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Intraductal papilloma of the submandibular gland

Showkat Mirza, F.R.C.S.(Ed.), Sunil N. Dutt, M.S., D.N.B., F.R.C.S. (Ed.),
Richard M. Irving, M.D., F.R.C.S.(orl), E. L. Jones, M.D., F.R.C.Path.*

Abstract
Salivary tissue intraductal papillomas are rare, benign tumours that predominantly affect minor salivary glands. We report a case of an intraductal papilloma arising in the unusual site of the submandibular gland. The tumour was completely excised and recurrence is not expected. A brief review of this histologically distinct lesion is presented.

Key words: Salivary gland neoplasms; Submandibular gland

Introduction

Salivary gland ductal papillomas are a group of rare, benign tumours that include the intraductal papilloma (IDP), inverted ductal papilloma and sialadenoma papilliferum. Castigliano and Gold[1] were the first to report a case of IDP, in a minor salivary gland. Abrams and Finck[2] introduced the term sialadenoma papilliferum, whilst White *et al*.[3] described the inverted ductal papilloma which is analogous to the inverted papilloma of the nasal cavity. These neoplasms may form a spectrum of neoplastic changes seen in salivary gland duct epithelium.[4] We report the case of an intraductal papilloma in the unusual location of the submandibular salivary gland.

Case report

A 76-year-old man presented with a four-month history of a painless, slow-growing left submandibular salivary gland swelling. An ultrasound scan showed a 3 cm diameter well-defined but complex cystic lesion within the left submandibular gland. The clinical suspicion was that of a pleomorphic adenoma, though malignancy was considered a possibility. An excision of the left submandibular gland was performed. The gland was found to be enlarged and to contain a cystic swelling.

Histopathology

The left submandibular gland (wt 15 g) measured 5 × 3 × 2.5 cm and showed a pale area 1 × 0.5 cm towards one edge. The cut surface of the gland was otherwise normal. Histologically it showed a thick-walled duct enclosing a unilocular intraluminal papillary epithelial tumour (Figure 1). Recent haemorrhage was present in the duct. The epithelium appeared stratified (Figure 2) and surrounded oedematous vascular stroma containing foam cells. The nuclei were regular and no mitoses were seen. In places the papilloma cells had an oncocytoid appearance with uniform small round or ovoid nuclei and abundant eosinophilic cytoplasm (Figure 3).

The appearances were those of a benign intraductal papilloma.

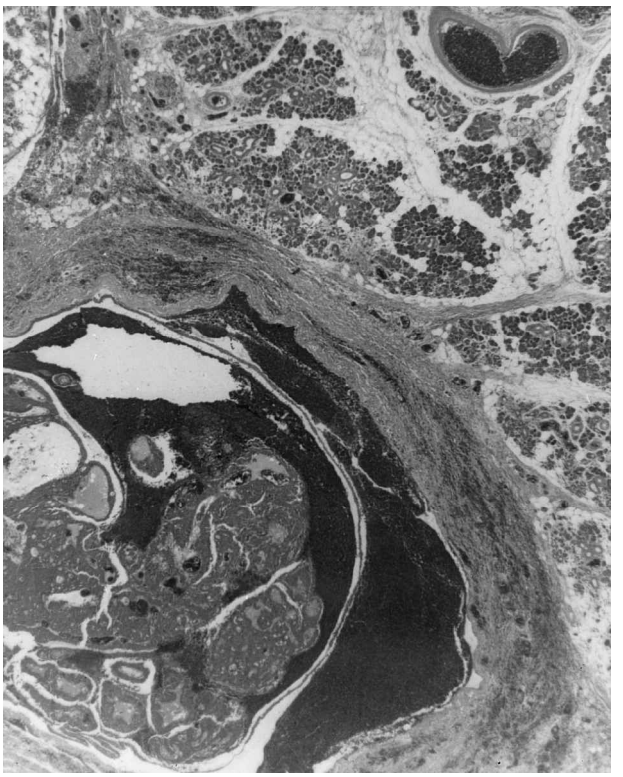

Fig. 1

The dilated duct has a thick fibrous wall and the lumen contains a branching papillary epithelium proliferation that nearly fills the lumen. (H & E; ×22).

Discussion

IDPs of the salivary glands are rare, benign tumours reported to be found primarily in minor salivary glands.[4,5] Locations include the palate, buccal mucosa, floor of the mouth,[6] nasal cavity,[7] and lip.[5] IDPs have also been

From the Department of Otorhinolaryngology – Head and Neck Surgery, Queen Elizabeth University Hospitals NHS Trust, Birmingham and Department of Pathology*, Birmingham University Medical School, Birmingham, UK.
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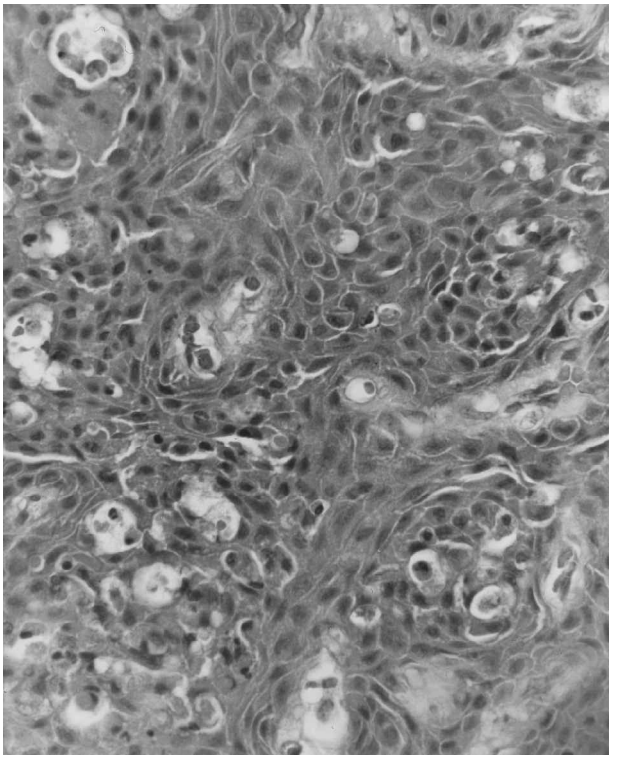

FIG. 2
Intraductal papilloma showing stratified squamoid epithelium supported by cores of fibrovascular tissue. (H & E; ×342).

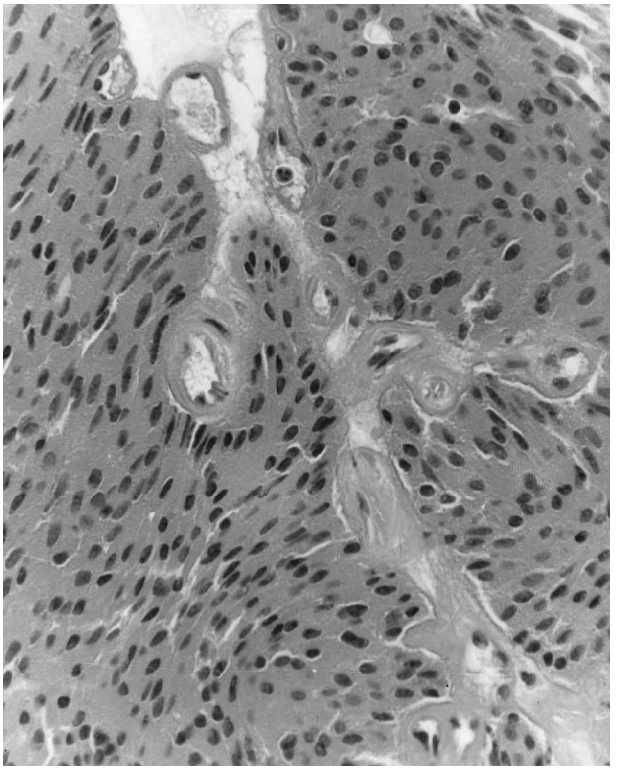

FIG. 3
Intraductal papilloma showing eosinophilic oncocytoid type epithelium with regular small nuclei (H & E; ×342).

reported in the parotid gland,[6,8–10] sublingual gland[11,12] and submandibular gland duct.[6]

If found in the minor salivary glands, these tumours present as asymptomatic, submucosal swellings that vary in size from less than 1 to 1.5 cm, in patients with an average age of 50 years.[6]

The histological features of ductal papillomas are essentially those of ductal proliferation and of papillary projections into the lumen of dilated-duct like structures.[4] The differential diagnosis rests between intraductal papilloma, inverted ductal papilloma, sialadenoma papilliferum or papillary cystadenoma. An intraductal papilloma comprises a cellular mass protruding into a dilated duct.[4] The mass contains a core of connective tissue with dilated glandular ducts and cysts, as well as one or two layers of cuboidal or squamous epithelium projecting into the ductal lumen.[13] An inverted papilloma shows intramural growth and a proliferation of sinonasal transitional papilloma-like epithelium into the supporting connective tissue.[3] The papillary cystadenoma has been described as a cystic adenoma in which multiple cystic spaces are filled with papillary projections.[14] Sialadenoma papilloferum is an exophytic mucosal lesion of minor salivary glands in which papillary projections of stratified squamous epithelium form in tortuous dilated excretory ducts.[2]

In IDPs the cells are bland and uniform with minimal nuclear atypia and absent mitotic activity. The morphology, in addition to the negative immuno-staining of the cells with the proliferation cell marker Ki-67, strongly supports the benign nature of the tumour.[7] Further immunohistochemical studies support a clonal neoplastic proliferation of ductal epithelium origin.[7,11]

Fine needle aspiration cytological findings of IDP are unique and may allow its specific diagnosis but appearances may vary from benign cyst[10] to adenoid cystic carcinoma.[8]

Treatment consists of excision which is curative. Recurrences do not seem to occur.[4,6,8]

The lesion is benign but has possibly been implicated in one case of papillary adenocarcinoma.[9] The authors reporting this case found an infiltrative growth with an atypical glandular ductal structure in the periphery of a papillary growth that was consistent with the appearance of an intraductal papilloma. They suggested that an intraductal papilloma may have been present initially which then developed into a carcinoma.

IDPs in the parotid gland can result in the formation of duct cysts. As there is a possibility a malignant tumour may develop from an IDP possibly manifesting only as a ductal cyst, the operative treatment of benign cysts is important. Therefore if surgery is not the treatment of choice due for example to the patient's overall condition being poor then long-term follow-up should be arranged.[10]

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Address for correspondence:
Mr Showkat Mizra,
7 Cemetery Road,
Royton,
Oldham OL2 5SP, UK.

Fax +44 (0)121 627 2299

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