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CASE REPORT

Case Report: Case report: a rare salivary gland tumor [version 1; peer review: 2 approved with reservations]

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Abstract

Salivary duct carcinoma is a distinctive primary neoplasm of the major salivary gland characterized by aggressive behavior with early metastasis, local recurrence and significant mortality. We report a 40 year old male with parotid swelling diagnosed as pleomorphic adenoma, who underwent parotidectomy with modified radical neck dissection and later, on routine histopathology, the swelling was reported as a salivary duct carcinoma, confirmed via immunohistochemistry. Given the relative low occurrence and known difficulty in making an accurate diagnosis using fine needle aspiration cytology, the possibility of salivary duct carcinoma in the appropriate clinical setting of elderly patients with parotid mass and facial palsy should be seriously considered.

Open Peer Review

Approval Status **??**

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1. **Pavel Dulguerov**, Geneva University Hospital, Geneva, Switzerland

2. **Giulio Cantu**, Istituto Nazionale Tumori, Milan, Italy

Any reports and responses or comments on the article can be found at the end of the article.

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Editorial note:

Please note that the refereeing status of this article was changed from "indexed" to "[v1; ref status: approved with reservations 2]".

When this article was first published, *F1000Research* was still in its beta phase; during this period articles that received any two of "Approved" or "Approved with Reservations" statuses from the reviewers were labelled as "indexed". When the journal was formally launched in January 2013, the requirements for indexing were tightened, and only articles that are given either two "Approved" or one "Approved" plus two "Approved with Reservations" statuses by the reviewers are labelled "indexed". The new criteria for "indexing" can still be met in the future if a new revised version receives the necessary approval status from the reviewers.

Introduction

Salivary duct carcinoma is a distinctive primary neoplasm of the major salivary gland first described by Kleinsasser *et al* in 1968¹. The term was selected because of its resemblance to ductal carcinoma of the breast. It is characterized by aggressive behavior with early metastasis, local recurrence and significant mortality. Nearly 85% of cases occur in the parotid gland followed by submandibular gland². The tumor has predilection for older men in the 6th to 7th decades of life³. A number of patients experience facial nerve palsy or paralysis and/or pain, and have cervical lymphadenopathy on presentation¹. Familiarity with this entity is necessary to avoid false interpretation.

Case report

A 40 year old Hindu male who had a 15 year history of smoking presented with a gradually increasing painless swelling on the left parotid region. On examination an 8 × 6 cm swelling was observed the A single Level II mobile lymph node of size < 1 cm was palpable. There was no facial palsy.

An ultrasonograph of the parotid region performed previously revealed a well defined hypoechoic mass (4.7 × 3.9 cm) with lobulation occupying the left temporomandibular joint with adjacent hypoechoic areas of varying sizes: 1.9 × 1.6 cm, 1.4 × 1.5 cm, and 1.1 × 1.2 cm. It was interpreted as a parotid mass. Fine needle aspiration cytology (FNAC) from the left parotid gland was done and reported as a pleomorphic adenoma. However, FNAC from the submandibular lymph node comprised of blood only.

A repeat FNAC at our institute (Figure 1) showed moderately cellular aspirates with few clusters of normal salivary gland tissue along with epithelial cells showing overcrowding, with a mild-to-moderate pleomorphic population of medium sized cells with the vesicular nuclei having evenly distributed chromatin without conspicuous nucleoli. Cytoplasm was eosinophilic with ill defined borders. It was interpreted as an epithelial neoplasm.

A CT scan of the face and neck showed that the left parotid gland had enlarged in size (9 × 6 × 4 cm), involving deep and superficial lobes, with replacement of normal glandular architecture by homogenous soft tissue. The adjacent musculo-fascial planes were preserved. Multiple enlarged discrete lymph nodes in the left

parotid were noted. The left internal jugular vein was compressed and no intraluminal thrombosis was seen.

In order to reach a diagnosis, a frozen Level II lymph node was performed. (Figure 2) On frozen section, the lymph node architecture was not seen. Cells were singly scattered having scanty cytoplasm, enlarged hyperchromatic nuclei and condensed chromatin. It was not possible to identify the type of malignancy and was therefore reported as a high grade malignant neoplasm.

On routine histopathology, neck nodes were resected. Six out of seven lymph nodes showed a metastatic neoplasm comprising of sheets and lobules of pleomorphic cells with coarse clumped chromatin separated by fibrous septa. Mitotic activity increased, rosette formation was noted, and it was reported as a poorly differentiated carcinoma with basaloid phenotype. The presence of a high mitotic rate and of focal large, polypoid nuclei suggested an origin from the sebaceous gland.

Figure 1. A hematoxylin and eosin stain of fine needle aspiration cytology of the parotid gland (40x).

Figure 2. A hematoxylin and eosin stain of a frozen level III lymph node (40x).

Finally, a total parotidectomy with modified neck dissection was performed. On gross examination the specimen comprised of:

- Single gray soft tissue piece with skin tag (15 × 14 × 4 cm).
- Skin (4.5 × 2 cm).
- Salivary glands (4 × 2 × 2 cm) were grossly unremarkable.
- Multiple lymph nodes at level II & III – (1.5 to 5.5 cm).
- A deep lobe parotid gland.
- Several gray soft tissue pieces (7 × 7 × 3 cm) with a level I lymph node.
- A single gray soft tissue piece (4 × 4 × 1 cm) (Figure 3).

Microscopic analysis (Figure 4, Figure 5) showed that the tumor comprised of slightly pleomorphic ovoid cells with vesicular nuclei

Figure 3. A gross photo of the parotidectomy.

Figure 4. A hematoxylin and eosin stain of the parotid gland tumor (10x).

Figure 5. A hematoxylin and eosin stain of the parotid gland tumor (40x).

arranged in sheets and a trabecular pattern separated by fibrous septa. Mitotic activity was not increased. Focal area showed an acinar and comedo pattern. Perineural and lymphovascular invasion were seen. Infiltration into the salivary gland tissue was noted. Eight out of ten lymph nodes showed metastatic carcinoma. In view of metastasis to a lymph node, a diagnosis of high grade malignant epithelial neoplasm was suggested, which was later confirmed via immunohistochemistry as salivary duct carcinoma.

Discussion

Salivary duct carcinoma (SDC) is an aggressive adenocarcinoma which resembles high-grade breast ductal carcinoma. It is also known as cribriform salivary carcinoma of excretory ducts, or high-grade salivary duct carcinoma. SDC represents 9% of salivary malignancies. The male: female ratio is at least 4:1 and most patients present after age 50⁴. The parotid is most commonly involved, but submandibular, sublingual, minor salivary gland, maxillary and laryngeal tumours have been reported². SDCs are usually firm, solid, tan, white or grey, with a cystic component. Infiltration of the adjacent parenchyma is usually obvious, but occasional tumours may appear to be circumscribed. SDC may also arise as the malignant component of a carcinoma ex-pleomorphic adenoma, so that the macroscopic features of pleomorphic adenoma may also be present. For SDC, perineural spread (60%) and intravascular tumour emboli (31%) are common⁴. SDC resembles intraductal and infiltrating mammary duct carcinoma, both architecturally and cytologically. The diagnostic “ductal lesion” comprises pleomorphic, epithelioid tumour cells with a cribriform growth pattern, “Roman bridge” formation, and intraductal comedonecrosis. Cytologically, these cells have abundant, pink cytoplasm and large pleomorphic nuclei with prominent nucleoli and coarse chromatin. The cytoplasm may also be densely eosinophilic, granular, or oncocytic. Mitotic figures are usually abundant. Goblet cells are not seen^{5,6}.

Immunohistochemistry

SDC is immunoreactive for low- and high-molecular-weight cytokeratin, and markers such as carcinoembryonic antigen (CEA), LeuM1, and epithelial membrane antigen (EMA)⁷. Strong nuclear

reactivity for androgen receptors (AR) is reported in all SDC. As well as being positive for GCDFP-15, they are negative for S-100 protein, myoepithelial markers as well as estrogen and progesterone receptors⁸. The MIB1 proliferative index is high. Most SDCs show positive distinct membrane staining for HER-2/neu protein. Metastatic breast and squamous carcinomas, oncocytic carcinoma and mucoepidermoid carcinoma come in differential diagnosis because they also show similar immunohistochemistry profiles^{9–11}.

SDC is one of the most aggressive salivary malignancies. Sites for distant metastasis include lungs, bones, liver, brain and skin⁴. Sixty-five percent of patients die from the disease, usually within 4 years of diagnosis (ranging from 5 months to 10 years)¹¹. The clinical course is characterized by early distant metastases. Tumour size, distant metastasis, and HER-2/neu overexpression are putative prognostic parameters for SDC, while expression of p53 protein, DNA aneuploidy, and proliferative activity do not correlate with outcome^{9,10}. The clinical outcome for the mucin-rich variant of SDC is similar to that of conventional SDC¹².

Conclusion

Given the known difficulty in making an accurate diagnosis of salivary duct carcinoma¹³, the identification of a tumor exhibiting variable nuclear grade with cribriform, papillary and comedo patterns in the appropriate clinical setting of elderly patients with parotid

mass and facial palsy should suggest the diagnosis of this uncommon tumor after excluding a metastatic carcinoma.

Consent

Written Informed consent for publication was obtained from the patient.

Author contributions

RS and CLP contributed to the conception and design of the study. RS collected and analyzed the data and wrote up the manuscript. RS and CLP both approved the manuscript.

Competing interests

No competing interests were disclosed.

Grant information

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Every report of rare cases must be appreciated, even if salivary duct carcinoma is not so rare (9% of salivary glands malignancies), and dozens of cases have been reported in the past.

Competing Interests: No competing interests were disclosed.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

Reviewer Report 01 November 2012

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In general, This is a well written case report of salivary duct carcinoma.

In this case, like most rare and poorly differentiated salivary neoplasms, the correct diagnosis was only possible on the final specimen despite FNA, frozen, and permanent section histopathology of the metastatic lymph nodes. While the immunohistochemistry is reviewed in the discussion, it is not

specified how it was used in this particular case. This report will not alter the clinical management of this disease but it highlights the most salient points in the clinical diagnosis of salivary ductal carcinoma.

Competing Interests: No competing interests were disclosed.

I confirm that I have read this submission and believe that I have an appropriate level of expertise to confirm that it is of an acceptable scientific standard, however I have significant reservations, as outlined above.

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