

Case Report

Diagnostic challenge of parotid multifocal adenomatous oncocytic hyperplasia: A case report

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Abstract Parotid glands have diverse histological findings thanks to abundant different types of cells presence in the gland. Our routine fine needle aspiration cytology might be inaccurate and misleading in cases where there are cell changes due to ongoing concurrent infection and chronic sialadenitis, which might have mimicked tumoural changes. We highlighted a rare case of multiple florid benign hyperplasia of oncocytic cells of the parotid gland that manifest as parotid swelling. The characteristic multifocality of the lesion and high rate of recurrences, although not a known malignant entity, resulting in a significant shift in the treatment plan for the patient.

Keywords: Oncocytes; oncocytoma; oxyphil cells; parotid gland.

Introduction

The parotid gland is the largest salivary gland in the human body and comprises different types of cells. Typically, the most common benign salivary lesion according to WHO classification is pleomorphic adenoma (El-Naggar *et al.*, 2017), with prevalence of 53.8% (Araya *et al.*, 2015), which is characterized by proliferation of the glandular cells along with their myoepithelial components. Surgical excision of the lesion is the most appropriate treatment plan. On the other hand, the most common malignant counterpart of the parotid lesion, which is mucoepidermoid carcinoma with prevalence of 31% (Xiao *et al.*, 2016), warrants complete removal of the gland to achieve tumour clearance, with consideration of adjuvant oncology treatment.

Case report

A 65-year-old Chinese gentleman presented with a left parotid swelling. It was gradually increased in size, together with signs of infection. However, the size of the swelling remained static after completion of antibiotic treatment. Otherwise, it was asymptomatic.

He had a strong family history of malignancy; two of his siblings suffered from colon and breast carcinoma, respectively. Examination-wise, the facial nerve was intact. There was left parotid swelling measuring 3 cm x 3 cm, non-tender, firm in consistency, not fixed to overlying skin, and with a smooth surface. The left level IIB lymph node, measuring 0.5 cm x 0.5 cm, was palpable. The Stenson's duct appeared normal with no stone, and there was no medialization of the lateral pharyngeal wall. Other examinations were unremarkable.

Computed tomography (CT) scan of the neck showed a well-defined cystic lesion with thin regular peripheral wall enhancement, confined to the superficial lobe of the left parotid gland and measuring 1.9 cm x 1.3 cm x 1.5 cm (Fig. 1). Otherwise, no obvious deeper extension to the paravertebral region or spinal involvement, and the adjacent left sternocleidomastoid muscle appeared normal. He was subjected to fine needle aspiration for cytology (FNAC) of left parotid mass, which was reported as atypical squamous cells with hyperchromatic and angulated cytoplasm (Fig. 2a, Fig. 2b). Scattered mucinoid cells positive for Periodic

Acid Schiff (PAS) stain were present. The background showed necrotic material. A diagnosis of suspicious malignancy, presumed to be low-grade mucoepidermoid carcinoma, was made. With benign-looking lesion on CT, but unfavorable cytology report, a diagnostic dilemma was faced. Superficial parotidectomy was commenced as part of the diagnostic work-up. During the operation, selective left neck dissection of level IIA and IIB, together with biopsy of deep lobe of left parotid, was carried out. Intraoperative frozen section of left level IIA lymph node showed no malignancy. The final histopathology report of the resected

left parotid gland revealed multifocal adenomatous oncocytic hyperplasia with chronic sialadenitis features. There were multiple nodules of oncocytic cells hyperplasia visible throughout the glands, the cells having small round uniform nuclei and abundant granular eosinophilic cytoplasm (Fig. 2c, Fig. 2d). In the centre of the organ, chronic inflammatory infiltrates with metaplastic changes of the ductal epithelium were observed (Fig. 2e). The biopsy of the deep lobe also showed similar multifocal lesions. Otherwise, the patient was well for almost five months postoperatively to date; and is under close watchful observation at our centre.



Fig. 1 CT image axial view in soft tissue setting showing a left parotid cystic lesion with minimal peripheral rim enhancement (arrow).

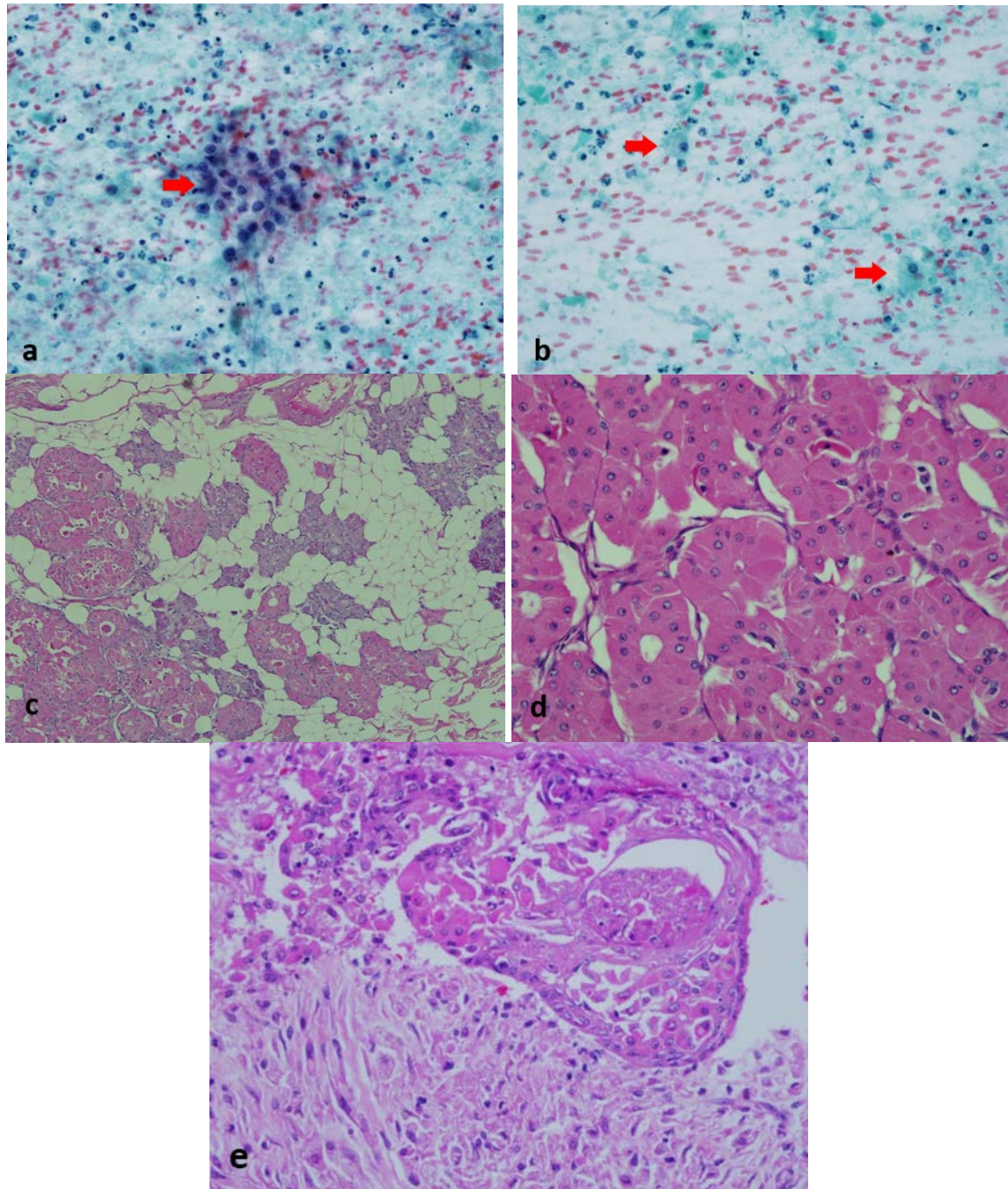


Fig. 2 Cytology and histopathology findings: (a) Atypical squamous cells seen with hyperchromatic and angulated cytoplasm (40x10 magnification). (b) Scattered mucinous like cells were present (40x10 magnification). (c) Multiple nodules of oncocytic hyperplasia seen throughout the gland (H&E, 10x10 magnification). (d) The oncocytes exhibit small round uniform nuclei with abundant granular eosinophilic cytoplasm (H&E, 40x10 magnification). (e) Metaplastic changes of the ductal epithelium are seen, surrounded by chronic inflammatory infiltrates (H&E, 40x10 magnification).

Discussion

Oncocytes are cells that are found in small aggregates, interposed in between acinar and ductal cells of the salivary gland (Schwartz and Feldman, 1969). Little is known about the function of these eosinophilic granular cytoplasmic cells, but, interestingly, similar cells are also present in many other organs, such as thyroid, parathyroid, adrenal cortex, kidney, pituitary, and breasts (Schwartz and Feldman, 1969). Thus, differential diagnosis of a parotid mass should also include possible metastases from these organs. These cells appear as the principal component in both benign and malignant salivary gland lesions, such as diffuse oncocytosis, multifocal adenomatous oncocytic hyperplasia, oncocytic metaplasia, oncocytoma, as well as oncocytic carcinoma.

This oncocytic nodular hyperplasia is an extremely rare condition, with less than ten cases reported in the literature (Politi *et al.*, 2005). It is not a true neoplasm, as opposed to oncocytoma, which also only accounts for less than one percent of salivary gland tumours and also has findings of multiple foci of oncocytes surrounding the glandular tissue, a common finding in endocrine organs, such as thyroid and parathyroid (Blanck *et al.*, 1970).

In the present case, the fine-needle diagnosis was suggestive of a possible low-grade mucoepidermoid carcinoma. Chronic sialadenitis is one of its close differential diagnoses. Both features could overlap, and it was impossible to differentiate them on fine-needle aspirate. Therefore, histopathological confirmation is necessary in this situation. FNAC of parotid gland lesions have 73% sensitivity and 97% specificity (Gudmundsson *et al.*, 2016). Mucoepidermoid carcinoma, together with acinic cell carcinoma, and epithelial-myoeplithelial carcinoma were reported as having the highest false-negative diagnosis (Gudmundsson *et al.*, 2016).

The atypical squamous cells seen on fine needle aspiration are actually immature metaplastic squamous cells present in chronic sialadenitis, as proven

by the histopathological findings of the gland. Most of chronic sialadenitis occurs due to obstruction of the salivary ducts by calculi, which were not present in this case. It is possible the obstruction was due to the presence of multiple nodules of oncocytic hyperplasia, which were seen surrounding the chronic sialadenitic area.

The disadvantage of this histological finding is the fact that although it was not even a tumour, oncocytic hyperplasia could occur bilaterally, with a high frequency of multifocality, as well as having a high tendency of recurrence. In a study involving 13 cases, five had bilateral diseases, which appeared synchronously in two of the patients, while four cases had recurrence between six months and five years post-operatively (Blanck *et al.*, 1970). In some cases, the patients had received radiotherapy instead of surgical removal, with the outcome of static disease but no observable eradication. However, no further elaboration of radiotherapy was mentioned.

In our patient, reflecting retrospectively, the salivary gland tissue complexity itself has put the role of FNAC to certain limit. Thus, correlation with histopathological examination with intraoperative frozen section microscopy, is necessary. We might embark on total parotidectomy to eradicate all foci of hyperplasia, but minus the unnecessary neck dissection. Now, as to where we stand, we have not only just partially removed the lesion, but also have unintentionally increased his risk of facial nerve palsy due to healing fibrosis, should re-operation be needed in case of recurrence. Perhaps, repeat FNAC is a better decision in cases of unmatched cytological and radiological findings. All in all, the present case has served as a valuable learning curve on how diverse the diseases of head and neck could be.

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