

Intranodal capillary-cavernous hemangioma: Report of a very rare case

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Abstract

Mixed type capillary-cavernous hemangioma is a rare vascular anomaly, with an intranodal localization being extremely rare. Its finding is often accidental but may be clinically symptomatic. The diagnosis relies on histopathology, showing a proliferation of capillaries and cavernous vessels filled with erythrocytes and lined by endothelial cells. Magnetic resonance imaging with enhanced and T2 STIR sequences is the most efficient imaging for diagnosing this type of hemangioma. Rarely, malignancy or another vascular lesion can be evoked as a differential diagnosis of mixed hemangioma. Treatment in this localization is often surgical, even if other possibilities exist for other areas. In this article, we describe a very rare case of intranodal capillary-cavernous hemangioma.

Keywords

Capillary hemangioma, cavernous hemangioma, vascular malformation, lymphadenopathy, intranodal tumor

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Introduction

Hemangiomas are frequent benign lesions, of which capillary-cavernous hemangioma (CCH) is one of the rarer types, considered as a vascular malformation between low- and high-flow lesion according to the last International Society for the Study of Vascular Anomalies (ISSVA) classification.¹ Such lesion can lead to functional impairment and aesthetic consequence.² Arising in a node is extremely rare, with only 10 intranodal CCH.^{3–9} They can be suspected of primary—especially malignant vascular tumor—or secondary malignancies.¹⁰

Pathological diagnosis relies on special stains and immunohistochemical features and is quite consensual. The challenge facing those vascular malformations is to use imaging—especially magnetic resonance imaging (MRI) with enhanced sequence—to avoid invasive or aesthetically harming sampling.² Starting from that, correlating pathology and imaging is useful for clinicians and radiologists. In this article, we report a case of incidental axillary intranodal mixed hemangioma and review cases of intranodal CCH.

Case report

We report the case of a 44 years old woman, who was being followed for a chronic tendinopathy of the right shoulder, in

need of a shoulder MRI (Figure 1). The imaging incidentally revealed an oval axillary mass measuring $4.5 \times 2.5 \times 1$ cm, with slightly lobulated wall, isointense in a T1 sequence, slightly heterogeneously hyperintense with low-signal intensity rows in a T2 sequence, and round millimetric hypointense structures in the lower part. Of note, there was a marked contrast enhancement in a T1 gadolinium sequence, without invasion of adjacent structures. Physical examination confirmed a mobile, well-delimited, painless axillary adenopathy. Ultrasonography (Figure 2) confirmed an oval lesion with regular delimitation, with a heterogeneous aspect comprising a hypoechogenic focus in the caudal portion and small calcifications combined with hypervascularization in Doppler imaging. Mammography performed at the same time was without any pathological lesions. Histological analysis after surgical excision showed an encapsulated mass of $3.9 \times 2.5 \times 2.0$ cm, with white to brown areas and cystic appearance on macroscopic examination. The final histological diagnosis of an intranodal

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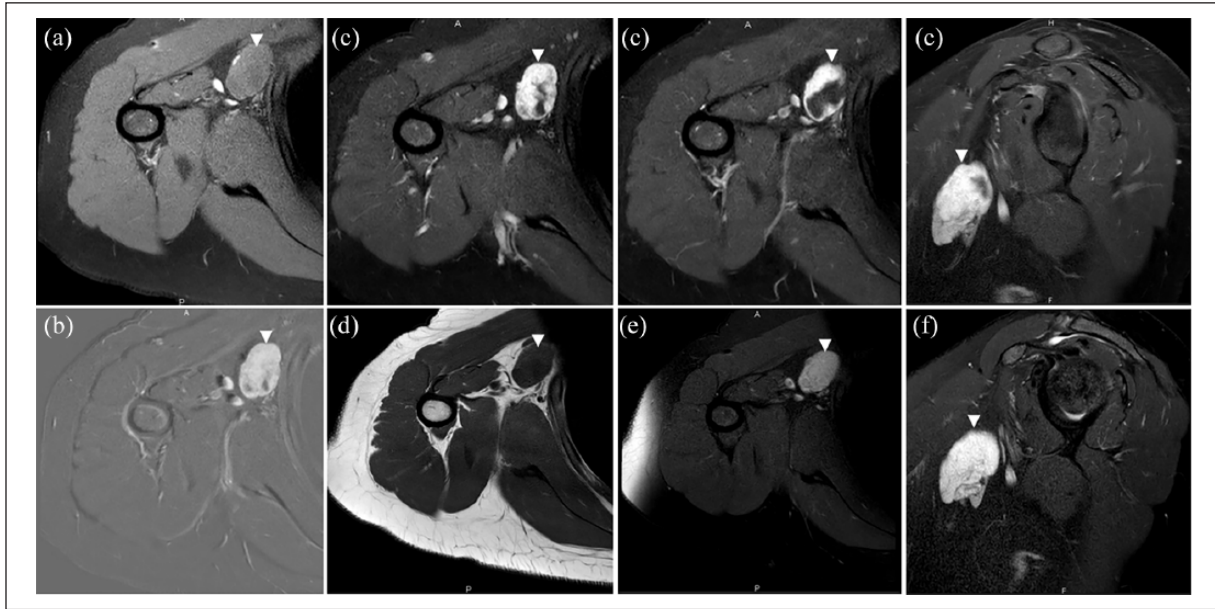


Figure 1. Shoulder MRI. (a) T1 FS axial, (b) subtraction T1 FS axial, (c) gadolinium T1 FS axial and sagittal, (d) T1 TSE axial, (e) T2 SPIR axial, and (f) sagittal. Oval axillary mass measuring $4.5 \times 2.5 \times 1$ cm, hypersignal in T2 sequence, and enhanced in T1-gadolinium sequence (white arrow).

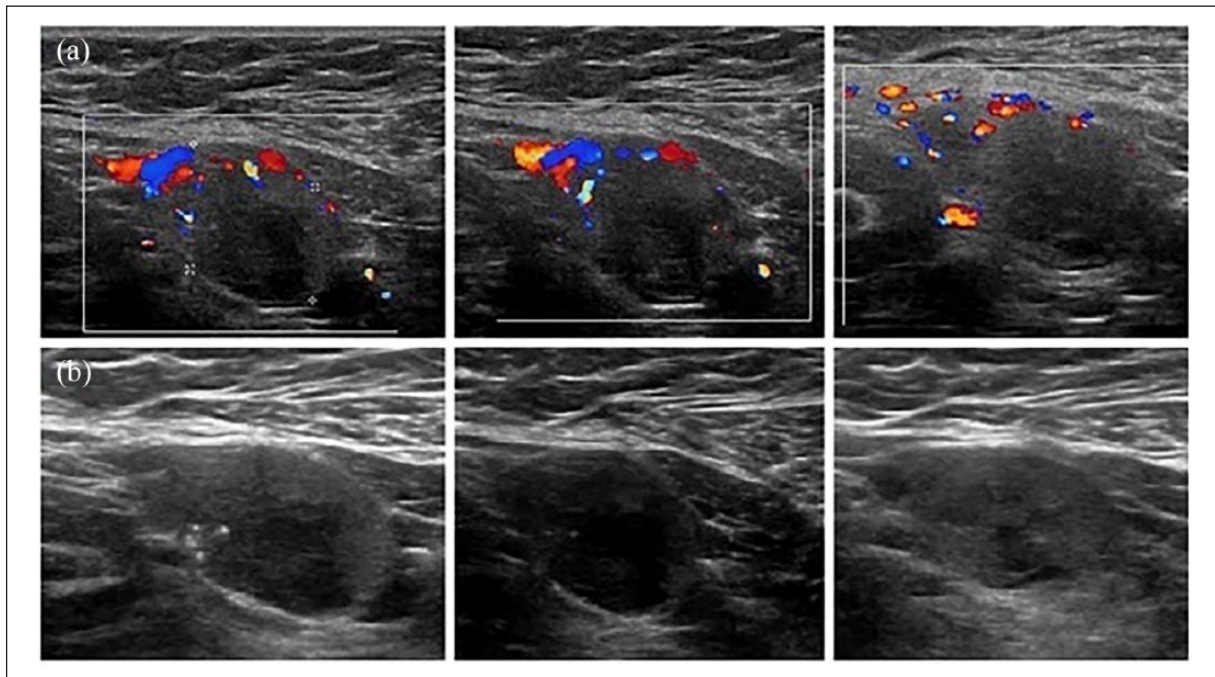


Figure 2. Axillar Doppler (pictures of (a) row) and sonography (pictures of (b) row). Heterogeneous with hypoechogenic focus, small calcifications, and hypervascularization.

hemangioma of CCH type was made following special stains and immunohistochemical confirmation (Figure 3).

Discussion

CCH is a rare entity, arising extremely rarely in lymph nodes. It is characterized by an abnormal morphologic development

of the embryonic vascular system, non-regressive. Many lesions are potentially present with bleeding complications in case of trauma, infection, or hormonal changes. Histologically, the lesion is characterized by a proliferation of smaller, more capillary-like as well as larger, cavernous blood vessels. Many classifications have been used, first that of Mulliken and Glowacki¹¹ in 1982, that of Hamburg in

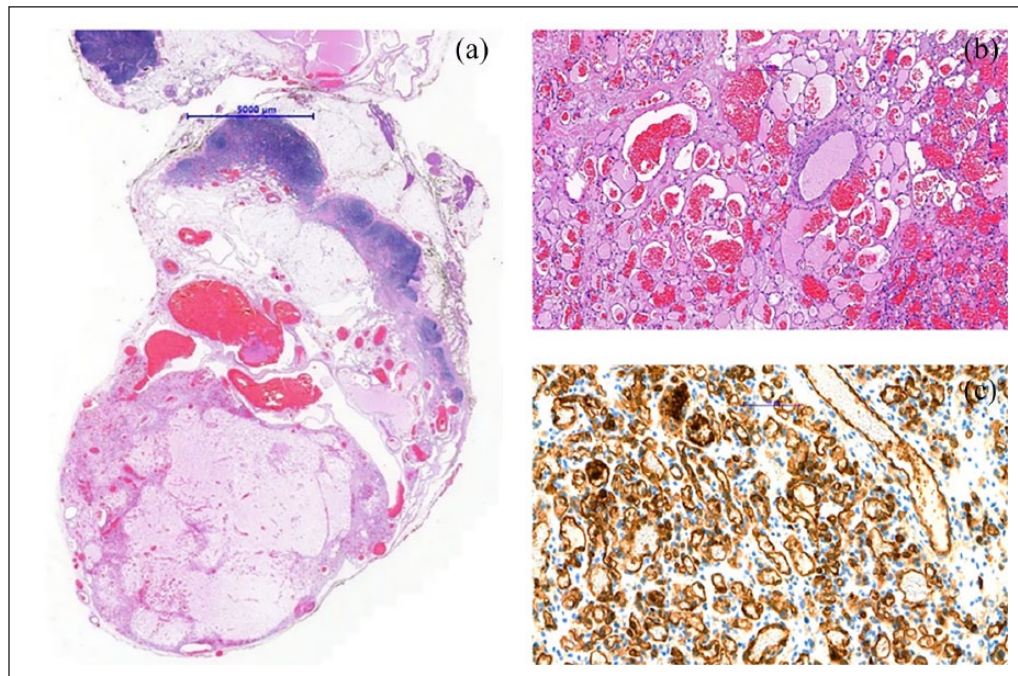


Figure 3. Histological analysis of the excised lymph node. Histological assessment of the lymph node in hematoxylin and eosin (H&E) staining, overview in panel (a). Little residual lymphoid tissue is present, the majority of the lymph node tissue is replaced by a vascular proliferation. Panel (b) shows in more detail in H&E the proliferation of here mainly capillary-like blood vessels lined by inconspicuous vascular endothelium (panel (c), CD31 immunohistochemistry, brown stain denoting positivity).

1988, reviewed by Chan et al.³ in 1992, and the last by ISSVA in 2014¹ proposing to rename capillary, cavernous, and mixed hemangioma as low-flow venous malformations. However, the question remains between whether these lesions should be classified as tumor-like/neoplastic or vascular malformations. Depending on the classification, the final diagnosis and classification relies on whether the lesion was present at birth, how it developed, its histological and hemodynamic characteristics, as well as its presentation using imaging techniques, especially MRI.¹² Differential diagnoses of an intranodal CCH are other types of tumor-like vascular lesions, benign ones including capillary, cavernous, mixed, lobular capillary hemangiomas, angiomyomatous hamartoma, vascular epithelioid tumor and lymphangioma, and malignant ones including hemangioendothelioma, Kaposi sarcoma and angiosarcoma.^{3,10} Vascular transformation of the sinus can usually be excluded histologically with ease. CCH can also be found in Maffucci syndrome (multiple enchondromatosis with soft tissue hemangiomas) and Klippel–Trenaunay syndrome (vascular malformation and soft tissue and bone hypertrophy). In our case, no sign of Klippel–Trenaunay or Maffucci syndrome was noted.

Improvement of imaging techniques in the last decades allows to distinguish capillary from cavernous type and hemangiomas from arteriovenous malformations with enhanced MRI,^{13,14} but mixed types can easily mimic malignancies and their recognition is of importance, for example, in our case, where sarcoma has been evoked. Classic MRI

findings of extra-nodal CCH are a lobulated septated lesion on T2 STIR sequence, isointense to muscle in T1-weighted sequence, with enhanced T1-weighted sequence making difference between venous and mixed malformations, showing early diffuse homogeneous or heterogeneous lesion enhancement depending on the time between injection and acquisition.^{2,14} Especially in the case of an axillary intranodal location in a woman, some special dynamic MRI features are recommended to make the difference between a metastasis and nonmetastatic lymph node,¹⁵ which is usually the first question asked in this situation. For superficial CCH, sonography can show calcifications and heterogeneity, and color Doppler hypervascularization.¹⁶ Those features were found in our case. No positron emission tomography/computed tomography (PET/CT) imaging of mixed hemangioma has been reported, whereas capillary type usually shows low F-18 fluorodeoxyglucose (FDG) uptake, and cavernous type usually shows high FDG uptake.^{17,18}

Among the ten cases of intranodal CCH reviewed, nine patients were women, and ages range from 11 to 75 years with a mean of 53 years. Only one was symptomatic, resulting in a small-bowel intussusception. One case was found in buccal mucosa, but eight were found on lymphadenectomy during oncologic surgery (mastectomies, hysterectomies, pulmonary lobectomy). Immunohistochemical staining reveals positivity of the endothelial cells for CD31, CD34, and factor VIII related antigen, with negativity for epithelial markers such as pancytokeratins and lymphatic markers such as D2-40.

Surgical excision of low-flow vascular anomalies is the rule in cases of functional impairment, aesthetically relevant changes, or psychological issues. In our case, excision was performed to exclude metastatic disease. Other therapies have been proposed to avoid surgical interventions, including Sirolimus, Pulse Dye Laser, and chemical or thermic sclerotherapy.^{19–21} Recognizing this type of complex vascular malformation is then essential to guide therapy, follow up, and avoid surgery. Nevertheless, there are currently no specific guidelines for treatment of CCH.

Conclusion

We reported a case of an intranodal CCH. Their presence in a node can be suspected of either primary or secondary lesion. When such lesion is suspected, clinician should use MRI with enhanced and T2 STIR sequences as the imaging of choice, either for intra- or extra-nodal CCH, which appear to have same MRI features according to our case. Evolution being a key element, MRI could also be used for the follow-up in case of a deep difficult-to-reach lesion, to detect growing and complication, knowing that no recurrence of CCH has been reported after treatment. Radiologist must then know this pathology to distinguish CCH from malignancy.

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Ethical approval

Our institution does not require ethical approval for reporting individual cases or case series.

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Informed consent

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References

1. Wassef M, Blei F, Adams DM, et al. Vascular anomalies classification: recommendations from the International Society for the Study of Vascular Anomalies. *Pediatrics* 2015; 136(1): e203–e214.
2. Ernemann U, Kramer U, Miller S, et al. Current concepts in the classification, diagnosis and treatment of vascular anomalies. *Eur J Radiol* 2010; 75(1): 2–11.
3. Chan JK, Frizzera G, Fletcher CD, et al. 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): 335–350.
4. Terada T. Capillary cavernous hemangioma of the lymph node. *Int J Clin Exp Pathol* 2013; 6(6): 1200–1201.
5. Dellacha A, Fulcheri E and Campisi C. A lymph nodal capillary cavernous hemangioma. *Lymphology* 1999; 32(3): 123–125.
6. Reich RF, Moss S and Freedman PD. Intranodal hemangioma of the oral soft tissues: a case report of a rare entity with review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2000; 90: 71–73.
7. Fabio P and Camillo D. Capillary cavernous hemangioma of lymph node. *Int J Surg Pathol* 2010; 18: 338.
8. Morgan DR, Mylankal K, El Barghouti N, et al. Small bowel haemangioma with local lymph node involvement presenting as intussusception. *J Clin Pathol* 2000; 53(7): 552–553.
9. Goto T, Akanabe K, Maeshima A, et al. Hemangioma in a pulmonary hilar lymph node: case report. *World J Surg Oncol* 2011; 9: 8.
10. Elgoweini M and Chetty R. Primary nodal hemangioma. *Arch Pathol Lab Med* 2012; 136(1): 110–112.
11. Mulliken JB and Glowacki J. Hemangiomas and vascular malformations in infants and children: a classification based on endothelial characteristics. *Plast Reconstr Surg* 1982; 69(3): 412–422.
12. Moukaddam H, Pollak J and Haims AH. MRI characteristics and classification of peripheral vascular malformations and tumors. *Skeletal Radiol* 2009; 38(6): 535–547.
13. Sklar EL, Quencer RM, Byrne SF, et al. Correlative study of the computed tomographic, ultrasonographic, and pathological characteristics of cavernous versus capillary hemangiomas of the orbit. *J Clin Neuroophthalmol* 1986; 6(1): 14–21.
14. Dobson MJ, Hartley RWJ, Ashleigh R, et al. MR angiography and MR imaging of symptomatic vascular malformations. *Clin Radiol* 1997; 52(8): 595–602.
15. Fusco R, Sansone M, Granata V, et al. Use of quantitative morphological and functional features for assessment of axillary lymph node in breast dynamic contrast-enhanced magnetic resonance imaging. *Biomed Res Int* 2018; 2018: 2610801.
16. Furuta T, Shimose S, Nakashima Y, et al. 2017 Novel sluggish speed signs on ultrasound is indicative of hemangiomas. *Acta Radiol*; 58(10): 1231–1237.
17. Hashimoto H, Kurata A, Fujiwara M, et al. Solitary pulmonary capillary hemangioma of adult cases: clinicopathologic characteristics as an unrecognized entity. *Am J Surg Pathol* 2016; 40(10): 1380–1389.
18. Li ZY, Li XC and Wu Q. PET-CT diagnosis of cardiac cavernous hemangioma with large pericardial effusion. *Eur Rev Med Pharmacol Sci* 2014; 18(21): 3256–3259.
19. Adams DM, Trenor CC 3rd, Hammill AM, et al. Efficacy and safety of sirolimus in the treatment of complicated vascular anomalies. *Pediatrics* 2016; 137(2): e20153257.
20. Wales L, Nasr H, Bohm N, et al. Paediatric venous malformation: treatment with endovenous laser and foam sclerotherapy. *EJVES Extra* 2007; 14: 6–7.
21. Blaise S, Charavin-Cocuzza M and Riom H. Treatment of low-flow vascular malformations by ultrasound-guided sclerotherapy with polidocanol foam: 24 cases and literature review. *Eur J Vasc Endovasc Surg* 2011; 41(3): 412–417.