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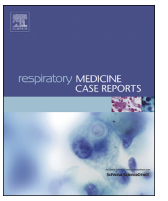

Case Report

Xanthogranulomatous pleuritis induced by recurrent biliothorax due to a biliopleural fistula: The first case report in the literature

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ABSTRACT

Xanthogranulomatous pleuritis is an extremely rare pathological entity, characterized by the infiltration of foamy cells and multinucleated giant cells within the pleural space. This condition often mimics infectious and neoplastic processes, presenting significant diagnostic challenges. This report details the first documented case of xanthogranulomatous pleuritis induced by recurrent biliothorax due to a biliopleural fistula, presenting a unique clinical scenario. We describe the clinical presentation, diagnostic hurdles, and both the surgical and medical management of this case. The discovery of biliothorax, evidenced by pleural fluid bilirubin levels that exceed serum bilirubin levels, underscores the importance of considering biliothorax in the differential diagnosis of recurrent pleural effusions, particularly in patients with a history of trauma. This case emphasizes the need for heightened awareness and a multidisciplinary approach in the diagnosis and treatment to effectively manage this complex condition and prevent recurrence.

1. Introduction

Xanthogranulomatous pleuritis is an exceedingly rare pathological condition [1,2]. It is an uncommon variant of pleural inflammation characterized by the accumulation of foamy cells, histiocytes, and multinucleated giant cells [1,2]. This condition is often misdiagnosed due to its similarity to other infectious and neoplastic pleural diseases [1]. To date, only a limited number of cases have been reported in the medical literature [1–3]. This case report aims to present a rare case of xanthogranulomatous pleuritis induced by recurrent biliothorax due to a biliopleural fistula, discuss its clinical features, diagnostic challenges, and treatment options, and emphasize the need for accurate recognition of this entity to avoid unnecessary invasive interventions. To the best of our knowledge, this represents the first documented case of such a presentation in the medical literature.

2. Case presentation

A 19-year-old male student presented to our respiratory department with complaints of recurrent pleuritic chest pain, fever, cough, and dyspnea. One year prior, the patient experienced a car accident followed by multiple episodes of right-sided pleural effusion, which had been treated with thoracentesis. The analysis of the effusions revealed an exudate and pleural biopsy results indicated reactive pleuritis without evidence of granulomas or malignancy. Despite interventions, the effusion recurred within a few weeks of

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each procedure. On admission, vital signs were reported as normal. Physical examination revealed decreased breath sounds and dullness to percussion over the right lower chest. Routine laboratory tests were within normal limits. Chest ultrasonography (USG) screening showed multiple septations with thick parietal pleura Fig. 1. Additionally, chest CT revealed a right-sided pleural effusion Fig. 2. The pleural fluid analysis confirmed an exudative type of pleural effusion, with the following values: protein at 3.8 g/dL (serum protein: 6.5 g/dL), lactate dehydrogenase at 800 IU/L (serum LDH: 190 IU/L), and bilirubin at 1.2 mg/dL (serum bilirubin: 0.8 mg/dL). Cytology analysis of the fluid was unremarkable. A 28 French chest drain (Smiths Medical, Minneapolis, MN) was surgically inserted into the right chest for therapeutic drainage of the recurrent effusion and remained in place following a limited thoracotomy performed under sterile conditions in the operating room. During this procedure, pleural decortication was carried out. Extensive adhesions and yellow plaque-like deposits were observed on the pleura and diaphragm Fig. 3A, indicating a severe inflammatory response. While exploring these features, a biliopleural fistula within the diaphragm was discovered Fig. 3B [Video]. This fistula was promptly closed with non-absorbable sutures. The chest drain was maintained for 14 days (two weeks) post-procedure to manage any residual effusion. Histopathological examination of the pleural peeling revealed diffuse infiltration of the pleura by lipid-laden macrophages, foamy histiocytes, multinucleated giant cells, and chronic inflammatory cells Fig. 4. Mesothelioma, carcinoma, and fungal infection were excluded by the negativity of immunohistochemical staining and PAS stain. Based on these histological find-

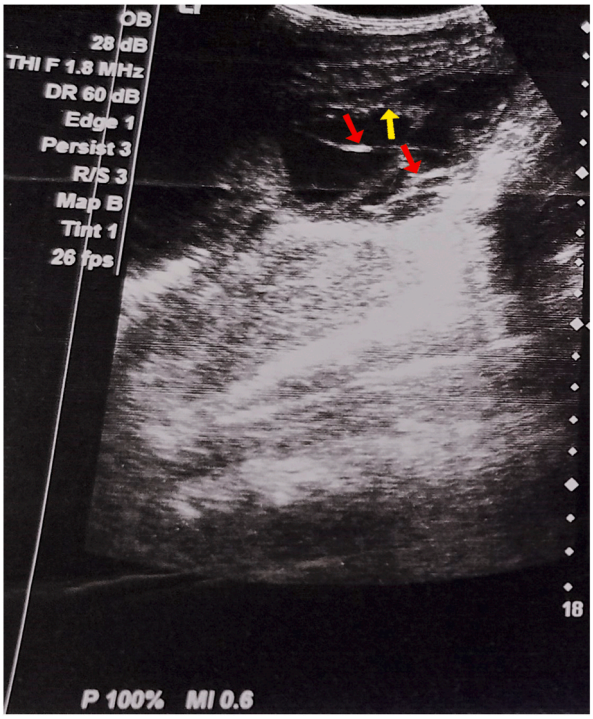

Fig. 1. Chest ultrasonography (USG) image demonstrates multiple septations (red arrows) with thick parietal pleura (yellow arrow).

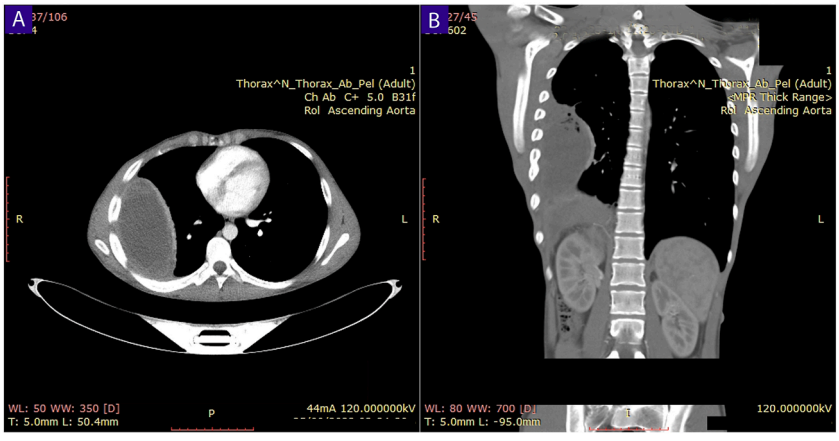

Fig. 2. Chest CT images. (A) Axial section and (B) Coronal section reveal a right-sided pleural effusion with multiple thick septations.

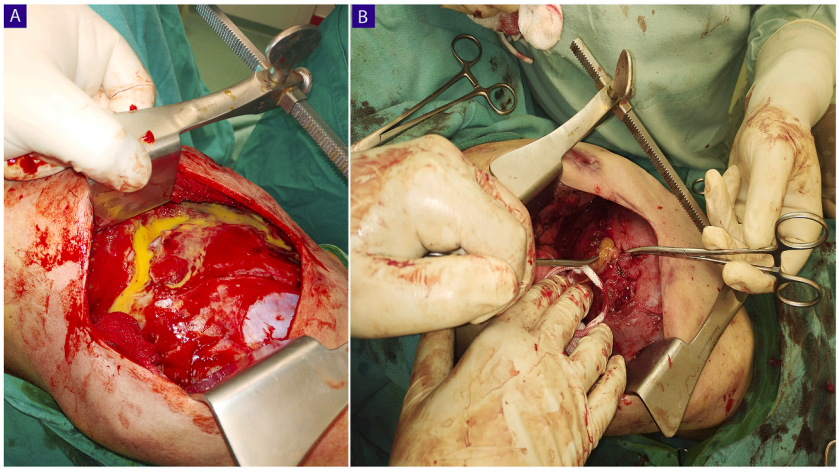

Fig. 3. Surgical images (A and B). (A) Depicts yellow plaque-like deposits in the pleura. (B) Illustrates the presence of a biliopleural fistula.
Video: The surgical video visually demonstrates the biliopleural fistula.

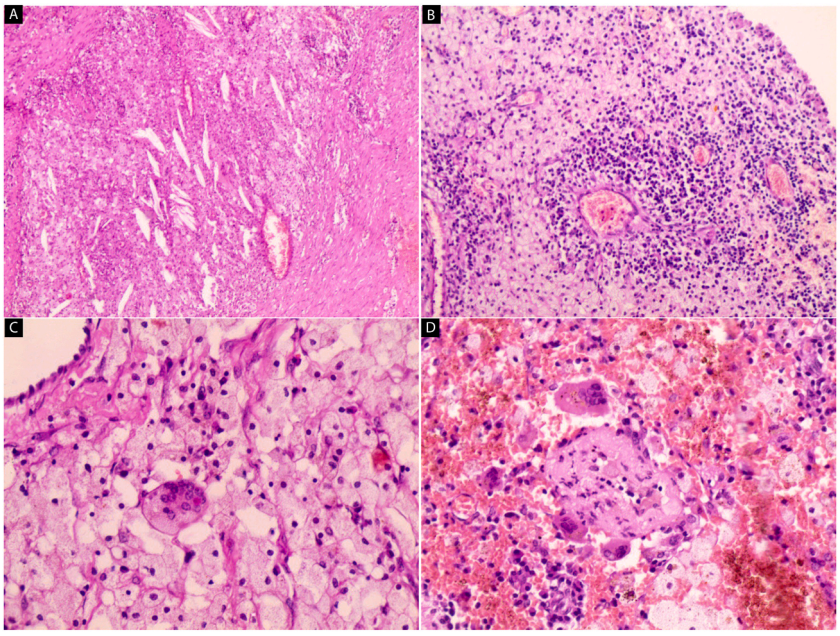

Fig. 4. Hematoxylin and eosin-stain (A–D). Microscopic images of the pleural peeling. (A and B) The low-power magnification shows diffuse infiltration of the pleura by lipid-laden macrophages, foamy histiocytes, and chronic inflammatory cells, cholesterol clefts (40×). (C and D) The high-power magnification shows foamy histiocytes and multinucleated giant cells (200×).

ings, the diagnosis of xanthogranulomatous pleuritis was made. After drain removal, an endoscopic retrograde cholangiopancreatography (ERCP) was performed as a subsequent procedure to place a stent in the bile ducts. Following surgical intervention, the patient was prescribed prednisolone 20 mg daily for 14 days to reduce inflammation and facilitate recovery. Subsequently, a chest x-ray was conducted to assess progress, revealing continuous improvement Fig. 5. The patient was discharged in good overall health, with an expanded lung and halted bile flow.

3. Discussion

Xanthogranulomatous pleuritis is an exceptionally rare pathological entity with only a few cases reported in the literature [1–3]. The clinical scenario of this case is dominated by the recurrent biliothorax, resulting from a biliopleural fistula, leading to xanthogranulomatous pleuritis. This specific combination is unique and previously unreported in the medical literature. The presence of biliothorax, evidenced by pleural fluid bilirubin levels that exceed serum bilirubin levels, suggests ongoing bile leakage into the pleural space due to a biliopleural fistula. To our knowledge, this is the first documented case of its kind in the medical literature, underscoring the rarity and diagnostic challenges associated with this condition. Xanthogranulomatous pleuritis is believed to be an un-

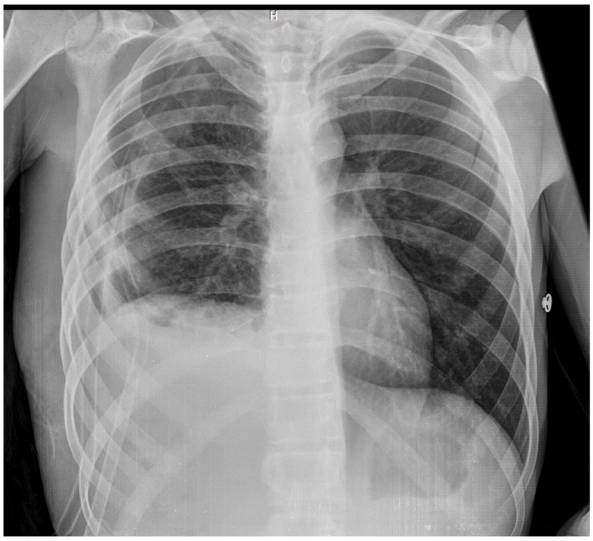

Fig. 5. Follow-up chest x-ray demonstrates significant improvement.

usual variant of chronic pleuritis and is characterized by the infiltration of the pleura by foamy macrophages and multinucleated giant cells [1,2]. The exact etiology of this condition remains unclear, and its occurrence in the pleura, specifically induced by biliothorax due to a biliopleural fistula, adds complexity to the understanding of the disease process. The mechanism by which bile enters the pleural space and triggers xanthogranulomatous pleuritis remains speculative. However, it is conceivable that the chronic exposure to bile constituents provokes an inflammatory response, leading to the distinctive histopathological findings observed in this case. The clinical presentation of xanthogranulomatous pleuritis is nonspecific, mimicking other pleural diseases, such as tuberculosis, malignancy, or empyema, making accurate diagnosis challenging [2]. The presence of recurrent pleural effusions, after trauma, and the unusual association with a biliopleural fistula complicated the diagnostic process, as in our case. Thus, including biliothorax in the differential diagnosis of recurrent pleural effusions, especially post-trauma, is a critical teaching point that extends beyond the pathological curiosity of xanthogranulomatous changes. Imaging modalities, including chest X-ray and CT scan, may reveal pleural effusion, thickening, or loculated fluid collections [5]. However, definitive diagnosis requires histopathological examination of pleural biopsy specimens, which typically demonstrate infiltrates of foam cells, histiocytes, and multinucleated giant cells. Immunohistochemical analysis may be useful to exclude infectious etiologies and confirm the presence of xanthogranulomatous inflammation [1–4]. The management of xanthogranulomatous pleuritis largely depends on treating the underlying cause if identified [3]. In cases where no specific etiology is identified, systemic corticosteroids have been shown to provide symptomatic relief and promote resolution of pleural inflammation [3]. A thoracoscopic pleural biopsy may be necessary for definitive diagnosis and to exclude other potential malignancies or infections [6]. In our case, the fistula was surgically corrected and the pleural effusion was drained. The discovery of a biliopleural fistula during surgical exploration provided the anatomical basis for the recurrent biliothorax observed in this patient. Repairing this fistula was essential, highlighting the critical role of surgical intervention in managing such complications. In addition to surgical repair, managing biliothorax typically involves supportive measures to control pleural inflammation and prevent further episodes. In this case, following the surgical correction of the biliopleural fistula and drainage of the pleural effusion, the patient was prescribed prednisolone 20 mg to reduce inflammation and aid in recovery.

4. Conclusion

This article presents an unprecedented case of xanthogranulomatous pleuritis associated induced by biliothorax due to a biliopleural fistula. The presence of biliothorax highlights a critical clinical entity that necessitates heightened awareness in the differential diagnosis of chronic pleural effusions, particularly in post-traumatic scenarios. The management of such cases relies heavily on accurate and timely diagnosis, where histopathological examination plays a crucial role as the gold standard. Early identification and appropriate management of biliothorax can significantly improve patient outcomes and prevent recurrent hospitalizations due to pleural effusion complications. The findings from this case underscore the importance of considering both xanthogranulomatous pleuritis and biliothorax in patients with unexplained recurrent pleural effusions. Further research is warranted to better understand the pathogenesis of xanthogranulomatous pleuritis and biliothorax, improve diagnostic approaches, and explore long-term outcomes for patients suffering from these complex pleural diseases.

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CRediT authorship contribution statement

Moatasem Hussein Al-janabi: Writing – review & editing, Writing – original draft, Visualization, Validation, Project administration, Methodology, Formal analysis, Data curation, Conceptualization. **Hussein Kaada:** Writing – original draft, Resources, Methodology, Investigation, Data curation, Conceptualization. **Ghina Ismail:** Writing – review & editing, Writing – original draft, Methodology, Formal analysis, Data curation. **Dommar Roumieh:** Resources, Methodology, Data curation, Conceptualization. **Zuheir Al-Shehabi:** Validation, Supervision.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.rmcr.2024.102065.

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