CellPress

Contents lists available at ScienceDirect

Heliyon

journal homepage: www.cell.com/heliyon



Case report

A rare case of squamous inclusion cyst in cervical lymph node



Y. Houcine a,b,c,*, A. Sassi a,c, M. Mlika a,b,c, H. Yassine a,b, F. Mezni a,b,c

- ^a Pathology Department, Abderrahmen Mami Hospital, Ariana, Tunisia
- ^b LR18SP06, Tunisia
- c El Manar University, Tunisia

ARTICLE INFO

Keywords: Oncology Pathology Health sciences Intranodal squamous inclusion cyst Ectopic tissues Lymph node Differential diagnosis Tuberculosis

ABSTRACT

A 38-year-old man, with no history of malignancy, was found to have a 2 cm jugular lymph node, for which a lymph node tuberculosis was suspected. The specimen revealed a cystic structure lined by mature keratinizing squamous epithelium with a prominent granular cell layer consistent with a squamous inclusion cyst in a lymph node, but a metastatic squamous cell carcinoma could not be excluded. This is the first case report of a rapidly enlarging squamous inclusion cyst in a jugular lymph node. Our case demonstrates the diagnostic challenges related to a squamous inclusion cyst in cervical lymph node and serves to inform the readers to consider this lesion in the differential diagnosis for similar situations.

1. Introduction

Benign glandular structures in or associated with lymph nodes is a well-documented phenomenon in axillary region. In fact, recently, there has been a sizable number of articles dealing with benign glandular inclusions in axillary nodes, focusing on their potential for being misdiagnosed as metastatic carcinoma [1, 2, 3, 4, 5, 6]. Therefore, the awareness of such an entity is all the more important in preventing overdiagnosis of a malignant lesion.

Two others localizations of such lesions have been reported: intramammary node [7] and peripancreatic node [8].

Conversely, the presence of benign squamous inclusions in lymph nodes is a very uncommon event, with only a few cases having been reported to date in axillary and submandibular region [9, 10].

We report here the morphologic features of histologically benign squamous inclusions in jugular nodes and discuss the differential diagnosis.

2. Case report

The patient was 38-year-old who presented with an enlarging jugulary mass. There was no past or family history of malignancy. At physical exam, a single, nontender, right jugular lymph node was palpable. The computerized tomography scan was normal. This scan indicated the absence of lymphadenopathy, organomegaly or solid tumor mass.

Fine needle aspiration was performed and part of the sample was sent to the microbiology laboratory for analysis. Cytologic examination revealed an acellular eosinophilic materialevoking a caseous necrosis. The laboratory analysis of the biopsy confirmed the absence of Koch Bacillus. Cytologic examination revealed an acellular eosinophilic material evoking a caseous necrosis. Behind these clinical, radiological and cytological results, and as we are endemic for tuberculosis clinical diagnosis of tubercular lymphadenopathy was considered first.

The lymph node which consisted of a single, capsulated node measuring $2.5~{\rm cm}\times 2.0~{\rm cm}\times 1~{\rm cm}$ was excised. The cut surface showed a cyst filled with whitish creamy caseum-like liquid in appearance. Routine H&E sections revealed a lymph node with normal nodal architecture (Figure 1). The single involved node was almost completely replaced by a cystic structure lined by mature keratinizing squamous epithelium with a prominent granular cell layer (Figure 2, Figure 3). This epithelium was limited to a thin layer of squamous cells or replaced by a stromal granulomatous reaction to the keratin (Figure 1).

The larger cystic inclusion, corresponding to the focus seen grossly, contained keratin debris.

As the computed tomography scan, ear nose and throat examinations were normal and the node histology identified normal nodal architecture but with mature keratinizing squamous epithelium, prominent granular cells and keratin debris, the diagnosis of squamous inclusion cyst in cervical lymph node was retained.

E-mail address: dryoldezhoucine@gmail.com (Y. Houcine).

^{*} Corresponding author.

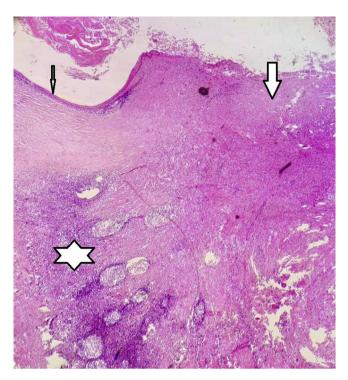


Figure 1. Benign squamous epithelium-lined cyst (Thin arrow), in a lymph node tissue (star), associated to a stromal granulomatous reaction to the keratin (thick arrow) (H&E).

2.1. Consent

A patient's consent to publication of information about him was obtained.

3. Comments

Benign squamous inclusions are foci of non-neoplastic ectopic tissue in lymph nodes. It is important to identify them for the differential diagnosis with lymph node metastases or Tuberculosis as in our case.

Cases reported in the literature describe predominantly 3 types of benign epithelial inclusions: glandular type, squamous type, and mixed glandular squamous type [7].

The pathogenesis of this rare phenomenon is not yet well established. In the literature, 2 main hypotheses are discussed: those of nodal implants from "transported" benign epithelium from the other structures via lymphatic vessels to lymph node and of epithelial rests resulting from an embryologic malformation; i.e. an embryologic error may have resulted in the admixture of epithelium and lymphoid tissue [7].

In adults, the most common cystic-appearing neck lesions occuring in a lymph node is metastases from squamous cell carcinoma, with a higher incidence in metastases associated with carcinomas of Waldeyer's ring [11, 12, 13]. These cystic metastases occured most frequently in jugular lymph node. Goldenberg et al confirmed a relationship among cystic lymph node metastasis, infection with HPV, and tonsil/tongue base squamous cell carcinoma. They suggested that in adults with a cystic neck node, a carcinoma of the tonsil or tongue base should be suspected [13]. In our case, the histologic findings were consistent with a squamous inclusion cyst in a lymph node, but a metastatic squamous cell carcinoma could not be ruled out.

Detection of metastases is regarded as one of the most important parameters to plan further surgical and/or medical therapy. This fact emphasizes the importance of a careful search for their presence as well as the avoidance of overdiagnosis.

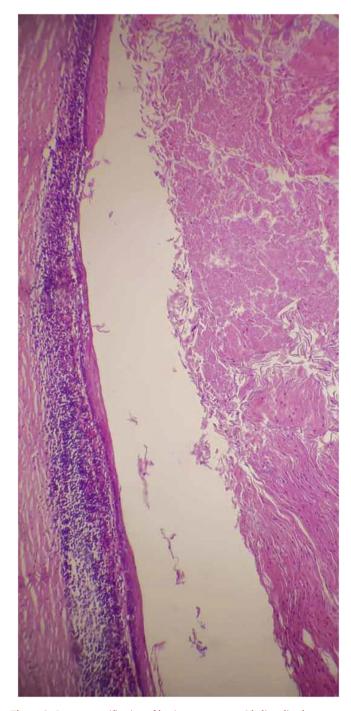


Figure 2. Lower magnification of benign squamous epithelium-lined cyst containing keratin debris (H&Ex 10).

Basically, a diagnosis of benignancy should be favored in the presence of a few squamous structures without cytologic atypia within the lymph node capsule, and/or cystically enlarged squamous structures in the nodal parenchyma without an associated desmoplastic stromal reaction.

Tuberculosis may present a major differential diagnosis of squamous cystic inclusion in cervical lymph node, especially in endemic country as our. In fact, the so-called cold abscesses in tuberculous lymph nodes occur most frequently in cervical lymph node [14]. They show, on echography, an absence of reaction in the surrounding tissue and therefore cannot be differentiated from noninfectious lymph nodes.

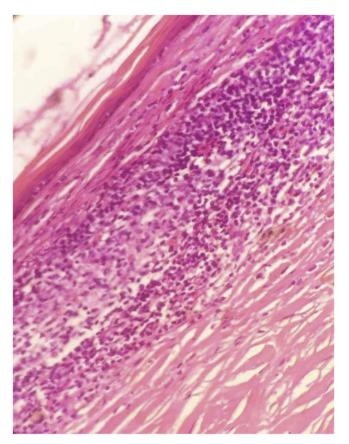


Figure 3. Higher magnification of benign cystic structure lined by mature keratinizing squamous epithelium with a prominent granular cell layer and surrouded by lymph node tissue (H&Ex 40).

Besides, on macroscopic examination, the cut surface of tuberculous lymph node can show necrotic yellow material (caseum) which mimics the keratine contents of squamous cyst inclusion. In this context, our patient had the lymph node excision with a suspected diagnosis of Tuberculosis.

Other differential diagnoses which can be considered, are non-nodal cystic neck lesions, with an emphasis on branchial cleft cysts in children and young adults. Venous/lymphatic malformations are congenital lesions most often recognized at birth or in very young children.

A complication to the diagnosis was the rupture of the squamous inclusion cyst within a lymphnode with extensive reactive changes. Aggregates of histiocytes as a reaction to cyst rupture, can have atypical appearance, which suggest malignancy which is a well-known pitfall in cytologic preparations.

Our case emphasizes that occasional non neoplastic nodal inclusions do occur. Epithelial inclusions may co-exist with micrometastases and thus careful attention is required to distinguish between benign inclusions in lymph nodes and metastatic malignancy by histological examination of the nodal tissue.

Declarations

Author contribution statement

All authors listed have significantly contributed to the investigation, development and writing of this article.

Funding statement

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Competing interest statement

The authors declare no conflict of interest.

Additional information

No additional information is available for this paper.

References

- [1] M. Bianchi, L. Tono, Contribution to the study of epithelial inclusions of the lymph nodes. Anatomo-clinical considerations on a case of glandular inclusions of the axillary lymph nodes, Arch. De Vecchi Anat; Patol; 30 (1959) 307–319 [Article in Italian].
- [2] C. Bramlett, R. Laucirica, D.L. Page, et al., Differentiation between metastatic low grade carcinoma of the breast and benign epithelial inclusion in axillary lymph nodes (abstract), Mod. Pathol. 8 (1995) 15A.
- [3] Y.B. Chen, J. Magpayo, P.P. Rosen, Sclerosing adenosis in sentinel axillary lymph nodes from a patient with invasive ductal carcinoma: an unusual variant of benign glandular inclusions, Arch. Pathol. Lab Med. 132 (2008) 1439–1441.
- [4] C. Chuang, D.G. Hicks, M. Berenson, et al., Benign inclusion of axillary lymph nodes: report of two cases and literature review, Breast J. 15 (2009) 664–665.
- [5] A.D. Corben, T. Nehhozina, K. Garg, et al., Endosalpingiosis in axillary lymph nodes: a possible pitfall in the staging of patients with breast carcinoma, Am. J. Surg. Pathol. 34 (2010) 1211–1216.
- [6] P. Colombo, L. Cattaneo, Axillary intranodal cysts associated with breast malignancy, Arch. Pathol. Lab Med. 128 (2004) 361–362.
- [7] F. Giovanni, C. Maria Luisa, R. Juan, Benign epithelial inclusions in axillary lymph nodes: report of 18 cases and review of the literature, Am. J. Surg. Pathol. 35 (8) (2011) 1123–1133.
- [8] S. Shozo, I. Hiroshi, H. Hitoshi, T. Akira, et al., Cystic lymphoepithelial lesions of the pancreas and peripancreatic region: report of two cases, Jpn. J. Surg. 29 (1999) 467-471
- [9] H. Saroona, F. Naveen, F. Saira, Squamous inclusion cyst in a sentinel axillary lymph node associated with breast malignancy, J. Coll. Phys. Surg .Pak. 22 (1) (2012) 50–52.
- [10] S. Bahadur, M. Pujani, S. Jetley, Epithelial inclusion cyst in a cervical lymph node: report of a rare entity at an uncommon location, Ann. Med. Health Sci. Res. 6 (2) (2016) 137–138.
- [11] D. Goldenberg, J. Sciubba, W.M. Koch, Cystic metastasis from head and neck squamous cell cancer: a distinct disease variant? Head Neck 28 (2006) 633–638.
- [12] C.G. Gourin, J.T. Johnson, Incidence of unsuspected metastases in lateral cervical cysts, Laryngoscope 110 (2000) 1637–1641.
- [13] D. Goldenberg, S. Begum, W.H. Westra, et al., Cystic lymph node metastasis in patients with head and neck cancer: an HPVassociated phenomenon, Head Neck 30 (2008) 898–903.
- [14] J. Fruehwald-Pallamar, Q.C. Li, F. Hasteh, S. Hauff, et al., Inclusion cyst in a cervical lymph node, Neurographics 2 (2012) 163–166.