CASE REPORT

Axillary intranodal palisaded myofibroblastoma: report of a case associated with chronic mastitis

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SUMMARY

Intranodal palisaded myofibroblastoma is a rare tumour of the lymph node that may be derived from myofibroblasts. The most usual area of presentation is the inguinal lymph nodes, but occurrence within other areas has also been reported. It is characterised by spindle cells, amianthoid-like fibres, and by the proliferation of hemosiderin-containing histiocytes in the lymph node. Although intranodal palisaded myofibroblastoma is benign, it is frequently confused with metastatic lesions, especially when it occurs in atypical sites. We herein report the second case of axillary intranodal palisaded myofibroblastoma occurring in a woman with a granulomatous chronic mastitis. The salient clinicopathological features of this unusual tumour are presented with emphasis to the pathogenesis of the tumour as well as to its histological and immunohistochemical characteristics. Clinicians and pathologists must be aware of this rare tumour to avoid a misdiagnosis of malignancy and assure patient a correct therapeutic management.

BACKGROUND

This is a rare tumour that may cause problems in the differential diagnosis and in the correct therapeutic management.

CASE PRESENTATION

A 25-year-old woman with an history of chronic mastitis treated with steroid therapy presented at our observation with a non-tender palpable left breast mass, predominantly in the subareolar region, without nodularity or cutaneous erythema. In addition, the patient referred a gradually increasing painless swelling in the left axilla present for about 4 months. Physical examination revealed a single, painless, firm nodule, and an ultrasonography confirmed hypoechoic rounded mass, measuring about 3×2.6 cm, and having solid components. Laboratory tests showed a previous Epstein-Barr virus (EBV) infection, demonstrated by viral capsid antigen (VCA) IgG and VCA IgM antibodies in the absence of Epstein-Barr nuclear antigen-1 IgG. Given the association between left breast mass and axillary nodule, a clinical diagnosis of breast cancer with lymph node metastasis was suspected. Accordingly, an incisional biopsy of the breast and a fine-needle aspiration cytology (FNAC) of the axillary nodule were performed. Histopathological examination of breast biopsy showed a non-caseating granulomatous inflammatory condition of breast lobules with microabscess formation diagnostic of granulomatous chronic

Figure 1 Fine-needle aspiration cytology showed small spindle cells with no atypia, suggesting a low-grade spindle cell tumour.

mastitis. Axillary nodule FNAC showed a predominant population of single, small spindle cells with no atypia, suggesting a low-grade spindle cell tumour (figure 1). The nodule was excised and the gross specimen received was a well-encapsulated lymph node with a nodular tumour mass of size 3×3 cm, firm in consistency with greyish white cut section. The low power microscopic examination revealed a thin peripheral rim of lymphoid tissue underneath the fibrocollagenous capsule (figure 2). The tumour mass was composed of interlacing fascicles of uniform spindle cells (figure 3). The cells were also arranged in palisading fashion with eosinophilic 'amianthoid' fibres characterised by an eosinophilic center surrounded by a paler eosinophilic, stellate-shaped periphery (figure 4). Extensive areas of interstitial haemorrhages were

Figure 2 Well-encapsulated lymph node with a nodular tumour mass and a thin peripheral rim of lymphoid tissue underneath the fibrocollagenous capsule.

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Figure 3 The tumour mass was composed of interlacing fascicles of uniform spindle cells.

also seen. Mitotic activity, including atypical mitoses were absent. Masson's trichrome revealed peripherally bristled 'amiantoid' fibres. The spindle cells were immunoreactive for vimentin, α -smooth muscle actin (figure 5), but not for desmin, h-caldesmon, S-100 protein, CD34, CD31, CD117, CD99, CK AE1/AE3, activin receptor-like kinase-1, glial fibrillary acidic protein and human herpes virus-8 (HHV-8). Notably, tumour cells showed strong nuclear expression of Cyclin D1 (40% of spindle cells). The Ki67 index was less than 1%. Immunohistochemical study of amiantoid fibres revealed that the center of these fibres was composed of collagen type I. On the basis of these morphological and immunohistochemical results, a final diagnosis of axillary intranodal palisaded myofibroblastoma was rendered.

OUTCOME AND FOLLOW-UP

Two years after the lymph node excision the patient is alive without disease.

DISCUSSION

Although schwannoma is the prototypical spindle cell tumour that characteristically shows nuclear palisading, other spindle cell tumours, including uterine or soft tissue leiomyomas,

Figure 4 The cells were arranged in palisading fashion with eosinophilic 'amiantoid-like' fibres.

Figure 5 The spindle cells were diffusely immunoreactive for α -smooth muscle actin.

angioleiomyomas and gastrointestinal stromal tumours may occasionally show a prominent nuclear palisading.¹ Among fibroblastic/myofibroblastic tumours, intranodal myofibroblastoma usually exhibits neoplastic cells which adopt a striking nuclear palisading. Interestingly, this morphological feature has also been recently reported in mammary myofibroblastoma.² Intranodal palisaded myofibroblastoma (IPM) is a rare neoplasm arising from the stromal component of the lymph node.³⁻¹¹ Almost all cases develop in lymph nodes of the inguinal region.³⁻⁹ To our knowledge, approximately 70 cases of IPMs have been reported in the literature, but only one has been reported in axillary lymph nodes.¹¹ Clinically, IPM presents as an asymptomatic, slowly growing tumour. It is more common in males and it is diagnosed at a wide age range. Its size varies from 0.6 to 5 cm, and predominant morphological features include the bland appearance of its constituent spindle cell population and the presence of acellular eosinophilic stellate areas (so-called amiantoid-like fibres).⁴⁻⁶ The immunohistochemical profile of the neoplastic cells along with their ultrastructural features is indicative of myofibroblastic or smooth muscle differentiation. IPM has recently been shown to have a strong expression for cyclin D1 and a low proliferating index of Ki-67.⁷ The overexpression of cyclin D1 presented in the study of Kleist *et al.*⁹ points to the proliferation regulatory pathway as one of the factors involved in the aetiological pathogenesis of IPM. Genetic evidence associated with HHV-8 and EBV suggests also that a viral mechanism of oncogenesis might be involved in the development of these lesions.⁶⁻¹⁰ In our case, there was no evidence of HHV-8 infections but the patient presented in the her clinical history a previous infection from EBV, supporting the hypothesis of a viral or immune-mediated mechanism in the pathogenesis of IPM, probably through the overexpression of cyclin D1 and overgrowth of intranodal differentiated smooth muscle or myofibroblastic cells. IPM pathological differential diagnosis includes a wide spectrum of lesions.⁵⁻⁷ The most important are represented by schwannoma and Kaposi sarcoma.¹ Moreover, haemangioendothelioma, intranodal leiomioma, leiomyosarcoma, dendritic reticulum cell tumour, inflammatory myofibroblastic tumour, primary and metastatic malignant spindle-cell tumours should also be considered in the differential diagnosis.¹ The clinical history, morphological and immunohistochemical features help in the correct diagnosis of IPM. When it arises at unusual sites, as our case, and is associated with clinical situations highly suspicious for malignancy, it may represent a potential

diagnostic pitfall for both clinicians and pathologists because of its ability to mimic metastatic lesions in the lymph node. IPMs are, in fact, benign tumours and a local excision is curative in all cases, as demonstrated by the fact that our patient is well 2 years after the surgery. Accordingly, both clinicians and pathologists must be aware of these rare lesions to avoid a misdiagnosis and assure patient a correct treatment, management and prognostic information.

Learning points

- Intranodal palisaded myofibroblastoma is a rare and benign tumour of the lymph node that may be derived from myofibroblasts.
- Genetic evidence associated with human herpes virus-8 and Epstein-Barr virus suggests that a viral mechanism of oncogenesis might be involved in the development of these lesions.
- Intranodal palisaded myofibroblastoma may represent a potential diagnostic pitfall for both clinicians and pathologists because of its ability to mimic metastatic lesions.
- The simple lymph node excision is curative.

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Competing interests None.

Patient consent Obtained.

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