Angiomyomatous Hamartoma of a Lymph Node - A Possible Late Complication of Radiotherapy? Case Report and Review of the Literature

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Abstract

Angiomyomatous hamartoma (AMH) of a lymph node is a benign vascular disorder of unknown etiology. We present a case of an inguinal AMH associated with previous lymph node dissection and radiotherapy to the same area. Pre- operative fine needle aspiration was not diagnostic. Surgical excision was successful, but the procedure got complicated by a high-output lymphatic fistula. Later, spontaneous closure of the fistula led to secondary lymphedema. AMH seems to be a rare nodal condition, commonly occurring in the inguinal region. The pathogenesis of AMH of a lymph node is still unclear. AMH may be related to previous radiotherapy or to chronically impaired lymphatic flow. Being a benign process, the need for surgery is unclear, as it may further impede drainage and probably needs a more tedious surgical technique (i.e. intra-operative lymphatic mapping with a dye, ties rather than diathermy etc). Recognition of this entity is important for the differential diagnosis of other angiomatous benign and malignant lymph node tumors, as well as for reconsideration of the need for surgery and the applied surgical technique.

Keywords: Angiomyomatous Hamartoma; Lymph Node; Radiotherapy

Introduction

Angiomyomatous hamartoma (AMH) of a lymph node is a benign vascular disorder of unknown etiology. It was first described by Chan et al. [1], as a distinctive lymph node lesion characterized by the replacement of the lymph node parenchyma with proliferating blood vessels, smooth muscle, and fibrous tissue in the absence of cellular fascicle formation. AMH was originally classified as a vascular tumor and was later included specifically in the group of smooth muscle proliferations of the lymph node, along with hilar smooth muscle proliferation, angiomylipoma, lymphangiomyomatosis, leiomyomatosis, and intranodal leiomyoma [2].

To the best of our knowledge, only 28 cases of AMH of a lymph node have been reported in the literature (Table 1) [1-14]. Patient median age 42 (ages 8 months to 82 years, standard deviation of 21), with a male-to-female ratio of 5:2. The disease mainly affected the inguinal and femoral lymph nodes, [1,3-9] although there are three reports of cervical lymph node involvement [10-12] and two of popliteal lymph node involvement [13,14]. It usually presented as a painless, unilateral inguinal lymphadenopathy. Although AMH of a lymph node is very rare, its recognition is important for the differential diagnosis of other angiomatous benign and malignant lymph node tumors.

We present a case of an inguinal AMH associated with previous lymph node dissection and radiotherapy to the same area. Surgical excision was successful, in that the tumor was completely excised measuring 7×11×2 cm, but the procedure was complicated by a high-output lymphatic fistula. Later spontaneous closure of the fistula led to secondary lymphedema.

Case Report

A 51-year-old man presented with a painful left inguinal mass that had been growing for two years. No lymphedema was noted. His past medical history was remarkable for seminoma of the left testicle at age 18 years which was treated by left orchiectomy, left retroperitoneal lymph node dissection, and radiotherapy. The patient had since been free of disease.

Ultrasound revealed a large subcutaneous ill-defined mass, 6×2×7 cm, with no hypervascularity and no lymphadenopathy. Magnetic resonance imaging demonstrated a 1×2.5 cm mass in the anterior aspect of the left thigh. Fine needle aspiration was not diagnostic. Surgical resection was recommended on the basis of the symptomatology.

Table 1: Cases of angiomyomatous hamartoma of a lymph node reported in literature.
Discussion

The pathogenesis of AMH of a lymph node is unclear. Some authors suggested that it represents a proliferative vascular and smooth muscle response either to chronic impairment of nodal lymphatic flow or to previous nodal inflammation [1,5]. Others proposed that it is caused by a malformation associated with congenital damage to the lymphatic area [7]. The histological features point to a disordered angiogenic process arising from hilar blood vessels, presumably the hilar vein and its branches [6]. The destruction of the nodal sinuses by these vascular and stromal alterations impairs the lymphatic circulation and may lead to lymphedema in some patients [1,6]. The continuous stimulation of the lymphatic vessels may cause vasoproliferation and, eventually, vascular transformation of the lymph nodes.

We describe a rare case of AMH of a lymph node in a 51-year-old man who had undergone previous surgery and radiotherapy in the same area. We speculate that there may have been a pathogenic relationship between these events, with the AMH representing a late complication possibly caused by chronic irritation or delayed lymphatic drainage.

The lymphatic fistula that occurred in our case has not been previously reported. It is possible that following the previous irradiation and AMH formation, a very large lymphatic vessel...
developed as a consequence of the chronically impaired lymphatic drainage through the groin, and this vessel was disrupted during surgery. This assumption is supported by the spontaneous closure of the fistula leading to chronic distal lymphedema.

Surgeons should be alert to the possibility of the rare diagnosis of AMH of a lymph node when confronted with a benign inguinal mass. AMH may be related to previous radiotherapy or to chronically impaired lymphatic flow. Being a benign process, the need for surgery is unclear, as it may further impair drainage. Core-needle biopsy to exclude malignancy may suffice, especially if the inguinal area was previously irradiated. If surgery is contemplated, one may consider intra-operative lymphatic mapping with a vital dye and using ties for the feeding lymphatics, rather than using heat energy resources for cutting out the mass.

References


