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CASE REPORT

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Pulmonary actinomycosis and polymicrobial empyema in a patient with ABPA and bronchocoele

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Abstract

We present a 43-year-old woman, with a history of allergic bronchopulmonary aspergillosis and a chronic bronchocoele, who was admitted to hospital with an infection of the bronchocoele, progressing to a pulmonary abscess and polymicrobial empyema, following dental extraction and regular *Lactobacillus* probiotic ingestion. Interval chest imaging following this procedure demonstrated worsening right upper lobe opacities and a right-sided pleural effusion. Bronchoscopies identified copious mucoid secretions and an infected bronchocoele with a right upper lobe airways impaction. Oral cavity organisms including *Actinomyces odontolyticus* were cultured on bronchial washings. *Streptococcus mitis* and *Lactobacillus rhamnosus* were cultured in pleural fluid. Treatment with endoscopic mucoid secretion suctioning; intercostal catheter insertion and therapeutic drainage; and antibiotic, glucocorticoid and anti-IgE therapy resulted in clinical and radiological improvement. Our case illustrates the potential pulmonary complications from oral cavity organisms following tooth extraction and probiotic use in patients with chronic lung disease associated with mucoid lesions and airways obstruction.

KEYWORDS
ABPA, *Actinomyces odontolyticus*, bronchiectasis, *Lactobacillus rhamnosus*, *Streptococcus mitis*

INTRODUCTION

Actinomyces odontolyticus, *Lactobacillus rhamnosus* and *Streptococcus mitis* are rare causes for pulmonary infections and are mostly reported in immunocompromised patients or those with a history of untreated periodontal disease. To our knowledge, we describe the first case of a patient with a history of allergic bronchopulmonary aspergillosis (ABPA) and a chronic bronchocoele, who subsequently developed an infected bronchocoele, complicated by pulmonary actinomycosis and *L. rhamnosus* and *S. mitis* empyema following tooth extraction and regular *Lactobacillus* probiotic use.

CASE REPORT

A 43-year-old woman was hospitalized after presenting with fevers, right-sided pleuritic chest pain and productive cough. Her medical history included previously treated pulmonary tuberculosis; childhood atopic asthma; and right upper lobe, right middle lobe and lingula lobe bronchiectasis. Prior spirometry and transfer factor for carbon monoxide (TLCO) were within normal limits with no significant bronchodilator response. Chest imaging performed 4 years prior to her admission (Figure 1A) demonstrated bronchiectasis and an uncomplicated bronchocoele in the right upper lobe. Twelve months prior to admission, eosinophil count was 1.02×10^9/L, total immunoglobulin E (IgE) level was 1070 IU/ml and radioallergosorbent (RAST) test to *Aspergillus* was positive. Her background symptoms (i.e., cough and exertional dyspnoea) were generally well controlled on regular combined fluticasone propionate/salmeterol 250 μg/50 μg inhaler. She denied any history of immunodeficiency, alcoholism, aspiration or gastroesophageal reflux disease.

The hospitalization followed a dental extraction 4 months prior for a cracked tooth. She was also regularly

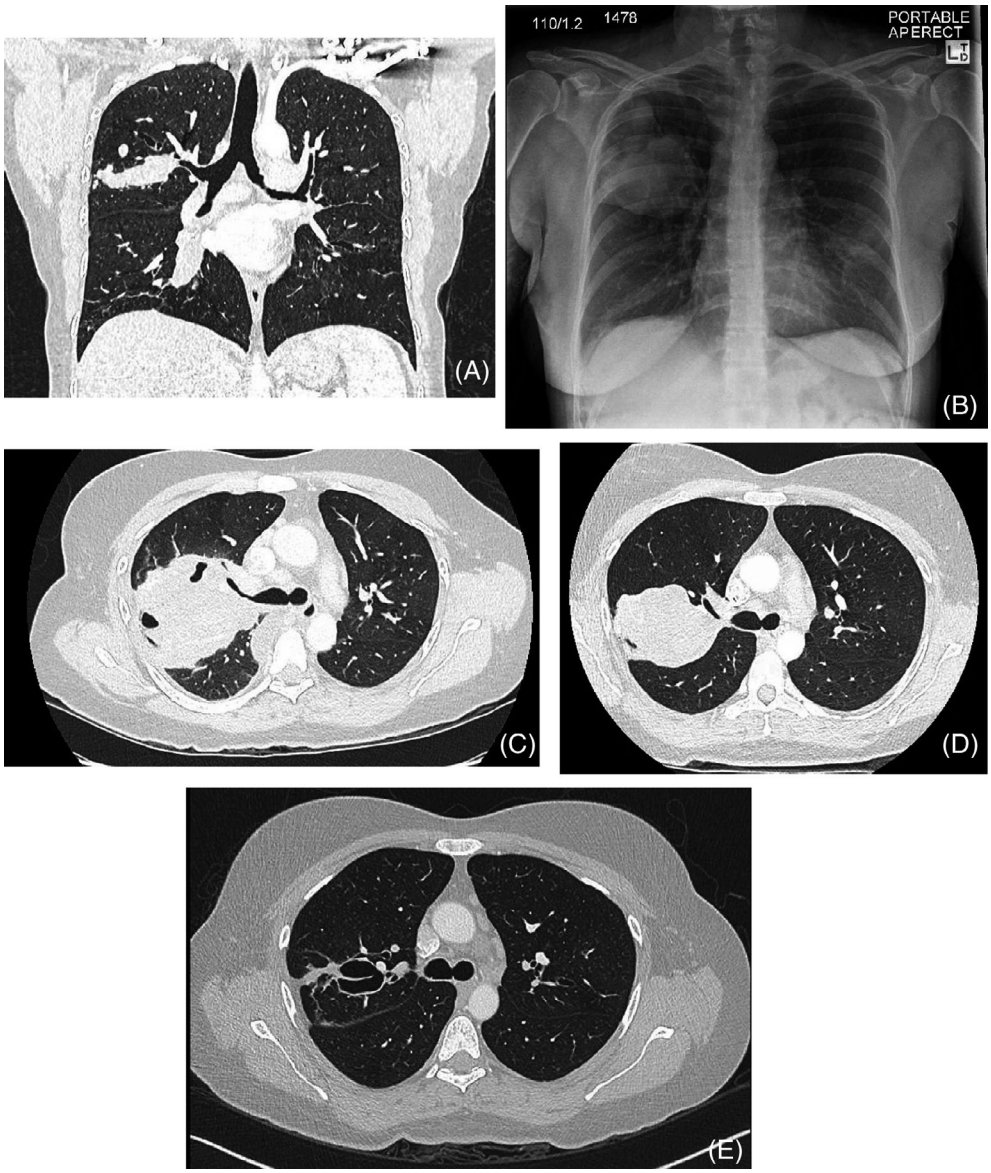

FIGURE 1 (A) Chest computed tomography (CT) scan. Presence of pre-existing bronchocoele in the right upper lobe 4 years prior to hospital presentation. (B) Frontal chest radiograph performed at the time of initial presentation demonstrating worsening of the right upper lobe opacity and existing bronchocoele following recent tooth extraction and probiotic use. (C–E) Serial chest CT (axial imaging—lung windows). (C) Worsening of the impacted bronchocoele at the time of initial presentation following tooth extraction and probiotic use. (D) Further progression of the impacted bronchocoele post-outpatient bronchoscopy. (E) Resolution of the impacted bronchocoele 12 months following hospitalization

ingesting *Lactobacillus* probiotic capsules. Chest imaging demonstrated significant enlargement of the known right upper lobe opacity, with another small bronchocoele in the lingula (Figure 1B,C). She was admitted for 2 days and treated with 9 days of empiric cephalosporins and discharged home for an urgent outpatient bronchoscopy with a weaning regimen of prednisolone, commencing at a dose of 37.5 mg daily over a 2-week period. The bronchoscopy performed 7 days following her hospital discharge identified impacted mucoid plugs in the right upper lobe and lingula. The lingula plug was successfully aspirated endoscopically with drainage of purulent secretions. The tenacious right upper lobe mucoid plug was unable to be aspirated. A right upper lobe endobronchial biopsy was performed. *Aspergillus fumigatus* was cultured on bronchial washings and *Aspergillus niger* complex and *A. odontolyticus* were cultured from the tissue sample.

Eight days following bronchoscopy, she represented to the emergency department with recurrent productive cough, pleuritic chest pain and fevers. She was afebrile, haemodynamically stable and had a respiratory rate of 18 breaths per minute with oxygen saturations of 96% on room air. Chest examination was unremarkable. Follow-up chest x-ray demonstrated persistence of the right upper lobe opacity, unchanged from her chest imaging performed at the time of initial presentation. Laboratory investigations

TABLE 1 Summary of data on reported cases of intrathoracic *Actinomyces odontolyticus*, *Lactobacillus rhamnosus* and *Streptococcus mitis* infection

Reference	Disease(s)	Age (years)/ sex	Underlying condition(s)	Presentation	Chest radiograph finding(s)	Diagnostic procedure
Pulmonary actinomycosis caused by *A. odontolyticus*						
8	Lung abscess	61/F	Rheumatoid arthritis, corticosteroid therapy	Fever, chest pain, dyspnoea	Pleural effusion, cavitating lesion	Abscess culture
9	Pneumonia	61/M	Lung transplant, immunosuppression	Chest pain	LUL infiltrate	Bronchoscopy brush culture
9	Mediastinitis	43/M	Heart-lung transplant, immunosuppression	Post-operative sternal wound	Bi-basilar infiltrate	Wound culture
10	Empyema	38/F	Periodontal disease	Weight loss, fever, chest pain, cough, dyspnoea	Pleural effusion	Pleural fluid culture
11	Pneumonia	52/F	Bronchiectasis	Weight loss, fever	LUL infiltrate with cavitation	Sputum culture, lung granule
12	Pneumonia, skin abscess	52/M	Alcoholism, periodontal disease	Weight loss, fever, cutaneous drainage	B/L cavitating apical infiltrates, pleural thickening	Abscess culture
12	Empyema necessitates	50/M	S/P pneumonectomy for aspergilloma, ETOH use, pulmonary TB	Fever, chest pain, dyspnoea	Left pleural empyema	Pleural fluid culture
14	Pericardial + pleural effusion	68/M	S/P resection of gastric polyp	Dyspnoea, fever	Pericardial + pleural effusion	Pericardial fluid culture
14	Chest wall erosion, spinal and calf abscesses, pleural effusion	58/F	Dental plate	Weight loss, fever, chest pain	Left anterior mid-lung shadow	Chest wall biopsy culture
14	Pneumonia, empyema	40/M	Alcoholism, smoker	Fever, chest pain, productive cough	RUL infiltrate, pleural effusion	Pleural fluid culture
15	Lung abscess	49/M	Alcoholism, smoker	Dyspnoea	Pleural effusion	Pleural fluid culture
16	Lung abscess	37/F	Sarcoidosis, PNL, newly diagnosed diffuse large B-cell lymphoma	Dyspnoea, fever, dry cough	LLL infiltrate, right lung mass, right hilar mass	Abscess culture
17	Lung abscess	64/F	Periodontal disease, appendicitis (age 33)	Fever, chest pain, bloody sputum	RML nodular shadow	Pleural fluid culture
18	Pneumonia	34/M	S/P gastric polypectomy, dental caries	Cough, sputum	LUL cavitating lesion	Bronchial washings
19	Pleural effusion	65/M	Smoker, periodontal disease, alcoholism	Cough, sputum, fever, dyspnoea	Pleural effusion, RUL + LUL consolidation	Bronchial washings
20	Lung abscess	60/M	Smoker, newly diagnosed lung squamous cell carcinoma	Hoarseness	Left hilum mass, LUL cavitating lesion, mediastinal lymphadenopathy	Bronchial washings
21	Pneumonia	69/M	Periodontal disease, renal transplant	Unknown	Unknown	Bronchial washings
22	Pneumonia	43/M	Asthma, chronic eosinophilic pneumonia	Catarrh, dyspnoea	Pulmonary infiltrates, left pleural effusion	Endobronchial biopsy culture, bronchial washings
23	Pneumonia	38/M	Smoker, newly diagnosed Hodgkin's lymphoma	Sputum, cough	LUL infiltrate	Bronchial washings
24	Empyema	68/M	Smoker, periodontal disease, alcoholism	Fever, chest pain, dyspnoea, sputum, cough	Right-sided consolidation, pleural effusion	Pleural fluid culture
25	Empyema	59/M	Pulmonary TB, asthma, obesity, periodontal disease	Cough, sputum chest pain, fever	Pleural effusion	Pleural fluid culture

(Continues)

TABLE 1 (Continued)

Reference	Disease(s)	Age (years)/ sex	Underlying condition(s)	Presentation	Chest radiograph finding(s)	Diagnostic procedure
Pulmonary infection caused by *L. rhamnosus*						
26	Lung abscess	79/M	COPD, dental caries	Chest pain, fever	Cavitating lung mass in the left lingula	Pleural fluid culture
Pulmonary infections caused by *S. mitis*						
27	Lung abscess	48/M	Smoker, chronic bronchitis	Fever, malaise, cough, sputum, haemoptysis, chest pain	RUL + LLL cavitating lung lesion	Abscess culture
28	Pneumonia, bacteraemia	Three cases varied	Cancer	Varied	Varied	Varied
29	Community-acquired lung abscess	84 cases varied	Varied	Varied	Varied	Varied
Pulmonary actinomycosis caused by *A. odontolyticus*, complicated by *L. rhamnosus* and *S. mitis*						
PR	Lung abscess, ABPA exacerbation	43/F	ABPA, asthma, pulmonary TB, bronchiectasis	Chest pain, fever, sputum	Pleural effusion, RUL infiltrate	Bronchial washings and pleural fluid culture

Abbreviations: ABPA, allergic bronchopulmonary aspergillosis; B/L, bilateral; COPD, chronic obstructive pulmonary disease; ETOH, alcohol; LLL, left lower lobe; LUL, left upper lobe; PNL, prednisolone; PR, present report; RML, right middle lobe; RUL, right upper lobe; S/P, status post; TB, tuberculosis.

revealed a C-reactive protein (CRP) of 102 mg/L and white blood cell (WBC) count of 14.7 × 10^9/L with neutrophilia of 12.4 × 10^9/L. She was hospitalized and commenced on intravenous ceftriaxone and azithromycin and maintained on oral prednisolone 25 mg.

Repeat computed tomography (CT) of the chest demonstrated significant enlargement of the obstructed bronchocoele in the right upper lobe with radiological evidence of a lung abscess, complicated by a right-sided empyema (Figure 1D). Treatment was broadened to piperacillin-tazobactam after several days with persistent fevers and elevated inflammatory markers (peak CRP of 451 mg/L and WBC count of 15.7 × 10^9/L). A small-bore intercostal catheter was inserted for therapeutic drainage of the right pleural effusion. Approximately 300 ml of brown pus was drained. Oral organisms, *L. rhamnosus* and *S. mitis*, were cultured on the pleural fluid. Biochemistry post-procedure revealed a CRP of 244 mg/L and WBC count of 10.0 × 10^9/L. Repeat bronchoscopy performed on day 12 of admission following a longer course of steroids showed that the previously impacted right upper lobe mucoid plugs were no longer occlusive and mucopurulent secretions were now draining from the right upper lobe. The oral cavity organism, *A. odontolyticus*, was again cultured on bronchial washings and antimicrobial therapy was subsequently changed to intravenous benzylpenicillin and teicoplanin. As *Aspergillus* species was previously cultured on bronchial washings and tissue sample, empiric voriconazole was also commenced. A third bronchoscopy was performed on day 19 of admission due to poor radiological resolution of the right upper lobe abscess. During this procedure, the previously occluded right upper lobe segment remained patent and only normal upper respiratory flora were cultured from these bronchial washings. Interval chest CT imaging performed on day 22 of admission revealed the right upper lobe lung abscess had reduced in size with a smaller recurrence of the right-sided pleural effusion. A second intercostal catheter (14 Fr) was inserted for therapeutic drainage and 90 ml of cloudy amber-coloured fluid was drained. *Lactobacillus rhamnosus* was again cultured on the pleural fluid. Laboratory investigations revealed a CRP of 51 mg/L and WBC count of 5.4 × 10^9/L. Steady clinical, biochemical and radiological improvement occurred over a 4-week inpatient stay from a combination of a prolonged course of intravenous antibiotics, oral prednisolone and antifungal therapy in combination with endoscopic aspiration of impacting mucoid plugs and therapeutic drainage of the right-sided empyema. She was subsequently discharged home with a further 2-week course of intravenous antibiotics, oral prednisolone and antifungal therapy.

She was followed up over a 12-month period. Voriconazole was ceased at 6 months due to side effects of liver function derangement, blurred vision, lethargy, skin and nail discolouration and hair loss. Prednisolone was weaned to 5 mg daily with elevation in blood eosinophil counts and increased cough and breathlessness when further weaning was attempted. Due to the requirement for maintenance oral corticosteroids, she was commenced on omalizumab (anti-IgE monoclonal antibody). Following four doses of omalizumab, she reported significant improvements in her respiratory symptoms. Her prednisolone has subsequently been ceased. At 12 months, there was significant clinical and radiological resolution of the right upper lobe lesion with no residual day-to-day respiratory symptoms (Figure 1E).

DISCUSSION

Aspergillus rarely causes infection in individuals with normal immunity and bronchial architecture. Abnormal airway anatomy and a predisposition to airway hypersensitivity reactions are key aspects in the pathogenesis of ABPA. *Aspergillus* can colonize the bronchial airways in susceptible individuals, causing bronchial inflammation, mucus impaction and inflammation resulting in further bronchiectasis, fibrosis and respiratory compromise.[1] Mucocoele formation may result from chronic mucous impaction. Our patient met the International Society for Human and Animal Mycology (ISHAM) 2013 Diagnostic Criteria for ABPA on the basis of her childhood asthma, positive RAST test to *Aspergillus*, elevated total IgE and presence of pulmonary opacities on chest radiograph and eosinophilia.[2] Oral antifungal agents and corticosteroids may be used in the treatment of an acute exacerbation of ABPA.[3] Anti-IgE therapy, such as omalizumab, has also been shown to improve outcomes in patients with ABPA and severe asthma.[4]

Actinomyces odontolyticus, *L. rhamnosus* and *S. mitis* are microorganisms that have all been individually identified as a part of the normal oropharyngeal flora and considered to have low pathogenicity in humans.[5–7] A structured search of the medical literature (Ovid MEDLINE and EMBASE) from January 1966 to December 2021 was conducted to identify English-language articles that reported on pulmonary infections caused by *A. odontolyticus*, *L. rhamnosus* and *S. mitis*. The findings of our literature review are summarized in Table 1. Twenty-one cases of pulmonary actinomycosis caused by *A. odontolyticus*,[8–25] one case of empyema caused by *L. rhamnosus*[26] and 89 cases of empyema caused by *S. mitis* were identified.[27–30] Of these cases, 84 were identified as a part of a retrospective audit examining the aetiology of community-acquired lung abscesses diagnosed at one Japanese healthcare service.[29] Only one case of pulmonary actinomycosis has been reported in a patient with a past history of bronchiectasis of unclear aetiology.[11] None of these microorganisms have been previously been identified in patients with ABPA.

A history of periodontal disease, aspiration and being immunocompromised were the most commonly reported risk factors identified in patients who developed pulmonary infections secondary to *A. odontolyticus*,[8–16] *L. rhamnosus*[31] or *S. mitis*.[32]

We propose a plausible cause where obstructive mucus impaction due to ABPA led to the formation of a chronic mucocoele in the right upper lobe, which acted as a nidus for infection and abscess formation from organisms originating in the oral cavity in which dental extraction may have acted as a seeding event based on the temporal proximity of these events and identified organisms. Silent aspiration could have also triggered this infection, although we had no reason to suspect this based on this patient's clinical history. A secondary empyema was additionally colonized by *Lactobacillus* species perhaps made more likely from high loads of this organism due to regular ingestion of *Lactobacillus*-containing probiotics. Our case study raises the need for discussion regarding the role of preventative antibiotic therapy in conjunction with dental procedures for at-risk patients with chronically occluded airways and presence of mucocoele. Aspiration risk should also be assessed in those who regularly consume *Lactobacillus*-containing probiotics. In addition, our case highlights the potential for ABPA patients to develop severe complications when mucoid impaction prevents drainage post infection.

CONFLICT OF INTEREST

None declared.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

ETHICS STATEMENT

The authors declare that appropriate written informed consent was obtained for publication of this case report and accompanying images.

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