Short reports

Endobronchial inflammatory pseudotumour exacerbating asthma

D Jayne, B Bridgewater, RAM Lawson

Summary

Primary pulmonary tumours are rare in the paediatric population. Of the benign tumours, the most common is the inflammatory pseudotumour (plasma cell granuloma). It can present as a variety of non-specific symptoms which may delay diagnosis. It usually occupies a peripheral pulmonary location and gives rise to symptoms due to its local mass effect and destructive invasion. We report the case of an endobronchial inflammatory pseudotumour presenting as an exacerbation of asthma in a 10-year-old girl. Appropriate investigation lead to pre-operative diagnosis and early complete surgical resection. A good prognosis is anticipated.

Keywords: inflammatory pseudotumour, plasma cell granuloma, endobronchial tumour

Asthma in the paediatric population is a common clinical entity. However, symptoms of airway obstruction may be caused by rarer pulmonary pathology. Therefore, when first-line therapy fails to control asthmatic symptoms, further investigation is necessary. This is illustrated in the case presented, where appropriate investigation of a 10-year-old asthmatic girl led to the uncovering of a rare benign bronchial tumour – an inflammatory pseudotumour (plasma cell granuloma).

Case report

A 10-year-old girl, who had been asthmatic since the age of five, presented with a four-month history of increasing cough and dyspnoea on exertion. She was initially treated with conventional bronchodilators and antibiotics but failed to show any improvement in her symptoms. A subsequent chest X-ray showed focal consolidation of the right upper lobe. Computed tomography (CT) confirmed an isolated right upper lobe collapse. No hilar lymphadenopathy was demonstrated. Bronchoscopy revealed an endobronchial tumour in the right main bronchus at the origin of the right upper lobe. Biopsies were taken and were consistent with an inflammatory pseudotumour.

An exploratory right thoracotomy confirmed the collapsed right upper lobe with a palpable tumour in the right main bronchus at the level of the upper lobe origin. A single enlarged hilar lymph node was subjected to frozen histologi-

cal examination and shown to be normal. A sleeve resection of the upper lobe was performed with the resection margins being macroscopically free of tumour.

Her post-operative course was uncomplicated and she was discharged home on the fifth day after surgery. Histology showed a pale nodular tumour obstructing the right upper lobe bronchus. Microscopically, the tumour was composed of predominantly spindle cells with a uniform infiltrate of lymphocytes. Plasma cells were numerous focally as were aggregates of foamy macrophages. These findings are consistent with a diagnosis of inflammatory pseudotumour of the fibrous histiocytoma type. Three months post-operatively the patient had made a complete recovery and was asymptomatic with no evidence of tumour recurrence.

Comment

Primary pulmonary neoplasms are rare in the paediatric population. Two-thirds of these are malignant or potentially malignant. One-third are benign, and although of these the inflammatory pseudotumour is probably the most common,¹ they remain extremely rare; in a series of 40 childhood primary pulmonary tumours reported since 1980 only one was an inflammatory pseudotumour.² They may occur in all age groups, and although reported in children as young as 12 months, the majority occur after the age of five years.³

Their usual presentation is as a solitary peripheral lesion. As such they may be asymptomatic or cause a range of symptoms including cough, haemoptysis, chest pain, recurrent infections and dyspnoea.⁴ In the case presented, the tumour occupied an unusual endobronchial position, causing obstruction of a major airway with symptoms identical to an exacerbation of pre-existing asthma. Their incidence in this site is variously reported as between 5% and 16%.^{1,4,5}

Although inflammatory pseudotumours are benign, non-neoplastic lesions, their natural history is unpredictable. The majority show a slowly progressive increase in size. However, approximately 5% of tumours behave in an aggressive manner.⁵ These may attain a considerable size with invasion of the neighbouring chest wall, mediastinum, diaphragm and pericardium.⁶ In addition, local tumour recurrence following incomplete surgical resection is well documented. Correct pre-operative diagnosis of these tumours is essential. The local

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infiltration and hilar lymphadenopathy with which they are associated can lead to a mistaken diagnosis of malignant disease. This can result in unnecessarily extensive resection being performed with serious implications in a young child. The importance of performing a chest X-ray in a child presenting with a history of increased cough and dyspnoea on exertion is well illustrated, and might have been performed at an earlier stage in this case. Both X-ray and CT scan usually show a solitary peripherally placed parenchymal mass with regular or irregular margins, with or without calcifications and occasional cavitation.⁷ Fine-needle aspiration cytology may be helpful,⁸ but complete exclusion of malignancy requires surgical biopsy. When endobronchial in position, as in this case, the tumours are readily accessible to endoscopic biopsy.

Although a variable tumour response to both corticosteroids and radiotherapy has been reported, the mainstay of treatment is by complete surgical excision to remove the destructive lesion, exclude malignancy and prevent recurrence. If this can be achieved

Inflammatory pseudotumours

- commonest type of benign pulmonary tumour in the paediatric population
- usually occur as solitary peripheral lesions, giving rise to a variety of respiratory symptoms
- rarely endobronchial in position
- may be locally invasive, mimicking malignant disease

then the long-term prognosis is excellent. Early diagnosis before significant local invasion has occurred enables a more conservative approach. This in turn relies on an increased awareness of these lesions presenting with common respiratory symptoms.

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Pneumonitis induced by sulphasalazine

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Summary

We describe a 65-year-old woman with eosinophilic pneumonitis induced by sulphasalazine. Laboratory findings revealed peripheral eosinophilia. The chest X-ray showed bilateral infiltrations, which disappeared after sulphasalazine was discontinued.

Keywords: sulphasalazine, rheumatoid arthritis, eosinophilic pneumonitis

Sulphasalazine is used in the treatment of rheumatoid arthritis and chronic inflammatory bowel disease.¹ Between 20% and 30% of treated patients exhibit adverse reactions (box).^{2,3} We present a patient with pneumonitis as an unusual adverse effect of sulphasalazine.

Case report

A 65-year-old woman was admitted because of progressive dyspnoea, nonproductive cough, high fever and headache. Her medical history revealed rheumatoid arthritis which had been treated by sulphasalazine (2 g daily) for the last six months. Arthritic signs were presently absent and the patient denied using other drugs. There was no history of chronic obstructive pulmonary disease, smoking or allergic reactions to drugs. On examination the patient was dyspnoeic. Her body temperature was 39°C. Crackles were heard at the lower zones of both lungs. Further examination was negative. Laboratory investigation revealed elevated erythrocyte sedimentation rate (84 mm after 1 h) and peripheral eosinophilia (15%). Haemoglobin, leucocytes and platelet counts, capillary blood gas analysis,

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