

Case Report

Primary intranodal hemangioma: a diagnostic differential for inguinal lymphadenopathy

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ABSTRACT

Primary intranodal hemangiomas presenting in the pelvis are extremely rare. They must be distinguished from malignant vascular lesions which are much more common in lymph nodes. Here, in this case report, we describe a case of a 21-year-old male presenting with painless groin swelling since childhood. Clinically, it was suspected as a reactive lymph node, however, the diagnosis was confirmed later on histopathological examination as primary intranodal hemangioma. Thus, diagnosing such rare entities is important in deciding the mainstay treatment for the patient. The approach to such a case, differential diagnosis, and a brief literature review are also presented.

Keywords: Lymphadenopathy, Hemangioma, Immunohistochemistry

INTRODUCTION

Lymph nodes often display significant vascularity in relation to various infections and other disease processes. However, primary vascular tumors originating in a lymph node are sporadic. Hemangioma, a benign vascular tumor, seldom occurs in lymph nodes.¹ It is imperative to accurately diagnose primary intranodal hemangiomas to avoid misidentifying them as potential malignant conditions, which are much more prevalent. These conditions can mimic lymph node metastasis or primary malignant lymph node tumors, necessitating different treatment plans and yielding distinct prognosis. Herein is described one such case of primary intranodal hemangioma arising in an inguinal lymph node of a 21-year-old male so as to create awareness among the dealing clinicians about this enigmatic entity.

CASE REPORT

A 21-year-old male presented to the surgery outpatient department with a chief complaint of painless swelling in the left inguinal region. The swelling had been present

since childhood and had gradually increased in size for the last two months. The patient had no history of trauma, fever, or tuberculosis contact. His medical history for any major disease or prior surgeries as well as family history of any cancer was non-contributory. On local examination of the swelling, it was firm, mobile, non-tender, and measured 2×2 cm in size. The overlying skin was normal. His systemic examination was unremarkable. On ultrasonography (USG), a well-defined homogenous, hyperechoic mass measuring 2×2 cm was identified in the left inguinal region.

Fine needle aspiration cytology (FNAC) was performed which yielded blood only with no diagnostic material. A repeat FNAC unfortunately gave similar results. Therefore, an excision biopsy was advised. The patient got the left inguinal mass removed surgically and the specimen was sent for histopathological evaluation. Grossly, it measured 2×2×2 cm. The cut section was grey-brown and showed foci of hemorrhage. Microscopic examination showed a well-circumscribed lesion comprising of the proliferation of vessels of varying caliber, prominently small-sized capillaries with few large vascular spaces filled with blood. No mitosis or

necrosis was seen (Figure 1). On immunohistochemistry (IHC), the endothelial cells were positive for smooth muscle actin (SMA), CD31, and CD34 and negative for epithelial markers, pan-cytokeratin, and lymphatic marker D2-40 (Figure 2). Based on the histological and immunohistochemical findings, a final diagnosis of primary intranodal hemangioma of the left inguinal lymph node was rendered. The postoperative period of the patient was uneventful. On follow-up after 2 weeks, there were no fresh complaints.

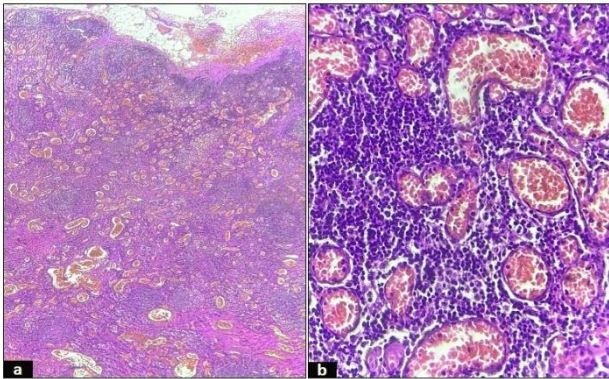


Figure 1: Photomicrographs showing (a) Nodal parenchyma effacement by proliferating vessels (H and E, x10); (b) Small, capillary-sized vessels admixed with few larger vessels (H and E, x40).

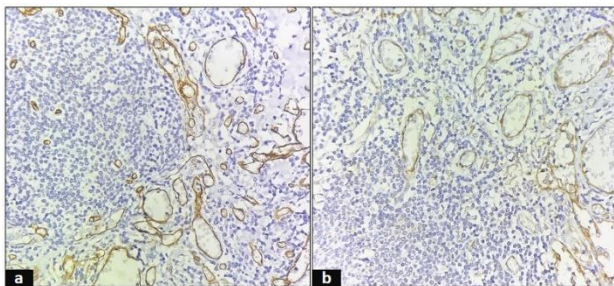


Figure 2: Endothelial cells exhibiting (a) SMA immunopositivity (IHC, x20); (b) Positive CD31 immunoexpression (IHC, x20).

DISCUSSION

Lymph node vascular neoplasms are rare, yet morphologically diverse, encompassing several entities with inconsistent and somewhat perplexing nomenclature. The variety of vascular proliferations found in lymph nodes includes benign vascular hyperplasias that coexist with various infections, as well as the unusual vascular transformation of sinuses. In their study on benign vascular tumors of lymph nodes, Chan and colleagues identified five histologic categories: (a) hemangioma and its variant; (b) angiomatous hamartoma; (c) hemangioendothelioma and its variant; (d) polymorphous hemangioendothelioma; and (e) lymphangioma.² These vascular tumors could be incidentally discovered during surgical exploration or

present as enlarged lymph nodes in a clinical setting. Mostly the lymph nodes involved are peripheral. Hemangioma in the inguinal lymph node is an exceptionally uncommon occurrence, with only a handful of cases documented in the existing literature.^{3,4} Its etiology is still debatable; however, a multifactorial origin is implicated, leading to clonal endothelial proliferation and aberrant downstream pathways, including hypoxia-inducible factor-1 α , vascular endothelial growth factor, phosphatidylinositol 3-kinase, and unchecked angiogenesis.^{5,6}

On further exploring the pertinent literature on primary intranodal hemangiomas, certain clinicopathological features of this condition draw special attention, which can help in its correct diagnosis and treatment. Clinically, the age range reported for the onset of intranodal hemangiomas varies widely, from 4.5 to 75 years. There is a higher prevalence among females; typically, only one lymph node is affected. However, males, particularly those in their young to middle years, have also been documented, as observed in our case. They can arise in peripheral and centrally located lymph nodes, such as supraclavicular, submental, cervical, axillary, inguinal, common iliac, and soft tissue lymph nodes. Most of the patients are asymptomatic. Nevertheless, a mass is often the major presenting symptom when superficial and peripheral nodes are involved.¹

On the other hand, a high index of suspicion and collaboration of investigations is required to clinch a definite diagnosis. Imaging techniques such as chest USG and magnetic resonance imaging (MRI), are extremely helpful in its assessment. USG often displays a well-defined mass with high vascularity. On the color doppler scan, SSS (sluggish speed signs) also show high sensitivity and specificity for diagnosing hemangiomas, which is an important diagnostic aid.⁷ MRI allows the characterization of hemangiomas and is also the most valuable means of diagnosing deep-seated ones.⁸ FNAC has been seldom beneficial for vasoformative lesions owing to paucicellularity and hemodilution. In our case too repeated FNAC, did not yield any diagnostic clue as it was heavily admixed with blood. However, researchers have suggested a good rationale for making cell buttons/cell blocks in such a scenario. It not only enhances diagnostic yield and cell preservation but also facilitates the use of IHC. The material obtained from these techniques is often more concentrated, resulting in a better understanding of tissue architecture.⁹

Macroscopically, the size of the affected lymph nodes can vary from 2 to 35 mm. Histology is identical to any soft tissue hemangioma. They can be capillary/cavernous/mixed, lobular capillary, cellular, and epithelioid. Capillary hemangiomas are well-circumscribed vascular proliferations having a single layer of flat endothelial cells lining them, giving the appearance of a conglomerate of capillary vessels. Under low magnification, the lesion has a lobular appearance in

some areas. In other areas, the vessels have a more anastomosing, slit-like appearance. Large, uneven gaps filled with erythrocytes and an occasional thrombi with partial organization are characteristic of the cavernous type. The stroma may be edematous or fibrosis might be prevalent. Lymph node architecture is usually well maintained. Germinal centers might become prominent. Lymph node hilum and medulla are mostly involved. The lobular capillary type can replace most of the nodal parenchyma and looks similar to a pyogenic granuloma. The cellular type consists of closely packed, solid to rarely canalized, vascular channels outlined by periodic acid-Schiff and reticulin stains. The epithelioid type is characterized by the presence of plump endothelial cells. There is no cytologic atypia, necrosis, mitosis, or extravasated erythrocytes in any of the types mentioned.² However, their main differential diagnosis includes lymphangioma, vascular transformation of the lymph node sinuses, bacillary angiomatosis, angiomatous hamartoma, hemangioendothelioma (epithelioid, composite, polymorphous), Kaposi sarcoma and angiosarcoma.¹ The distinction of lymph node hemangioma requires the use of immunohistochemical studies, particularly when dealing with patients in the context of neoplasia. The endothelium of most hemangioma subtypes exhibits positivity for the following markers: SMA, CD31, CD34, and factor VIII-related antigen.^{1,10} SMA will be additionally positive in the spindle cells in angiomatous hamartoma. Nevertheless, it is uncertain whether the differentiation of hemangioma subtypes, primarily determined by microscopy, has any impact on patient prognosis.

Surgical excision is the primary and curative treatment of choice, with other current modalities including chemical or thermic sclerotherapy.^{4,9,10} No recurrences have been documented, which suggests a good prognosis for these cases.

CONCLUSION

Primary intranodal hemangioma, although rare, should be considered in the differential diagnosis of localized lymphadenopathy, particularly of the inguinal region. Recognizing its typical morphology is vital for avoiding diagnostic errors. Preoperatively radiological and cytological investigations can aid in its diagnosis, but it is the histopathology that is confirmative. However, more insight is required to understand the genesis and behavior of hemangiomas occurring at such atypical sites.

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REFERENCES

1. Elgoweini M, Chetty R. Primary nodal hemangioma. *Arch Pathol Lab Med.* 2012;136(1):110-2.
2. Chan JK, Frizzera G, Fletcher CD, Rosai J. Primary vascular tumors of lymph nodes other than Kaposi's sarcoma. Analysis of 39 cases and delineation of two new entities. *Am J Surg Pathol.* 1992;16(4):335-50.
3. Oswal R, Jagtap SV, Vartak SA, Jagtap SS, Oswal S. Intranodal capillary-cavernous hemangioma- an uncommon histopathological lesion. *Int J Health Sci Res.* 2021;11(4):114-7.
4. Jeong S, Park SB, Lee EJ, Kim MK, Park HJ, Lee ES. Intranodal hemangioma in the pelvic cavity: a case report. *Curr Med Imaging.* 2020;16(9):1182-4.
5. Marchuk DA. Pathogenesis of hemangioma. *J Clin Invest.* 2001;107(6):665-6.
6. Tang R, Xian D, Xu J, Peng H, Pan S, Zhong J. Proanthocyanidins as a potential novel way for the treatment of hemangioma. *Biomed Res Int.* 2021;2021:5695378.
7. Furuta T, Shimose S, Nakashima Y, Kubo T, Ochi M. Novel sluggish speed signs on ultrasound is indicative of hemangiomas. *Acta Radiol.* 2017;58(10):1231-7.
8. Vilanova JC, Barceló J, Smirniotopoulos JG. Hemangioma from head to toe: MR imaging with pathologic correlation. *Radiographics.* 2004;24(2):367-85.
9. Sharma S, Mannan R, Bhasin T. Cytological diagnosis of deep-seated cellular hemangioma of the parotid gland by using cell button technique. *J Cytol.* 2016;33(3):174-6.
10. Terada T. Capillary cavernous hemangioma of the lymph node. *Int J Clin Exp Pathol.* 2013;6(6):1200-1.

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