Case Reports

Endobronchial inflammatory pseudotumor of the lung

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ABSTRACT

Inflammatory pseudotumor (also called plasma cell granuloma, histiocytoma and x-anthofibroma) is a benign, slow growing lesion which may present with cough, dyspnea, hemoptysis and unresolving pneumonia or can be discovered radiographically as a localised lesion. It has been reported in individuals up to 70 years old, but approximately two-thirds have developed in individuals under 30 years of age. The sex incidence is approximately equal. Inflammatory pseudotumors of the lung are usually peripheral lesions but may occasionally be endobronchial. We report the case of an endobronchial inflammatory pseudotumor in a 17-year-old girl who presented with unresolving right-sided pneumonia. Appropriate radiological, bronchoscopic and histopathological investigations lead to accurate pre-operative diagnosis and early complete surgical resection through a limited right main bronchus incision (bronchotomy). The patient made good postoperative recovery and an excellent prognosis is anticipated.

Keywords: Endobronchial, inflammatory, pseudotumor, plasma cell granuloma.

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The endobronchial inflammatory pseudotumor is a rare and little described variant of inflammatory lung pseudotumor. The complex histomorphology and proliferative capacity of these luminal bronchial neoplasms may result in radiological, bronchoscopic and even histopathological diagnostic difficulties. We present a case of endobronchial inflammatory pseudotumor in a 17-year-old female patient. The clinical, radiological and histopathological features are described and compared with those of similar reported cases. The management options are also discussed together with a thorough review of the relevant medical literature.

Case Report. A 17-year-old Saudi female was referred to King Khalid University Hospital because of a history of unresolving right-sided pneumonia. She also complained of shortness of breath and intermittent vomiting for the last 2 months and productive cough with occasional hemoptysis for the last 6 months. Her past medical history was not otherwise significant and there were no records of a previous major surgery. The patient was, however, being treated in the referring hospital for a persistent right-sided pneumonia. On admission, her biochemical and hematological profiles were normal and her electrocardiogram (ECG) was reported to be within normal limits. Her chest x-ray showed right middle and lower lobes atelectasis with some areation in the right upper zone and significant mediastinal shift to the right side (Figure 1). The left lung was normal. Computed tomography examination (CT scan) showed a mass in the right main bronchus which was protruding to the lower trachea and causing complete obstruction of the right main bronchus and bronchus intermedius with atelectasis of the right lung (Figure 2). The patient subsequently underwent right rigid bronchoscopy under general anesthesia which showed that 90% of the lumen of the lower tracheal segment was

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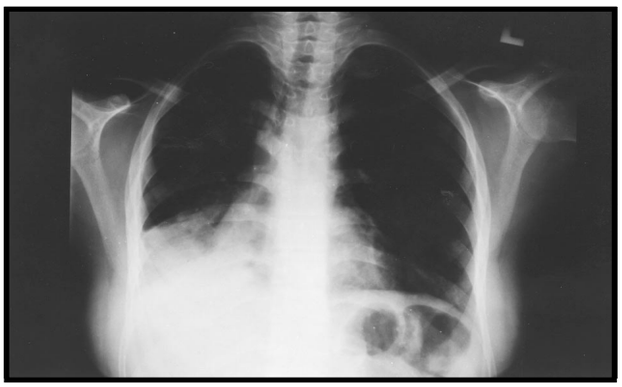

Figure 1 - Endobronchial inflammatory pseudotumor of lung. Pre-operative chest x-ray showing right and middle lower lobes atelectasis with significant mediastinal shift to the right side.

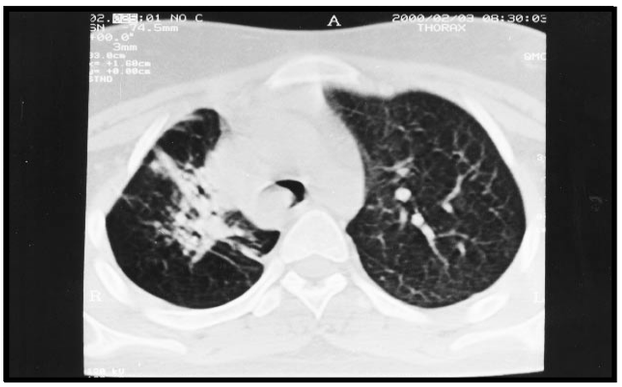

Figure 2 - Endobronchial inflammatory pseudotumor of lung. Computed tomographic scan showinbg complete obstruction of the right main bronchus by a tumor mass and atelectatic right lung.

occluded by a polypoidal mass which was protruding from the right main bronchus to the lower end of the trachea. A biopsy taken from the described polypoidal mass showed histological features consistent with an endobronchial inflammatory pseudotumor of the lung (Figure 3a and 3b). The patient subsequently underwent a thoracotomy and limited right main bronchoscopy through which the polypoidal mass was easily and completely excised without the need to perform any lobectomy. Following the excision of the tumor mass, the right main bronchus and bronchus intermedius were assessed intraoperatively and were seen to be normal. The base of the tumor was cauterised and the bronchotomy closed with simple sutures. The postoperative period was uneventful with a return to complete areation of the right lung and of the mediastinum to its normal position. The patient made a good recovery and was discharged on the 6th postoperative day. A check-up bronchoscopy after 6 months was also reported to be normal.

Discussion. Inflammatory pseudotumor of the lung is a rare and usually benign lesion which is variously named by different authors. In the case presented, the tumor occupied, and was restricted to, an unusual endobronchial position causing obstruction to a major airway which lead to unresolving pneumonia and atelectasis. The incidence of endobronchial inflammatory pseudotumor is rare and variously reported as between 5% and 16%.[1-3] In 1988, Matsubara et al[4] divided this entity into 3 distinct histological types: organizing pneumonia type, fibrous histiocytoma type and lymphoplasmacytic type, although there was considerable overlap between the various histological features. Our own case could be regarded as a "mixture" of the fibrohistiocytic and

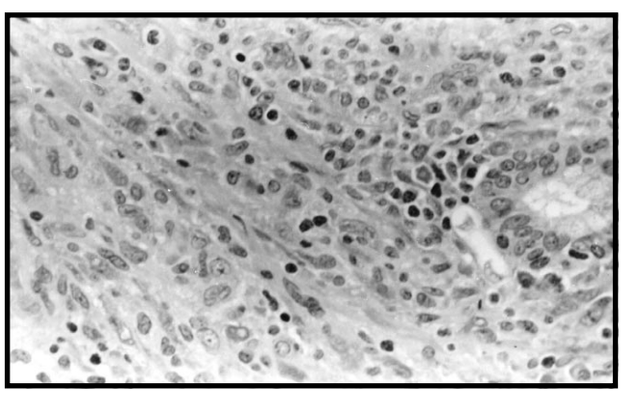

Figure 3a - Endobronchial inflammatory pseudotumor of lung. Histopathological section showing fibrohistiocytic and mononuclear inflammatory cell infiltration with an entrapped bronchial gland (right side). Hematoxylin & Eosin stain X 200.

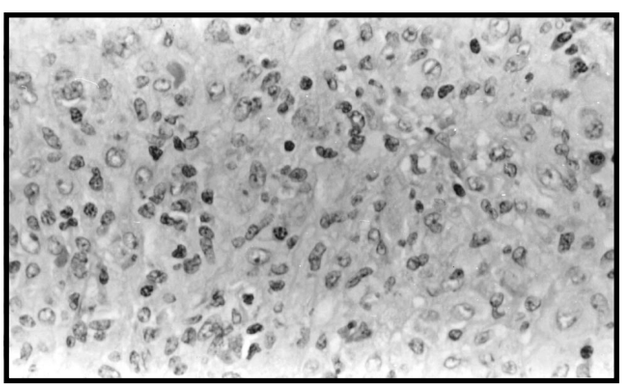

Figure 3b - Endobronchial inflammatory pseudotumor of lung. Histopathological section showing proliferating fibrohistiocytic cells with several scattered plasma cells. Hematoxylin & Eosin stain X 200

lymphoplasmacytic types (Figures 3a and 3b). The etiology of inflammatory pseudotumors of the lung is controversial.[5] In 1990, Barbareschi et al[6] demonstrated by immunohistochemical analysis that inflammatory pseudotumors are due to a mixed histiocytic-myofibroblastic reactive proliferation which evidently support the inflammatory theory of these lesions. Park et al[7] also suggested that the inflammatory pseudotumor of the lung could be a post inflammatory lesion associated with mycoplasma pneumonia infection. This finding together with those of Pilozzi et al[8] which suggested that accumulation of non-proliferating histiocytes induced by monocyte chemotactic protein I (MCP-I) is at least one of the pathogenic events occurring in inflammatory pseudotumor of the lung; are in keeping with the reactive inflammatory etiology of these lung lesions. Clonal cytogenetic changes in a clinically and pathologically typical case of pulmonary inflammatory pseudotumor were, however, found by workers from the University of California,[9] USA. Their findings in addition to the reported aggressive biological behavior of these tumors[10,11] and their association with other malignancies[12] support the idea that pulmonary inflammatory pseudotumor might be a true neoplasm rather than a purely inflammatory or reactive lesion. The different clinical presentations and biological behavior of inflammatory pseudotumor of the lung indicate that the choice of the type of surgical management depends on the site, size, number of lesions and the presence or absence of mediastinal involvement. We agree with the opinion of Dewar and Connett[13] that localized intraluminal tracheal pulmonary pseudotumor could be removed with a rigid bronchoscope. In the case presented, we did however, and in accordance with the recommendations of most authors,[14-16] excise the polypoid mass by simple bronchotomy preserving as much normal lung tissue as possible. In our experience such an excision has proven to be both curative and diagnostic, as frozen section and later paraffin sections histopathological examination could be carried out on the resection specimen. Bronchoscopic follow-up at 6 month intervals is, however, indicated in such cases.

In conclusion, in children and young adults presenting with a single and localized endobronchial obstructive lesion, the possibility of intraluminal pulmonary inflammatory pseudotumor should be considered, investigated and confirmed by histopathological examination. Local excision by simple bronchotomy is feasible in single and relatively small lesions. Bronchoscopic follow-up at 6 month intervals is, however, recommended in these patients to avoid undetected recurrences.

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References

1. Hancock BJ, Lorenzo M, Youssef SD, Marcotte JE. Childhood primary pulmonary neoplasms. J Pediatr Surg 1993; 28: 1133-1136.
2. Bahadori M, Liebow A. Plasma cell granulomas of the lung. Cancer 1973; 31: 191-208.
3. Berardi RS, Lee SS, Chen HP, Stines GJ. Inflammatory pseudotumours of the lung. Surg Gynecol Obstet 1993; 156: 89-94.
4. Matsubara O, Tan-Liu NS, Kennedy RM. Inflammatory pseudotumours of the lung. Progression from organising pneumonia to fibrous histiocytoma or to plasma cell granuloma in 32 cases. Hum Pathol 1988; 19: 807-814.
5. Storck M, Liewald F, Heymer B, Dienemann H, Sunder-Plassmann L. Inflammatory pseudotumours of the lung and trachea. Zentralbl Chir 1995; 120: 650-656.
6. Barbareschi M, Ferrero S, Aldovini D, Leonardi E, Colombetti V, Carboni N et al. Inflammatory pseudotumour of the lung. Immunohistochemical analysis on four new cases. Histol Histolpathol 1990; 5: 205-211.
7. Park SH, Choe GY, Kim CW, Chi JG, Sung SH. Inflammatory pseudotumor of the lung in a child with myocoplasma pneumonia. J Korean Med Sci 1990; 5: 213-223.
8. Pilozzi E, Stoppacciaro A, Rendina E, Ruco LP. Monocyte chemotactic protein-1 in the inflammatory pseudotumour of the lung. Mol Pathol 1998; 51: 50-52.
9. Snyder CS, Dell'Aquila M, Haghighi P, Baergen RN, Suh YK, Yi ES. Clonal changes in inflammatory pseudotumor of the lung: A case report. Cancer 1995; 76: 1545-1549.
10. Boman F, Champigneulle J, Boccon-Gibod L, Merlin JL, De Miscault G, Schmitt C et al. Inflammatory myofibroblastisc tumor of the lung with endobronchial, infiltrating multifocal and recurrent form. Ann Pathol 1995; 15: 207-210.
11. Batsakis JG, El-Naggar AK, Luna MA, Goepfert H. 'Inflammatory pseudotumor': What is it? How does it behave? Ann Otol Rhinol Laryngol 1995; 104 (4 Pt 1): 329-331.
12. Stappaerts l, Van Schil PE, Van Marck E, Vermiere P. Endobronchial inflammatory pseudotumor in a patient with B-cell chronic lymphocytic leukaemia and malignant pleural mesothelioma. Acta Clin Belg 1996; 51: 261-264.
13. Dewar AL, Connet GJ. Inflammatory pseudotumor of the trachea in a ten-month-old infant. Pediatr Pulmonol 1997; 23: 307-309.
14. Messineo A, Mognato G, D'Amore ESG, Antioniello L, Guglielmi M, Cechetto G. Inflammatory pseudotumors of the lung in children: Conservative or aggresive approach? Medial and Pediatric Oncology 1998; 31: 100-104.
15. Daudi FA, Less GM, Higa TE. Inflammatory pseudotumors of the lung: Two cases and a review. Can J of Surg 1991; 34: 461-464.
16. Scully R, Mark E, McNeely W. Case records of the Massachusetts General Hospital. Case 20-1994. N Engl J Med 1994; 20: 1439-1446.