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Angiomyomatous hamartoma in an inguinal lymph node

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1. Introduction

Angimyomatous hamartoma (AMH) is a very rare, benign disease of lymph node characterized by parenchymal replacement of nodal structure with fibrous tissue containing proliferative blood vessels and smooth muscle [1]. It is mostly seen in inguinal lymph node but involvement of popliteal and cervical lymph nodes has also been reported [2,3]. The histopathological examination provides the correct diagnosis. Tubercular lymphadenitis is the common cause of lymph node swelling in the developing nation. It is only after pathological examination, the correct diagnosis of lymphadenitis in majority of cases is obtained. Reporting of this rare and benign etiology of lymphadenitis is essential to aware the importance of pathological diagnosis.

2. Case report

A 12-year-old female child from eastern Nepal presented with the complaints of right inguinal mass for one year. The swelling was associated with mild pain on and off and slowly increased in size. There was no history of trauma, weight loss and fever. Patient had no history of chronic illness. Her birth history was uneventful. She had normal developmental milestones. On examination, the swelling was 4.5 × 4 cm in size, firm, mobile and mildly tender. Provisional diagnosis of tubercular lymphadenitis was made, and laboratory investigations were ordered to confirm tuberculosis. All the laboratory investigations in the line of tuberculosis were negative. Then radiological investigation was ordered. Ultrasonographic scan of right inguinal mass suggested the differential diagnosis of benign tumor and advised for tissue biopsy. The abdominal and pelvic magnetic resonance imaging was normal.

In tissue biopsy, the inguinal mass measured 1.5 × 1 cm and was firm, gray-white appearance on cut surface. Under microscopic examination, a capsulated lymph node was identified with partially preserved architecture and the parenchyma was partially replaced by fibrous tissue containing numerous irregular blood vessels (Fig. 1). There was presence of interspersed spindle cells and few lobules comprised of benign adipocytes from hilum to cortex (Fig. 2).

Immunohistochemistry with a primary antibody against smooth muscle actin demonstrated smooth muscle cells in the blood vessel walls and in the stromal tissue (Fig. 3). The vascularity of the lesion was highlighted by CD34 antibodies (Fig. 4). Thus, the diagnosis of angimyomatous hamartoma of right lymph node was made and surgical removal of the node was done. After one year of follow up, patient has no complaints.

3. Discussion

Evaluation of inguinal swelling is pediatric population is a challenge, since differential diagnosis including congenital, neoplastic and infective lesions should be considered. Clinical examination along with radiological investigation is insufficient most of the times and histopathological examination of the specimen obtained from lesion is required.

In our case the absence of fever and normal laboratory findings ruled out the infective origin of inguinal swelling. Investigation for tuberculosis, brucellosis and typhoid fever was done in this case. Similarly tests for rheumatoid arthritis and systemic lupus erythematosus were negative which ruled out common causes of autoimmune origin of swelling. Normal clinical examination along with normal blood counts and organ function tests ruled out hematological malignancies. Magnetic resonance imaging of abdomen and pelvis ruled out the malignant primary lesions in these areas.

Ultrasonographic scan is the most cost effective non invasive and cheaper option for evaluation of palpable body surface swelling. Due to variability of sonographic appearance, the definitive diagnosis is not provided in most of the cases. In our case, the radiologist suggested the histopathological examination for the correct diagnosis. After pathological examination, the diagnosis of AMH of right inguinal node was made. Microscopically this AMH is characterized by an extensive replacement of nodal parenchyma by mixture of blood vessels, smooth muscle and sometimes adipose tissue.

The pathological differential diagnosis is lymphangiomyomatosis, leiomyomatosis and angiomyolipoma of the lymph node [4]. Lymphangiomyomatosis is microscopically characterized by the presence of smooth muscle cells forming fascicles around anastomosing ectatic vascular spaces, resulting in a pericytomatous pattern [5]. Leiomyomatosis is composed of a proliferation of compact bundles of smooth

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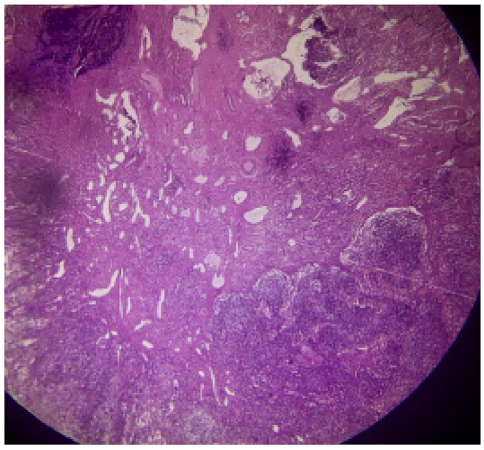

Fig. 1. Preserved architecture and the parenchyma of lymph node which is partially replaced by fibrous tissue containing numerous irregular blood vessels.

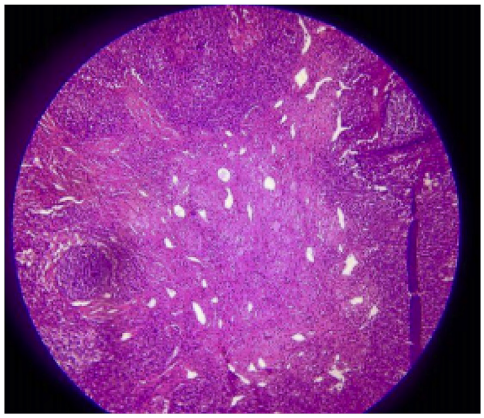

Fig. 2. Presence of interspersed spindle cells and few lobules comprised of benign adipocytes from hilum to cortex.

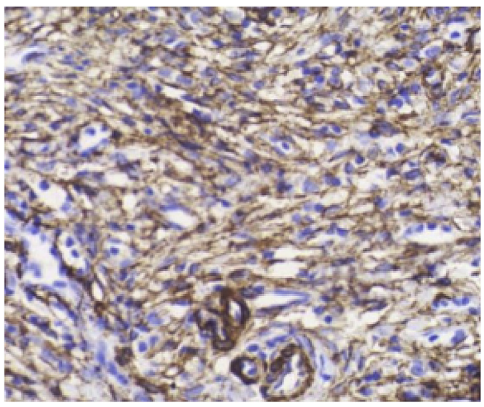

Fig. 3. Immunohistochemistry with a primary antibody against smooth muscle actin demonstrated smooth muscle cells in the blood vessel walls and in the stromal tissue.

muscle cells with an insignificant vascular component [6]. Angiomyolipoma usually show an epithelioid appearance of smooth muscle cells, hypercellularity, pleomorphism, prominent perivascular arrangement and positivity for melanoma associated antigen HMB-45 [7]. The exact etiology of AMH is still not understood. The long term impairment of nodal lymphatic flow or previous nodal inflammation may lead to vascular and muscle proliferative response [4,8].

The treatment of choice for this benign disorder is surgical removal of the lymph node. The child in the index case completely recovered after removal of the diseased lymph node.

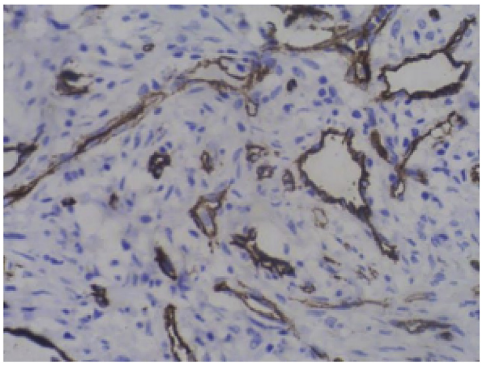

Fig. 4. The vascularity of the lesion highlighted by CD34 antibodies in immunohistochemistry.

4. Conclusion

Pathological diagnosis of AMH is essential to distinguish from angiomatous benign and malignant tumors of lymph nodes since AMH of lymph node can be surgically cured.

Patient consent

Consent to publish the case report was not obtained. This report does not contain any personal information that could lead to the identification of the patient.

Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

Conflict of interest

The following authors have no financial disclosures: DP, KG, AS, SP and VP.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.epsc.2019.02.004.

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