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Infarcted Warthin's Tumour of the Parotid Gland: A Case Report

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Introduction

Warthin's tumor is the second most common salivary gland neoplasm [1]. It usually occurs as a benign tumor in the superficial tail of the parotid gland [1-7]. Tobacco smoking has been associated with an 8-fold increase of Warthin's tumor incidence [5]. Other risk factors include males between 50 and 70 years, Epstein-Barr viral infection [1,5]. Patients typically present with an asymptomatic, slow-growing, hard, and fluctuant mass [1,5]. Warthin's tumor is usually unilateral, however, up to 14% of cases present with bilateral masses. Multicentricity and multifocal lesions are found in about 20% of cases [1,5]. Warthin's tumor is also known as Papillary Cystadenoma Lymphomatosum. This name reflects the histopathological features of the tumor, which include papillary epithelial projections, cystic spaces lined by oncolytic epithelium, and a dense lymphoid stroma [1,2]. Infarction is a rare occurrence in Warthin's tumor and is estimated to occur in 6% to 7% of cases [2,8].

Infarcted tumors often present with increasing pain, rapid enlargement, and occasional fevers, which reflects tissue necrosis and inflammation [2,4]. Infarction may trigger metaplastic changes that can mimic malignancy and complicate diagnosis [2,4]. Hence, during fine needle aspiration (FNA) it is crucial to be meticulous in diagnosing Warthin's tumor and to distinguish it from malignancy [6]. FNA has been the gold standard for diagnosing Warthin's tumor with a substantial sensitivity and specificity of 93% and 95% respectively [6]. If left untreated, multiple complications may arise including acute parotiditis, massive enlargement with cosmetic deformity, and in rare cases malignancy [1,5]. Therefore, early identification and surgical excision remain as the standard management method to prevent the progression of Warthin's tumour [1,5].

Case Presentation

A 49-year-old male patient presented with a long-standing right parotid mass that had been stable initially. Two weeks prior to presentation, he noted a sudden increase in size associated with severe, agonizing pain. The patient had a history of chronic smoking.

Imaging

Ultrasound examination revealed three oval-shaped hypoechoic lesions with cystic components in the right parotid gland. The largest lesion was at the inferior margin, and it measured 3.8 × 1.8 × 2.5 cm. An adjacent lesion measured 1.8 × 1.5 × 2.3 cm, and the smallest lesion measured 0.9 × 0.5 cm. Internal vascular flow was preserved, and the left parotid gland appeared unremarkable. Computed tomography (Figure 1a,b) of the neck demonstrated multiple lesions in the right parotid gland, with the largest measuring 3.5 cm and one intraglandular lesion measuring 2.5 cm. All lesions showed heterogeneous enhancement, suggesting necrosis. Multiple cervical lymph nodes were noted, likely reactive, and the palatine tonsils were enlarged. Other findings were consistent with changes related to chronic smoking.

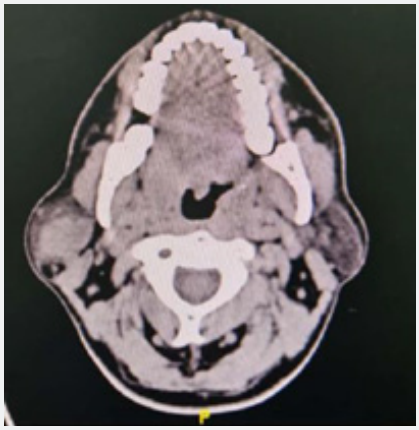

Figure 1a: Horizontal Plane.

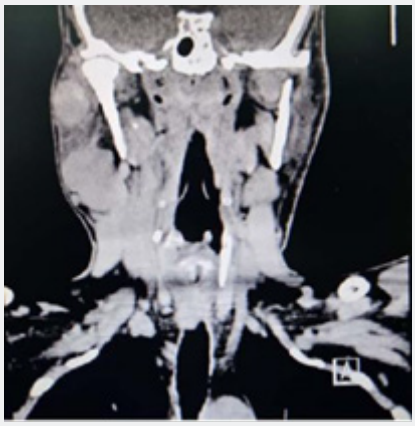

Figure 1b: Coronal Plane.

Cytopathology

FNA of the largest lesion revealed monolayered sheets and singly dispersed oncolytic cells associated with lymphoid tissue and proteinaceous debris, consistent with Warthin's tumor.

Surgical Intervention

A modified Blair's incision was performed, and a right superficial parotidectomy was undertaken. Multicentric masses from the tail, mid, and upper portions of the parotid gland were excised. A drain was placed and secured, and postoperative management included a seven-day course of Augmentin.

Histopathology

Gross examination revealed three masses weighing a total of 53.0 g. The largest mass measured 7.0 × 4.5 × 5.0 cm and was lobulated; the other masses measured 2.5 × 2.5 × 1.5 cm and 2.0 × 1.0 × 1.0 cm. Cut surfaces were yellow-white and hemorrhagic. Sections were processed in nine cassettes (cassettes 1-2: small pieces; 3-9: larger pieces). Microscopic evaluation (Figure 2a-c) confirmed Warthin's tumor with areas of infarction and fibrosis. Three benign intracarotid lymph nodes were also identified. The patient's postoperative course was uneventful.

Discussion

Warthin's tumor is typically a slow-growing, painless parotid mass seen in older males with a history of smoking [1,5]. The patient of this case, a 49-year-old male, presented with a sudden increase in tumor size and severe pain over the two weeks prior to surgery. Six weeks prior to excision, FNA confirmed the diagnosis of Warthin's tumor. Similarly, other cases have of infarcted Warthin's tumor were strongly associated with rapid tumor enlargement and agonizing pain. A similar case was reported by Yorita et al., involving a 69-year-old Japanese man who presented with a slow-growing right buccal mass of one year's duration. The lesion showed rapid enlargement and severe pain one month before surgery, resembling the clinical course in the present case [2]. Histopathological examination confirmed Warthin's tumor with areas of coagulative necrosis and squamous and mucinous non-oncolytic epithelial metaplasia [2]. The authors emphasized that the absence of MAML2 gene rearrangement helped distinguish reactive metaplasia from mucoepidermoid carcinoma, thereby preventing diagnostic error of malignancy [2].

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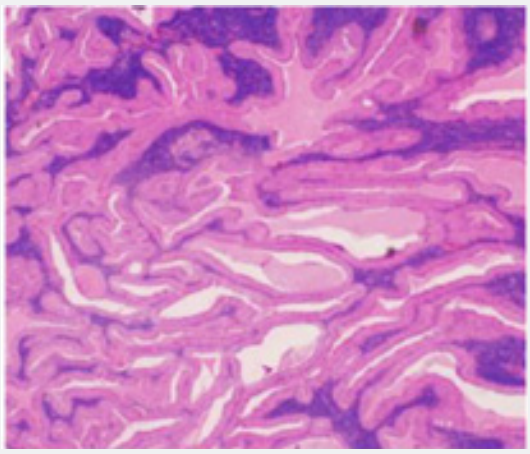

Figure 2a: Papillary cystadenoma lymphomatosum (Warton's tumor), characterized by papillary cystic structures lined by oncotic cells forming two layers in a lymphoid background.

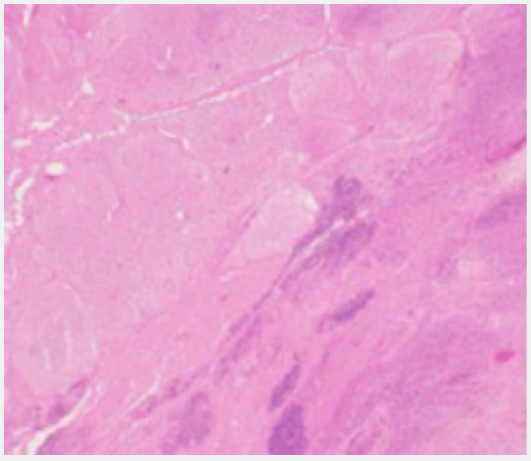

Figure 2b: This image shows an outline of the papillary structures of Wharton's tumor, which underwent necrosis in the form of pink ghost cells (anucleated cells). Remaining lymphoid stroma can be seen.

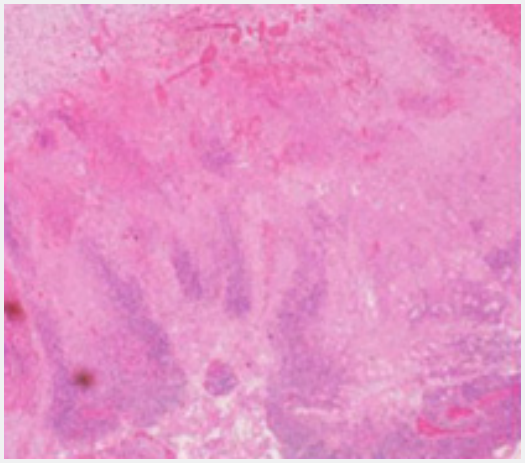

Figure 2c: Areas of hemorrhage and congestion with foci of inflammation.

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Jha and Gupta reported a case of a 54-year-old male with multiple comorbidities and a history of chronic smoking who presented with a left parotid swelling, slowly growing for the past year [8]. Histological evaluation revealed inflammatory cells, oncolytic sheets, squamous metaplasia, and cystic macrophages with no cytological atypia [8]. The authors attributed the infarction and metaplastic change to a later revealed history of repeated FNAs [8]. They emphasized that the WHO (2022) classification recognizes Warthin tumors with squamous metaplasia as a distinct benign entity, which has aided to distinguish them from mucoepidermoid and squamous cell carcinomas [8]. Therefore, key distinguishing features of carcinoma must be assessed: squamous cell carcinoma shows cytologic atypia, increased mitoses, and invasive growth, whereas mucoepidermoid carcinoma is characterized by mucin-producing epithelial cells [8].

Conclusion

This case presents a typical patient of infarcted Warthin's tumor with features of sudden enlargement and pain in a previously slow-growing parotid mass. It is essential to be meticulous in the identification of this benign entity to prevent misdiagnosis of cancer. Hence, histopathological evaluation, preoperative and postoperative, becomes highly crucial in confirming the diagnosis and guiding appropriate management.

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