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Angiomyomatous hamartoma in the inguinal lymph node: A case report

İnguinal lenf düğümünde anjiyomatöz hamartom: Olgu sunumu

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ABSTRACT

Angiomyomatous hamartoma is a rare tumour of lymph nodes. This report describes a case of angiomyomatous hamartoma in the inguinal lymph node. The patient was a 33-year-old woman who underwent surgery because of a right inguinal mass. The excised specimen consisted of a grossly enlarged lymph node covered with fatty tissue, measuring 4.5 cm in diameter. On microscopic examination, the lymph node parenchyma was replaced by haphazardly dispersed thick-walled vessels and smooth muscle cells in a fibrous background. This process extended to the cortex from the hilum, and there was a thin cortical lymphoid tissue. Immunohistochemical actin staining indicated smooth muscle cells dispersing into the fibrous background. Although angiomyomatous hamartoma of lymph nodes is very rare, its recognition is important for differential diagnosis from angiomatous malignant tumors of lymph nodes.

Key words: Angiomyomatous hamartoma, inguinal, lymph node

ÖZET

Anjiomyomatöz hamartom lenf düğümlerinin nadir görülen bir tümörüdür. Bu çalışmada, inguinal lenf düğümünde anjiomyomatöz hamartom saptanan bir olgu sunulmaktadır. Otuz üç yaşında kadın hastada eksizyonel biyopsi ile alınan sağ inguinal kitlenin, makroskopik olarak 4.5 cm çapında, yağ dokusu ile kaplı bir lenf düğümü olduğu saptandı. Mikroskopik incelemede, fibröz zeminde gelişigüzel yerleşmiş kalın duvarlı vasküler yapılar ve arada dağılmış düz kas hücrelerinin lenf düğümü parankiminin yerini alarak hilustan kortekse doğru uzandığı izlendi. Kortekste ince bir kortikal lenfoid doku mevcuttu. Zeminde dağılmış düz kas hücreleri yapılan immünhistokimyasal çalışmada aktin ile pozitif boyandı. Lenf düğümlerinin anjiomiyomatöz hamartomu çok nadir görülmekle birlikte, lenf nodlarının anjiomatöz malign tümörlerinin ayırıcı tanısında önemlidir.

Anahtar sözcükler: Anjiomyomatöz hamartom, inguinal, lenf düğümü

INTRODUCTION

Primary vascular tumors other than Kaposi sarcoma are rare in lymph nodes (1). Angiomyomatous hamartoma is a benign vascular disease of lymph nodes with unknown etiology. This rare disease particularly involves inguinal lymph nodes. Few cases of femoral or cervical lymph node involvement have been reported (1-6).

We report here a case of angiomyomatous hamartoma in the inguinal lymph node in a 33-year-old woman.

CASE REPORT

A 33-year-old woman complaining of swelling that had persisted for 10 years in her right inguinal region was admitted in our clinics. On examination, a hard, mobile mass with a diameter of 3 cm was found in the right inguinal region, and the mass was excised with a clinical diagnosis of soft tissue tumor. Gross examination of the excised material demonstrated a mass

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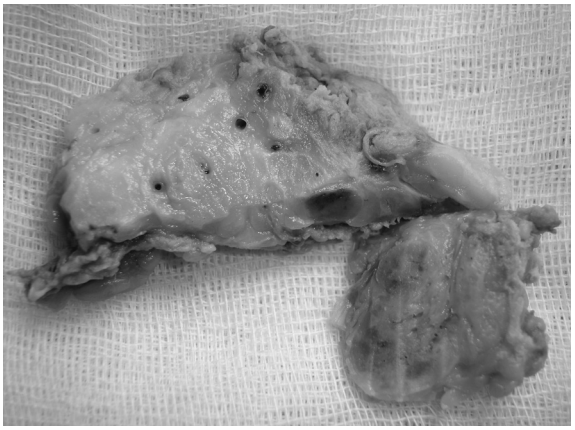

Figure 1. Section of angiomyomatous hamartoma. Lymph node is replaced by fibrous tissue, fat tissue and blood-filled vascular structures.

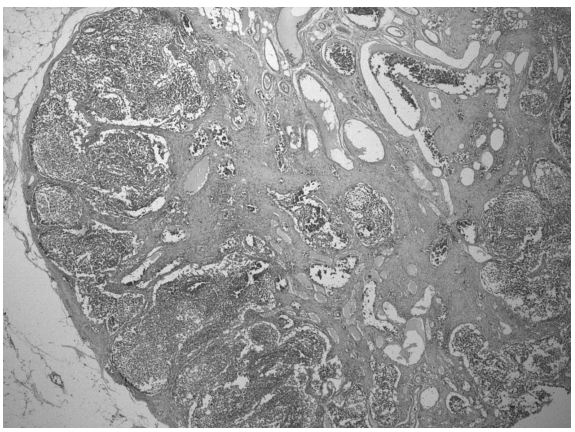

Figure 2. Lymph node parenchyma is replaced by fibrous tissue, randomly distributed smooth muscle cells, and numerous thick-walled vascular structures (HE x25).

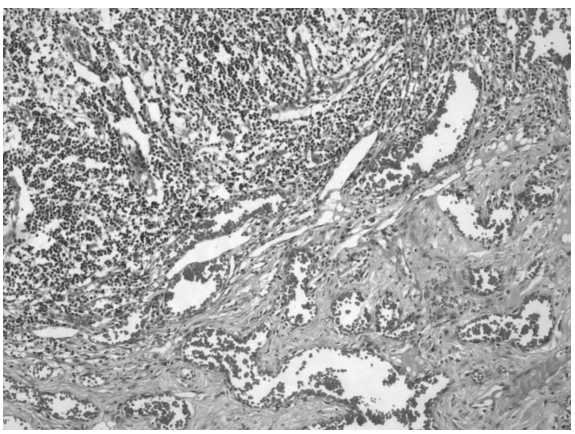

Figure 3. Abnormal and haphazardly arranged vessels in the medulla and their focally distribution pattern in the cortex of the lymph node (HE x100).

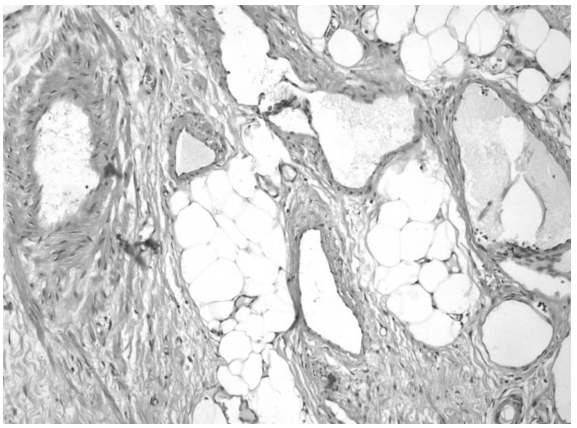

Figure 4. Angiomyomatous hamartoma with angiomatous and fat component (HE x200).

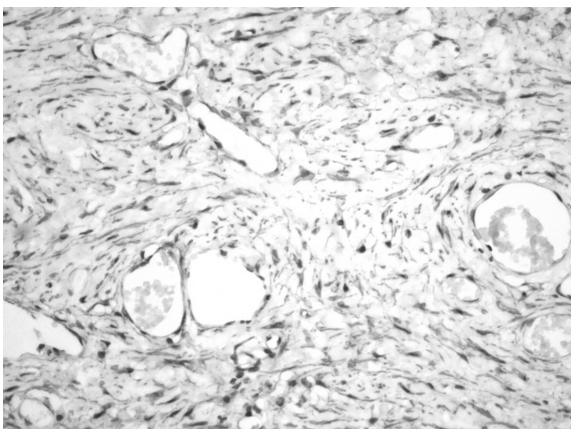

Figure 5. SMA immunoreactivity in smooth muscle cells in the lymph node parenchyma (SMA x100).

of 4.5 cm in diameter within adipose tissue, and section of the mass revealed yellow-white colored tissue with occasional spots of blood-filled vascular structures (Figures 1 and 2). Microscopic examination of the lymph node specimen revealed that parenchyma was replaced with fibrous tissue including randomly distributed smooth muscle cells and numerous thick-walled vascular structures (Figure 3). Occasional adipose tissue components were observed (Figure 4).

DISCUSSION

Angiomyomatous hamartoma was first defined by Chan et al. (1) in 1992. It predominantly involves inguinal lymph nodes of midd-

le-aged patients, but it has been reported in the femoral and cervical lymph nodes (1-6). Chan et al. reported 12 patients with ages ranging from 10 to 80 years (median 41.5 years); 10 of the 12 cases were males.

Angiomyomatous hamartomas were described as lesions that extended from the hilus to the cortex and comprised thick-walled vascular structures distributed within a collagenous stroma, and smooth muscle cells that were randomly distributed in or in close proximity to vascular structures, but not arranged in a fascicular fashion (1). In some cases, angiomyomatous hamartoma included adipose tissue (2,4). Thus, it should be differentiated from lymph node involvement of angiomyolipoma. The smooth muscle cells of angiomyolipoma had a prominent perivascular arrangement and expressed melanoma-associated antigen HMB-45 (7). Our case had an adipose tissue component, but the smooth muscle cells did not show HMB-45 immunoreactivity.

Angiomyomatous hamartoma should be differentiated from lymphangiomatosis, which usually involves intrathoracic and intraabdominal lymph nodes, with smooth muscle cells arranged in bundles and groups around the ectatic vascular structures (1). Nodal leiomyomatosis typically involves intraabdominal lymph nodes. It is characterized by proliferation of smooth muscle cells and lacks prominent vascular proliferation particularly resembling uterine leiomyoma.

The pathogenesis of angiomyomatous hamartoma has not yet been explained. Two possible mechanisms have been suggested. According to these hypotheses hamartomatous lesion is either acquired or it represents a reparative reaction against previous nodal inflammation (1). The present case had no history of surgery or inflammation. Recurrences and metastases of angiomyomatous hamartomas have not been reported (1-4). However, a secondary lesion after tumor resection may develop due to impaired lymphatic transport (5).

Recognizing angiomyomatous hamartoma as a rare and benign vascular tumor of lymph nodes is important in discriminating it from other benign and malignant vascular lesions of lymph nodes.

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