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Case report

Xanthomatous pleuritis

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A B S T R A C T

This case presentation relates to a 21 year young male, cachectic in appearance, who presented with progressive shortness of breath, and dull pain on the lower part of the chest ongoing over three months. The patient received antitubercular drugs by local physician for suspected tuberculosis with no significant improvement. He was referred to our Respiratory Department for further evaluation of his chest pain and breathlessness. His X-ray showed moderate pleural effusion on the left side which confirmed by USG chest screening which showed multiple septation with thick parietal pleura and USG abdomen was normal. Moreover, the pleural effusion aspiration was performed and pleural fluid examination revealed exudative in character and thoracoscopic view of pleura was reddish with exudatives. Pleural biopsy histological examination showed mixed cell infiltrate with predominantly foamy macrophage along with plasma cells, lymphocytes and eosinophils with capillary proliferation. This is the first care report of xanthomatous pleuritis in the literature.

1. Case presentation

This case presentation relates to a 21 year young male, cachectic in appearance, who presented with progressive shortness of breath, and dull pain on the lower part of the chest ongoing over three months. The patient received antitubercular drugs by local physician for suspected tuberculosis with no significant improvement. He was referred to our Respiratory Department for further evaluation of his chest pain and breathlessness. On admission, the patient had chest pain on the left side and shortness of breath on exertion. His X-ray chest showed moderate pleural effusion on the left side (Fig. 1) which was confirmed by USG chest screening which showed multiple septation with thick parietal pleura and USG abdomen was normal. As the clinical suspicion for tuberculosis was very high, we proceeded to thoracoscopic biopsy and drainage of fluid. Moreover, the pleural effusion aspiration was performed and pleural fluid examination revealed exudative in character and thoracoscopic view of pleura was grayish with red background (Fig. 2). Pleural biopsy histological examination showed mixed cell infiltrate with predominantly foamy macrophages along with plasma cells, lymphocytes and eosinophils with capillary proliferation (Fig. 3). The diagnosis of xanthomatous pleuritis was therefore made based on histopathological findings.

2. Treatment

The patient was advised to stop antitubercular drugs. X-ray chest was repeated after 3 months which showed improvement. Follow up X-ray was essential to exclude the possibility of recurrence of disease (Fig. 4) which showed marked improvement. We empirically started antibiotic amoxicillin for 6 weeks in view of pus inside the pleural cavity. This is the first case report on xanthomatous pleuritis so treatment is not available in the literature however a few case reported on xanthogranulmatous appendices, xanthomatous cholecystic diseases and kidney stone where treatment were removal of the cause.

3. Discussion

Xanthomatous inflammation is a rare form of chronic inflammation, manifested by the presence of lipid-laden macrophages admixed with lymphocytes, plasma cells, neutrophils, and often multinucleated giant cells with or without cholesterol clefts [1]. It was initially described in the kidney by Osterlind in 1944 [2]. It has also been reported in other organs, such as gall bladder, prostate, epididymis, ovary, urinary bladder, kidney, appendix, and others [1,3 and 4]. The exact etiology of xanthomatousinflammation is uncertain. Proposed etiologies include defective lipid transport, immunologic disturbances, infection by low-virulence organisms, and lymphatic obstruction [4]. Cozzutto and Carbone noted that hemorrhage plays a major role in the development of foamy macrophages, postulating that the ingested erythrocytes and platelets at the bleeding site overwhelm the lysosomal system of the macrophages causing deposition of phospholipids which results in a foamy appearance of the macrophages [1]. The inflammatory infiltrate

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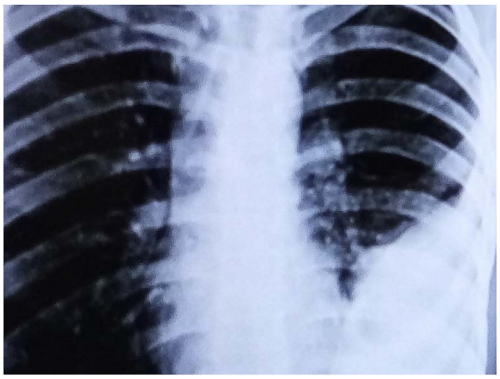

Fig. 1. Left pleural effusion.

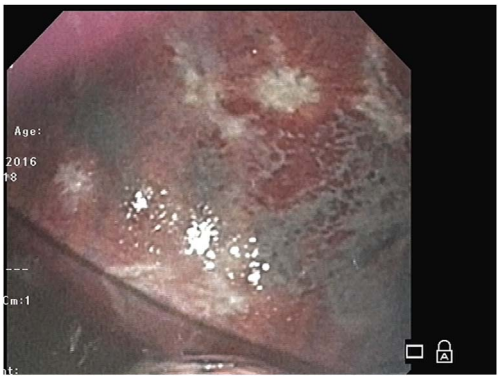

Fig. 2. Thoracoscopic view with pigmented parietal pleura.

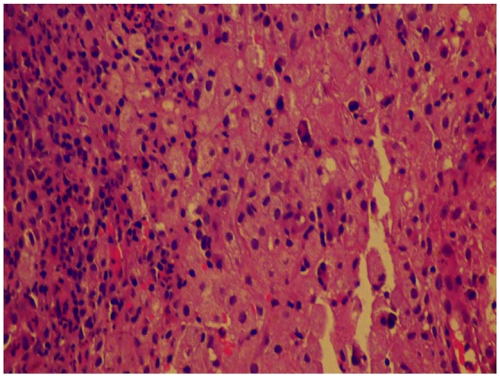

Fig. 3. HPE showed foamy macrophage with plasma cells and eosinophil.

are predominantly composed of foamy macrophages along with lymphocytes and plasma cells. Histopathological differential diagnoses are other type of chronic inflammation including tuberculosis. No granuloma is identified in this case. Z-N stain was also negative. Gene Xpert study for tuberculosis was also negative. Histopathological findings did not suggest any evidence of lymphoma or metastatic carcinoma. Hence we did not perform immunohistochemistry in our case. Xanthomatous inflammation in pleura is histomorphologically similar to xanthomatous inflammation in Gallbladder, Kidney and Appendix. Etiology of xanthomatous pleuritis could be longstanding inflammation or malignancy. In our case no tumor deposit is identified. Further follow-up of our case did not show any evidence of malignancy. Patient improved on conservative management. There are a few uncommon causes of pleural effusion where chronic inflammatory cells are present for example; IgG4 related diseases (6), solitary fibrous tumor of the pleura (7), mesothelioma and erdheim chester disease (8).

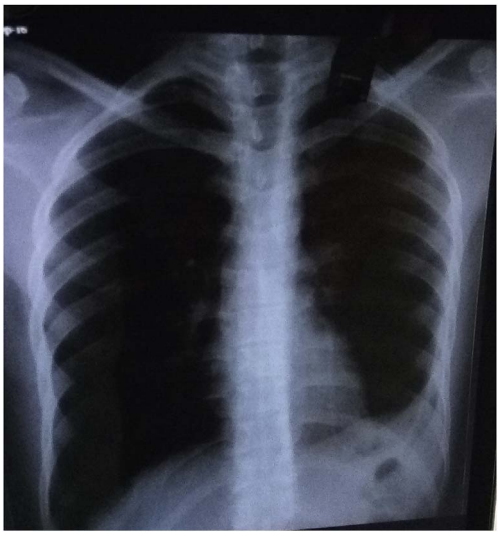

Fig. 4. Left pleural thickening.

4. Conclusion

Xanthomatous is a rare clinical entity, in pleural effusion. We would consider thoracoscopic pleura biopsy where pleural fluid character is exudative and naked eyes appearance of parietal pleura pigmented.

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