Intractable pain due to rectus abdominis intramuscular hemangioma

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Intractable pain due to rectus abdominis intramuscular haemangioma

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Abstract

Haemangiomas are tumours of vascular origin accounting for approximately 7 % of all benign tumours. Three types of haemangioma have been described according to the vessel type involved: capillary, cavernous and mixed. Intramuscular haemangiomas (IMHs) are infrequent, accounting for less than 1 % of all haemangiomas and are mostly located in the extremities and the trunk. Intramuscular haemangiomas of the rectus abdominis muscle are extremely rare, with only one previous case reported in the literature to the best of our knowledge. In this report, we present the case of a patient with intractable pain related to IMHs of the rectus abdominis and we analyse diagnostic assessment and surgical management of the condition.

Keywords

Haemangiomas Intramuscular haemangiomas Rectus abdominis muscle Rectus abdomis haemangiomas

Introduction

Haemangiomas are tumours of vascular origin accounting for approximately 7 % of all benign tumours [1]. Three types of haemangioma have been described according to the vessel type involved: capillary, cavernous and mixed [1]. Intramuscular haemangiomas (IMHs) are infrequent, accounting for less than 1 % of all haemangiomas and are mostly located in the extremities and the trunk [1, 2]. Intramuscular haemangiomas of the rectus abdominis muscle are extremely rare, with only one previous case reported in the literature to the best of our knowledge [3]. In this report, we present the case of a patient with

intractable pain related to IMHs of the rectus abdominis and we analyse diagnostic assessment and surgical management of the condition.

Case report

A 37-year-old woman was admitted to our clinic with a history of a painful mass lesion localised on the right subcostal line. She had noticed the mass approximately 3 months before her admission. The pain was severe, constant and did not change with movement nor revert with morphine.

Seventeen years earlier, the patient underwent the surgical removal of an angioma localised in the right-hand chest wall, but unfortunately, no documentation of the previous surgical procedure was available. She also underwent a Roux-en-Y gastric bypass for morbid obesity 1 year before her admission, with good weight loss results: she weighed 140 kg (BMI 50 kg/m^2) before the gastric bypass and 72 kg, BMI 26 kg/m^2 on admission.

At the physical examination, the patient showed a solid immobile mass lesion approximately 10 × 4 × 3 cm in her right-hand subcostal line. The mass was not tender and did not change in size during movement; no pulsation or bruits were observed, and the overlying skin appeared normal except for a purple hemispherical nodule measuring 1 cm in diameter.

The patient underwent an extensive investigation to study the nature of the mass. At the ultrasound study of the thoraco-abdominal wall, there were multiple confluent hypoechogenic extrafascial images suggestive of venous vessels with slow blood flow. The echo-colour-Doppler study of subcutaneous tissues confirmed the presence of venous vessels, with thrombosis and no arterial flow (Fig. 1).

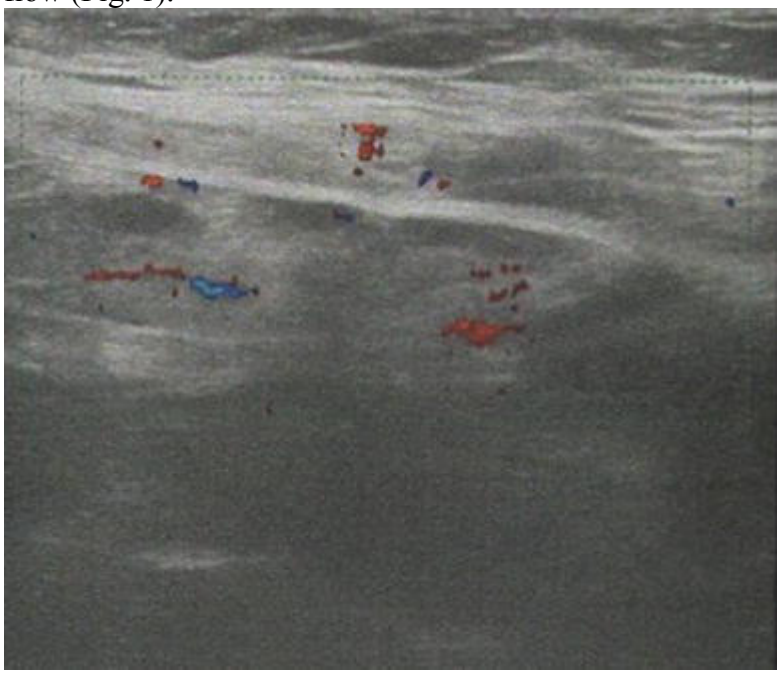

Fig. 1

Echo-colour-Doppler study of the subcutaneous tissues, showing the presence of venous vessels, with thrombosis and no arterial flow

Computerised tomography (CT) of the abdomen and chest revealed an isodense 6 × 5 cm mass lesion, with several nodular structures with small calcifications. On post-contrast images, the lesion contrasted minimally and homogeneously; it did not cause erosion or overgrowth on the underlying bone structure (Fig. 2). Magnetic resonance imaging (MRI) showed a mass with polycyclic contours; it showed multiple cluster structures with well-defined contours occupying most of the right rectus abdominis muscle and involving subcutaneous tissue. The mass was weakly hypointense in T1-weighted images and non-homogeneously hyperintense in T2-weighted images with dappled images hypointense in every sequences, related to calcifications; with contrast injection, the mass showed a non-homogeneous enhancement (Fig. 3).

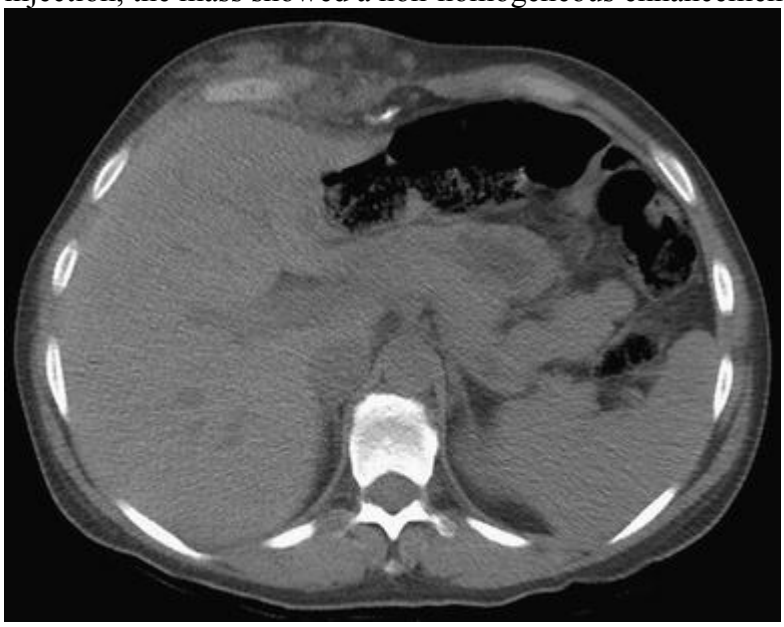

Fig. 2

CT of the abdomen and chest showing an isodense lesion, with several nodular structures

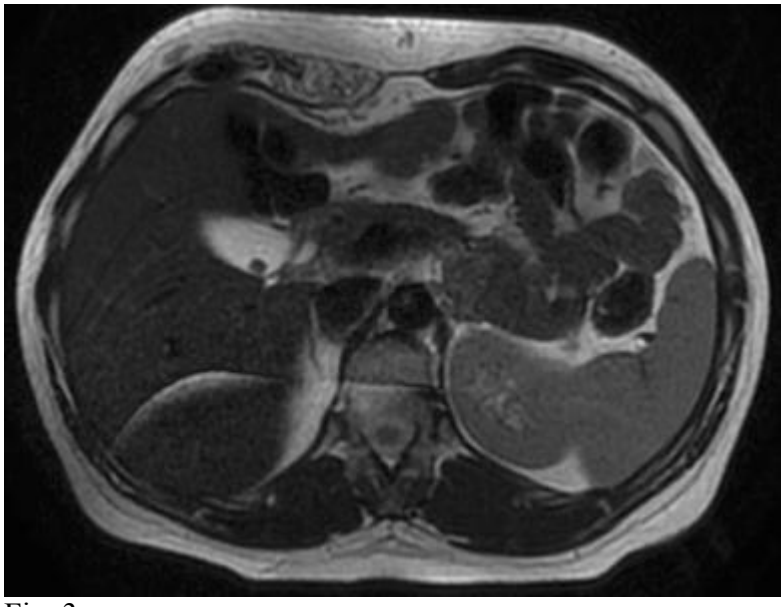

Fig. 3

MRI of the abdomen and chest showing a mass with multiple cluster structures occupying most of the right rectus abdominis muscle and involving subcutaneous tissues

Given the severe pain reported by the patient, unresponsive to pluri-pharmacological treatment, and the patient's increasing anxiety related to the unknown origin of the lesion, a multidisciplinary discussion between general surgeons, plastic surgeons and radiologists indicated surgical management.

The patient underwent surgical resection of the mass without complications. The operation was performed by means of a midline xipho-umbilical incision with concomitant excision of a cutaneous flap. The mass was completely removed en bloc with ligation of its vascular peduncle and macroscopic negative margins. The mass was found around and behind the rectus abdominis muscle on the median side and was completely removed, preserving most of the rectus abdominis muscle (Fig. 4). The abdominal wall was closed with the positioning of a polypropylene mesh. The postoperative course was uneventful.

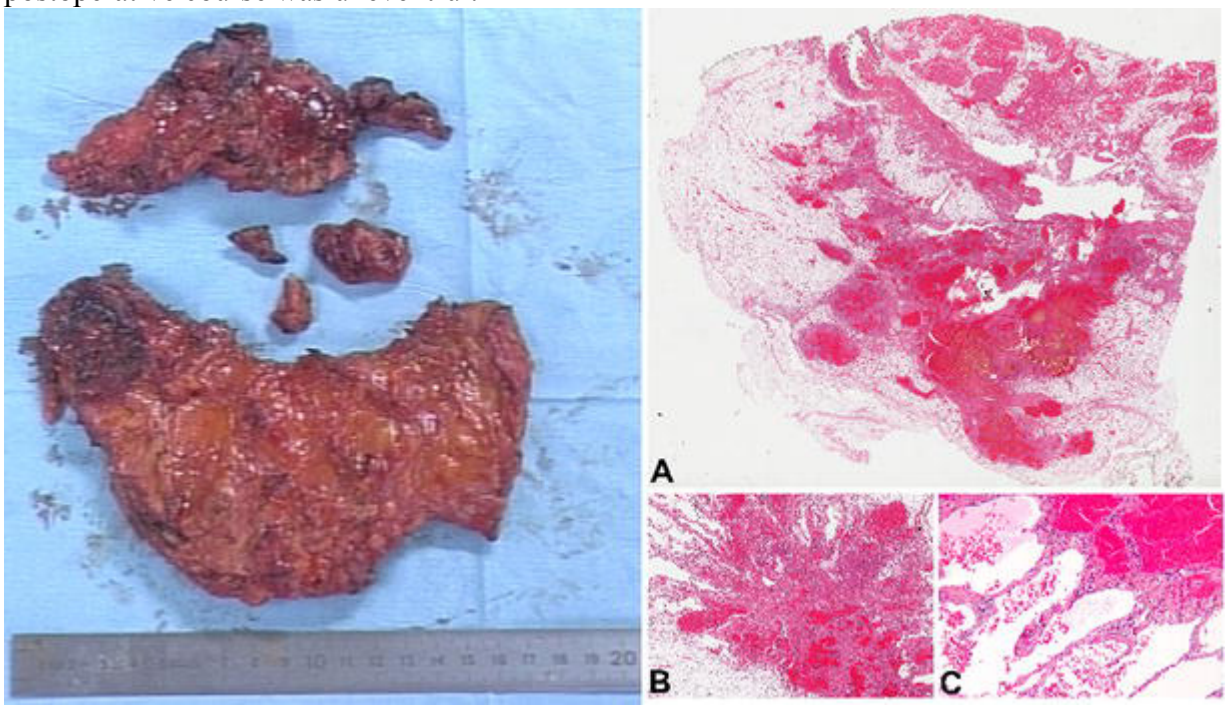

Fig. 4

Left, intraoperative image of the surgical specimen. *Right,* representative micrographs of the soft-tissue mass diagnosed as hemolymphangioma displaying two types of vascular spaces arranged in a roughly lobular pattern. **a** Low-power magnification (×2) showing the subcutaneous-intramuscular location of the lesion. **b**, **c** ×4 and ×20 magnifications, respectively, illustrating congested blood vessels and dilated lymphatic spaces lined by flattened endothelium

Macroscopically, the lesion was haemorrhagic with an irregular contour, and the skin and subcutaneous tissues were unremarkable. Histological examination of the specimen was consistent with a cavernous haemangioma with no mitotic activity and no necrosis (Fig. 4).

Discussion

Haemangiomas are tumours of vascular origin representing approximately 7 % of all benign tumours [1]. Intramuscular haemangiomas are infrequent, accounting for less than 1 % of all haemangiomas [4, 5]. Intramuscular haemangiomas are mostly located in the extremities and the trunk, probably due to the large muscle bulk of these regions [1, 5]. In a series of 110 patients [6], most lesions were in the lower extremities (66 %) followed by the upper extremities (27 %) and the trunk (6 %), while the thigh was the most common single location (32 %).

Intramuscular haemangiomas were first described by Liston in 1843 [7], while in 1972, Allen and Enzinger, in the first large study on 89 patients [8], suggested a classification based on the size of the predominant vessel type involved (small-vessel, <140 mm in diameter; large-vessel, >140 mm in diameter and mixed type). They correspond to capillary, cavernous and mixed type, respectively. These three histopathological types show clinical behaviour differences: the capillary type, characterised by predominantly capillary structures with proliferative activity, occurs more frequently, representing 68 % of IMHs and shows invasion of the surrounding tissue with a typically short clinical history; the cavernous type, characterised by large vessels with occasional mitotic activity, represents 26 % of IMHs and usually has a longer clinical history; the mixed type, and the least frequent with an incidence of 6 % is characterised by having both capillary and cavernous structures but resembles the cavernous type clinically [4–6, 9]. Moreover, capillary haemangiomas are smaller and located mainly in the trunk and upper limbs while the cavernous type prefers the lower limbs [1].

The aetiology of these lesions is unknown, although trauma or hormonal changes are considered important factors in the proliferation of embryonic vascular tissue. Also, a congenital theory involving an abnormal development of embryonic vascular structures has been proposed on account of the high incidence of haemangiomas in the years up to early adulthood [4, 10]: it has been estimated that 90 % of IMHs occur before the third decade of life [1, 4, 5, 10, 11]. With regard to gender distribution, some authors have reported the absence of sex predilection [4, 10], others a female predominance [9, 12] while others again a slight male preponderance [11].

The clinical presentation of IMHs is usually a palpable, localised mass with distinct margins and rubbery consistency, characterised by slow growth. Pulsations and other

signs associated with arteriovenous vascular malformations, such as bruits and thrills, are typically absent [11]. Symptoms are related to location of the mass and pain, which is reported to be present in 50–60 % of cases, is commonly secondary to neural compression [11].

Because of the rarity and vague presentation, more than 90 % of IMHs are misdiagnosed before surgery [4]. The main differential diagnosis is arterial malformation, which usually presents a thrill and murmur on auscultation, and a feeding artery evident during arteriogram. Furthermore, the vessels in haemangiomas have a normal architecture, whereas vascular malformations show dysplastic vessels.

Several radiological techniques can be helpful in the diagnosis of IMHs, for example, ultrasonography, CT, MRI or angiography [1], while plain radiography is usually not diagnostic [11]. Computerised tomography is useful in defining form, size and anatomic relationship of the tumour but generally does not allow a precise differentiation between the lesion and muscle or surrounding tissue [11]. Echo-colour-Doppler can reveal the vascular nature of the lesion [11]. Magnetic resonance imaging is very helpful, representing the method of choice in defining the vascular nature of the tumour [4] and providing soft-tissue delineation and spatial involvement of the lesion better than CT [11]. Intramuscular haemangiomas are characterised as iso-intense or hypo-intense with increased signal intensity due to fat presence on T1-weighted images, and markedly hyper-intense mass lesions containing tubular structures with blood flow on T2-weighted images in MRI [1, 4]. A preoperative angiography can be helpful in the operative planning and embolization of the tumour, although the latter alone has been reported as an inadequate treatment if not followed by surgery [1]. Angiography can be useful in detecting feeding arteries in large IMHs but may fail to do so in small tumours [11]. Aspiration cytology or open biopsy from these lesions is generally not recommended because of the risk of insufficient sampling or bleeding [1, 11]. In any case, the definitive diagnosis is made by histological study of the surgical specimen.

Reported treatment options for IMHs include simple observation, injection of sclerosing agents, corticosteroid treatment, embolization and surgical excision [1]. The use of corticosteroids, radiotherapy and sclerosing agents has been proposed both as an alternative and adjunct to surgical excision to reduce tumour bulk in the case of massive organ involvement.

Nevertheless, when suitable, surgery is the treatment of choice to exclude malignancy and for adequate treatment. Furthermore, IMHs do not undergo spontaneous regression and

may be locally destructive with time because of pressure exerted on neighbouring structures. Surgery should be planned according to patient's age, symptoms and eventual cosmetic, functional or neurological deficits, depth of invasion and vascular structure of the tumour [1]. To improve surgical results, a surgical approach of ligation of the feeding and draining veins of the intramuscular cavernous haemangioma with subsequent thrombosis of the haemangioma has recently been reported [13].

Indications for surgical resection include accelerated growth, intractable pain, functional impairment, risk for local skin necrosis, thrombocytopenia, cosmetic deformity and suspicion of malignancy. In Bella et al. [6], the main indication for surgical excision in most cases was symptomatic relief from increasing pain and discomfort. In our case, surgical indication was based on the pain reported by patient, which was unresponsive to pharmacological treatment and leading to severe impairment of daily activities and worsening of quality of life.

Because of the infiltrative nature of IMHs, normal muscle must be removed beyond the gross limits of the tumour to prevent recurrence. Reported local recurrence rates range between 18 and 61 % [6, 12] and have been attributed to incomplete excision, not to the histopathological type [6, 9]. On the other hand, Allen and Enzinger [8] have reported a difference in recurrence rates according to vessel type involved, with mixed type having the most recurrences (28 %) compared with the small-vessel (20 %) and large-vessel type (9 %).

Bella et al. [6] have reported a series of 110 consecutive patients with IMHs treated initially either with observation or surgical excision. In the 41 patients observed, there was a progressive increase in failures over time: 76 % of patients followed with observation were surgery-free up to 2 years, 66 % up to 5 years and 59 % up to 10 years. For patients treated with surgery, 86 and 73 % were recurrence-free at 2 and 5 years, respectively. Moreover, there were substantial differences in local recurrence when stratified by surgical margins: 93 % of patients were recurrence-free at 5 years when the excision was marginal and wide, 65 % when intralesional without any gross tumour remaining and 33 % when intralesional with gross tumour remaining. In this study, surgical margins and, to a lesser extent, tumour size were the only independent factors predicting local recurrence-free survival. Other factors such as age, gender, duration of symptoms and tumour location did not affect tumour recurrence. As the risk of recurrence is not completely understood, a meticulous and prolonged follow-up, based on clinical and radiological examination, are

strongly recommended in order to ensure the immediate diagnosis of eventual local recurrence [4].

In conclusion, the rarity of IMHs may render an accurate preoperative diagnosis difficult. A high index of suspicion is required, and complete surgical excision is mandatory.

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