Case Report

Salivary gland hamartoma with eosinophilic cell adenoma: a clinicopathologic analysis of two cases

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Abstract: Hamartoma of the salivary gland is a rare, non-neoplastic lesion originating in the salivary gland. Hamartomas accompanied by eosinophilic cell adenoma are much rarer. Here we present two cases of salivary gland hamartoma with adenomatous oncocytic hyperplasia, and the related medical literature on both topics was reviewed simultaneously. We hope to raise awareness of the special hamartomas as one of the differential diagnoses of salivary masses.

Keywords: Hamartoma, adenomatous oncocytic hyperplasia, salivary gland

Introduction

Hamartoma is characterized by an overgrowth of local tissue and structural disorder located between the deformity and the tumor. It is a common lesion observed in various parts of the body [1-4]. Salivary gland hamartoma that occurs with adenomatous oncocytic hyperplasia is a special type of hamartoma originating in the salivary gland [1, 2], and only 6 cases have been reported in the English-language literature prior to this report [1-3, 5-7]. To the best of authors’ knowledge, this is the second case report of salivary gland hamartoma with a histologic feature of oncocytic adenoma [2].

In the current report, we present two unusual cases of hamartoma and their complex formation under the microscope. We hope the presentation of such cases can provide clinicians with information regarding the differential diagnosis of salivary tumors.

Case reports

Case 1

In case 1, a 34-year-old man, whose tumor was located in the left parotid gland, was admitted to our hospital. The patient stated that a peanut sized lump was found three years ago, which gradually grew, and now it is about cherry-sized. A physical examination disclosed a round tumor with a volume of about 1.5 cm × 1.5 cm × 1.5 cm in the left parotid gland area behind the left earlobe. The mass showed a smooth surface, medium hardness, no obvious tenderness, no pulsation, clear boundaries, and adhesion to deep tissues. An imaging examination revealed a smooth surface, medium hardness, no obvious tenderness, no pulsation, clear boundaries, and adhesion to deep tissues. An imaging examination revealed an abnormal signal in the left parotid gland, with a high possibility of hamartoma.

An MRI showed that the lesion was located in the left parotid gland, demonstrating a heterogeneously high signal on the T1 and T2 series (Figure 1), and a low signal on the T2-fs series (Figure 2). The maximum cross-section of the mass was about 1.3 cm × 1.0 cm. DWI showed mixed, slightly higher signal changes, and heterogeneous enhancement was observed after we injected a contrast medium. Enlarged lymph nodes could be seen in the surrounding soft tissue. No obvious abnormality was found on a computed tomographic (CT) scan in the right parotid gland.

Then the patient underwent a radical surgery of the tumor. Gross examination showed a round tumor with a volume of about 2 cm × 1.7 cm × 1.5 cm, a smooth surface, and a thin fibrous membrane. It was partly grayish yellow and
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Under microscopic examination, the tumor appeared to be made up of eosinophilic adenoid epithelial cells, adipose tissue, sporadic lymphocytes, and the remnants of salivary gland epithelial cells. Eosinophil and lipocytes were the main components, occupying about 40% and 50% respectively. The eosinophilic cells showed adenomatoïd hyperplasia, with a flaky and nest-like distribution (Figure 4). The cells were large, square or polygonal, and were arranged into luminal structure with eosinophilic secretions in certain areas (Figure 5). The boundary between the adipocyte components and the eosinophils was unclear, and some adipocytes were scattered between fibrous connective tissues and eosinophilic components (Figure 6), and many lymphocytes and a few

Figure 1. MR showed a parotid gland mass with heterogeneously high signal on T2 series (red arrow).

Figure 2. MR showed a parotid gland mass with low signal on T2-fs series (red arrow).

Figure 3. Excised biopsy specimen. Gross examination showed a round tumor with a smooth surface, and a thin fibrous membrane; partly grayish yellow and partly grayish red in color.

Figure 4. The eosinophilic cells were adenomatoïd hyperplasia, with a flaky and nest-like arrangement (hematoxylin and eosin stain; original magnification, × 80).

partly grayish red in color, with a soft texture (Figure 3).
neutrophils and eosinophils had infiltrated the stroma. Dilated glandular cavities with eosinophilic secretions could be seen in local tissue (Figure 7). Inflammatory granulomas and foreign body giant cells appear in some areas (Figure 8), with occasional necrotic fat tissue and calcified cholesterol crystals. The mass was diagnosed as hamartoma with adenomatoid acidophilus hyperplasia.

Case 2

In case 2, a 44-year-old man, a mass in the submandibular gland was found 4 years ago, with an indolent and laxly process. A physical examination showed a round mass of about 4.5 cm × 3.5 cm × 3 cm located in the inferior border of left mandibular angle and mandible, and the anterior border of the sternocleidomastoid muscle. The mass was soft, with no pulsation or tenderness.

Imaging examination: CT showed an irregular soft tissue mass on the lateral side of the left submandibular gland area, with a maximum cross-section of 5.0 cm × 4.1 cm, and the density of the lesion was not inhomogeneous (Figure 9). On a contrast-enhanced scan, the arterial phase was markedly inhomogeneous (Figure 10), the delayed phase was further enhanced, the medial margin of the lesion was not clear, and the mass was locally wrapped around the left mandible without any obvious bone destruction. No obvious enlarged lymph nodes were found in bilateral neck. It was viewed as an adenoma according to its imaging characteristics.

A complete surgical resection was performed, and a pathological examination followed. The gross examination showed an irregular tissue with a volume of 6 cm × 4.5 cm × 4 cm, in a
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Gross examination showed an irregular tissue with a complete envelope, grayish-white and grayish-yellow.

The cross section was golden yellow, hard and tough, and close to the membrane. The rest of the tissue was grayish-white and grayish-yellow, with a tough texture (Figure 11).

Under microscopic, typical sebaceous glands and epidermoid cysts are seen in some areas (Figure 12). The tumor was mainly composed of alveolar eosinophils, which were separated by fibrous connective tissue into lobes or nests (Figure 13). The squamous epithelial nests were interspersed between the tumor cells. The pathological diagnosis was hamartoma, mostly composed of eosinophilic adenomas.

Discussion

Hamartoma is a tumor-like morphological lesion characterized by a mixture of normal tissue components and one or more other abnor-
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Figure 13. The alveolar eosinophils were separated by fibrous connective tissue into lobes or nests (hematoxylin and eosin stain; original magnification, × 80).

mal components, including fibrous connective tissue, fat cells, glands, blood vessels and lymphoid tissue [1-5]. Hamartomas occur mostly in organs such as the lungs, kidneys, liver, spleen, and other organs but rarely occur in the salivary gland [1-4, 6]. Unlike malignant tumors, hamartomas do not possess the ability to grow perpetually, their proliferation is self-limiting, and they are rarely malignant [2, 4].

We have reported two special hamartomas in the parotid gland and the submandibular gland. A microscopic examination of the tumors revealed that the hamartoma was composed of scattered salivary gland elements, mature adipose tissue, an adenomatous eosinophil nest, proliferating lymphoid tissue, and an epidermis cyst [7]. Hamartomas of the salivary glands featuring oncocytic hyperplasia and sebaceous metaplasia are rare, and our case marks the second report of this disease [2]. Analysis and recognition of such diseases could surely provide help for the diagnosis of special cases in frozen sections and accumulated experience for routine pathological diagnosis.

Salivary gland hamartoma with eosinophilic adenoma and sebaceous metaplasia is very rare, and differential diagnosis should be made from the following lesions: (1) Oxyphilic adenoma: The tumor is composed merely of eosinophils, and no squamous epithelial nests or sebaceous gland structures are seen [8]. (2) Sebaceous adenoma: Tumor cells arranged into a sebaceous gland cell nest, a tubular structure, squamous metaplasia, and microcapsules often appear [9]. (3) Acinic cell carcinoma: Generally occurs in young men, with an average age of 25 years. Typical acinar cell carcinomas generally have a complete envelope with a gray-white, fragile cross section. In microscopic observation, the morphology of normal granular and basophilic salivary acinar cytoplasm typically appear [10].

This particular type of hamartoma is similar to a general benign tumor and can be completely removed by surgery [11]. Both of the patients involved in this study recovered well and no recurrence has been found postoperatively.

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Disclosure of conflict of interest

None.

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