SHORT REPORT

Infantile chest hamartoma—case outcome aged 11

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Abstract

Chest wall hamartoma of infancy is a rare lesion, usually presenting in the first year of life. Recent literature has recommended conservative management of asymptomatic children, yet most continue to undergo surgical resection irrespective of their symptom status. We report a case of spontaneous regression of a chest wall hamartoma of infancy, supporting recommendations for conservative management in asymptomatic children.

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Hamartoma of the chest wall in infancy is a rare lesion, usually presenting in the neonate or infant with a mass or respiratory symptoms. Most reported cases have been managed by surgical resection.^{1–6} We report expectant management and long term follow up of a case showing uncomplicated spontaneous resolution and rib remodelling.

Case report

A previously healthy 7 month old male was admitted to hospital with respiratory syncytial virus positive bronchiolitis. Coincidentally a protuberance of the right lower rib cage was

Figure 2 CT scan of chest, confirming a 5 cm × 3 cm right sided extrapleural mass containing calcification.

noted for the first time. Chest x ray at this stage showed a peripheral soft tissue lesion in the right lower zone with a well defined inner margin (fig 1). There was associated deformity of the adjacent 7th rib with splaying of the 7th and 8th ribs, indicating a mass component. Ultrasound scan confirmed the presence of a solid lesion. Skeletal survey showed no other abnormalities and blood parameters were normal.

Computed tomography (CT) scan of the chest (fig 2) confirmed a 5 cm × 3 cm extrapleural mass containing calcification, arising from one rib but involving adjacent ribs because of its bulk. A trephine biopsy was performed and the histopathology showed a cartilaginous and vascular hamartoma of infancy (mesenchymoma), considered to be benign. At 10 months of age the mass had increased in size on chest x ray but the patient remained asymptomatic; five months later the chest x ray showed slight reduction in mass size. At this stage he had typical asthmatic symptoms intermittently, consistent with his family history and responding well to inhaled terbutaline. At 11 years of age he remains well with the chest x ray showing total resolution of the lesion and only minor residual deformity of the 7th rib (fig 3).

Discussion

Chest wall hamartoma in infancy is a rare lesion, which may be present at birth, or present in early infancy, either as an asymptomatic mass or with some respiratory compromise. Histologically, the lesion is composed of cartilage, smooth muscle, and respiratory

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Figure 1 Chest x ray showing a peripheral soft tissue lesion in the right lower zone with a well defined inner margin. There is associated deformity of the adjacent 7th and 8th ribs, indicating a mass component.

Figure 3 Chest x ray ten years later showing only minor residual rib deformity.

epithelium forming a disorderly mass. A review of the English language literature has revealed 59 reported cases, with only scanty information available in standard paediatric or pathology textbooks.

Of those cases reported up to 1991, wide en bloc surgical excision was the recommended course of management.¹ This treatment is complicated by inevitable chest wall defect postoperatively, often requiring synthetic grafts for closure, and is associated with a risk of developing scoliosis in later life.² There are two early reports^{3,4} describing three infants who did

not have radical surgery. However, one of these children received chemotherapy as an alternative to resection³; one had only short term follow up of one year, remaining well without further growth of the tumour,⁴ and the oldest child remained well 16 years later but with evidence of residual tumour. Subsequently Gwyther and Hall,⁵ in 1991, were the first to recommend a conservative approach to the management of such lesions in otherwise asymptomatic children. However since then, of the 18 cases reported, only one has been managed conservatively and that patient has no recorded long term follow up.⁶

To our knowledge our patient is the first in whom conservative management in association with documented complete spontaneous regression of the lesion in the long term has been described. Our experience provides support for the recommendation of a conservative approach to management in an asymptomatic patient, thus avoiding the significant risks and complications of major surgery.

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