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Oncocytic Cystadenoma of the Parotid Gland Presenting as a Cystic Lesion: A Case Report of Diagnostic Discordance

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Abstract

Cystic lesions of the parotid gland represent a recognised diagnostic challenge, as both cross-sectional imaging and fine-needle aspiration cytology (FNAC) may have reduced diagnostic accuracy in these cases. We report the case of a 57-year-old man with a seven-month history of painless swelling anterior to the right earlobe. Ultrasonography and contrast-enhanced computed tomography both favoured a lymphoepithelial cyst or a necrotic lymph node, while FNAC, despite repeated aspirations, yielded atypia of undetermined significance (Milan category III), with a recommendation for excision biopsy to exclude malignancy. Right conservative superficial parotidectomy with antegrade facial nerve dissection was performed.

Histopathological examination revealed a multiloculated cystic lesion lined by oncocytic epithelium, consistent with an oncocytic cystadenoma, a diagnosis not suggested by any of the preoperative investigations. The postoperative course was uneventful, with no facial nerve deficit. Milan category III cytology in a progressively enlarging cystic parotid mass should prompt consideration of diagnostic surgery rather than provide reassurance regarding the absence of malignancy.

Categories: General Surgery
Keywords: facial nerve, milan system, oncocytic cystadenoma, parotid gland, superficial parotidectomy

Introduction

Salivary gland neoplasms account for a small proportion of head and neck tumours, with the majority arising in the parotid gland. The fifth edition of the World Health Organization (WHO) Classification of Head and Neck Tumours recognises more than 30 distinct salivary gland entities, several of which exhibit overlapping cytomorphological features. This diversity underlies the well-documented limitations of preoperative diagnosis in this region [1,2].

Cystadenoma is one of the rarer benign epithelial neoplasms. The WHO recognises papillary, mucinous, and oncocytic variants, with the latter characterised by multicystic growth lined by oncocytes [1].

Two features make cystic parotid lesions particularly challenging to diagnose. First, radiological findings have limited discriminatory value: a thin-walled, minimally enhancing cystic lesion in the superficial lobe often results in a relatively narrow differential diagnosis, predominantly comprising non-neoplastic conditions. Second, fine-needle aspiration cytology (FNAC) of cystic lesions predominantly samples fluid, and the resulting hypocellular or degenerative atypical smears commonly fall into category III (atypia of undetermined significance, AUS) of the Milan System for Reporting Salivary Gland Cytopathology. This category has the widest reported variation in risk of malignancy (ROM) [3-6].

We report a case in which ultrasonography, contrast-enhanced computed tomography (CECT), and FNAC all failed to suggest the eventual diagnosis, highlighting how clinical decision-making under diagnostic uncertainty ultimately led to both definitive diagnosis and treatment.

Case Presentation

A 57-year-old man presented with a swelling anterior to the right earlobe that had been present for seven months. The swelling had an insidious onset and had gradually increased in size. There was no associated pain, fever, sudden increase in size, swelling elsewhere, or dryness of the eyes or mouth. He had a 10-year history of type 2 diabetes mellitus and systemic hypertension, both of which were being treated. However, he had been non-compliant with his antihypertensive medication for one month before presentation.

On examination, he was afebrile, with a blood pressure of 170/100 mmHg. A spherical swelling measuring approximately 3 × 2 cm was noted anterior to the right earlobe, with normal overlying skin and elevation of the earlobe. The swelling was firm, non-tender, and non-compressible. The overlying skin was pinchable, and the curtain sign was positive, localising the lesion to the parotid gland. There was no cervical

lymphadenopathy, and facial nerve function was intact (House-Brackmann grade I). Haematological indices were within normal limits.

Ultrasonography performed at the referring institution demonstrated a 12 × 10 mm cystic lesion in the right parotid gland, which was reported as either a lymphoepithelial cyst or a reactive lymph node. FNAC, performed with multiple repeated aspirations, yielded moderately cellular smears containing clusters of salivary acinar cells and occasional fibrous stromal fragments against a haemorrhagic background. Some clusters exhibited an increased nuclear-to-cytoplasmic ratio and mild nuclear atypia. The cytological impression was AUS (Milan category III), and excision biopsy was recommended to exclude malignancy.

CECT of the neck demonstrated a well-defined cystic lesion measuring 1.8 × 1.5 × 1.8 cm in the superficial lobe of the right parotid gland, with minimal peripheral wall enhancement, no internal enhancement, and medial abutment of the external carotid artery. A separate 4 × 3 mm nodule was considered to represent an intraparotid lymph node. An incidental hypoenhancing nodule measuring 3.4 × 3.7 mm, with a calcific focus, was also identified in the left thyroid lobe, for which ultrasound correlation was recommended.

Glycaemic and blood pressure control were optimised with multidisciplinary input before surgery. The working diagnosis was an indeterminate cystic neoplasm of the superficial lobe of the right parotid gland with Milan category III cytology. Surgical excision was undertaken to establish a definitive diagnosis and provide treatment.

Right superficial parotidectomy was performed under general anaesthesia. Through a modified Blair incision, flaps were elevated in the subplatysmal plane inferiorly and in the superficial musculoaponeurotic system (SMAS) plane anteriorly. The greater auricular nerve was preserved. The main trunk of the facial nerve was identified at the stylomastoid foramen using the classical anatomical landmarks of the tragal pointer, posterior belly of the digastric muscle, and tympanomastoid suture. The superficial lobe was then dissected in an antegrade fashion, with all five facial nerve branches identified and preserved (Figure *1*). A 2 × 2 cm tumour was excised en bloc with a cuff of surrounding glandular tissue. A closed-suction drain was placed, and the wound was closed in layers.

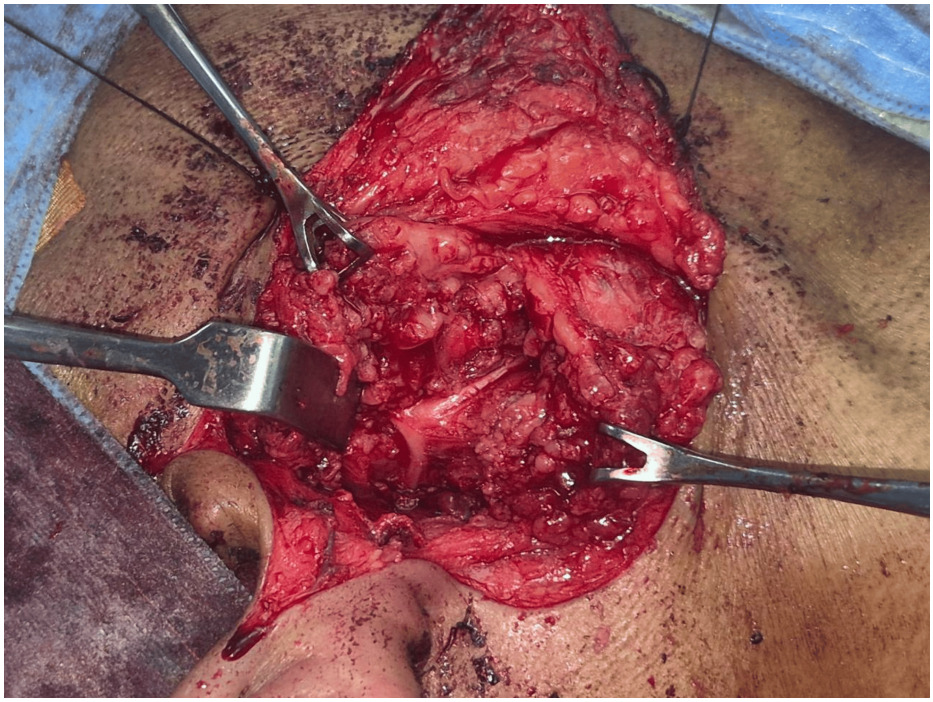


FIGURE 1: Intraoperative view of the main trunk and branches of the facial nerve following antegrade dissection.

The postoperative course was uneventful, with no evidence of facial nerve palsy. The patient was discharged on the second postoperative day.

On gross examination, the specimen consisted of salivary gland tissue weighing 9.9 g and measuring 5.5 × 4 × 1.9 cm. It contained a cystic nodule measuring 2 × 2 × 1.5 cm, which extruded yellowish fluid on sectioning. The cut surface revealed a multiloculated cyst with septations and a wall thickness of 0.2 cm (Figure *2*). All resection margins were free of the lesion, with the closest margin measuring 0.1 cm.

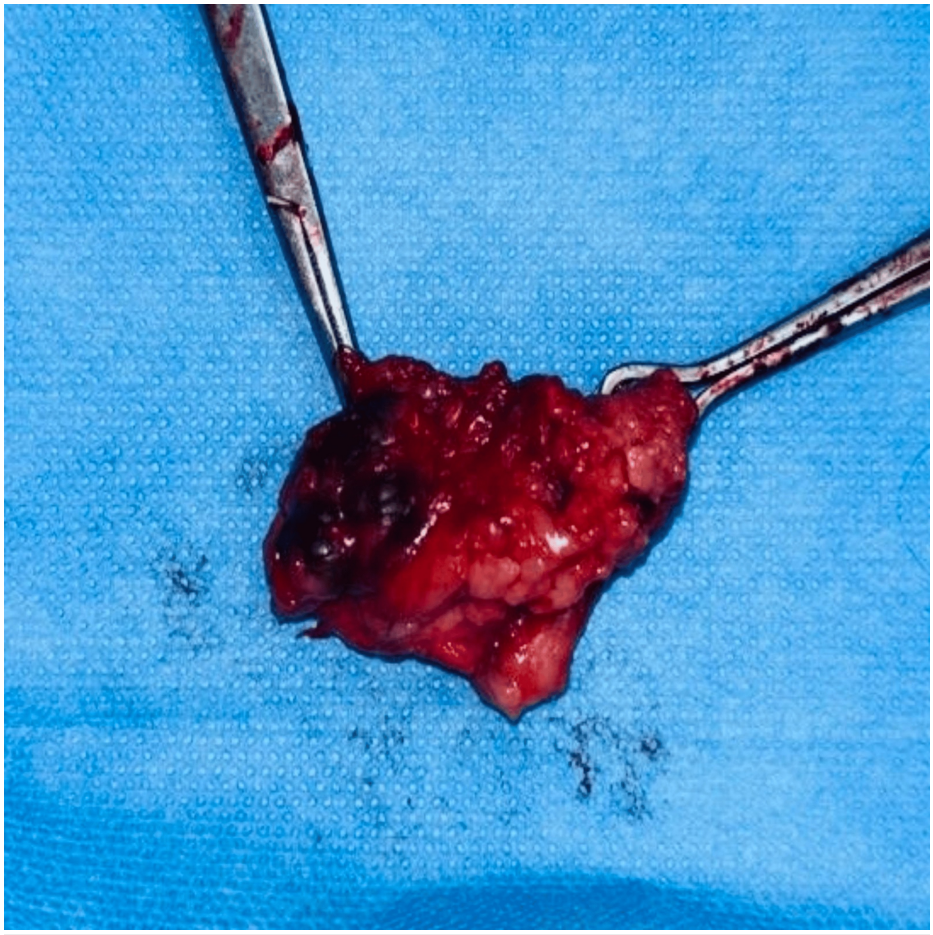


FIGURE 2: Gross appearance of the salivary gland specimen, weighing 9.9 g.

Microscopic examination revealed a multicystic lesion lined by oncocytic epithelium, without lymphoid stroma, cytological atypia, or evidence of invasion. The findings were reported as an oncocytic neoplasm, favouring oncocytic cystadenoma (Figures *3*, *4*).

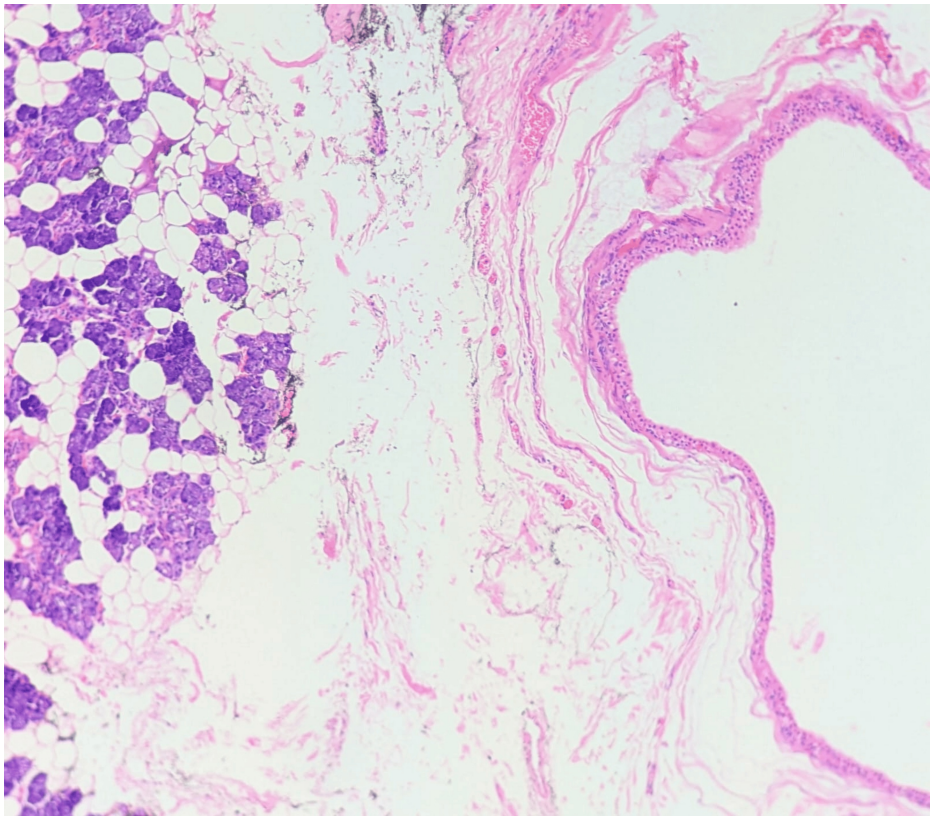


FIGURE 3: Parotid gland tissue with an adjacent cyst lined by oncocytic epithelium (haematoxylin and eosin stain, ×10 magnification).

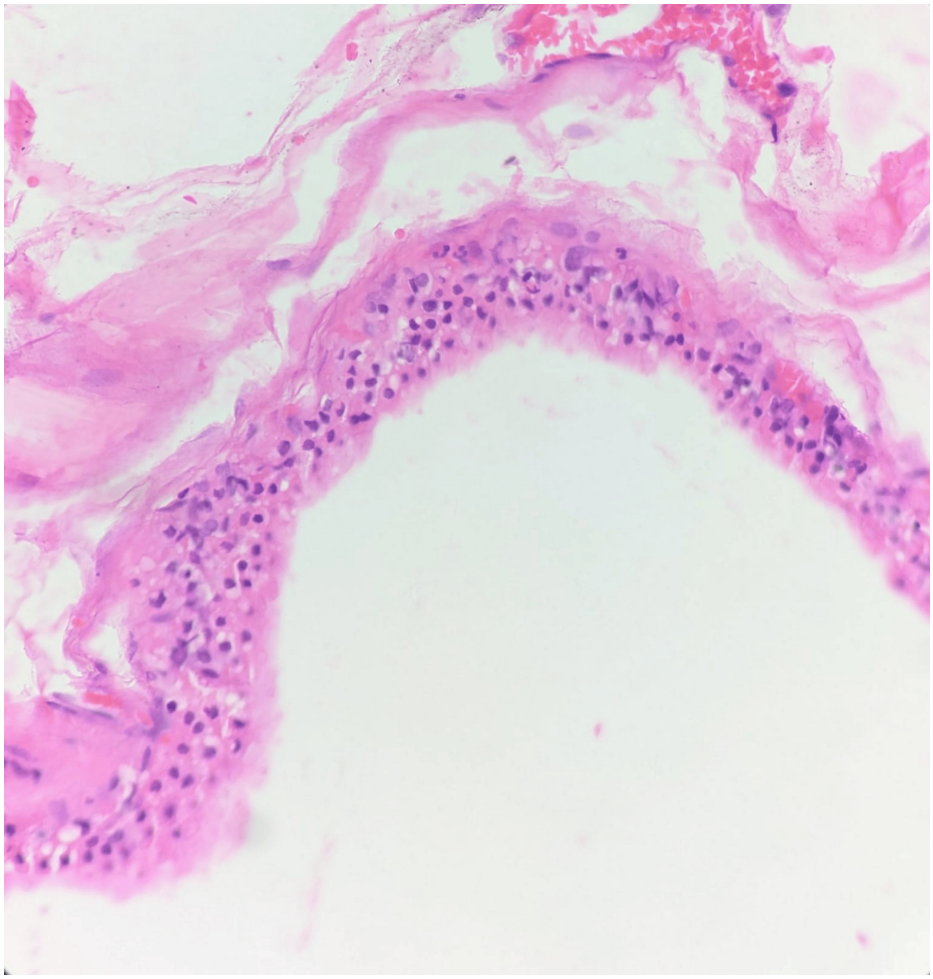


FIGURE 4: The oncocytic lining epithelium shows abundant eosinophilic cytoplasm and eccentrically placed nuclei (haematoxylin and eosin stain, ×40 magnification).

Discussion

The instructive value of this case lies in the systematic discrepancy between the preoperative assessment and the final histopathological diagnosis. Clinical examination estimated the lesion to measure 3 × 2 cm, ultrasonography measured it at 12 × 10 mm, and CECT demonstrated dimensions of 1.8 × 1.5 × 1.8 cm, whereas the excised lesion measured 2 × 2 × 1.5 cm. Palpation through the overlying parenchyma and SMAS may account for the larger clinical estimate. More importantly, no preoperative modality suggested the type of neoplasm ultimately identified. Both imaging reports proposed non-neoplastic entities, although the differential diagnosis of cystic parotid lesions also includes benign and malignant tumours with macrocystic changes [7,8]. Cytology could only establish the presence of atypia, without excluding malignancy. Nevertheless, the appropriate operation was performed because the decision was guided by the extent of diagnostic uncertainty rather than by the most probable diagnosis.

FNAC is the mainstay of preoperative salivary gland assessment but performs substantially less well in cystic than in solid lesions [8]. The needle preferentially samples cyst fluid, resulting in frequently paucicellular smears, while the epithelial cells retrieved may exhibit degenerative and reactive nuclear changes that are difficult to distinguish from genuine neoplastic atypia [8,9]. The findings in this case, comprising acinar cell clusters with an increased nuclear-to-cytoplasmic ratio and mild atypia against a haemorrhagic background despite repeated aspirations, are characteristic of this diagnostic challenge. In practice, the cyst wall, rather than the fluid alone, should be targeted for sampling under ultrasound guidance following decompression. The patient should then be re-examined: a residual mass raises suspicion, while complete disappearance does not exclude cystic mucoepidermoid carcinoma [8,9].

The Milan system classifies salivary gland FNAC findings into six categories, each associated with an implied risk of malignancy (ROM) and a corresponding management recommendation [3]. Category III is a category of last resort, used when the findings are neither convincingly non-neoplastic nor sufficient to establish a

neoplastic diagnosis. It also carries the widest variation in reported ROM. Studies involving parotid masses have reported figures approaching 50%, with FNAC demonstrating low sensitivity but high specificity for malignancy [4-6]. Recommended management includes repeat aspiration or surgical excision, depending on the clinical context. In this patient, repeat aspiration was the less favourable option: multiple aspirations had already been performed, the cystic nature of the lesion increased the likelihood of another non-diagnostic sample, and the patient's age and seven-month history of progressive enlargement raised, rather than lowered, the pretest probability of neoplasia.

Enucleation of a parotid lesion of unknown histology would not have been an appropriate approach. Had the lesion proved to be a low-grade mucoepidermoid carcinoma, a genuine possibility given its cystic morphology and category III cytology, enucleation could have resulted in tumour involvement at the resection margin and necessitated revision surgery, potentially placing the facial nerve at greater risk. Formal superficial parotidectomy with antegrade facial nerve dissection is therefore an appropriate index operation for an indeterminate lesion of the superficial lobe. It is diagnostic, as it allows removal of the entire lesion with assessable margins; curative for benign neoplasms confined to the superficial lobe; and oncologically appropriate as an initial resection should histopathological findings prove unfavourable [10,11]. Preservation of the greater auricular nerve and elevation of the anterior flap in the SMAS plane may reduce the risk of lobular anaesthesia and Frey syndrome, respectively [12,13].

Cystadenoma is a rare benign epithelial salivary gland neoplasm that affects the major and minor salivary glands with broadly similar frequency. The oncocytic variant is characterised by a multicystic architecture lined by oncocytes, which are large cells with abundant granular eosinophilic cytoplasm reflecting mitochondrial accumulation [1,8,14,15]. The gross findings in this case were consistent with this diagnosis. The principal differential diagnosis is Warthin tumour, which is characterised by lymphoid stroma with germinal centres, the most useful distinguishing feature [8,16]. Oncocytoma is typically solid rather than multicystic, while nodular oncocytic hyperplasia is unencapsulated and contains entrapped normal parenchyma. Cystadenocarcinoma and cystic mucoepidermoid carcinoma are distinguished by their malignant features, including invasion and, in the case of mucoepidermoid carcinoma, the presence of mucous, intermediate, and epidermoid cells. The reported wording, "favouring oncocytic cystadenoma," indicates some degree of diagnostic uncertainty. However, the findings support a benign diagnosis, and the lesion was completely excised.

Conclusions

Cystic parotid lesions present a diagnostic challenge, as ultrasonography, computed tomography, and FNAC may all have reduced diagnostic accuracy in this setting. In this patient, all three modalities yielded non-neoplastic or indeterminate findings, and none suggested the final histopathological diagnosis. Nevertheless, the management approach was appropriate because the operative decision was guided by what could not be excluded rather than by the most likely diagnosis. Milan category III cytology in a progressively enlarging parotid mass in a patient in the sixth decade of life should prompt consideration of diagnostic surgery rather than reassurance or repeated sampling. Superficial parotidectomy with antegrade facial nerve dissection provides a definitive diagnosis, is curative for benign disease, and preserves oncological options should malignancy be identified, while carrying a relatively low risk of permanent morbidity.

Additional Information

Author Contributions

All authors have reviewed the final version to be published and agreed to be accountable for all aspects of the work.

Concept and design: Shruthi Ravishankar, Abirami Kailasam, Narayanan Cunnigaiper

Acquisition, analysis, or interpretation of data: Shruthi Ravishankar

Drafting of the manuscript: Shruthi Ravishankar

Critical review of the manuscript for important intellectual content: Shruthi Ravishankar, Abirami Kailasam, Narayanan Cunnigaiper

Supervision: Narayanan Cunnigaiper

Disclosures

Human subjects: Informed consent for treatment and open access publication was obtained or waived by all participants in this study. **Conflicts of interest:** In compliance with the ICMJE uniform disclosure form, all authors declare the following: **Payment/services info:** All authors have declared that no financial support was received from any organization for the submitted work. **Financial relationships:** All authors have declared that they have no financial relationships at present or within the previous three years with any

organizations that might have an interest in the submitted work. **Other relationships:** All authors have declared that there are no other relationships or activities that could appear to have influenced the submitted work.

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