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Case Report

A rare case of sclerosing polycystic adenosis of parotid gland

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ABSTRACT

The parotid gland tumors are classified according to their morphological and histological patterns. The most common site of presentation of a salivary neoplasm is the parotid gland. Sclerosing polycystic adenosis (SPA) is a rare disease of the salivary glands, first described by Smith et al at 1996. We report a case of 61 year gentleman admitted in KVG Medical college and hospital with complains of swelling in right side of face since 6 months. Local examination of right parotid region showed solitary swelling with ipsilateral mandibular lymphadenopathy. EBV profile was positive. CT head and neck revealed an enlarged right sided parotid gland resembling features of Pleomorphic adenoma. Fine needle aspiration of gland showed features of chronic sialo-adenitis. A provisional diagnosis of Pleomorphic Adenoma was made. Patient underwent superficial parotidectomy. Histopathology of the operated specimen had features suggestive of Sclerosing polycystic adenosis of parotid gland. Patient was followed up for a period of one year and he had no recurrence. To conclude, Sclerosing polycystic adenosis of the parotid gland is a rare benign salivary gland lesion with histologic analogies to sclerosing adenosis of the mammary gland. Complete surgical excision is the reference treatment, to reduce the risk of recurrence and/or evolution.

Keywords: Parotid, polycystic sclerosing adenosis, EBV, pleomorphic adenoma

INTRODUCTION

The parotid gland tumors are classified according to their morphological and histological patterns. The most common site of presentation of a salivary neoplasm is the parotid gland. The 85% of these neoplasms are benign tumors, being the pleomorphic adenoma the most frequent type.[1] Sclerosing polycystic adenosis (SPA) is a rare disease of the salivary glands, first described by Smith et al. at 1996.[2]

The main location of the SPA is the parotid gland, but there are reported cases in the submandibular and accessory salivary glands too.[2] The SPA is similar to the fibrocystic changes, sclerosing adenosis and adenoid tumors of the mammary gland.[3,4]

CASE REPORT

We report a case of 61 year old gentleman admitted in KVG Medical college and hospital with complains of swelling in right side of face since 6 months. General physical examination was unremarkable. Local examination of right parotid region showed Solitary swelling of size 8×5 cms, obliterating the hollow between mandible and mastoid process, raising the right ear lobule, globular in shape, smooth surface. The swelling is firm to hard in consistency, non tender, on contracting masseter, there was restricted movement of the swelling. On bi-digital exam, deep lobe not palpable. Opening of Stenson's duct normal. A solitary submandibular LN of size 1x1cm palpable, rounded borders, firm, non tender, mobile. Systemic examination was unremarkable. All routine

investigations were with in normal limit, EBV profile was positive.

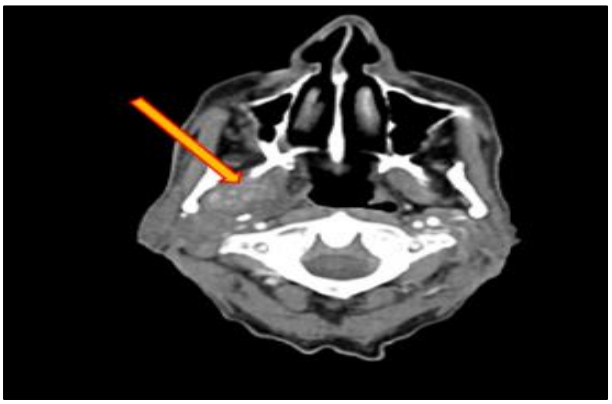

Figure 1: CT Head and neck showing enlarged right parotid gland with mandibular lymphadenopathy.

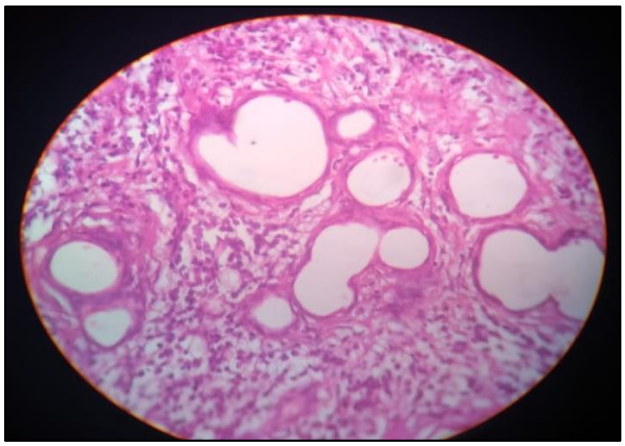

Figure 2: The microscopic study reveal a parotid gland tissue with periductal inflammation and fibrosis with intraluminal cellular debris and polymorphic nuclear cells.

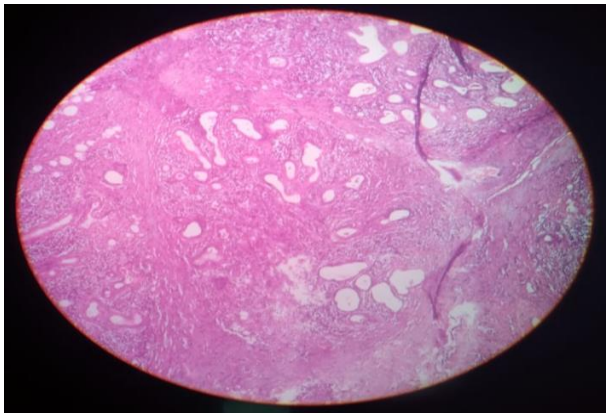

Figure 3: The glandular parenchyma appears in continuity with a dense-hyaline connective tissue with undefined borders that forms a sclerosing-hyaline stroma mass with inflammatory cells and interposed myoepithelial elements.

Chest X-ray was normal. CT head and neck revealed an enlarged right sided parotid gland with well-defined margin, a smooth border, an inhomogeneous aspect and intermediate or high signal intensity resembling Pleomorphic adenoma. Fine needle aspiration of gland showed features suggestive of chronic sialo-adenitis. A provisional diagnosis of Pleomorphic Adenoma was made. Patient underwent superficial parotidectomy under general anaesthesia with no post-operative complications. Histopathology of the operated specimen had features suggestive of Sclerosing Polycystic Adenosis of Parotid Gland. Patient was followed up for a period of one year and he had no recurrence.

DISCUSSION

There have been about 51 cases of sclerosing polycystic adenosis (SPA) in the salivary glands published. All cases of SPA have been unilateral like this case report.[5] The clinical picture is nonspecific, presenting in most cases as a progressively growing intra-parotid asymptomatic mass, although in some cases the tumor has been associated with pain and/or paresthesias in the region.[2,6] The pathogenesis of SPA is unknown, although there has been a possible association with the Epstein–Barr virus.[7] Microscopically the SPA consists of lobes circum- scribed by hyaline connective tissue. Moreover, within the fibrous tissue there is hyperplastic ductal and acinar epithelium with no differentiation.[7,8] The differential diagnosis should include acinar cell carcinoma, sclerosing sialadenitis, salivary ductal carcinoma and adenocarcinoma.[6] The treatment in most cases consists of excision of the tumor with a superficial parotidectomy with adequate surgical margins and facial nerve sparing, which has demonstrated a prolonged survival.[2,4] Recurrences are rare and usually due to incomplete surgical excision or occult multifocal disease rather than a real recurrence.

CONCLUSION

Sclerosing polycystic adenosis of the parotid gland is a rare benign salivary gland lesion with histologic analogies to sclerosing adenosis of the mammary gland. Complete surgical excision is the reference treatment, to reduce the risk of recurrence and/or evolution.

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