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BRONCHOPLEURAL FISTULA AND HYDROPNEUMOTHORAX IN PULMONARY AND PLEURAL TUBERCULOSIS: CASE REPORT

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Abstract: Tuberculosis (TB), which is highly prevalent worldwide, continues to be a public health problem, especially in Brazil. The failure to eradicate *Mycobacterium tuberculosis* infection continues to cause unusual complications and structural and functional pulmonary alterations. Dyspnea, cough and fever are the initial symptoms of pulmonary tuberculosis. Extrapulmonary symptoms can also occur, especially when the infection spreads to other structures. In pleural tuberculosis, chest pain associated with breathing is common. This case refers to a patient with pulmonary tuberculosis who developed bronchopleural fistula, pleural tuberculosis and hydropneumothorax. These complications are rare, with few cases reported in the medical literature.
Keywords: Hydropneumothorax; bronchopleural fistula; *mycobacterium tuberculosis;* pleural diseases

INTRODUCTION

The infection caused by the bacterium *Mycobacterium tuberculosis* mainly affects the lungs and can also affect other organs, spreading via the hematogenous route or extending to an adjacent structure [1]. The infection can be primary or by reactivation, developing after a period of dormancy and appearing at sites of hematogenous spread or contiguity [1]. In this scenario, 15% of infections occur in extrapulmonary sites, with the cervical lymph nodes and pleura, respectively, being the most affected sites [2]. In Brazil, pleural TB accounts for around 8.7% of all cases in the country [3].

The diagnosis of pulmonary TB can be made through signs and symptoms, such as cough, fever, weight loss and night sweats, in conjunction with an alcohol-acid-resistant (BAAR) smear and chest X-ray [4]. However, TB can still present in peculiar ways, especially in the younger generation, followed by unpredictable complications [5]. In extrapulmonary forms, symptoms vary according to the site affected, which can make diagnosis difficult. In pleural TB, the clinical picture can be acute or subacute with chest pain associated with breathing, malaise, fatigue, dyspnea and exudative pleural effusion, usually unilateral [3].

Because they are rare complications, there is little medical literature on bronchopleural fistula and hydropneumothorax in TB [5,6]. The diagnosis of bronchopleural fistula is made using a combination of clinical and imaging findings that confirm an air leak from the bronchial tree into the pleural space. Chest CT scans enable the location, size and characterization of fistulas and hydropneumothorax [7]. In addition to managing complications, antimicrobial treatment for TB is divided into a two-month active phase (rifampicin, isoniazid, pyrazinamide and ethambutol) and a four-month maintenance phase (rifampicin and isoniazid), according to the particularities of each case, such as age and possible antimicrobial resistance [8,9,10].

Due to the complexity and long duration of the available regimens, adherence to treatment becomes challenging. Several interventions focus on providing patient education, with the aim of favoring treatment success and avoiding complications [11]. In a recent systematic review, at least 10 to 15% of TB survivors had severe pulmonary impairment [12]. Taken together, patients who smoke have an increased risk of TB, recurrent TB and increased mortality [13]. The aim of this case report is to present bronchopleural fistula and hydropneumothorax as rare complications of TB, with increased morbidity and mortality, following reactivation of the disease in a young smoker from southern Brazil.

CASE PRESENTATION

A 29-year-old man from Porto Alegre, in the south of Brazil, who smoked and was immunocompetent, came to the emergency room with pleuritic pain, dyspnea, a low-secretive cough, night sweats and a weight loss of 10 kg in two months. No fever at the time of admission. No allergies or other comorbidities. A year earlier, he had been treated for pulmonary tuberculosis for six months, and denied poor adherence to medication. The patient was initially admitted to another hospital, where primary measures were taken, such as a chest X-ray and the start of anti-tuberculin treatment. In addition, a right chest drain was placed with purulent secretion coming out, due to the bronchopleural fistula and the hydropneumothorax, with no local phlogistic sign from the surgical wound and no collections.

He was then transferred in stable condition to a tertiary hospital for further assessment of complications and possible treatment. On physical examination during hospital admission, pulmonary auscultation showed an abolished vesicular murmur in the right hemithorax, with some crackles at the base of the left hemithorax, but the patient was eupneic on room air. The rest of the physical examination was normal. General and specific tests were carried out. C-reactive protein was initially 200, with a decrease during the nine days he was hospitalized. The blood count showed slight leukocytosis (11,850/mm3) and anemia (hemoglobin 9.2g/dl). Hemoglobin remained low until discharge (8.0 g/dl), but the leukocytosis resolved. Other tests were within normal limits. Serologies for human immunodeficiency virus (HIV), syphilis, hepatitis B and hepatitis C were negative.

The test for acid-fast bacillus (BAAR) was positive. The culture was also positive for *M. tuberculosis*. The GeneXpert rapid molecular test was positive, with sensitivity to rifampicin. A chest CT scan showed nodular lesions with excavations in both lungs and thickening of the pleura due to granulomatous involvement, complete fibroatelectasis of the right lower lobe with bronchiectasis and foci of calcification in between, a fistulous air pathway from the middle lobe to the pleural cavity on the right and multiple areas of bilateral air trapping, suggesting bronchiolopathy. There was also mediastinal lymph node enlargement on the right, measuring up to 1.5 cm in the shortest axis, probably reactive. Next, we'll analyze the chest CT images:

Figure 1

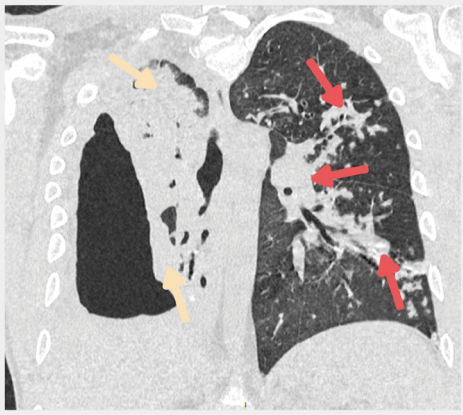
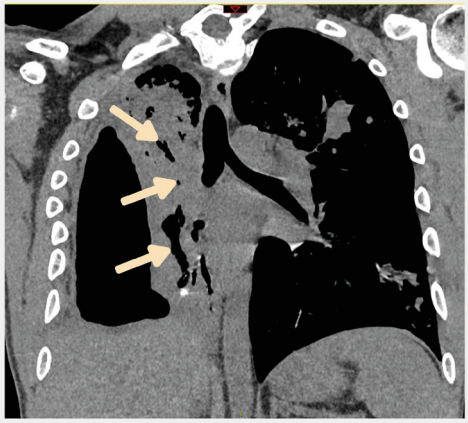


Figure 1, on the left, shows centrilobular lesions in red, with a sprouting tree pattern, demonstrating endobronchial dissemination. On the right, shown in yellow, there are consolidations in the upper lobe and pleural thickening.

Figure 2



In figure 2, the yellow arrows point to the path of the bronchopleural fistula on the right.

Figure 3

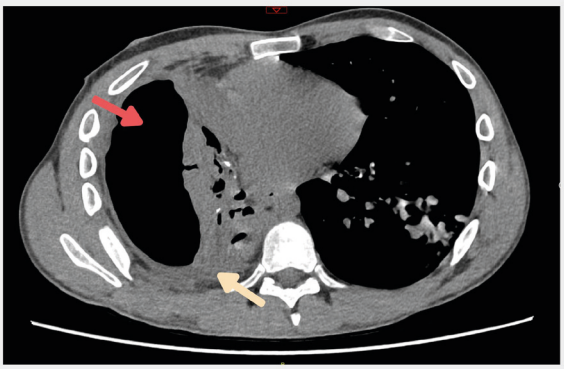
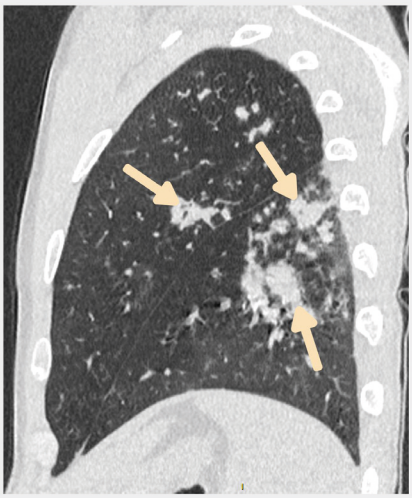


Figure 3 shows the right hydropneumothorax. The yellow arrow points to the liquid component and the red arrow to the air component.

Figure 4



Finally, figure 4 shows more centrilobular lesions on the left, with a sprouting tree pattern, highly suggestive of pulmonary TB.

As discussed with the hospital's infectology team, the patient was treated conservatively with a chest drain, as well as continuing rifampicin, isoniazid, pyrazinamide and ethambutol. The appearance of the drained secretion remained the same for around 20 days. The thoracic surgery team decided not to intervene surgically, due to the presence of active disease. The plan was to complete the TB treatment and, after proving that the condition had resolved, to assess the possibility of pneumonectomy. In total, the patient was hospitalized for twenty days. After discharge, he continued with the chest tube and medications, and remained under outpatient follow-up with the service's clinical medicine and thoracic surgery teams.

DISCUSSION

TB is classified as the second leading cause of infectious death in the world, second only to COVID-19 [14]. In Brazil, there were 80,000 new cases of the disease in 2023. Although there was a small decrease in the number of cases during the COVID-19 pandemic in 2020, the numbers rose again in the following two years [15]. In the south of the country, the scenario is even more alarming, since the incidence in the city of Porto Alegre, capital of the state of Rio Grande do Sul, was 83 cases per 100,000 inhabitants last year, more than double the Brazilian rate [16].

In this context, there are also the challenges of the higher incidence in prisons and co-infection with the human immunodeficiency virus (HIV). The city of Porto Alegre, located in the south of Brazil, is the capital with the highest number of TB and HIV co-infections [16]. Together, poor adherence to treatment and drug resistance make it difficult to eradicate the disease. Despite this, the incidence of TB decreased in 2023, suggesting at least a slowdown in the increase or stabilization [17, 15].

TB can develop in its primary form or through reactivation of *Mycobacterium tuberculosis.* In the first five years after infection, reactivation occurs in up to 15% of individuals [18]. In most cases, the bacteria remain dormant indefinitely and, when immunity wanes,

reactivation can occur. The appearance of the X-ray differs between primary tuberculosis and reactivation. If further investigation is required, computed tomography (CT) can be used for better visualization and confirmation of the diagnosis [1].

Due to endogenous reactivation, the risk of TB recurrence is higher in patients who have previously treated the disease than in the general population [19]. Treatment regimens combining rifampicin, isoniazid, pyrazinamide and ethambutol are almost completely effective, as long as patients are susceptible to the drugs and take them in the correct doses and at the correct time. However, the TB treatment abandonment rate reached 12.5% in the state of Ceará, in northeastern Brazil, between 2001 and 2017 [3]. A systematic review showed that the factors associated with TB treatment abandonment in the country are male gender, black race/ethnicity, age between 19 and 49 years, co-infection with the human immunodeficiency virus (HIV), low schooling (<8 years), use of tobacco, alcohol and illicit drugs [20]. In addition, another review of clinical trials of new tuberculosis treatment regimens showed that 94% of relapses occurred in the first 18 months after completion of treatment. All this evidence is in line with that found in the case reported here, with relapse one year after treatment in a young male patient who smoked [21].

Although pulmonary TB is the most common type, pleural TB also has a significant incidence, from 3% to 5% in non-endemic areas and 30% in endemic areas with a high proportion of HIV-positive individuals, as in our location in southern Brazil [22]. In a recent study, patients with pleural TB were younger and had a higher proportion of men, which is in line with the case reported by us [14]. Biopsy, with the specific histopathological finding of granulomas, has been used less frequently. Generally, pleural fluid or sputum is analyzed by direct examination, culture, polymerase chain reaction (PCR) tests, as well as checking levels of adenosine deaminase and gamma interferon [23].

The risk of TB increases 2 to 2.5 times in patients who smoke and is significantly associated with recurrent TB and increased mortality [13]. This case report falls within this estimate. A recent systematic review found an association between smoking and poor TB treatment outcomes, such as loss to follow-up, disease severity and drug resistance. Therefore, smoking cessation can be influenced during treatment, due to the possible positive effects on both treatment and relapse of the disease, as well as on future lung disease [13].

In addition, this case highlights a rare event of bronchopleural fistula, possibly resulting in hydropneumothorax. Bronchopleural fistulas challenge successful management and are associated with high morbidity and mortality, with an incidence of 1.9% and a mortality rate ranging from 18% to 50% [24]. The clinical picture can manifest as cough, fever, dyspnea, as well as subcutaneous emphysema and tension pneumothorax. Existing therapy methods include sclerosing and tamponade agents, occlusive devices and endoscopic intervention. The location of the bronchopleural fistula may require multiple images and bronchoscopies. It is worth noting that bronchoscopy is the preferred initial choice for small fistulas [24]. The bronchopleural fistula in our patient was evident on a chest CT scan, so there was no need to perform a bronchoscopy. Given the lack of standardized treatment recommendations for bronchopleural fistulas, individual treatment strategies should be formulated, taking into account the location, size, existing complications and comorbidities [24]. For some patients, more invasive surgery, such as decortication, thoracoplasty or pleuropneumonectomy, may be necessary to definitively close the fistula. Conservative

medical treatment is an alternative treatment approach. In a recent study, fistulas were resolved within 36 days with chest tube drainage and treatment of the underlying tuberculosis [25]. The need for correct treatment is due to the fact that untreated patients can develop respiratory compromise, subcutaneous emphysema or even death [7]. In patients with a suspected infectious cause, this can include HIV testing, cultures for acid-fast bacilli, PCR for TB, gamma interferon assay [7].

Only isolated case reports have been documented of TB-related hydropneumothorax and there has been a scarcity of large case studies [26]. Hydropneumothorax is a rare variant of pneumothorax, consisting of free fluid and air within the pleural space. The appearance of hydropneumothorax requires rapid management. Its origin can be explained by the hypothesis that the bronchi and the pleural space were connected through the rupture of a cavity, thus creating a bronchopleural fistula. In this way, the pleural space is contaminated with caseous material, requiring urgent correction due to its tendency to produce pleural fibrosis with failure to expand [1].

Hydropneumothorax has been associated with malignancy, infection, chest trauma, rheumatological diseases affecting the lung parenchyma, after placement of a chest tube and after thoracentesis [27]. In this condition, dyspnea and sudden unilateral chest pain are known to be common symptoms, as well as fever and weight loss, probably due to TB being the main etiology. Asymmetrical expansion of the hemithorax is usually observed, associated with decreased air intake. Intercostal drainage and suction are commonly used to drain air and fluid from the pleural space, providing symptom relief.

The diagnosis of hydropneumothorax is suspected by X-ray and computed tomography is the method of choice. Ultrasound is not the best method for determining the nature of a hydropneumothorax [28]. With regard to treatment, the insertion of a drainage tube remains the ideal course of action, associated with antimicrobial therapy for TB [25]. In a study in India, the intercostal drainage tube was inserted in all 57 patients and drainage was required for more than 30 days in a third of the individuals [25].

Therefore, the complications of the clinical case reported were treated conservatively with chest tube drainage for the hydropneumothorax. In addition, antibiotics were administered to treat the superimposed bacterial infection. In summary, we describe a rare case of pulmonary and extrapulmonary tuberculosis in an immunocompetent patient who developed serious pulmonary complications. The management of the patient required a multidisciplinary approach, including clinicians and radiologists.

CONCLUSION

Pulmonary *tuberculosis* is the best known form of *M. tuberculosis* infection. Extrapulmonary tuberculosis usually occurs in a single system, with the pleura being the most affected site. Hydropneumothorax is one of the rare complications of the disease, and is even more serious in conjunction with bronchopleural fistula. Prolonged antituberculosis therapy is essential to control the infection, together with local drainage when indicated. Early diagnosis of the disease is therefore extremely important, as it facilitates the start of appropriate treatment and reduces the rate of complications.

NOTES

The authors have declared that there are no conflicts of interest.

HUMAN ETHICS

Consent was obtained from all participants.

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