Case Report

A Rare Case of Benign Synchronous Neoplasm of Parotid Salivary Gland

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Abstract

Introduction

Salivary gland neoplasms represent 2–6% of all head and neck neoplasms. Salivary gland neoplasms arising from the major salivary glands are predominantly benign. Pleomorphic adenoma is the most common benign neoplasm of the salivary gland, followed by warthin's tumour. The synchronous occurrence of both of these neoplasms is rarely described. We present a case of unilateral salivary gland neoplasm with two distinct histological lesions, which were found incidentally during gross examination and confirmed microscopically.

Case report

A 72-year-old man with complaints of left parotid swelling, which progressively increased in size over the past 6 months. Ultrasound imaging studies and FNAC show features of pleomorphic adenoma, for which the patient underwent surgery, and the specimen was sent for histopathological examination. In the gross examination, a tan-brown area measuring 0.6 cm in greatest dimension was noted in the parotid salivary gland, in addition to the encapsulated grey-brown neoplasm. Histologically, the tan-brown lesion was found to be an intranodal warthin's tumour along with a pleomorphic adenoma.

Conclusion

Synchronous neoplasm is a rare entity that can easily be missed in clinical, imaging and even in cytological examinations. Proper histopathological examination of the resected specimens helps in identifying the lesions that were left unrecognized even by imaging studies due to their proximity to the existing neoplasm, as in our case. The use of VTIQ in combination with B-mode ultrasound may be beneficial in detecting the nature of the lesion prior to surgery.

Keywords:

synchronous neoplasm, intranodal warthin's tumour, pleomorphic adenoma

Introduction

Salivary gland neoplasms are comparatively less common, and they represent 2-6% of all head and neck neoplasms. Salivary gland neoplasms arising from the major salivary glands are predominantly benign (~70%). The rest of the benign neoplasms are seen involving the minor salivary glands (~25%) [1]. The presence of two histologically different neoplasms within the same salivary glands is rare. These synchronous neoplasms lie closely within the same salivary gland and are usually undetected by imaging studies. This becomes a challenge for both radiologists and surgeons in the diagnosis and management of salivary gland neoplasms [2]. We present a case of unilateral salivary gland neoplasm with two distinct histological lesions, which were found incidentally during gross examination and confirmed microscopically.

Case Report

In December 2023, a 72-year-old man was referred to the Department of General Surgery at Apollo Loga Hospital, Karur, for surgical management of a left parotid swelling that had progressively increased in size over the past 6 months. The medical history was unremarkable. On examination, the mass was firm, non-tender, and 4 X 3 cm in size with restricted movement. There were no features of nerve, deep lobe, or nodal involvement. An ultrasound of the neck showed a left parotid mass lesion with increased vascularity and no significant lymphadenopathy. Based on the size of lesion and affordability of the patient, direct FNAC was done and the smears showed tight clusters of benign ductal epithelial cells admixed with myoepithelial cells in the background of fibromyxoid stroma. These features favor pleomorphic adenoma, Category IVA, Benign, The milan system for reporting salivary gland cytopathology (MSRSGC).

Based on the FNAC report, radiological studies, and clinical examination, a provisional diagnosis of pleomorphic adenoma was made. The patient was counseled for surgical management, even though the small risk of recurrence and carcinoma ex pleomorphic adenoma were explained. The postoperative complications of facial nerve injury, the development of a sialocele, and Frey's syndrome were discussed. The decision was then made to excise the lesion along with the superficial parotid lobe.

The next day, the patient underwent left superficial parotidectomy. Under the aseptic condition, the lesion was accessed using a modified Blair's incision. Skin, subcutaneous tissue incised, flap anteriorly raised in a subplatysmal plane up to the anterior margin of the masseter muscle. Left sternocleidomastoid and digastric muscles were delineated. The cartilaginous part of the external auditory canal is identified with a tragal point. The main branch of the facial nerve is identified and preserved. All tissue superficial to the branches of the facial nerve was removed carefully. All branches were carefully dissected out and separated from parotid tissue. The Tumour adherent to the anteromedial margin along with the duct was excised in toto. A few lymph nodes at level 3, were excised and sent for biopsy. Hemostasis was secured by abGel. A 10Fr-size drain was kept, and the wound closed in layers with 3-0 Vicryl and 3-0 Monocryl. The Dressing was done, and the patient was shifted to the post-operative ward.

The specimen was then sent for histopathological examination. The Specimen was sent in three containers, comprising the superficial parotid lobe, the excised tumor, and level 3 lymph nodes. The Superficial parotid lobe measures 7.5x5x1.8cm. The external surface was unremarkable. The cut surface had a small circumscribed tan-brown area measuring 0.6 cm in the greatest dimension (Fig. 1). The excised grey-brown capsulated tumor measures 3x3x2cm. The cut surface appears tan-white, bosselated, and glistening (Fig. 1). Two lymph nodes were identified, measuring 1x0.9x0.6cm and 0.9x0.5x0.4cm, respectively. Microscopically, the sections from the superficial parotid specimen show normal parotid tissue with few reactive lymph nodes and an intranodal neoplasm comprising papillary cystic structures lined by bilayered oncocytic epithelium surrounded by

lymphoid stroma (Figs. 2 and 3). Sections from the excised parotid tumour show a well-circumscribed neoplasm comprising epithelial (ductal) and myoepithelial components. The epithelial components form the inner layer of cysts and tubules. The outer layer was formed by spindled and stellate-shaped myoepithelial cells scattered in a chondromyxoid stroma (Fig. 4). Sections from level 3 lymph nodes show reactive changes. These histological appearances were consistent with a co-existing pleomorphic adenoma and an intranodal warthin's tumour of the unilateral parotid salivary gland.

The patient recovered uneventfully from the operation and was discharged on the 3rd postoperative day. During follow-up, the patient had mild LMN paresis, which was improved by physiotherapy.

Figure 1: The gross image of parotid salivary gland (left) with tan-brown area(*) and an excised parotid tumour with the cut-surface exhibiting grey white glistening appearance (right).

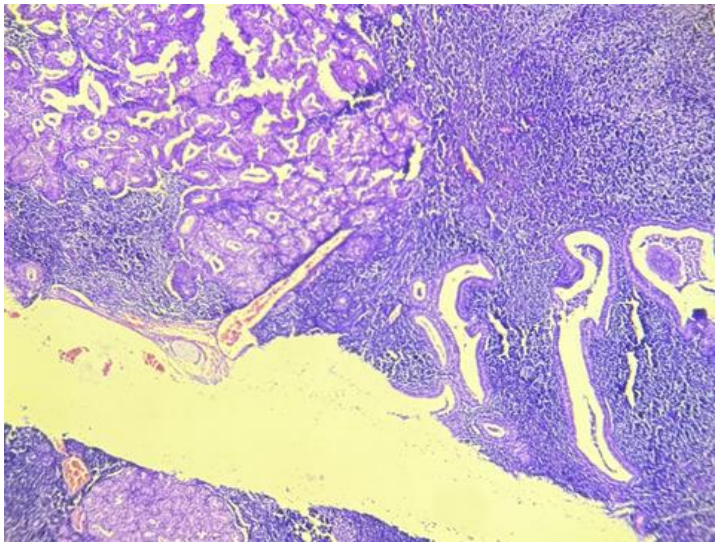

Figure 2: Low magnification image of Intra-nodal Warthin's tumour (H&E 10X)

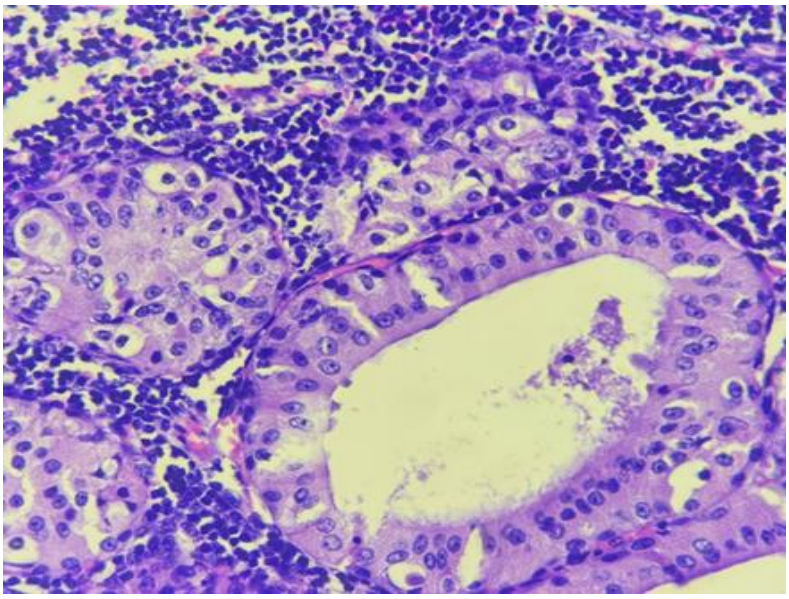

Figure 3: Higher magnification shows bilayered oncocytic epithelium with lymphoid stroma (H&E, 40X)

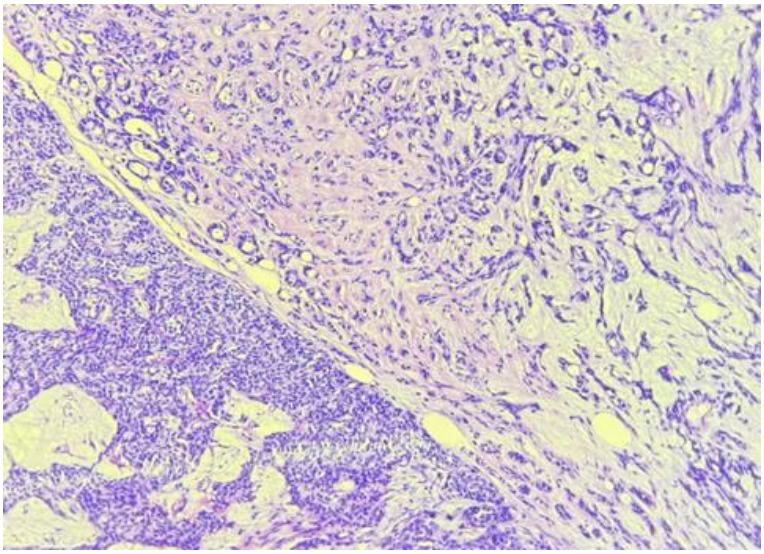

Figure 4: Low magnification image of excised parotid neoplasm showing features of pleomorphic adenoma. (H&E, 10X)

Discussion

Pleomorphic adenoma is the most common benign neoplasm of the salivary gland followed by Warthin's tumour or papillary Cystadenoma lymphomatosum.[3] The concurrent occurrence of both these neoplasms is rarely described.

In the last 30 years (1992-2023), only seven cases of synchronous pleomorphic adenoma and warthin's tumour involving the unilateral parotid salivary gland were reported [2,4-8]. Horisk et al had a similar case in which an MRI scan reported a single bilobed lesion, but in histopathological examination, it was two distinct lesions with different microscopic features [2]. Klamminger et al in their case report, suggested that the timely and accurate histomorphological assessment of all specimens helps in identifying multiple tumours and aids in further management[4]. Two cases were reported by Heine et al, the author suggests the use of virtual touch imaging quantification (VTIQ) in combination with B-mode ultrasound in the detection of two distinct

lesions preoperatively. VTIQ measures tissue stiffness quantitatively and can differentiate benign and malignant lesions [5]. A similar case was reported by Herce et al., where clinically it was a single lesion, and in a fine needle cytological examination, the lesion was reported as pleomorphic adenoma. Later, in a computed tomography scan, two separate masses were identified adjacent to each other, and a final impression of warthin's tumour was given. A superficial parotidectomy was done, and the specimen was sent for histopathological examination. The parotidectomy specimen showed two different neoplasms: a pleomorphic adenoma and an intranodal warthin's tumour, as in our case [6]. Franzen et al., in their case report, suggested that proper preoperative and intraoperative examination of glandular and periglandular lymph nodes aids in the detection of multiple neoplasms [7]. Godden et al,. had a similar case with two distinct lesions, which were reported as pleomorphic adenoma with warthin's tumour. In addition, one of the lymph nodes showed an intraparotid warthin's tumour [8].

In statistical surveys of unilateral synchronous pleomorphic adenoma and warthin's tumour of the parotid gland, Ethunandan et al., reported two cases out of 606 patients; Bein et al,. had one case in 196 parotidectomy specimens, three cases were reported among 2055 patients by Yu et al, and three cases in 341 parotidectomy surgeries of by Zeebregts et al [9-12].

Although clinical and radiological studies are generally useful in the diagnosis of all neoplasms, the difficulty in the diagnosis of multiple synchronous neoplasms involving the single salivary gland is well known. Even FNAC fails to detect the presence of multiple neoplasms due to the small sample size. A definite diagnosis requires a proper histopathological examination, as in the present case.

Cases of synchronous neoplasm presenting as three different histological types [13] and multiple bilateral warthin's tumours with pleomorphic adenoma [14] were reported. Curry et al. reported nine cases of synchronous benign neoplasms with malignant tumours; they also reported the first case of salivary duct carcinoma with pleomorphic adenoma [15]. Another case of synchronous malignant tumours with benign neoplasm was also reported i.e., concurrent secretory carcinoma with warthin's tumour by Kaleem et al [16]. A very rare case of mucoepidermoid carcinoma with carcinoma ex pleomorphic adenoma in the submandibular salivary gland was reported by Edward Wai-Hei et al [17].

Total or subtotal parotidectomy is the treatment protocol for all benign neoplasm of salivary gland. The presence of additional synchronous benign neoplasm doesn't alter the treatment protocol. But the presence of unilateral warthin's requires examination of the contralateral side for any lesion to rule out bilaterality of warthin's tumour.

Conclusion

Synchronous neoplasm is a rare entity that can easily be missed in clinical, imaging, and even in cytological examinations. Proper histopathological examination of the resected specimens helps in identifying the lesions that were left unrecognized even by imaging studies due to their proximity to the existing neoplasm, as in our case. The use of VTIQ in combination with B-mode ultrasound may be beneficial in detecting the nature of the lesion prior to surgery.

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