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CASE REPORT
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Coexistence of sclerosing polycystic adenosis and dysgenetic polycystic disease of parotid, Report of a case

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Abstract

Sclerosing polycystic adenosis (SPA) is a rare benign salivary gland lesion. Dysgenetic polycystic disease (DPD), which is a histologically similar lesion, may cause a lattice-like gross appearance with bilateral enlargement of the entire salivary glands. In this report, we present a case of SPA in the right parotid and coexistent DPD involving the both parotid.

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Full Text

Introduction

Sclerosing polycystic adenosis (SPA) is a rare benign salivary gland lesion which is characterized by morphological similarity to fibrocystic changes, sclerosing adenosis, and intraductal epithelial proliferations of the breast. SPA occurs over a wide age spectrum and is mostly unifocal.[1],[2] The lesions are well circumscribed and are composed of densely sclerotic lobules with prominent cystic, metaplastic, and hyperplastic changes in the ductal and acinar elements. The differential diagnosis of SPA includes a variety of non-neoplastic and neoplastic salivary gland lesions also including dysgenetic polycystic disease (DPD).[3] The presence of cystically dilated ducts and apocrine metaplasia are the most frequent shared features.

DPD is a congenital disease, frequently seen in females and is characterized by multiple epithelial-lined cystic spaces arising from acini and intercalated ducts.[4],[5]

In this paper, we present a coexistence of SPA of the parotid with bilateral DPD which were not documented previously.

Case Report

A 35-year-old woman applied to hospital with a slowly growing, painless, hard parotid mass, noticed recently without other meaningful clinical symptoms. Clinical history revealed that she had been succesfully treated for acute lymphoblastic leukemia with chemotheraphy and radiotheraphy to the head of possible brain involvement. Then total thyroidectomy was performed for thyroid papillary carcinoma and this was followed by radioactive iodine treatment.

Ultrasonographic and magnetic resonance imaging [Figure 1] examinations revealed a solid, hypoechoic lesion 2.3 cm in largest diameter within the deep posterior part of the irregular looking right parotid gland. In magnetic resonance imaging small cystic changes and calcified densities within both enlarged parotid glands [Figure 1]. Additionally, small microcalcifications were identified in the right and left parotid parenchyma. A right total parotidectomy was performed.{Figure 1}

Grossly, the parotidectomy specimen measured 6 × 3 × 2.5 cm in size. The cut surface showed a firm, well demarcated nodular lesion 2.3 cm in it's largest diameter with a rubber consistency. The salivary gland around the mass had a sponge like parenchyma and lattice-like appearance.

Microscopically; the solitary lesion was a well encapsulated nodule consisting of proliferating ducts in a sclerotic collagenous stroma [Figure 2]. The duct proliferations were composed of sclerosing adenosis-like closely packed areas, cribriform structures, solid islands, cystic changes and apocrine metaplasia. It was diagnosed as a representative example of SPA. Histologically, similar to gross apparence the salivary gland parenchyma outside the lesion demonstrated multiple lobules of cystic spaces in varying size with a lattice-like appearance. The cystic changes involved the acini more frequently than the ducts and were lined by cuboidal or flattened epithelial cells with vacuolated cytoplasm reminiscent of lactational change of the breast. Cystic ducts of the DPD communicates with the normal lobular structures of the salivary glands. The lumina of these cysts contained pale eosinophilic secretion, globular dens spherules with concentric lamellation, mineralization, large sialoliths, and macrophages [Figure 3]. This coexistent widespread cystic change was consistent with a diagnosis of DPD. Abdominal MRI also revealed splenic subcapsular cysts of varying size.{Figure 2}{Figure 3}

Immunohistochemically there was diffuse CK 7 immunoreactivity in the ductal epithelial cells and smooth muscle actin and p63 positivities in the myoepithelial layer around the ducts which were highlighted the presence of biphasic pattern. Weak estrogen receptor positivity was found within the proliferating ducts and Ki-67 immunoreactivity was less than 2%.

Discussion

SPA was grouped under the nonneoplastic epithelial lesions of salivary glands in the 2017 WHO classification.[6] However, the presence of an uninterrupted thick capsule, a centrifugal growth pattern and a totally different histological pattern from the surrounding tissue seen in the present SPA case may support the possibility of neoplasia.[7] Rarely, SPA may be

associated with other salivary gland lesions, though the coexistence of SPA and DPD as in our case has not been reported previously. In the present case, morphologically, it was not difficult to distinguish the localized SPA from DPD which involved entirely bilateral salivary glands.

Histological features of DPD may resemble those of polycystic conditions affecting the other organs. However, there is no report about the association of DPD involving other organs except for a simultaneous multiple, major, and minor salivary gland involvement.[8]

Except splenic cysts in the present case, there is no reported association.[9] Immunohistochemical staining results in our case showed a dual epithelial and myoepithelial differentiation in SPA and DPA, consistent with the previous reports.[2] This finding is similar to the proliferative breast lesions.

The previous chemotheraphy and radiotheraphies may be contributing factors for the development of SPA in the present case. The coexistence of SPA and DPD in this report may well suggest that the widespread cystic changes in the both salivary gland provided a ground for the formation of SPA. SPA cases were usually treated with excision of the salivary gland and the recurence rates were low.[10] We believe that the management of the DPD should be tailored according to the clinical symptoms. The cystic lesions of the left parotid will be followed-up in the present case.

Ethical approval

This article does not contain any studies with animals performed by any of the authors.

Informed consent

The samples of the patients whose consent forms taken before were evaluated, retrospectively.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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