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A unique parotid adenocarcinoma

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Abstract
A large number of different types of benign and malignant tumours of the salivary glands are recognized, and rare and unusual variants arise from time to time that can be difficult to interpret. We report an apparently unique parotid neoplasm that does not fit any of the currently recognized diagnostic groups and can best be termed lymphadenocarcinoma. Clinical and pathological features are described, and its possible relationship to recognized tumour types is examined.

Key words: Parotid gland; Neoplasms; Adenocarcinoma

Case report

A 38-year-old man presented with a mass in the left parotid gland of one month's duration. The mass was painless, about 1.5 cm across, and mobile. There were no signs of facial nerve deficit, and no other abnormalities were evident. The only significant past medical history was of a cholecystectomy for gallstones. A fine needle aspirate of the parotid mass was equivocal for malignancy. The aspirate contained two populations of cells, one epithelial and the other lymphocytic. In part, densely eosinophilic epithelial cells were syncytial, while in other areas pale stained cells with prominent nucleoli were loosely cohesive.

Within two weeks of presentation, a total left parotidectomy with sparing of the facial nerve was performed. The patient made an uneventful recovery, and in the subsequent two years he has remained well, without local recurrence or sign of metastatic spread. He continues to have follow-up examinations every six months. The local pathologist recognized the tumour as an unusual type, and sent slides from the specimen to the Armed Forces Institute of Pathology, Washington, DC, for consultation.

Grossly, the tumour was 1.5 cm in diameter and 2 cm from the inferior margin of the superficial lobe specimen. Sections prepared from the tumour were stained with haematoxylin and eosin (H & E), periodic acid-Schiff with; and without; diastase digestion, and mucicarmine. The tumour was a well-circumscribed but lobulated mass within the parotid gland. About two-thirds of the periphery of the tumour had a fibrous connective tissue capsule that separated it from the adjacent parotid parenchyma (Figure 1). This fibrous capsule varied from a few microns to half a millimetre in width. The fibrous capsule probably represented the fibrous septa that normally separates lobules of the parotid gland

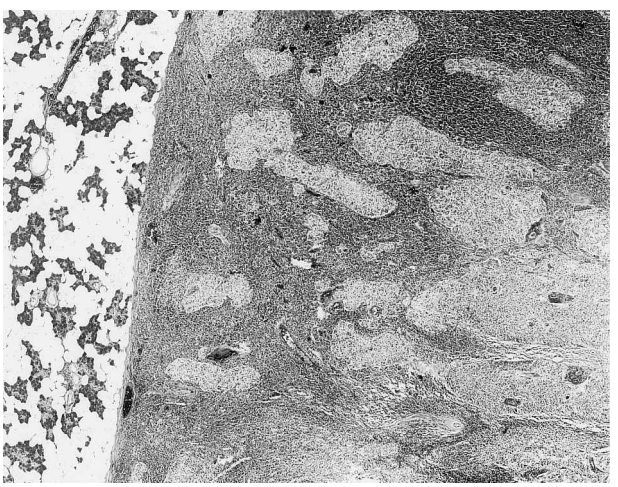

Fig. 1
Irregular-shaped nests of pale-staining neoplastic epithelium stand out in the dense stroma of dark-staining lymphocytes. In this portion of the tumour, it is well demarcated from adjacent normal parotid gland parenchyma at left (H&E; ×30).

Fig. 2
Pale-stained neoplastic epithelium with dark-stained lymphoid stroma infiltrates the fat and glandular tissue of an adjacent lobule of parotid gland at upper right (H&E; ×30).

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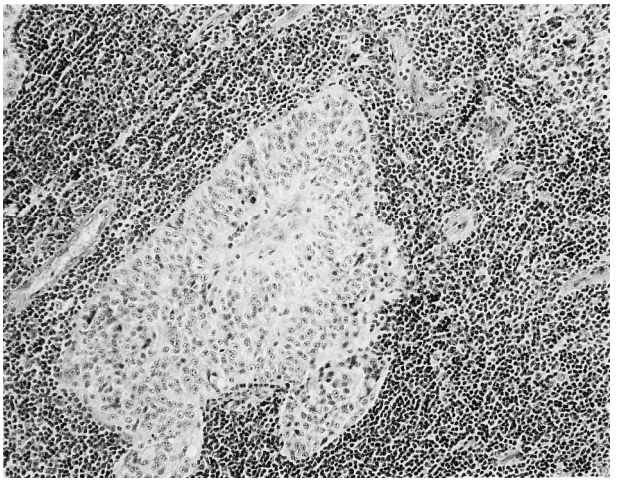

FIG. 3

Island of neoplastic epithelium in lymphoid tissue. The epithelial cells have poorly defined cell boundaries, pale stained cytoplasm, and fairly uniform round nuclei with small nucleoli. The epithelial does not form a glandular architecture (H&E; ×30).

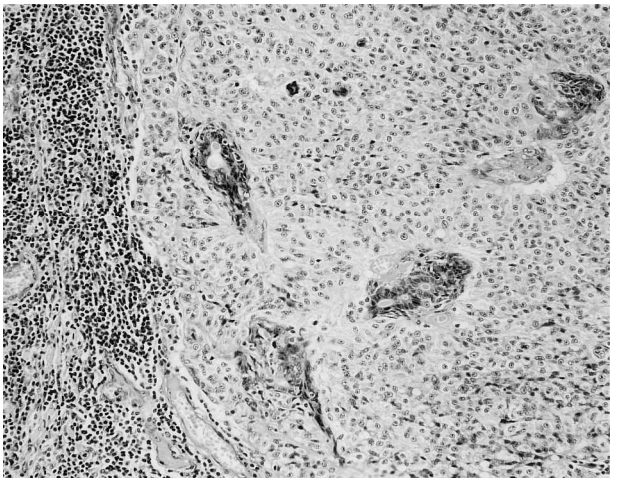

FIG. 4

Glandular lumens surrounded by cuboidal cells are present in a few foci of the tumour. The glandular structures are rudimentary (H&E; ×150).

because foci of normal parotid glandular tissue were within the capsule and infiltrated by the tumour (Figure 2). The tumour was composed of variously sized and irregularly shaped nests and sheets of fairly uniform epithelial cells within a very dense population of small lymphoid cells. The dense lymphoid tissue occupied the peripheral one-half to two-thirds of the tumour while the stroma of the central portion of the tumour was of a dense collagenous tissue. There were rare germinal centres in the lymphoid tissue, but architectural features of a lymph node, such as subcapsular sinus and medullary cords, were absent. The lymphoid tissue accompanied nest of tumour epithelium where it infiltrated the adjacent normal parotid tissue. The neoplastic epithelial cells had pale to moderately intense eosinophilic cytoplasm with poorly delineated cell boundaries (Figure 3). Their nuclei were mildly pleomorphic, had vesicular to stippled chromatin, and one to two small, basophilic nucleoli. In several scattered foci, cuboidal cells surrounded glandular lumens and were themselves surrounded by the more numerous myoepithelial-appearing cells (Figure 4). In one lobule of neoplastic epithelium there were two foci of three or four mucous cells with abundant basophilic cytoplasm, that stained with mucicarmine, but no subaceous cell differentiation was present. Mitotic figures were absent. There were also eosinophils admixed amongst the lymphoid cells and in foci within the neoplastic epithelium.

Discussion

This tumour does not fit well into the current classification scheme for parotid tumours.[1] Its most striking features were nodules of epithelial cells scattered in a dense lymphoid stroma, the mixed nature of the epithelial cell component, the central sclerosis, the eosinophilic infiltrate of the lymphoid component, and the infiltration of adjacent normal parotid tissue.

Possibilities in the differential diagnosis of this tumour can be identified by reference to the lymphoid component. The parotid contains about five to 20 lymph nodes per gland, and in addition there are aggregates of lymphoid tissue that do not have a fully developed nodal architecture.[2] Lymphoid tissue within the parotid can be nodal or extra-nodal normal tissue, an inflammatory infiltrate, a component of a parotid tumour, or a lymphoproliferative condition in its own right. Reactive lymphoid infiltrations include chronic sialadenitis, sarcoid, cystic lymphoid hyperplasia associated with human immunodeficiency virus (HIV) infection, and Sjögren's syndrome.[3,4] Salivary gland tumours in which a prominent lymphoid stroma is an integral diagnostic component include Warthin's tumour, sebaceous lymphadenoma and lymphadenocarcinoma, and lymphoepithelial carcinoma. In addition, tumours that are not typically associated with a lymphoid infiltrate may, in some cases, elicit a lymphoid response that has been described as tumour-associated lymphoid proliferation (TALP).[5] Metastatic carcinoma can involve parotid lymph nodes.[5] Finally lymphoproliferative conditions of the parotid include low-grade B-cell lymphomas derived from mucosa-associated lymphoid tissue (MALT), which arise at a higher frequency in patients with a history of Sjögren's syndrome.[1] The increased risk has been calculated to be 44-fold higher than controls.[6]

Sebaceous lymphadenoma is the salivary gland tumour that most closely resembles the tumour we have described.[1,7] This rare, benign tumour is characteristically well-encapsulated, or at least well-demarcated. It has a dense lymphoid stroma in which are embedded scattered islands of duct-like structures, which have a squamous or cuboidal lining with foci of sebaceous cells.

Interestingly, one of us (GLE) has on occasion examined tumours nearly identifical to sebaceous lymphadenoma but without evidence of any sebaceous component.[1] The epithelial cells are bland, as in our patient's tumour, but do not have infiltrative behaviour, as our specimen does. Extremely rarely, there is transformation of sebaceous lymphadenoma into sebaceous lymphadenocarcinoma, a tumour type for which only three cases have been reported.[8,9] In these examples the malignant cells are free of lymphoid stroma, have numerous mitotic figures, and exhibit sebaceous features with ductal differentiation. All of these features are lacking in our case.

Lymphoepithelial carcinoma is a undifferented salivary carcinoma associated with a dense lymphoid stroma and has the distinction of having an racial predilection for persons of Mongolian ancestry.[10] The undifferentiated epithelial component has varied cell shapes, abundant cytoplasm, indistinct cell boundaries, and frequent mitoses. The lymphoid component is dense and often has germinal

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centres. While the tumour is infiltrative, such as the specimen we are reporting, the epithelial component is clearly different.

Metastases to the parotid gland can come from primary tumours in the head and neck and more distant sites.[11,12] Those from the head and neck are most numerous and are predominantly cutaneous squamous cell carcinomas and melanomas. Those from distant sites represent about 20 per cent or parotid secondary tumours, and are most frequently from the lung, kidney or breast. Metastatic tumours tend to have haphazardly distributed epithelium and exhibit characteristics of the primary tumour, as well as possessing mitoses. Lymphoid tissue is an inconstant feature, and can be either nodal or reactive in origin. Our specimen contains epithelial cells that were not identifiable as deriving from another tumour, was not mitotically active, and the epithelial component was fairly evenly spaced as discrete islands in the lymphoid component. Furthermore the patient has remained well in the subsequent follow-up period, with no evidence of any other malignancy.

In conclusion, we describe an unusual salivary neoplasm, that, after discussion amongst colleagues at the Armed Forces Institute of Pathology, we have called a lymphadenocarcinoma. While there is certainly insufficient data to predict biological behaviour, we believe there may be some potential for local spread and metastasis because of the focal infiltrative growth. However, there were no mitoses noted, and microscopically we would consider it low-grade. The patient will remain under long-term observation.

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Mr D. Hill takes responsibility for the integrity of the content of the paper.
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