Hemosiderotic fibrohistiocytic lipomatous lesion: case report and review of the literature

Lesão lipomatosa fibrohistiocítica hemossiderótica: relato de caso e revisão da literatura

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ABSTRACT

CONTEXT: Lesions of the adipose tissue are the most common type of soft-tissue lesion among adults.

CASE REPORT: We describe the case of a 33-year-old female patient with a soft-tissue lesion in her left knee that was diagnosed as a hemosiderotic fibrohistiocytic lipomatous lesion. This type of lesion, which was described for the first time in 2000, prefersentially affects the ankle region of middle-aged women with a history of previous local trauma. Lesion recurrence is common, caused by incomplete resection, although there have not yet been any reports of metastases. After a review of the literature, we describe the clinical, radiological, morphological and immunohistochemical characteristics, along with their main differential diagnoses.

KEY WORDS:
Hemosiderin.
Histiocytes.
Soft tissue neoplasms.
Lipoma.
Venous insufficiency.

PALAVRAS-CHAVE:
Hemossiderina.
Histiócitos.
Neoplasias de tecidos moles.
Lipoma.
Insuficiência venosa.

RESUMO

CONTEXTO: Lesões de tecido adiposo são as mais comuns de partes moles em adultos.


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INTRODUCTION

Hemosiderotic fibrohistiocytic lipomatous lesion (HFLL) is a rare benign fibrolipomatous lesion, first described as an entity in 2000 by Marshall-Taylor and Fanburg-Smith. It accounts for 0.2% of benign lipomatous lesions.1

HFLL is usually superficial, solitary and circumscribed but not encapsulated. It generally affects the feet and ankles (80 to 92% of the cases), although it may appear in other locations, such as cheeks and hands.1,3 The mean age of the patients is 50.6 years and 80% are women.1,3 In 88% of the cases, there is an association with trauma.1,2 Despite the good prognosis, recurrence occurs in 50% because of incomplete resection.1,5 Venous stasis is implicated in the pathogenesis as an excessive tissue response.2 Mali’s acroangiodermatitis and vascular transformation of the lymph node sinuses, two lesions related to venous stasis, are differential diagnoses.2

Macroscopically, HFLL is a yellowish lesion, slightly darker than lipomas, measuring between 1 and 19 cm.1-4 Microscopically, it consists of mature adipose tissue without atypia, associated with fusiform cells that are accompanied by inflammatory infiltrate composed of lymphocytes, plasmocytes, histiocytes and mast cells, and abundant hemosiderin pigmentation.1-4 Cells with slight atypia,1,3 floret-like cells1 or osteoclast-like cells3 can occasionally be seen. In 20% of the cases, there are small vessels with hyalinization.1

HFLL is positive in most cases (77-100%) for CD34, vimentin (100%) and calponin (100%), focally positive for lysozyme and KP-1 and negative for protein S-100, desmin, smooth-muscle actin, CD68, HMB-45, epithelial membrane antigen, cytokeratins and caldesmon.1,3 Among the differential diagnoses, various lipomatous lesions should be considered, such as adiponecrosis, fibrolipoma, fusiform cell lipoma and liposarcoma, and also fibrohistiocytic/myofibroblastic lesions such as fibromatosis, nodular fasciitis, pseudo-Kaposi’s sarcoma, fibrohistiocytoma, dermatofibrosarcoma protubersans and pleomorphic hyalinizing angiectatic tumor. The clinical-morphological-immunohistochemical association is sufficient for correct diagnosis.1,3,4

CASE REPORT

Our patient was a 33-year-old white woman with pain and tumor on the posterior face of the left knee. Three years earlier, she had twisted this knee and, since then, she had been presenting pain and progressive swelling. On physical examination, there was a large-volume soft tumor accompanied by varicose veins on the posterolateral face of the left knee. The varicose veins were painful but without signs of inflammation. She also presented difficulty in flexing the left knee, with pain on touching and when walking. The patient was HIV-positive without signs of AIDS.

Ultrasound showed an expansive heterogeneous mass, laterally to the left popliteal fossa, of 8 cm in diameter. Tomography revealed a heterogeneous lesion with lacy highlighting, affecting muscle and subcutaneous tissues in the distal third of the left thigh. Magnetic resonance characterized it as lipoma.

The patient underwent surgery with an incision measuring 1.5 cm on the posterior face of the knee. The lesion was excised with free margins. The material consisted of soft friable brownish-yellowish fragments that together measured 10 x 8 x 6 cm and weighed 80 g. Under the microscope, proliferation of fusiform cells without atypia was observed, with mature adipocytes. The specimen was permeated with abundant hemosiderin pigment, along with small vessels with hyalinized walls (Figures 1 and 2). At the center, there was an old hemorrhage. There was no invasion of vessels and nerves. The margins were difficult to assess because the material was fragmented. Immunohistochemistry was positive for vimentin and CD34, and negative for cytokeratin, desmin, smooth-muscle actin, HHF-35, protein S-100 and CD 99.

Fifteen months later, the patient presented with a swelling adjacent to the previous surgical scar measuring 2 x 1.5 x 0.5 cm. On re-excision, the lesion had the same features, except for more ganglionic and multinucleated cells. There were no free margins. Six months after the second excision, the patient continued to show no signs of recurrence or metastasis.
**DISCUSSION**

We only found 29 cases of HFLL in PubMed1-8 (Table 1). Our case had some unusual characteristics and some interesting associated factors that may have been implicated in the pathogenesis, which is still a matter for discussion.

Among the 29 cases described, 22 (75.86%) were women, of mean age 50 years, and 25 cases (86.2%) were presented in the ankle. Except for one case in a child and a 32-year-old patient described by Browne and Fletcher,3 our patient was younger than the mean age in the literature. Moreover, our lesion is the first described in the knee. Among the others, 25 cases affected ankles, two affected the hands, one occurred in the cheek and one occurred in the thigh.18

One unusual morphological characteristic seen in our case was the hyalinization of the walls of small blood vessels, which has been seen in only 27.5% of the cases (eight cases) in other series.1-4

The idea that this lesion may be associated with venous stasis was first broached by Marshall-Taylor and Fanburg-Smith in 2000, while this concept was dismissed, given that none of the ten patients discussed rare entity that usually affects middle-aged women's feet and ankles. Complete resection is necessary in order to avoid local recurrence.

There is still much discussion regarding its reactive or neoplastic nature. This has generated controversy, and studies of greater extent are therefore needed to reach a definitive conclusion.

**REFERENCES**


**CONCLUSION**

Hemosiderotic fibrohistiocytic lipomatous lesion is a recently described rare entity that usually affects middle-aged women's feet and ankles. Complete resection is necessary in order to avoid local recurrence.