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Case Report
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Angiolymphoid Hyperplasia with Eosinophilia Involving Rectus Abdominis Muscle: A Case Report

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Abstract

Introduction: Angiolymphoid Hyperplasia with Eosinophilia (ALHE), also called epithelioid hemangioma, is a rare benign vascular tumor with less than one thousand cases reported worldwide in the literature. It mainly manifests as papules pink to red-brown in the skin in the head and neck. The etiology is unknown, but the histology of the tumor is characteristically referred to as a local proliferation of blood vessels with accompanying tissue infiltration of lymphocytes and eosinophils. The most frequent clinical manifestations are pruritus, pain and bleeding of the tumor, but it can also be asymptomatic. The most effective treatment is surgical excision, even though is high recurrence rates, of almost 50%, with a mean disease-free of 4.2 years.

Case Presentation: The authors present a 42 years-old man with a medical controlled HIV infection with an ALHE in the substance of the right rectus abdominis muscle with complaints for as long as one year of pain and loss of appetite (as a consequence of the latter symptom). Two surgical excisions were needed to be done because the first revealed as an incomplete one by the pathology exam.

Results: Complete tumoral excision was achieved in the second surgery, but the follow-up was only one year, which is insufficient to consider that the patient is disease-free.

Conclusion: Knowing this disease is important as this benign lesion has hard to identify margins leading to a high recurrence rate with surgery, even though it is considered the most effective treatment.

Keywords: Angiolymphoid hyperplasia with eosinophilia; Vascular neoplasms; Rare diseases; Muscle; Skeletal; Rectus abdominis; Excision

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Introduction

Angiolymphoid Hyperplasia with Eosinophilia (ALHE) also called epithelioid hemangioma is a rare benign vascular tumor, first described by Wells and Whimster in 1969 [1]. There are approximately 900 cases described worldwide in the literature [2]. Usually, it manifests in young to middle-aged Caucasian adults, with no clear sex prevalence, as a solitary or multiple pink to red-brown dome-shaped papules or skin nodules, more frequently encountered in the head and neck [2]. The most common locations are ear and periauricular area (36.6%), face (28.2%) and scalp (17.3%) [2], even though there are reported cases in many tissues and locations, such as skeletal muscle [3]. The etiology is currently unknown, but an immunologic allergic reaction may contribute to the pathogenesis [4]. The histology of this tumor has two main characteristics, the proliferation of blood vessels of varying sizes lined by plump endothelial cells and the response of the inflammatory cells, mainly in form of lymphocytes and eosinophils [4]. Also, some patients may develop peripheral blood eosinophilia and markedly elevated serum IgE levels, features typically seen in Kimura's disease, which lays the major differential diagnosis with ALHE. Kimura disease differs from the ALHE as it typically courses with a large subcutaneous mass in the periocular or submandibular region and lymphadenopathy and, as referred, serum eosinophilia.

The most frequent clinical manifestation is pruritus over the lesion (36.8%), but can also course with bleeding (25.3%), pain (20.2%), or even be asymptomatic (15.4%) [2]. A comprehensive review of the literature revealed only three cases of this tumor in individuals infected by the Human Immunodeficiency Virus (HIV) [5-7]. There could be a predisposition of these patients to ALHE because of both persistent viral infection and the depression of immunosurveillance.

The most effective treatment is surgical excision [2], as with complete tumoral resection, recurrence rarely occurs. One-third of cases that are incompletely excised do recur, either at the same site or along the course of the affected vessel [4]. Other modalities of treatment can be employed; however, with lower rates of success. Such as laser treatment with failure rates of 50% to 66.7%, radiotherapy with 60% of failure rate, and intralesional corticosteroid and oral retinoids with even less success [2,4].

In this article, the authors present a case report of an ALHE located in the right rectus abdominis muscle that the only treatment was surgical excision.

Case Presentation

The authors present a case report of a 45 years-old white male, with a medical history of HIV infection detected 25 years ago (with normal T cell counts and virus load in undetectable levels, treated with highly active antiretroviral therapy) and active smoking (5 pack-year units), presented to the plastic surgery department of our tertiary hospital with an ALHE located in the inferior third of the right rectus abdominis muscle. The patient was presented 3 months before to the general surgery department of the same hospital with localized pain in the right hypochondrium associated with a subcutaneous mass for as long as one year. Reduced appetite lead to a loss of 7 kg of weight in the same period. The tumoral excision was done and sent to the pathology exam. Its report revealed a vascular, multinodular neoplasm, constituted by epithelioid endothelial cells, with spaces filled with blood and areas of hemorrhage; mild to moderate chronic inflammatory infiltrate, rich in eosinophils. In the immunohistochemical study, there was positivity for the Cluster of Differentiation (CD) 31, focal for the FLI1 and pancytokeratins AE1/AE3, and negativity for the CD34. The diagnosis made was an ALHE with incomplete resection.

The study proceeded with Computed Tomography (CT) scan that showed that in the substance of the right rectus abdominis muscle at the level of epigastrium there was a captant nodular lesion with 27 mm × 11 mm, and another lesion inferiorly with the same characteristics with 12 mm long (Figure 1).

The patient was submitted to another surgical excision with a vertical median incision of the two superior thirds of the right rectus abdominis muscle after ligation of the upper and lower deep epigastric artery and comitant veins and ligation of the intralesional subcostal nerve (Figure 2 and 3). The surgical specimen includes the neoplasm of 2 cm × 1 cm, 5 cm × 2.5 cm and also the satellite lesion in suprafascial dependence.

Pathology confirmed that the lesions were ALHE and that was a complete excision. In the first year of postoperative time, no recurrence was noted (Figure 3). After that, the patient has lost the follow-up.

Discussion

The authors presented a case report of a middle-aged Caucasian patient as typically onsets the ALHE [2]. The most preponderant clinical manifestation was pain, which in this case resulted in a loss of appetite leading to a weight loss of 7 kg. Its abdominal musculature location is uncommon but skeletal muscle involvement has already been described [3]. Although the HIV infection causes various degrees of immunosuppression of the host, there is no augmented risk of developing ALHE described in the literature, even though

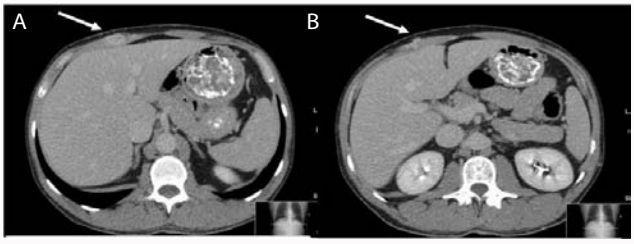

Figure 1: A) The pre-operative CT scan showing a right rectus abdominis muscle at the level of epigastrium a captain nodular lesion; B) another lesion just inferior to the latter one with the same characteristics.

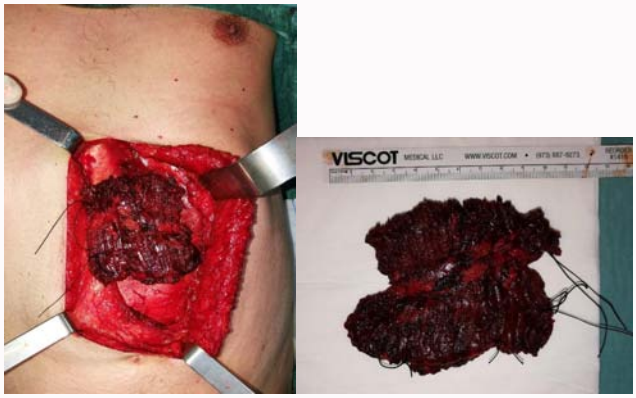

Figure 2: Surgical specimen - segmental resection of 10 cm × 10 cm of the right rectus abdominis muscle including both neoplasms.

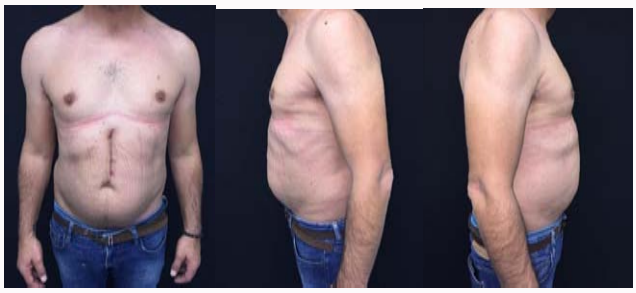

Figure 3: Three months of postoperative time with no abdominal bulging.

there are at least three cases reported worldwide of these conditions in association. Only after one year the onset of symptoms the tumor was excised and diagnosed. Because of its benign nature, no rapid growth or metastasis was found in the thoracoabdominal CT scans.

Two surgical excisions were performed for complete tumoral excision. The second surgery, as described, was needed because of the previous incomplete tumoral resection. The recurrence after surgical excision is unfortunately high (40.8%) [2], possibly because of the difficulty in identifying the margins in this highly vascular lesion. In this case it happened at the same site with two nodules identified in the CT scan, possibly in one of the deep inferior epigastric vessels branches or comitants. There was no need for medical therapy as the lesions were only two and operable. The tumoral resection was proven complete by the pathology exam, but the follow-up of only one year is insufficient to consider that the patient is free of recurrence, as the mean disease-free after surgical excision of ALHE is 4.2 years [2].

Conclusion

ALHE is a rare neoplasm affecting mostly the skin surface of

the head and neck region. Awareness of the existence of this tumor in other tissues and locations is important because ALHE surgical excision tends to be difficult as the tumoral margins are not well demarcated. It is also important to follow-up these patients because there is almost 50% of recurrence after surgical excision with a mean disease-free time of approximately 4 years.

Author Contributions

Gustavo Neves Pereira: Responsible for planning, collecting and interpreting data and drafting the manuscript. Diogo Ribeiro: Responsible for collecting and interpret data and review of the final version. Carlota Ramos: Responsible for collecting and interpret data and review of the final version. Joana Vaz: Responsible for collecting and interpret data and review of the final version. Luís Saraiva: Responsible for critical review, important intellectual content and approval of the final version.

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