow rapid control of inadvertent bleeding. In cases where the lobar bronchus and pulmonary branch vessels are densely adherent, occluding the main pulmonary artery then amputating the lobar hilum for initial lung removal, usually allows safe separation of the airway and vessels followed by independent closure. Finally, parenchymal sparing sleeve resections for patients with significant involvement of the right upper lobe hilum can be utilized to avoid pneumonectomy.

Table 3 summarizes the publications on surgical management of broncholithiasis. Potaris and colleagues

reported their experience with 47 patients with broncholithiasis requiring surgical intervention (20). The majority of patients were found to have right-sided lesions. Pulmonary resection was required in 64% of patients, while 34% underwent broncholithectomy without pulmonary resection. We previously reported our experience in 49 patients who underwent surgery for complications of histoplasmosis, 13 of which had broncholithiasis (19) and were not candidates for bronchoscopic removal due to broncholith adherence. Indications for surgery included either recurrent pneumonia or hemoptysis. Twelve of these patients underwent lobectomy or bilobectomy. In Cerfolio's series, only 5 of 50 patients underwent surgery with thoracotomy and lymph node curettage, removal, or both (1). Potaris and colleagues documented the largest number of surgical cases and reports an intraoperative complication rate of 12.8% and a major postoperative complication rate of 15% (20). While the reported surgical morbidity appears high, to date there have been no operative deaths in published series. Given the potential for complications and the small but definite risk of recurrent broncholithiasis (20), surgery should be reserved for cases not amenable to conservative or bronchoscopic management.

Conclusions

The management of broncholithiasis can be challenging and should be individualized for symptomatic patients. A

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multidisciplinary approach with assessment by thoracic radiologists, interventional pulmonologists, and thoracic surgeons prior to implementing any treatment strategy is usually optimal for patient care. In cases of mobile or loosely attached broncholiths, therapeutic bronchoscopy is often effective with minimal risks. Bronchoscopic intervention in cases of partially eroded broncholiths does present risks of hemorrhage, broncholith dislodgement, and fistula formation, but can be considered by experienced operators at specialized centers capable of managing potential complications. In patients with acceptable operative risk levels, surgery remains the mainstay of treatment in cases with significant distal parenchymal destruction, severe hemoptysis, or where bronchoscopic intervention failed.

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None.

Footnote

Conflicts of Interest: The authors have no conflicts of interest to declare.

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