Abstract

Educational Objectives: At the conclusion of this presentation, participants should be able to diagnose and treat sclerosing polycystic adenosis (SPA) of the salivary gland.

Objectives:
- Discuss a case of sclerosing polycystic adenosis of the salivary gland
- Review the literature on sclerosing polycystic adenosis

Study Design: A single case report of a patient with sclerosing polycystic adenosis is described.

Methods: A 74 year old female presented to head and neck surgery with a 4 year history of a slowly growing, painful left sided neck mass. Two years prior to presentation, patient received a fine needle aspiration (FNA) which was negative for malignancy. The mass persisted, and continued to enlarge. At presentation, the patient had a firm 3.5cm mass in the tail of the left parotid. An FNA suggested well differentiated adenocarcinoma.

Results: Patient received a superficial parotidectomy and super selective neck dissection (level 2). Final pathology revealed a 3.5cm well circumscribed tumor consistent with sclerosing polycystic adenosis.

Conclusions: Sclerosing polycystic adenosis is a rare inflammatory process that causes fibrocystic changes in the salivary gland. Apocrine like metaplasia and epithelial atypia are common pathological features. To our knowledge, a total of 51 cases have been described in the English literature.

Introduction

Sclerosing Polycystic Adenosis is a rare, reactive, inflammatory lesion of the salivary glands, resulting in fibrocystic changes and adenosis, similar to that of the mammary glands. Lesions present as slow growing masses in salivary gland parenchyma. They are discrete, pale, and rubbery nodules. The tumors are not encapsulated but are well defined. Pathologically, they display dense sclerotic lobules and cystic change with hyalinized collagen separation. Apocrine like metaplasia, epithelial atypia and ductal, acinar hyperplasia are commonly observed. A distinguishing feature of this lesion is focal cystic spaces within the fibrotic stroma. Most of the observed cases occur in the parotid gland. Our study reports a typical case of SPA occurring in the parotid gland.

Case

A 74 year old female presented to head and neck surgery at Kaiser Permanente Medical Center at Oakland with a 4 year history of a growing, painful left sided neck mass. Two years prior to presentation, the patient received a fine needle aspiration (FNA) which was negative for malignancy. The mass persisted and continued to enlarge. The patient now had a firm 3.5cm mass in the tail of the left parotid with no overlying erythema. An FNA suggested a well differentiated adenocarcinoma. The patient was scheduled for surgery and a superficial parotidectomy and a selective neck dissection (level 2) was performed. Final pathology revealed a 3.5cm well circumscribed tumor consistent with sclerosing polycystic adenosis (SPA).

Discussion

Sclerosing polycystic adenosis is a rare, benign tumor of the salivary glands. This entity was first described in 1996. About 80% of SPA present in the major salivary glands, specifically the parotid gland. However, cases have been observed in the minor salivary glands of the nasal septum, buccal mucosa, hard palate, floor of mouth, and retromolar pad. SPA has also been reported in the lacrimal gland.

SPA is equally common in men and women and there is a wide age distribution in reported cases.

Usually, parotid SPA are deep-seated, slow growing round palpable masses. Pain and tenderness may or may not be present. The masses are multinodular with cysts of 1-2 mm in diameter. SPA may be multifocal.

Histologically, SPA displays acinar cells with robust eosinophilic structures similar to zymogen granules. Ductal epithelial atypia is common and epithelial cells exhibit various cells of apocrine, foamy, vacuolated and mucinous nature. The lobular architecture typically includes atypical nests of myoepithelial cells. However, infiltrative carcinoma growth pattern does not occur.

SPA is considered benign. However, ductal carcinoma in situ has been reported in 1 case. Most cases of SPA are treated with localized surgical resection with clear margins. Recurrence has been reported in up to one third of cases. Recurrence generally occurs because of inadequate surgical resection and the multifocality of SPA. Within the literature, there have been no reported deaths or metastases attributed to SPA.

There is a high chance of misdiagnosis because of the rarity of the disease if clinicians and pathologists are unfamiliar with SPA. For example, this case was initially diagnosed as a well differentiated adenocarcinoma. Differential diagnosis includes pleomorphic adenoma, benign polycystic disease, sclerosing sialoadenitis, and malignant glandular neoplasias such as mucoepidermoid carcinoma, acinic cell carcinoma, adenocarcinoma NOS and salivary duct carcinoma.

The case presented in this study was a 3.5 cm tumor in the tail of the parotid gland. The presentation in the parotid gland, along with the histological appearance of cysts and lobular architecture with collagen separation are characteristic of SPA.

Conclusion

Sclerosing polycystic adenosis is a rare inflammatory process that causes fibrocystic changes in the salivary gland. Apocrine like metaplasia as well as epithelial atypia are common features. To our knowledge, a total of 51 cases have been described in the English literature.

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