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CASE REPORT

Coexisting subdural and intercostal haemangiomata

R. Aguiló, C. Montesinos, M. Llobera

Coexisting subdural and intercostal haemangiomata. R. Aguiló, C. Montesinos, M. Llobera. ©ERS Journals Ltd 1994.

ABSTRACT: Haemangioma in the chest wall is a very rare entity, seldom described in the literature. We report the case of a 23 year old woman presenting with independent, cervical subdural and intercostal haemangiomata. She suffered several episodes of recurrent, self-limiting subdural haemorrhage before diagnosis was made by magnetic nuclear resonance, aortography and selective intercostal angiography. Surgical excision was performed in both locations with excellent results.

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Thoracic Surgery Unit, Hospital "Son Dureta", Palma de Mallorca, Spain.

Correspondence: R. Aguiló
c/ Andrea Doria, nº 54
Cirugía Torácica
Hospital "Son Dureta"
Palma de Mallorca 07014
Spain

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Case report

The patient was a 23 year old woman with no personal or familial risk factors. She first presented at another hospital 4 yrs earlier, because of a self-limiting episode of acute headache, neck rigidity and hyperreflexia. Diagnosis of subdural haemorrhage was made by direct examination of the cerebrospinal fluid. Spontaneous amelioration of symptoms was observed within a few days. The same clinical picture was repeated one year later. On both occasions, no neural focality or lesion could be demonstrated by means of physical examination, computed tomography of the head, or cerebral angiography.

A third episode occurred 2 yrs later. It was investigated in our hospital by magnetic nuclear resonance of the central nervous system. An intrarrachidian, subdural arteriovenous malformation (A-VM) in the lower cervical higher thoracic region was then observed. When performing the arteriography, it was demonstrated that blood supply to the lesion was dependent on a branch from the left thoracic T5 intercostal artery (fig. 1). In addition, a second, unsuspected, asymptomatic, left-sided chest wall haemangioma was discovered, this one being supplied by the left thoracic T7 intercostal artery (fig. 2). On physical examination, no chest wall mass was palpable nor local thrill audible; and no other cutaneous or mucous vascular malformation or nevus could be detected. Preoperative chest X-ray did not demonstrate any pathological sign or image.

The patient was first operated upon by a neurosurgical team. After full recovery, she was referred to our thoracic surgical unit because of the chest wall haemangioma. Though asymptomatic, surgical excision was indicated on the basis of the well-documented intercostal vascular malformation that could bleed either spontaneously or because of minor trauma (as had been the case for the subdural

Fig. 1. – Selective left T5 arteriography. A complex nidus of arteriovenous communications is seen in the lower cervical region.

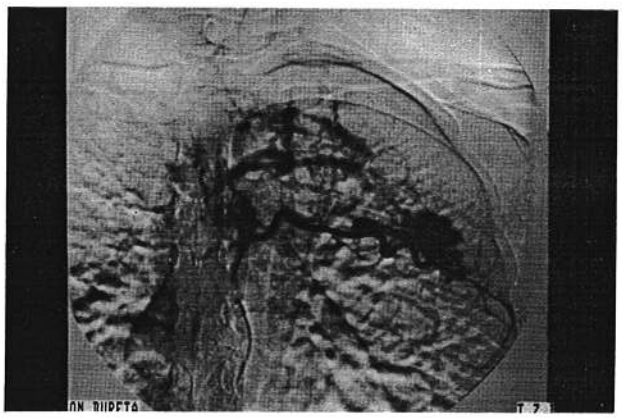

Fig. 2. – Selective left T7 arteriography. Enlarged feeding artery; arteriovenous angioma; and enlarged, varicose draining veins.

lesion), and was capable of provoking repeated and potentially serious episodes of intrapleural haemorrhage. At thoracotomy, the lesion was located at the seventh left intercostal space. It had the appearance of a very richly vascularized, "strawberry" tumour, 4×5 cm in size. When the pleural space was opened, neither pleural bulging nor lung paremchyma attachments to the lesion were observed; and ribs were not affected. Excision of the intercostal soft tissues beyond the limits of the lesion and ligature of the intercostal vessels was easily performed. Recovery presented no problems. Since then, no new episodes of internal bleeding have occurred.

Discussion

Haemangiomata are thought to be the most common of all human birth defects [1]. Single or multiple, they are often diagnosed as part of some complex systemic malformative syndromes, and sometimes as an isolated entity.

In the latter case, it is suspected that most remain undiagnosed, unless they become symptomatic. In reported series of solitary A-VM [2, 3], up to 50% are located within the skeletal muscle masses of the limbs, and up to 60% of the rest in the lung parenchyma; no cases have been reported in the chest wall.

In the literature reviewed, less than 2% of all skeletal muscle haemangiomata are found to be located in the intercostal musculature, so that only very few isolated cases have been reported [4]. Haemangiomata have never been included in any series of differential diagnosis of chest wall tumours. We have not found any case of double, coexisting intercostal and subdural haemangiomata.

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