

Case series of Tuberculous Peripleuritis in a Public Hospital in the Autonomous City of Buenos Aires during the Period from 1983 to 2021

Serie de casos de peripleuritis tuberculosa en un hospital público de la Ciudad Autónoma de Buenos Aires durante el período de 1983 hasta 2021

Baloco, Oscar¹ ; Sívori, Martín² ; Jajati, Mónica¹ ; Serrano, Mariela¹; González, Claudio¹; Rey, Darío³ 

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Correspondencia

Oscar Baloco. Urquiza 609,
CABA (1405). E-mail:
oscarbalocoe@hotmail.com

The endothoracic fascia and the extrapleural space are sites of pathological processes closely linked to the pathology of the lung parenchyma and pleura, including tuberculosis (TB).¹⁻² Understanding the anatomical structure will prevent the confusion that normally occurs with pleural localization, since the pathogenesis and some diagnostic methods differ from pleuropulmonary forms of tuberculosis, as well as the duration of treatment.¹⁻² The first reference is from Boyer in 1846, but Wunderlich in 1861 reported the initial description as “peripleuritis,” hence it has been known as the “Wunderlich’s disease.”¹ In 1867 Billroth and Verneuil emphasized the importance of the lymph nodes in the pathogenesis of this presentation.¹ Kauffmann supported the lymphatic pathogenesis of cold abscesses of the thoracic wall.¹ In 1939, Skarby provided a brilliant radiological work that was fundamental for understanding this condition.³ In our country, the first description of 30 cases of tuberculous peripleuritis was in 1945, by Prof. O. Vaccarezza.⁴ So peripleuritis was defined as “the inflammation (of different degrees) of the tissues located between the parietal pleura and the thoracic wall.”⁴ It is also known by other synonyms such as cellulitis or endothoracic fasciitis, epipleuritis, peripleural abscess, or cold abscess in the Anglo-Saxon literature.¹⁻² The main cause of peripleuritis is infectious, with *Mycobacterium tuberculosis* being the most common bacterium. However, other etiologies have been confirmed, such as fungal infections by *Paracoccidioides brasiliensis* and *Actinomyces israelii*, as well as other non-infectious causes like lymphomas, myelomas, benign or malignant tumors, and trauma.¹⁻²

The objective of this communication was to understand the incidence and demographic characteristics of tuberculous peripleuritis (TBPP), its association with parenchymal lesions, its coinfection with HIV, and the compliance and treatment withdrawal rates recorded in a public hospital during the period 1983-2021.

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¹Pulmonology University Center Dr. J. M. Ramos Mejía, Pulmonology and Tisiology Unit, Hospital Dr. J. M. Ramos Mejía, Faculty of Medicine, University of Buenos Aires (UBA), Autonomous City of Buenos Aires.

²Pulmonology University Center Dr. J. M. Ramos Mejía, Faculty of Medicine, University of Buenos Aires, director of the Pulmonology Specialization Career, Faculty of Medicine, UBA.

³Pulmonology University Center Dr. J. M. Ramos Mejía, Faculty of Medicine, University of Buenos Aires, Director of the Pulmonology Specialization Career, Hospital General de Agudos Dr. Enrique Tornú.

MATERIALS AND METHODS

A retrospective analysis was conducted on cases of TBPP that were confirmed through biopsy or imaging and were reported through the respective program form and documented in the medical records archive and the computer system of the Ministry of Health of the Government of the Autonomous City of Buenos Aires (SIGEHOS).

The following data were considered: demographic background, BCG (Bacille Calmette-Guérin) vaccination history, coinfection with HIV, presence or absence of concurrent lung involvement, treatment adherence, toxicity, pharmacological resistance, and mortality.

RESULTS

4,076 cases of TB were reported, 11 of which were TBPP (0.27 %, or an incidence of 269.8 cases of TBPP per 100,000 cases of TB).

The median age was 42 years (IQR, interquartile range of 23-96); and males accounted for 72.7 % (n = 8) of the sample. 36.3 % (n = 4) had been BCG-vaccinated, and 18 % (n = 2) had coinfection with HIV (from 1989 to 2021).

With regard to pulmonary presentation, 54.5 % (n = 6) of the patients showed associated parenchymal lesions: 50 % (n = 3) had solitary non-cavitary lesions, and the rest included: 1 with solitary cavitary lesions, 1 with bilateral non-cavitary lesions, and 1 with bilateral cavitary lesions (Figure 1).

The diagnosis was confirmed by biopsy in 72.7 % (n = 8) of the cases and was clinically and radiologically determined in the remaining cases. The anatomical areas involved were the internal mammary lymph node chain in 63 % (n = 7), the paravertebral in 27 % (n = 3), and intercostal in 9 % (n = 1) (Figure 2).

Seven patients (63.6 %) completed treatment, three patients (27 %) discontinued treatment, and one patient (9 %) passed away during the treatment. In terms of follow-up, one patient showed rifampicin resistance (patient with concurrent lung involvement), and another patient (9 %) experienced reversible hepatotoxicity due to pyrazinamide.

DISCUSSION

A case series of TBPP has been described in a multispecialty public hospital in the city of Buenos Aires. Its incidence was extremely low. It predominated in middle-aged men without BCG vaccination and with non-cavitary unilateral lung involvement. Coinfection with HIV was considerable, which could be attributed to the profile of our institution. In almost three out of four patients, a diagnosis was made through biopsy, predominantly involving the internal mammary chain. Treatment

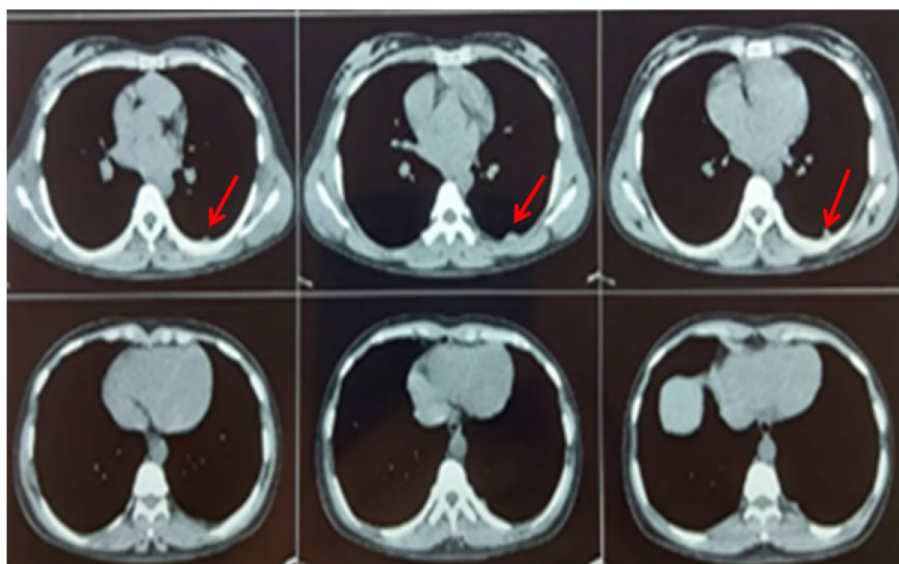


Figure 1. Computed axial tomography with mediastinal window with the Skarby sign (marked with red arrow)



Figure 2. Case of peripleuritis with involvement of internal and posterior intercostal mammary lymph node chain

withdrawal was reported in at least one every four patients, which was associated with polysubstance consumption and homelessness.

Although TBPP was first described in the mid-19th century, in our country, it was Prof. O. Vaccarezza who first described his series of 30 cases.^{1,4} Defined as “the inflammation (of different degrees) of the tissues located between the parietal pleura and the thoracic wall,” it is often an underdiagnosed form of presentation of TB.¹⁻² Some years later, Professors Juan Carlos Rey and Pedro Rubenstein described another case series.⁵⁻⁶

To understand its clinical presentation, it is necessary to recognize the normality of the peripleural anatomical space.^{1,2} The endothoracic fascia is an anatomical structure consisting of the apposition of numerous fibrils oriented in different directions and in a heterogeneous way. This explains how within the endothoracic fascia, purulent collections can adopt different arrangements and sizes. The lymph nodes of the thoracic wall are divided into four groups: 1) Posterior parietal nodes located in the costovertebral angle within the thickening of the endothoracic fascia; 2) Anterior or internal mammary nodes, which are also situated in the corresponding thickening of the fascia; 3) Intercostal nodes located between the intercostal muscles and the lateral wall of the

thorax, directly receiving lymphatics from the parietal pleura; 4) Diaphragmatic nodes.^{1,2} Given the heterogeneous arrangement of the fibrils, weak points are generated in the thoracic wall, allowing the opening of peripleuritic abscesses from the inside to the outside. They herniate and lead to the formation of cold abscesses. These can be intermuscular, emerging over the anterolateral areas of the chest, or intramuscular, following the paths of the perforating nerves. Others may become externalized as caseous-purulent forms.^{1-2,7-8} Tuberculous peripleuritis can originate in the pulmonary parenchyma, the pleura, or the thoracic wall. The process of primoinfection that develops in the lung, from the bacillus nesting in the alveoli to the primary infiltration, allows the observation of the contamination of the pleural serosa.^{1-2,7-8} In the secondary period of Ranke, the hematogenous route and contiguous spread through traumatic or iatrogenic means would be the mechanism of pleural infection, the starting point of peripleuritis when the lymph node in the extrapleural zone becomes infected, resulting in tuberculous adenitis. In the early stages of Ranke, peripleuritis manifests in its caseous-purulent form, while in the tertiary period, the fibrosclerotic form predominates.^{1-2,7-8} In chest images, it can appear as juxtacostal radiopacities or areas of higher attenuation in tomographies, with the free edge

directed towards the lung, convex in its central portion, and concave at both ends (Skarby Sign).³

Despite the low incidence of presentation compared to more traditional forms of TB, our series of TBPP has a significant number of cases in modern times if compared with the number reported on a national level more than 50 years ago, especially considering that most of those cases were diagnosed in the pre-antibiotic era of TB treatment.⁴⁻⁶

In conclusion, in this report, the incidence of TBPP was extremely low among patients diagnosed with TB, predominantly in middle-aged men without BCG vaccination and with non-cavitary unilateral lung involvement. Coinfection with HIV was considerable (18 %), and this could be attributed to the profile of our institution. A case of TBPP had already been reported in a patient with HIV.⁹ In almost three out of four patients, a diagnosis was made through biopsy, the preferred procedure in these cases, mostly involving the internal mammary chain. Treatment withdrawal was reported in at least one every four patients, which was associated with polysubstance consumption and homelessness. Due to the significant incidence of TB in our country and the low clinical-radiological suspicion of peripleuritis, this form of

clinical presentation should be considered for early diagnosis and treatment.

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