ABSTRACT

Primary lymph node hemangioma is a rare entity with only a few cases having been reported in the literature. This article describes a case of a 68-year-old female patient with breast cancer who underwent modified radical mastectomy with a subsequent histopathological evaluation that revealed invasive ductal carcinoma histological grade III according to Nottingham's Combined Classification. Among the 14 resected lymph nodes, the presence of vascular proliferation (intranodal) was observed in one of them, consistent with primary nodal hemangioma. Thus, knowledge about this clinical entity is important in order to establish the correct differential diagnosis with malignant primary neoplasms and metastasis, in which therapeutics and prognosis are very different.

KEYWORDS: Hemangioma; lymph node excision; breast neoplasms; breast ductal carcinoma.

RESUMO

Hemangioma linfonodal primário é uma entidade rara, sendo que poucos casos foram descritos na literatura. Neste artigo foi relatado o caso de uma paciente de 68 anos com neoplasia mamária à direita e que foi submetida à mastectomia radical modificada com posterior avaliação histopatológica, que evidenciou carcinoma ductal invasor de grau histológico III, conforme Classificação Combinada de Nottingham. Dentre os 14 linfonodos ressecados, foi observada em 1 deles a presença de proliferação vascular (intranodal) consistente com hemangioma nodal primário. Dessa forma, o reconhecimento dessa entidade clínica torna-se imprescindível para a realização de diagnóstico diferencial de neoplasias malignas primárias ou metastáticas, que apresentam terapêuticas e prognósticos totalmente distintos.

PALAVRAS-CHAVE: Hemangioma; excisão de linfonodo; neoplasias da mama; carcinoma ductal de mama.
INTRODUCTION

Primary lymph node benign tumors are rare, and lymph node hemangiomas are even more uncommon, with no more than 60 cases described in the literature\(^1\). Lymph node hemangiomas are benign vascular tumors or hamartomas characterized by the presence of vascular proliferation containing blood cells, varying in size, which alter the cytoarchitecture of the lymph node\(^1\). An accurate diagnosis of lymph node hemangiomas is important to differentiate possible malignant tumors, which may manifest as lymph node metastases or primary malignant lymph node tumors, which require different therapeutics and prognosis\(^2\). This subject becomes especially relevant when the tumor is located in lymph nodes derived from axillary dissection in the proaeproteic of mammary neoplasia, as described in this case.

CASE DESCRIPTION

A 68-year-old female patient, asymptomatic, with a mammary nodular neoplasm of the lower quadrant, positioned at “5 hours” with approximately 3 cm in diameter. Previous history indicated the presence of systemic arterial hypertension and five pregnancies, with four vaginal deliveries and one cesarean section, in addition to one abortion. Still in relation to the patient’s gynecological-obstetric history, menarche took place at age 13 and menopause at age 45. A nodulation of approximately 1 cm was observed in the right axillary region, in addition to the nodule described above, during physical examination. Mammary ultrasonography showed a macrolobulated nodular image in the right lower quadrant, positioned at “5” hours, measuring 13 x 12 x 11 mm, in the right breast — BI-RADS 4A. Follow-up was performed with core biopsy in the mammary nodule, which revealed an invasive ductal carcinoma, histological grade III, according to the Nottingham Combined Classification (poorly differentiated tumor). After immunohistochemical analysis, an estrogen- and progesterone-receptor-negative tumor was noticed, with Ki-67 rate at 90%. Analysis by fluorescence \textit{in situ} hybridization (FISH) was inconclusive. Modified radical mastectomy with subsequent histopathological evaluation was the chosen method, with confirmation of invasive ductal carcinoma. The product of the right axillary dissection at primary level did not reveal presence of metastasis in the 31 resected lymph nodes. However, within these lymph nodes, the presence of vascular proliferation in the hilar and medullary intranodal region was observed in one of them, with blood cells inside (Figure 1). Given the histopathological evaluation, the intranodal finding corroborated the diagnosis of primary lymph node hemangioma (Figures 2 and 3). The patient continued their treatment of mammary carcinoma with chemo and radiotherapy, being considered cured of the nodal hemangioma due to resection.

DISCUSSION

Primary lymph node hemangioma affects a wide age range (4.5 to 75 years of age), predominantly females\(^1\). The processes for the evaluation of sentinel lymph node and mammary axillary dissection, as well as lymph nodes generated from inferior genital resections, may contribute to the predominance of females, since their diagnosis is often incidental, as can be observed in this report. In some cases, there is a description of lymph node mass palpation, especially when the most superficial chains are affected. The reports mention tumors up to 35 mm\(^2\).

Figure 1. Lymph node (HE-4x magnification): Lymph node hemangioma — lymph node dissection with vascular proliferation in the medullary region.

Figure 2. Lymph node (HE-40x magnification): Lymph node hemangioma — multiple proliferation of vessels with erythrocytes on the inside.
The pathophysiological mechanism of hemangiomas' formation in nodal sites is still uncertain. In general, the microscopic analysis does not contribute to diagnosis. Microscopically, hemangiomas may be divided into four histological types: capillary/cavernous, capillary lobular, cellular and epithelioid. Capillary/cavernous types are distinguished due to their greater involvement in the hilar or medullary region with preservation of the parenchyma. The differentiation degree may vary and the grouping of very close capillary or cavernous vessels, bounded by flat endothelial cells, with or without blood cells, may occur, as observed in this case. The capillary lobular subtype may occupy the entire parenchyma, acquiring the appearance of pyogenic granuloma. The cellular subtype, on the other hand, is consisted of extremely united cells, often with no channeling. Channeling may be marked by Schiff’s periodic acid and reticulin stains. Finally, the epithelioid is characterized by large endothelial cells. In terms of immunohistochemistry, the endothelium of the four hemangioma subtypes is positive for the following markers: smooth muscle actin, CD31, CD34 and factor VIII-related antigen. The differentiation of hemangioma subtypes is basically at the discretion of microscopy, and the impact of this sub-differentiation on the prognosis of patients is uncertain.

Imaging scans are of little help in the diagnosis of nodal hemangioma, although they are extremely common in the prognosis of lymph node investigation. The most classic presentation of hemangioma on axillary ultrasonography is a solid, well-delimited, hypoechogenic mass with a multilobular margin. However, hemangioma may also present other echogenic aspects (hype or iso) and intranodal microcalcifications. In terms of vascularization, it is expected that few vessels with a single vascular pole are found. The presence of multiple poles and of intense vascularization draws attention to the differential diagnosis of malignancy.

Among the benign differential diagnoses of lymph node hemangioma, there are: bacillary angiomatosis, lymphangioma, angiomatous hamartoma and epithelioid hemangioendothelioma. Among the malignant ones, there are: compound hemangioendothelioma, polymorphic hemangioendothelioma, Kaposi’s sarcoma and Dabska’s tumor. Lymph node malignancies occur, as observed in this case. The capillary lobular subtype may occupy the entire parenchyma, acquiring the appearance of pyogenic granuloma. The cellular subtype, on the other hand, is consisted of extremely united cells, often with no channeling. Channeling may be marked by Schiff’s periodic acid and reticulin stains. Finally, the epithelioid is characterized by large endothelial cells. In terms of immunohistochemistry, the endothelium of the four hemangioma subtypes is positive for the following markers: smooth muscle actin, CD31, CD34 and factor VIII-related antigen. The differentiation of hemangioma subtypes is basically at the discretion of microscopy, and the impact of this sub-differentiation on the prognosis of patients is uncertain.

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Given the range of differential diagnoses, including aggressive neoplasms, surgical excision should occur in all patients with suspected hemangioma due to growth of lymph node mass\textsuperscript{1,3}. The treatment of primary lymph node hemangioma consists of surgical resection, which is considered curative\textsuperscript{2,3,6}. The prognosis is excellent and the recurrence of these tumors is not described in the literature\textsuperscript{1}.

**CONCLUSION**

Therefore, it can be concluded that the knowledge on primary lymph node hemangioma is important in order to establish a differential diagnosis among the various lymph node pathologies, especially in view of the possible malignancies and the different therapy and follow-up approaches for each one.

### REFERENCES