Case report

Juxtacardiac costal osteochondroma presenting as recurrent haemothorax

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Abstract

A 21-year-old man presenting with a recurrent spontaneous haemothorax was found to have an osteochondroma arising out of the left 4th rib. This was penetrating the apical part of the heart. Surgical excision was uneventful. © 2001 Published by Elsevier Science B.V.

Keywords: Osteochondroma; Exostosis; Haemothorax

1. Introduction

Osteochondromata are benign developmental abnormalities in which a portion of the epiphyseal growth plate cartilage becomes separated from the main epiphysis. This results in the laying down of an abnormal bony spur which is directed away from the epiphysis [1]. Osteochondromata are also known as exostoses. Costal exostoses involve the ribs in the region of the costochondral junction or at the vertebral end of the rib. Haemothorax is a well described presentation of costal osteochondromata [2-5]. Documented sites of intrathoracic injury from an osteochondroma include the diaphragm [3,4] and the lung [6]. There have been no previous reports of haemothorax following intrapericardial penetration by a costal osteochondroma.

2. Case report

A 21-year-old male was admitted with sudden left pleuritic chest pain. Chest aspiration yielded 500 ml of frank blood. A residual pleural collection was present during subsequent radiological assessment. He had previously been perfectly healthy and there was no history of chest trauma. After thorough investigation no cause for the haemothorax could be established.

At outpatient assessment 1 week later he remained asymptomatic. Chest X-ray now revealed an area of calcification in the left mid-zone adjacent to the left heart border. Eight days later the patient was admitted with another episode of pleuritic left chest pain which came on whilst lifting a heavy load. Chest X-ray showed a pleural collection. CT scanning demonstrated a lesion arising from the costochondral junction of the left 4th rib, projecting towards the cardiac apex. When viewed with mediastinal CT settings it stopped well short of the pericardium, but with lung settings the lesion appeared in continuity with the cardiac silhouette (Fig. 1). Surgical treatment was recommended.

Via a short left anterolateral thoracotomy (see Fig. 2), the section of the 4th rib bearing the lesion was divided anteriorly and posteriorly and cautiously elevated. Unexpected resistance to its removal was encountered together with an awareness that the spur was considerably longer than expected (Fig. 2). Although traversing the general pleural cavity, its tip was firmly anchored in the apical region of the heart. As the osteocartilaginous spur was carefully extracted, a cavity on the anterior pericardium leading directly towards the cardiac apex was exposed which was immediately occluded by the operating surgeons index finger. The cavity was surrounded by fibrofatty tissue which appeared to be acting as an insulating layer between the osteochondroma and the myocardium. Within the cavity a circumferential ridge could be felt outwith the fatty layer. This was taken to represent the point where the lesion had penetrated the apical pericardium. This circumferential ridge was marked by a corresponding groove on the external surface of the osteochondroma (see Fig. 2). On gradual withdrawal of the occluding finger we were pleased to find that there was no bleeding. After a further period of observation over a 5-min period, the cavity had visibly closed down and would no longer admit a finger tip. No attempt was made to excise the fibrofatty tissue at the cardiac apex. No abnormality of the intercostal vessels had been encountered during mobilisation and removal of

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the rib segment. After aspiration of residual blood and haematoma which lay freely in the pleural cavity, the chest was closed. The patient made a speedy recovery and was discharged home on his 3rd post-operative day. Histological examination of the excised rib showed the features of an osteochondroma. He remains well at follow-up 1 year later with no further episodes of haemothorax.

3. Comment

Osteochondromata usually present in childhood or adolescence. Presentation in adult life is, however, well documented, especially in the case of lower limb arterial penetration by an osteochondromatous spur [7]. It is suggested that in adult life the cartilaginous tip is resorbed leaving only a sharp bony spike. The mean age of patients presenting with vascular complications of osteochondromas (chiefly arterial penetration) was 23 years in a review of 97 published cases [7]. Thirty-four percent of these patients had a history of trauma or vigorous exercise preceding the onset of symptoms.

The finding of fibrofatty tissue around an osteochondroma is common when these lesions occur in the limbs [1] as a response to friction between the exostosis and the surrounding muscles and tendons. In our case the mechanical stimu-
lus to its development would have been the friction between the osteochondroma and the heart.

Despite extensive investigations at initial presentation no diagnosis was made. Probably the lesion was masked by residual pleural haematoma. This is a potential diagnostic trap in cases of spontaneous haemothorax and reinforces the need for careful follow up radiology once the haemothorax has cleared.

In non-traumatic haemothorax differentiation has to be made between actual haemothorax, for example due to rupture of an intrathoracic arterial aneurysm or dissection and a blood-stained pleural effusion, for example due to pleural malignancy, tuberculosis or pulmonary embolism. Pleural fluid haemoglobin level has been recommended in differentiating these two situations with a cut-off value of <2 g/dl for blood-stained effusions and of >7 g/dl for haemothorax [8]. In the conditions alluded to above, it is most likely that the initial clinical and radiological features will suggest the true diagnosis, rendering pleural fluid haemoglobin measurement un-necessary. Pulmonary arteriovenous malformation has been documented as a cause of spontaneous bleeding into the pleural space [9,10]. Despite careful investigation, in some cases no cause can be established [8]. One of us (KB) has encountered spontaneous haemothorax requiring intercostal intubation due to spontaneous rib fracture (due to severe coughing) in one case and to a primary mediastinal osteosarcoma which had invaded the aorta in the other.

Although we could not prove the exact anatomical source of our patient’s haemothorax, the potential for actual cardiac penetration was very obvious at operation. Whether this was the source of his haemothorax or whether there were small blood vessels in the depths of the apical cavity which had been eroded remains a matter for speculation. In future, if we were to encounter a similar case, we would have cardio-pulmonary bypass facilities available at the time of surgical resection.

References