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European Journal of Cardio-thoracic Surgery 18 (2000) 622–624

EUROPEAN JOURNAL OF CARDIO-THORACIC SURGERY

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Case report

Intercostal arteriovenous hemangioma

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Received 12 April 2000; received in revised form 24 July 2000; accepted 15 August 2000

Abstract

We report a case of a 46-year-old man who presented with a chest wall tumor in the right hemithorax. He underwent thoracotomy to remove the mass, which was found to be an arteriovenous hemangioma arising from the intercostal muscle. Arteriovenous hemangioma is a rare tumor and chest wall is an extremely rare site for this tumor. This tumor should be considered in the differential diagnosis of the chest wall tumors. Complete surgical excision offers the best treatment. © 2000 Elsevier Science B.V. All rights reserved.

Keywords: Arteriovenous hemangioma; Chest wall tumor

1. Introduction

Skeletal muscle hemangiomas comprise approximately 0.7% of all benign hemangiomas [1] and the intercostal musculature is the location in only 1.4% of skeletal muscle hemangiomas [2]. Arteriovenous hemangiomas (AVH) represent a histologic variant of hemangiomas and it is regarded by some as an arteriovenous malformation causing varying degrees of shunting [3].

Herein, we report a case of a 46-year-old man who was found to have an arteriovenous hemangioma, arising from the intercostal muscle.

2. Case report

A 46-year-old man was referred to our clinic with an abnormal chest roentgenogram during an investigation for epigastric pain and dyspeptic complaints. He had a 20-year history of duodenal ulcer. Physical examination revealed only epigastric tenderness. Laboratory data were within the normal limits. Chest roentgenogram demonstrated a smooth, well-circumscribed 4-cm mass in the right upper lung field. Computed tomography (CT) showed a soft tissue mass in close contact with the pleura and minimal cortical thickening at the adjacent rib (Fig. 1). Liver scan revealed a 7-mm hypodense lesion in the posterior part of the right lobe, which was consistent with a hemangioma and this finding was also confirmed by ultrasonography. Endoscopy showed an active ulcer and severe edema in the bulbus. After a 2-week course of anti-ulcer treatment, the patient underwent thoracotomy. Bronchoscopy, which was performed prior to thoracotomy under general anesthesia, showed no abnormality. At thoracotomy, we found a 4-cm, well-demarcated but not encapsulated soft tissue mass, located extrapleurally, at the second intercostal space in the anterior axillary line. There was neither adhesion of the tumor to the lung tissue nor to the adjacent ribs. Frozen sections revealed a benign tumor. Total excision was performed. On histologic examination, in some areas irregular arteries and veins were observed in close association with one another; in other areas the lesion resembled a cavernous hemangioma (Fig. 2). The lesion was diagnosed as an arteriovenous hemangioma. Post-operative course was uneventful. The patient remains well and disease-free 4 years after surgery.

3. Discussion

Chest wall tumors originating from vasculary structures are very rare. To our knowledge, there has been only two reports of AVH arising from the intercostal muscles in the world literature [4,5].

AVH, so-called cirsoid aneurysm, racemose hemangioma or arteriovenous aneurysm can be divided into two types; those which occur in deep locations with varying degrees of

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PII: S1010-7940(00)00556-X

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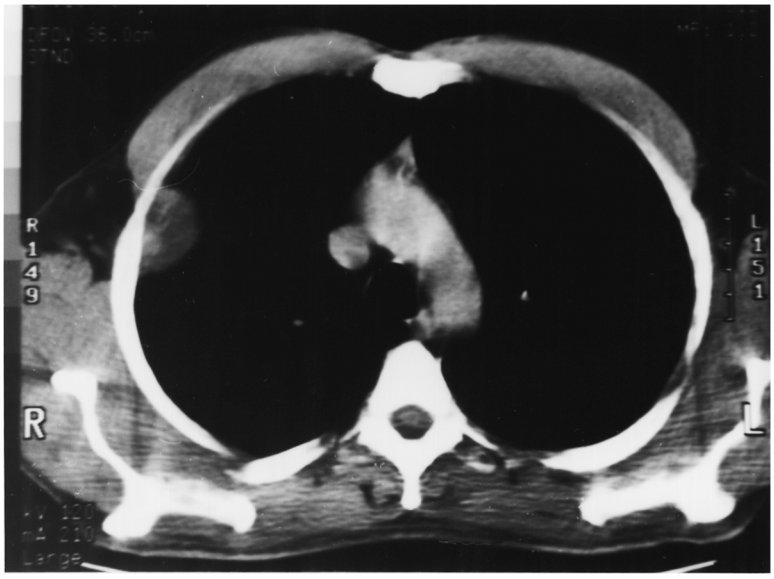


Fig. 1. CT shows the pleural-based soft tissue mass with minimal cortical thickening of the adjacent rib.

arteriovenous shunting and those occur superficially in the dermis with no significant shunting [3]. Many etiologic mechanisms have been postulated including the partial persistence of the fetal capillary bed, multicentric hamartomatous proliferation of vessels of the dermal subpapillary plexus [6] or trauma [7] but no definite cause has been identified. Association with chronic liver disease was also reported [8]. In our patient, no history of trauma or chronic liver disease was present.

Tumors with large shunting or close to skin may have pulsation, a continuous thrill or bruit over the mass. There may be pain resulting from pressure on nearby nerves. Our patient had a deep-seated AVH. He was asymptomatic and we did not notice any significant physical finding that is associated with AVH.

Radiographically, bone and soft-tissue hypertrophy and overgrowth or atrophy of the adjacent bony structures may be present as observed in our case as cortical thickening of the rib. Arteriography is helpful in the diagnosis of this tumor. It demonstrates large tortuous vessels of both the

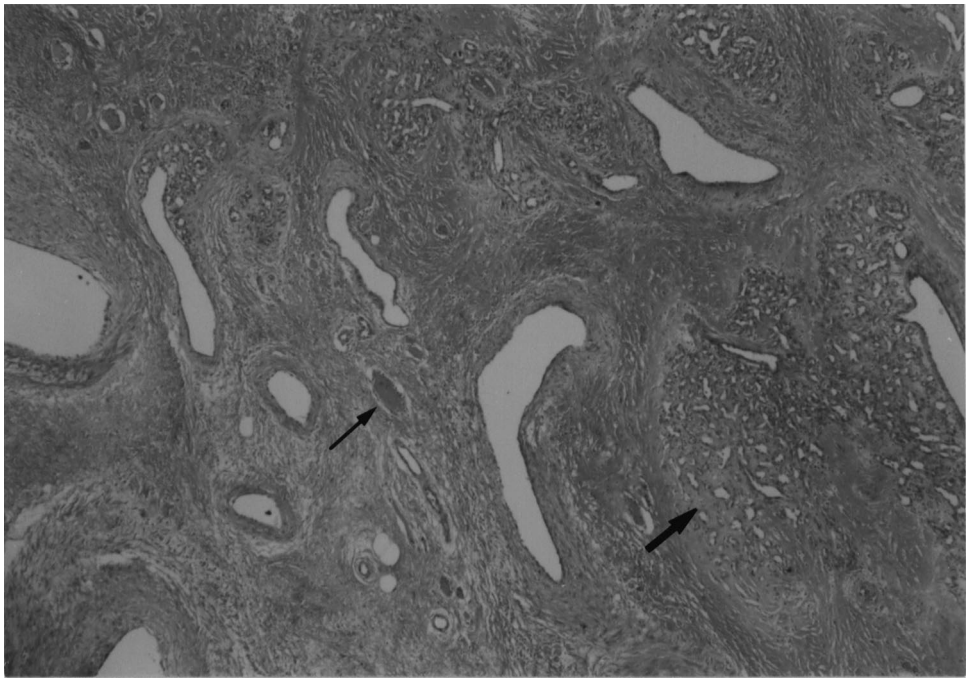


Fig. 2. Microscopic appearance of irregular arteries, veins with cavernous hemangioma-like areas (thick arrow). Residual skeletal muscle is observed between vascular structures (thin arrow). (H&E ×40).

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arterial and venous types with early filling of the draining veins [3]. Unfortunately we did not perform arteriography as we have never suspected a tumor of vascular origin.

Prompt treatment after the diagnosis is of importance as AVH is prone to bleed spontaneously or following a minor trauma. Treatment is difficult, particularly in huge tumors located adjacent to the main vessels and often dangerous. Ligation or embolization of the feeding arteries is palliative and subject to recurrence because arteriovenous communications are small and numerous [9]. However, these procedures may prevent cardiac failure, reduce the size of the tumor prior to surgery and minimize the risk of intraoperative complications like excessive bleeding. Partial extirpation usually causes rapid deterioration, disturbed wound healing and secondary bleeding [10]. Complete surgical excision is mandatory to achieve a cure. We did not encounter any difficulty in excising the tumor for it was small in size and well-demarcated.

Although a rare entity, arteriovenous hemagioma should be considered in the differential diagnosis of chest wall tumors and complete surgical excision will offer the best treatment.

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