Case Report

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Pathology Section

A Rare Case of Benign Oncocytic Cystadenoma of Parotid Gland

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ABSTRACT

Benign oncocytic cystadenoma of the parotid gland is a rare salivary gland tumour, constituting less than 1% of all salivary gland neoplasms. It is characterised by cystic spaces lined with bilayered oncocytic epithelium, composed of eosinophilic granular cytoplasm due to abundant mitochondria. While these tumours are typically asymptomatic, they can present as painless, slow-growing swellings in the preauricular region, often leading to misdiagnosis as other benign salivary gland tumours. Authors hereby, present the case of a 71-year-old female with recurrent right-sided parotid swelling. Eight years prior, she was diagnosed with low-grade mucoepidermoid carcinoma of the right submandibular gland. One year ago, she underwent total parotidectomy for a recurrent swelling, which was diagnosed histopathologically as a mucus retention cyst. Three months ago, she developed another swelling in the same region, prompting excision and biopsy. Histopathology revealed a cystic lesion lined by bilayered oncocytic epithelium with papillary projections, focal lymphoid aggregates and no evidence of malignancy, confirming oncocytic cystadenoma. Oncocytic cystadenomas present diagnostic challenges due to their rarity and overlap with other salivary gland lesions. Surgical excision is curative, with a low recurrence rate. Awareness of this rare entity is essential for accurate diagnosis and management.

Keywords: Cystic neoplasm, Parotid tumour, Salivary gland

CASE REPORT

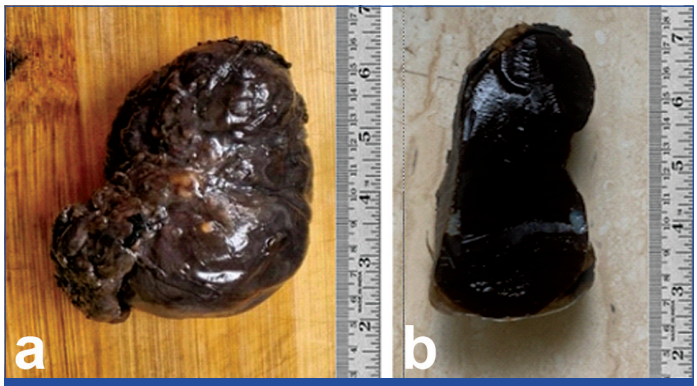
A 71-year-old female presented with recurrent swelling in the right parotid region, noticed over the past three months with a gradual increase in size. The swelling was not associated with pain, facial weakness, or constitutional symptoms such as fever, weight loss, or night sweats. Her medical history revealed a complex course of recurrent salivary gland pathology over the past eight years, involving multiple interventions.

Eight years prior, she had undergone Fine Needle Aspiration Cytology (FNAC) for a swelling in the right submandibular region. The cytology was reported as low-grade mucoepidermoid carcinoma, which led to subsequent surgery. Following this, she remained asymptomatic for several years, with no evidence of local recurrence or metastasis. However, approximately one year ago, she developed a swelling in the right parotid region, prompting further clinical evaluation. Given her prior history of salivary gland neoplasm, a decision was made to proceed with total parotidectomy. The Histopathological Examination {(HPE) Haematoxylin and Eosin (H&E)} of the excised parotid tissue revealed a mucus retention cyst, a benign lesion often attributed to obstructed salivary ducts. This diagnosis was reassuring and the patient was advised to undergo routine follow-up.

Despite the surgical intervention, she again noticed a swelling in the same region three months ago. The swelling was found during examination, presenting as a well-defined, mobile swelling measuring 6×3 cm, with no associated lymphadenopathy. An excision biopsy was performed and the specimen was sent for HPE.

On gross examination, the excised tissue appeared as a single, grey-brown to grey-black soft-tissue mass, measuring 6×3×2.8 cm [Table/Fig-1a]. Upon sectioning, the cut surface revealed a thin-walled cyst filled with brownish viscous material, suggesting a cystic salivary gland lesion [Table/Fig-1b]. No solid nodular growth or haemorrhagic/necrotic areas were observed.

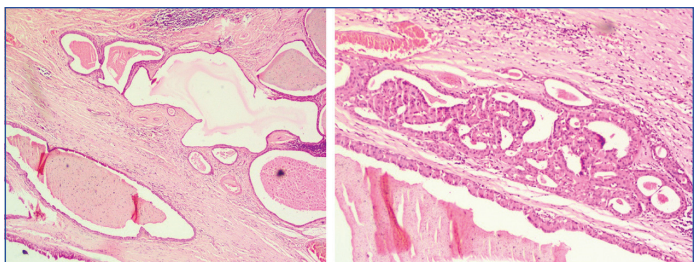
On microscopy, sections revealed a thin fibrocollagenous cyst wall lined by bilayered oncocytic epithelium [Table/Fig-2]. The cyst wall in some regions showed multiple smaller cysts, also lined by similar oncocytic epithelium, interspersed with focal lymphoid aggregates, suggesting a reactive inflammatory component [Table/Fig-3]. The



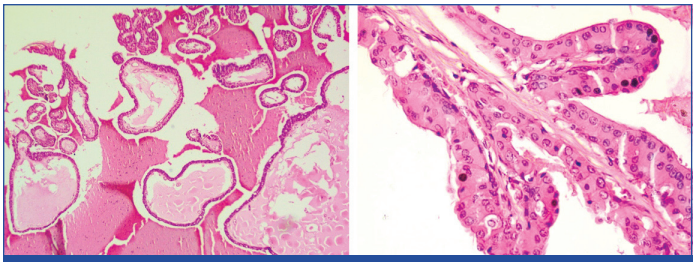
[Table/Fig-1]: a) Single grey-brown to grey-black soft tissue mass measuring 6×3×2.8 cm. External surface-smooth, congested; b) Cut surface- showed thin-walled cyst filled with brownish viscous material.

oncocytic cells displayed moderate eosinophilic granular cytoplasm, with uniform round to slightly oval nuclei and evidence of reverse polarity. In focal areas, the epithelium formed small papillary projections containing slender fibrous cores, lined by similar bilayered oncocytic epithelium [Table/Fig-4]. These papillary structures were delicate and well-formed, without any atypia [Table/Fig-5].

A distinct aggregation of foamy macrophages and lymphocytes was identified in one area, possibly indicating prior localised inflammation or a response to cystic degeneration. There were no mitotic figures, necrosis, atypia, or cellular pleomorphism, ruling out malignant



[Table/Fig-2]: Multiple cysts lined by oncocytic epithelium with fibrocollagenous cyst wall (H&E x40). **[Table/Fig-3]:** Small cysts lined by oncocytic epithelium with reverse polarity and focal lymphoid aggregates in the wall (H&E x100). (Images from left to right)



[Table/Fig-4]: Focal areas show papillary projections lined by bilayered oncocytic epithelium with hyalinised cores (H&E x100).
[Table/Fig-5]: High-power appearance of papillary projections with slender fibrous core lined by bilayered oncocytic epithelium with moderate eosinophilic Cytoplasm. No atypia noted (H&E x400). (Images from left to right)

transformation or recurrence of the previously diagnosed low-grade mucoepidermoid carcinoma.

Based on the gross and histopathological findings, the lesion was diagnosed as a benign oncocytic cystadenoma of the parotid gland. The patient returned for follow-up six months later and upon examination, there was no residual swelling or any lymph node enlargement.

DISCUSSION

Oncocytic cystadenoma is a rare benign neoplasm of the salivary glands, with a strong predilection for the parotid gland [1,2]. It is characterised by cystic spaces lined by bilayered oncocytic epithelium [3-5]. While oncocytic metaplasia is a common reactive change in aging salivary gland tissue, the formation of a distinct cystic neoplasm is uncommon and is often confused with other benign and malignant cystic salivary gland lesions [6]. These tumours are rare, accounting for less than 1% of all salivary gland neoplasms [7]. Despite their benign nature, the rarity and unique histological features of oncocytic cystadenomas make them a subject of clinical interest [8].

The clinical presentation of oncocytic cystadenoma is often nonspecific, as patients usually present with a slow-growing, painless mass persisting for years without any overt symptoms [7-9]. In the present case, the patient's history of multiple recurrent cystic salivary gland lesions, including a previous diagnosis of mucus retention cyst, complicated the diagnostic evaluation [2]. The bilayered oncocytic epithelium, papillary projections and multiple smaller cysts with focal lymphoid aggregates helped to differentiate oncocytic cystadenoma from other salivary gland lesions [10,11].

Histologically, oncocytic cystadenomas comprise multicystic spaces with a thin fibrocollagenous cyst wall [2]. The epithelial lining consists of oncocytic cells exhibiting moderate eosinophilic granular cytoplasm, reflecting their high mitochondrial content [3]. This bilayered epithelium typically comprises an inner cuboidal or columnar oncocytic layer, with a flattened basal layer, sometimes interspersed with lymphoid aggregates [6]. In some cases, papillary infoldings can be seen, as was observed in this case, featuring slender fibrovascular cores lined by oncocytic epithelium [9]. This feature may mimic oncocytic papillary cystadenoma or Warthin's tumour, necessitating careful histological evaluation [5]. The presence of foamy macrophages and chronic inflammatory infiltrates, as seen in this case, suggests chronic inflammation or prior cystic rupture, further supporting the diagnosis [11].

Importantly, there is no increased mitotic activity, nuclear pleomorphism, necrosis, or invasive growth, thus ruling out malignancies such as malignant oncocytoma, oncocytic carcinoma, or recurrent mucoepidermoid carcinoma [10]. The cystic spaces may contain intraluminal crystalloids, although this feature is not exclusive to oncocytic cystadenomas, adding to the diagnostic complexity [7].

Diagnosing oncocytic cystadenoma can be challenging [5]. Significant differentials include mucus retention cysts, which lack the oncocytic epithelium [3,4] and are lined by flattened epithelium, with the lumen filled with mucin and no papillary projections or oncocytic metaplasia [6]. Warthin's tumour is another oncocytic lesion that often presents as a multicystic tumour with lymphoid stroma [7]. These tumours have a double-layered oncocytic epithelium resting on a dense lymphoid background, often accompanied by germinal centre formation [4]. Papillary oncocytic cystadenomas exhibit extensive papillary projections with tall columnar oncocytic epithelium and pronounced nuclear atypia [5]. This case demonstrated only focal papillary structures rather than the prominent papillary fronds typical of oncocytic papillary cystadenomas [9].

The pathogenesis of oncocytic cystadenomas remains unclear, but some studies suggest that they arise due to chronic inflammation, salivary gland ductal obstruction, or oncocytic metaplasia in ageing glandular tissue [3]. These tumours predominantly occur in elderly patients, which aligns with this patient's age of 71 years. This rare salivary gland tumour, although benign, is recognised for its tendency to recur, which corresponds with the patient's history of multiple cystic salivary gland lesions. As seen in the present case, the recurrent nature of salivary gland cystic lesions raises concerns about the risk of regrowth following incomplete excision [1,5,7]. While oncocytic cystadenomas are benign, they may recur if remnant cystic epithelium persists after surgical excision [4]. Most studies and literature indicate that enucleation and inadequate resection are the main causes of recurrent swelling. Recurrent swelling after total parotidectomy, although rare, could be due to intraoperative rupture or tumour spill, or surgical dissection close to the tumour/capsule leading to microscopic spillage of the cells [10].

On imaging studies, including ultrasound and Magnetic Resonance Imaging (MRI), they typically appear as well-circumscribed cystic lesions [2,12]. FNAC may reveal oncocytic cells, but distinguishing between various oncocytic lesions can be challenging [7-9].

Surgical excision remains the treatment of choice for oncocytic cystadenomas [9]. Complete removal typically results in a favourable prognosis with a low risk of recurrence [7,8].

CONCLUSION(S)

Oncocytic cystadenoma of the parotid gland is a rare benign tumour that poses diagnostic challenges due to its overlapping features with other salivary gland lesions. A thorough understanding of its histopathological and immunohistochemical characteristics is essential for accurate diagnosis and appropriate management. Further research is needed to elucidate the pathogenesis of these tumours and to establish standardised diagnostic and treatment protocols.

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