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Left Intercostal Pain Due To Angioleiomyoma: Case Report

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ABSTRACT
Angioleiomyoma is a solitary smooth muscle cell tumour arising from the media of arteries and veins. We present a case of a 66-year-old woman who presented a left intercostal neuralgia in opposite to the D10-D11 intercostal space resisting to symptomatic treatment. A CT scan plus an MRI revealed an oval nodular formation, hyposignal T1 and a discrete hypersignal T2, centred in arch of the 10th left rib. After a thoracic surgery, Immunohistological studies added to a anatopathological examination confirmed the diagnosis of angioleiomyoma.

Keywords: *Angioleimyoma, intercostal pain, benign tumour, histopathological examination, immunostaining.*

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I. INTRODUCTION

Angioleiomyomas are formed by proliferation of smooth muscle cells in the vascular wall and are usually subcutaneous or appear as skin nodules in the extremities. Angioleiomyomas developing in organs other than the skin are very rare. The diagnosis of angioleiomyoma should not be based only on imaging. it requires histopathological examination withimmunostaining.

II. CASE REPORT

A case of a 66 year old woman, with a previous history of left intercostal ZONA in 1998 with anotion of trauma by a road accident in February 2020 responsible for intercostal pain for 15 days relieved by symptomatic treatment.

In November 2020, the patient had left flank pain radiating to the umbilicus. A digestive cause was ruled out. One month later, she presented left intercostal neuralgia opposite to D10, which was relieved by symptomatic treatment (paracetamol, pregabalin,vitamin B1).

Six months later (June 2021), the patient presented the same symptoms, a left intercostal neuralgia resisting to symptomatic treatment from where the indication of a dorsal medullary MRI which turned out to be normal.

In August 2021, a rheumatological consultation revealed a located pain in the area of the 10th left rib reproducing the intercostal neuralgia with a VAS pain of 9/10. A biological Analysis showed an absence of an inflammatory syndrome.

A thoracic CT scan performed on August 26th, 2021 showed no lesion. However, a complementary MRI by axial slices on the last ribs showed a well- limited nodular formation, oval, hyposignalin T1 and discrete hypersignal in T2, centered on the arc of the 10th left rib measuring 30/15mm, without costal lysis, Fig 1.

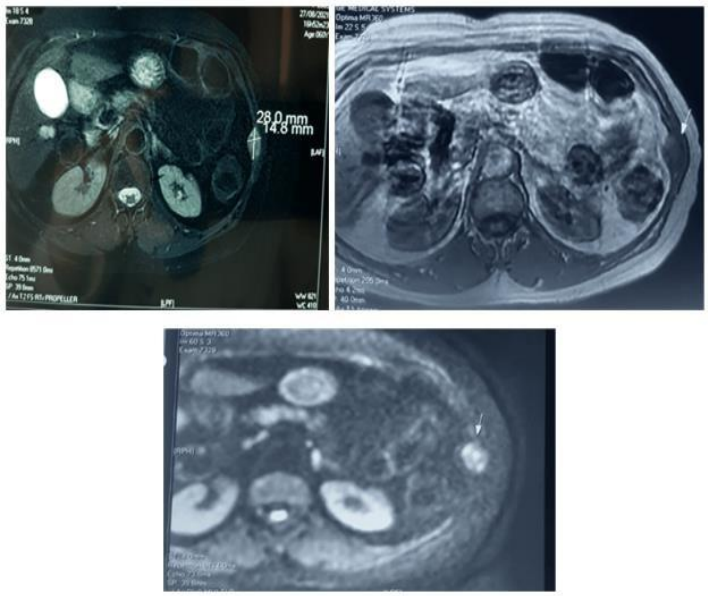

Fig 1: A thoracic CT scan and a complementary MRI showed a well-limited nodular formation, oval, hyposignal in T10, discrete hypersignal in T2, centered on the arc of the 10th left rib measuring 30/15mm, without costal lysis

In September 2021, the patient underwent thoracic surgery which showed a 3 to 4 cm/2.5 cm mass opposite to the 10th and 11th intercostal nerves, suggesting an intercostal neuroma.

The histological study was in favour of a benign cystic tumour suggestive of a hemangioma. Animmunohistochemical test was performed which showed a benign angioleiomyoma without suspicious character, Fig 2 & Fig 3.

During the follow-up, the patient described a total disappearance of her painful crisis and was highly satisfied with the surgery.

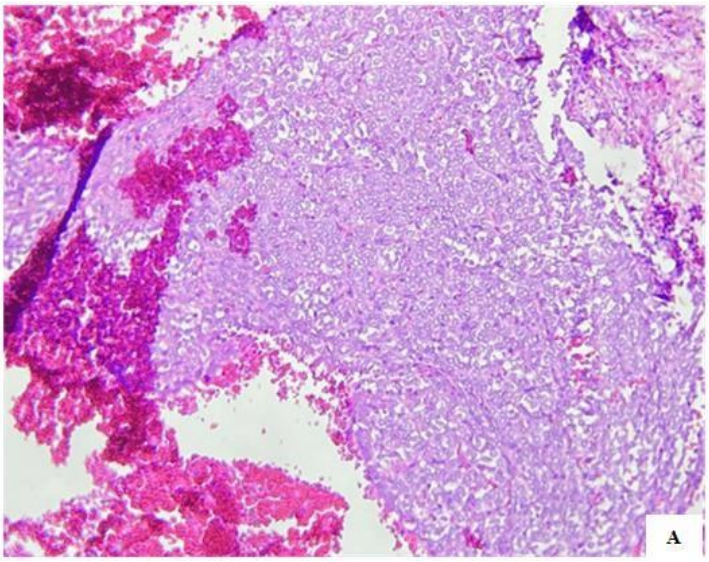

Fig 2: A-Histologic image Hemateine eosine staining showing the compact vascular component (Gx20). B-Histologic imageHemateine eosine staining showing the compact leiomyomateuse component (Gx20). C-Histologic image Hemateine eosinestaining showing proliferation with double vascular ant smooth muscle components delimiting vascular lakes (Gx10)

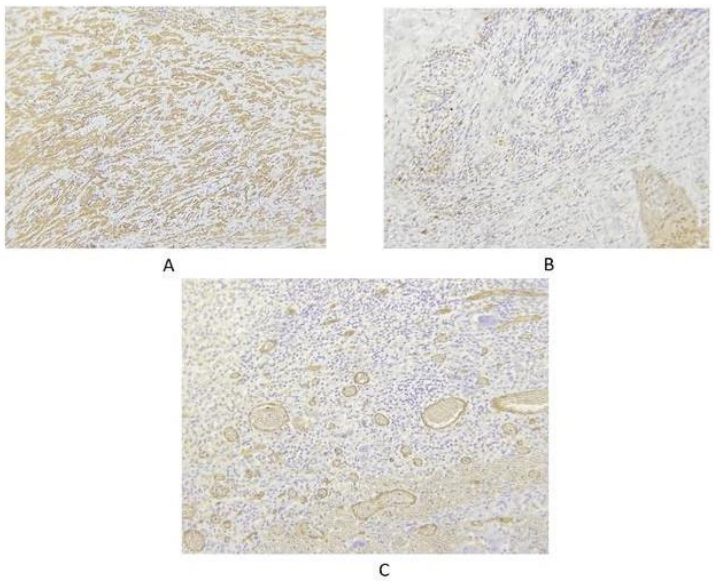

Fig 3: A-Diffuse expression AML. B-focale expression P100; C-vascular contigent CD34 expression

III. DISCUSSION

Angioleiomyoma (ALM), also known as vascular leiomyoma or angiomyoma, is a rare benign tumour that originates in the tunica media of bloodvessels [7]. It is mainly composed of mature smooth muscle cells with significant vascular components. It is classified by the World Health Organization (WHO) in 2013 as a benign pericytic (perivascular) tumour. It usually occurs in the subcutaneous tissue of the limbs, presented as a small, isolated, painful solid mass [1].

The incidence peaks in people between the ages of 40 and 60, mainly in women [6]. A majority of these tumors are generally <2 cm [5].

Hachisuga *et al*., reported that women were 1 to 1.7 times more frequently affected than men in a review of 562 cases. The clinical manifestations and physical examination of angioleiomyoma are non- specific. Most clinical symptoms are related to the location of the disease and the local compression occupying the space. Pain is considered the most significant clinical feature. In the Hachisuga study, pain was present in 58% of angioleiomyoma cases. Although suspected aetiologies such as trauma, infection and hormones have been put forward, the definitive aetiology of angioleiomyoma (ALM) is still unclear. In our case, the patient presented left intercostal neuralgia with severe pain. According to the classification of angioleiomyomas in Morimoto's theory, the tumour has been classified into three histological types: capillary orsolid, cavernous and venous. The solid subtype is characterised by small, slit-like vascular channels surrounded by bundles of smooth muscle. Cavernous tumours consist of fewer smooth muscle bundles and dilated vascular channels. Finally, venous angioleiomyomas consist of non-compact smooth muscle bundles and thick-walled vascular channels. Preoperative diagnosis of an angioleiomyoma is difficult before histopathology. Preoperative diagnosis of angioleiomyoma is difficult before histopathology. So, complete surgical resection is the only way to guarantee curative results. Angioleiomyoma can be correctly diagnosed by microscopy with conventional H&E staining. Special staining of smooth muscle cells, such as actin, desmin or myosin, and vascular endothelium, such as factor VIII or CD31, can help differentiate angioleiomyoma from haemangioma, angiofibroma, fibroma, angiomyolipoma and angiomyosarcoma, Fig 2 & 3 [2].

Its non-specific clinical and imaging presentation is a cause of diagnostic delay, detrimental to the patient's quality of life [4]. Effectively, the 10 months diagnosis delay has caused a lot of harm to our patient.

The most common location of this tumor is the subcutaneous tissue. They appear as painful solid subcutaneous nodules [8]. Morover, other locations have been described, such as intercostal, palmar and next to the elbow [2].

We report a case of a 66-year-old woman with a suspected nerve sheath tumour originating from the intercostal nerve. The mass was adjacent to but outside the intervertebral foramen, located at the 9th intercostal space. Although the mass bled easily, it was removed uneventfully by video-assisted thoracoscopic surgery.

There was a notion of Trauma 3 years ago responsible for the growth of the mass. Compression of the intercostal nerve by the mass seems to have caused left flank and back pain [3]. Another case was reported of a 56-year-old patient, in whom MRI showed a palmar mass located in the distal third of the forearm measuring 3.3 cm/1.6 cm/2.4 cm, hyperintense on fat- saturated images. Histopathological examination showed a benign spindle cell tumour containing cavernous vascular structures. The spindle cells were immunoreactive to smooth

muscle actin (SMA) [4]. We also report the case of a 72-year-old woman who presented with right elbow pain that had been evolving for 6 years. Specific MRI sequences showed a well- defined mass on the posterolateral side of the elbow close to the distal part of the triceps. It is a hypointense lesion on T1 with homogeneous enhancement after injection of gadolinium Histologic examination reported a tumor composed of vascular channels and smooth muscle consistent with an angioleiomyoma, subtype solid (or capillar) [5]. All these cases showed a marked improvement after surgical treatment

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