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Case Report

Küttner's tumour of the parotid gland- an uncommon entity

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ABSTRACT

Chronic sclerosing sialadenitis also known as 'Küttner's tumour' (KT) is a benign chronic inflammatory condition first described by Küttner, clinically presenting as a firm swelling, often simulating a neoplasm. The condition is usually misrecognised and the correct diagnosis is made after the excision of the gland. Recent evidence suggests that Küttner's tumor may be characterized by IgG4-related inflammation. An accurate preoperative diagnosis can be made by awareness of the condition, imaging and histopathopathology features, thereby potentially avoiding excision of an otherwise benign condition. Küttner's tumour most commonly affects submandibular gland. Küttner's tumour of the parotid and minor salivary glands are extremely rare. We report a case of a 69 year old man with this interesting and uncommon entity affecting the parotid gland.

1. Introduction

Küttner tumor (KT) is a chronic sclerosing benign inflammatory condition commonly affecting salivary glands in middle aged adults. Submandibular gland is the most common location for KT. KT of the parotid and minor salivary glands are extremely rare.[1,2]

2. Case Report

A 69-year-old man presented with infraauricular swelling since 10 years showing sudden increase in the size of the swelling and pain since one month. There was no history of trauma, alteration in the taste or dryness of the mouth. On local examination, a firm non- tender lesion was present in the right infraauricular area. There were no palpable local lymph nodes. His facial nerve function was intact. Systemic examination and blood investigations were normal.

Ultrasonography (USG) of right parotid region (Figure 1) showed diffusely heterogeneous solid cystic lesion in the superficial lobe of right parotid gland with prominent vascularity. USG guided fine needle aspiration cytology showed features consistent with acute inflammation.

Contrast enhanced computed tomography (CT) of the neck (Figure 2) showed large ill- defined hypodense heterogeneously enhancing solid cystic lesion in the superficial lobe of right parotid gland measuring ~5.3 x 4.8 x 5.6cm. Cystic component showed multiple enhancing internal septations. There was no evidence of calcification or hemorrhage within the lesion. Fat plane with the right sternocleidomastoid muscle, pterygoid and masseter muscles were preserved. Right parapharyngeal space was normal. There were no significant lymph nodes. Features were suggestive of acute sialadenitis. However histopathological examination (HPE) correlation was recommended to exclude the possibility of malignancy.

Patient underwent right total conservative parotidectomy under general anesthesia. Microscopic examination (Figure 3) showed salivary gland ducts and acini, extensive

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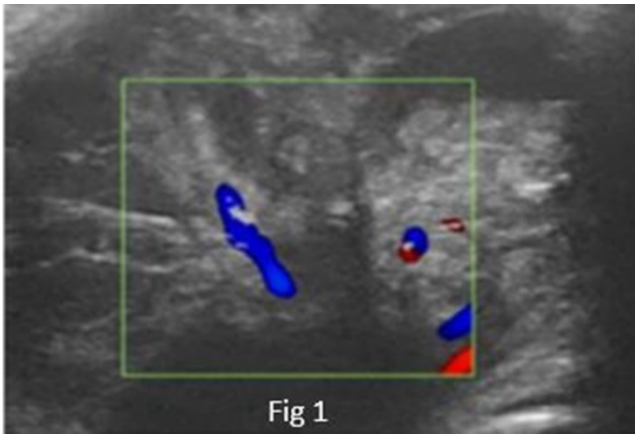

Fig. 1: Ultrasonography of right parotid region shows diffusely heterogeneous solid cystic lesion inthe superficial lobe of right parotid gland with prominent vascularity

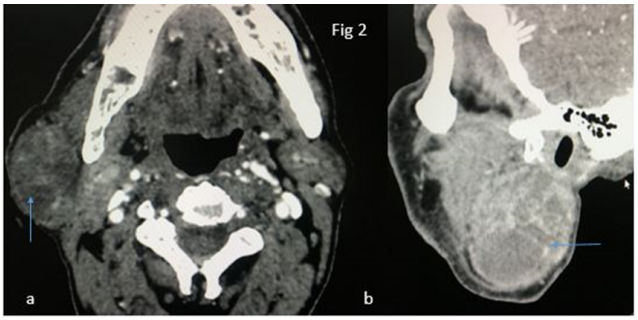

Fig. 2: CECT neck Axial (**a**) and Coronal (**b**) views in venous phase shows ill- defined heterogeneously enhancing solid cystic lesion (asterisk) in the superficial lobe of right parotid gland

areas of necrosis, hemorrhage and dense acute inflammatory cell infiltrate. Ductal dilatation, squamous metaplasia of the ductal epithelium, periductal fibrosis was seen. Acinar atrophy was seen with acini lined by benign looking epithelium. Extensive areas of hyalinisation and fibrosis was seen. Dense lymphoplasmacytic infiltrate was seen in the stroma. Lymphoid tissue was seen arranged in the form of lymphoid follicles with germinal centre formation. There was no evidence of malignancy. All the features were suggestive of KT with superadded acute inflammation. No recurrence was noted in the subsequent follow up.

3. Discussion

KT is a benign inflammatory condition most commonly involving submandibular gland in middle aged adults. However there are only a few publications in the literature.[1,2] KT of the parotid gland is very rare.[2]

Etiology of KT is not well understood. Sialolithiasis, secretory dysfunction with ductal inspissation, duct abnormalities, infectious agents, autoimmune reaction

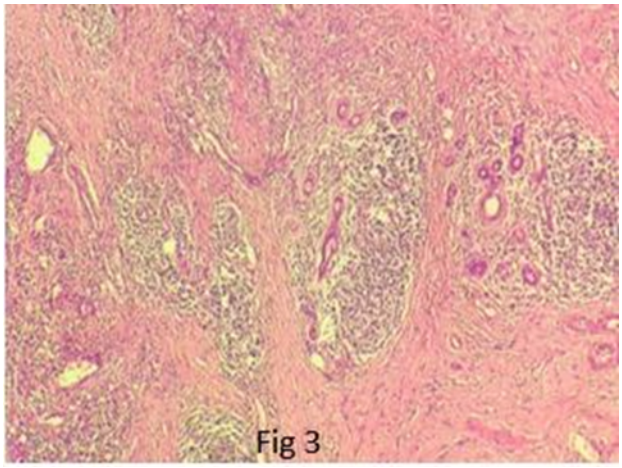

Fig. 3: Medium power view of the tumor shows residual salivary ducts with areas of chronic inflammation including follicle formation and peripheral fibrosis

have been suggested as possible mechanisms.[3] Recent studies have shown that KT is a part of the spectrum of IgG4-related disease.[4]

On Ultrasonography (USG), diffuse form shows multiple hypoechoic lesions with a heterogeneous background associated with duct dilatation, calculi and prominent vascularity. Focal form appears as heterogenous hypoechoic mass with radially oriented vessels.[5,6]

CT should be the first line of investigation if the mass appears to be related to sialolithiasis. Imaging findings are not specific and not a good predictor of histological diagnosis. Solid homogenous attenuation or heterogenous appearance with mixed solid cystic areas can be seen. Enhancement pattern is variable on imaging. MRI is especially useful for assessing status of ductal system. Relationship of the inflammatory mass to the floor of the mouth is important for surgical approach.[6] On MRI, diffusely involved gland shows mild T2 hyperintensity with T1 intermediate signal intensity.[4,7] MRI signal intensities may help distinguish KT from benign tumors as mean signal intensity ratios and the mean apparent diffusion coefficient values for KT and malignant tumors were lower than those of benign tumors.[7]

Diffusely infiltrative pattern points towards KT, but aggressive salivary malignant tumors can also show infiltration. Malignant tumors can show metastatic lymphadenopathy, but KT usually does not.[7]

Differential diagnosis includes sialadenitis, Mikulicz's disease (MD), Sjogren syndrome (SS), and neoplasms of salivary glands.[5] KT, MS, SS should be considered in bilateral lesions. They can be differentiated by clinical features and serology. HPE shows the presence of IgG4-positive plasma cell infiltration in KT and MD but not in SS.[8]

A definite diagnosis is made by HPE. The cytological features are paucicellular to moderately cellular, scattered ductules enveloped by collagen bundles or a lymphoplasmacytic infiltrate, isolated fragments of fibrous stroma and scanty acini.[4]

Sialadenectomy is generally required for symptomatic KT without extra-salivary involvement.[5] However, steroids can be helpful in regression of KT with extrasalivary involvement or those who refuse surgery. Observation should suffice for asymptomatic KT in whom the preoperative diagnosis is unequivocal.[8] Prognosis is good and do not tend to recur.[5]

4. Conclusion

KT is an under-recognised entity. An accurate preoperative diagnosis should be made by awareness of clinicoradiological and pathological features, thereby potentially avoiding operative excision of an otherwise benign condition. KT may also be a manifestation of a systemic IgG4-related disease and an autoimmune process should be excluded. We present a rare case of KT affecting parotid gland.

5. Source of Funding

None.

6. Conflict of Interest

None.

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