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Case Report

Cartilaginous choristoma of the tongue

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ABSTRACT

Cartilaginous choristoma of soft tissue in the oral cavity is rare. The choristoma is a tumor-like mass of normal cells in an abnormal location. In this report, a case of cartilaginous choristoma of the tongue in a 60-year-old female patient was reported. Approximately 10 mm × 10 mm pedunculated healthy colored and elastic-hard mass was observed in the front of circumvallate papillae in the median dorsum of tongue. The mass was surgically removed. Examination of the hematoxylin and eosin-stained sections of the mass revealed that under the squamous epithelium, a hyaline cartilaginous nodule was formed and surrounded by thin fibro-fatty tissue. Neither salivary gland tissues, epithelial components with a ductal structure, nor mesenchymal myxoid and osseous tissues were observed. Moreover, inflammation was minor at that lesion, and there was no cellular atypia. Immunohistochemical analysis revealed that the mass was positive for S-100 protein, and negative for cytokeratin and epithelial membrane antigens. Mixed salivary gland tumor was excluded. According to these findings, the final pathological diagnosis confirmed that the mass was a cartilaginous choristoma.

1. Introduction

Cartilaginous choristoma in the oral and maxillofacial region is rare. Choristoma is a tumor-like mass of normal cells in an abnormal location [1]. Choristoma in the oral cavity consists of various tissues, such as salivary gland, thyroid gland, cartilage, bone, sebaceous tissue, glial tissue, and gastric mucosa [1]. This study reported a case of cartilaginous choristoma on the tongue of a 60-year-old female patient.

2. Case report

A painless mass at the dorsum of tongue had been observed in a 60-year-old female for 5 years before visiting our department. The patient had undergone surgical excision of follicular adenoma of the thyroid at the Department of Endocrine Surgery in our university hospital 3 years ago. After consulting her attending physician at the department, because the mass was observed to gradually grow, the patient was referred to our department. The patient had no history of trauma and chronic inflammation, and no other medical history. Clinical examination showed an approximately 10 mm × 10 mm pedunculated healthy colored and elastic-hard mass, which was found in the front of circumvallate papillae in the median dorsum of tongue. The mass was surgically removed under general anesthesia to prevent the pharyngeal reflex. Gross examination of the cutting surface of the mass revealed that the pedunculated mass included a white tan colored, solid smooth nodule, with a size of 8 mm × 7 mm (Fig. 1). The surgically resected mass was then fixed with 10% neutral-buffered formalin and then prepared for histological and immunohistochemical analyses. Immunohistochemical analysis was performed using a rabbit polyclonal anti-S-100 protein antibody at a dilution of 1:400 (Dako, Glostrup, Denmark), which is used as a marker of chondrocyte, a mouse monoclonal anti-human cytokeratin antibody (clone AE1/AE3, Dako) at a dilution of 1:200, and a mouse monoclonal anti-human epithelial membrane antigen (EMA) antibody (clone E29, Dako) at a dilution of 1:100. The latter two antibodies were used as epithelial-cell markers. Examination of the hematoxylin and eosin-stained sections of

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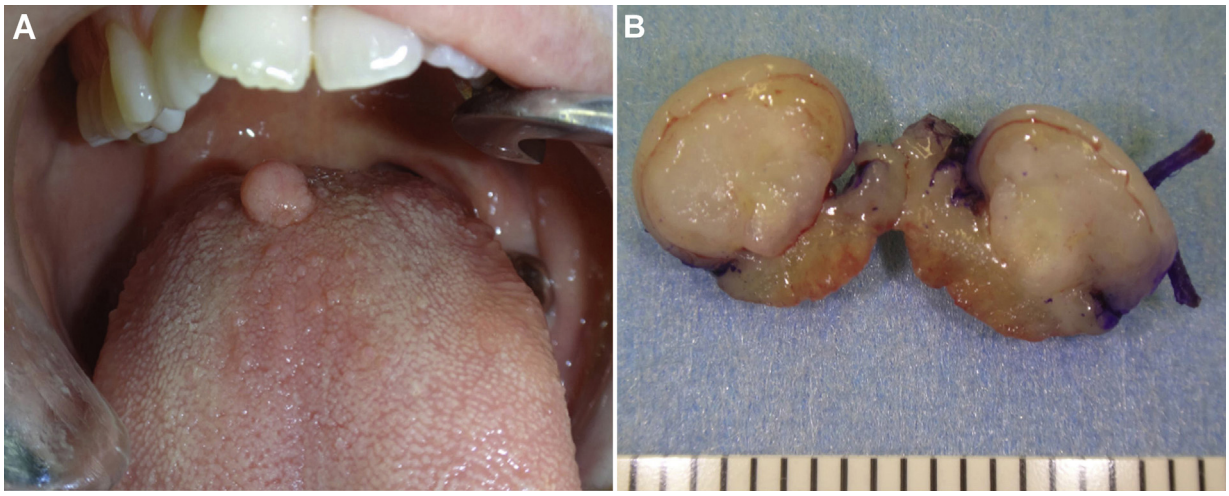

Fig. 1. Macroscopic observation of a mass on the tongue. (A) Pedunculated healthy colored and elastic-hard mass. The mass was located on the front of circumvallate papillae in the median dorsum of tongue. (B) The cross-sectional surface of the lesion showed a pedunculated, white solid nodule with a size of 8 mm × 7 mm.

the mass revealed that under the squamous epithelium, a hyaline cartilaginous nodule was formed surrounded by thin fibro-fatty tissue (Fig. 2A and B). Neither salivary gland tissues, epithelial components with a ductal structure, nor mesenchymal myxoid and osseous tissues were observed. Moreover, inflammation was minor at that lesion, and there was no cellular atypia. Immunohistochemical analysis revealed that the mass was positive for S-100 protein and negative for cytokeratin and EMA (Fig. 2C–E). Mixed tumor of the salivary gland was excluded, because the mass was negative for cytokeratin and EMA, which are makers for pleomorphic adenoma [2]. According to these findings, the final pathological diagnosis confirmed that the mass was cartilaginous choristoma.

3. Discussion

Although cartilaginous choristoma in the oral cavity is rare, cartilaginous choristoma is one of the most frequently observed choristomas of the oral cavity [1]. Cartilaginous choristomas in the palate [3], buccal mucosa [4], gingival [5], oral vestibule [6], lower lip [7], and tongue including dorsum [2,8], ventral surface [9], lateral margin [8] are reported. In a review of cartilaginous choristoma, the age at diagnosis ranges widely from 10 to 80 years, with a mean of 47 years. Fifty-eight percent of the patients are female, and the mass size ranges from <1 cm to several centimeters [1]. They occur most frequently in the tongue [1,8]; the middle dorsal is the most frequently observed location of cartilaginous choristoma on the tongue [2,8]. Histologically, cartilaginous choristoma is composed of a tumor-like mass of hyaline cartilage surrounded by dense, fibrous connective tissue suggestive of perichondrium, by loose fibrous connective tissue, or occasionally by myxoid tissue suggestive of primitive mesenchyme. The chondrocytes may be small and lying within clearly defined lacunar spaces in the hyaline intercellular substance, or may be large, mature, and lying in the calcified intercellular substance. The cartilage is usually more mature at the center of the mass and less mature at the periphery [1]. Moreover, a review of cartilaginous choristoma shows the following five possible origins of the occurrence of choristoma of the tongue: (1) from metaplastic chondroid tissue due to trauma or chronic inflammation, (2) from cartilaginous embryonic rests, such as Meckels cartilage, (3) from pluripotent cells, (4) from neoplasm or teratoma with the preponderance of cartilage, and (5) from the mixed tumor of the salivary gland with the predominance of cartilage [7]. In the case of this report, because the patient had no history of trauma and chronic inflammation of the tongue, metaplasia appears to be ruled out. Some cartilaginous masses have been reported as “chondroma” [3,7,9,10]. Differentiation between choristoma and chondroma is somewhat controversial. Chou et al. [1] suggest that (1) the mass is the non-neoplastic developmental lesions of cartilage rather than neoplasms, because the pattern of tumor-like growth and abnormal location fit the criteria of choristoma, and (2) “cartilaginous choristoma” is a more appropriate term. The present case was considered to be non-neoplastic, because there was no cellular atypia and the lesion contained no different types of tissue such as hair. Furthermore, no components of mixed tumor were found by immunohistochemical examination. Taken together, the possible origin of cartilaginous choristoma from cartilaginous embryonic rests or from pluripotent cells were considered in the present case. However, van der Wal and van der Waal have found neither bone nor cartilage in 130 cadaver tongues [10]. Ectopic cartilage formation in the tongue surely seems rare. The possible origin of cartilaginous embryonic rests in cartilaginous choristoma is still controversial. Treatment of the cartilaginous choristoma is simple excision. No recurrence of this lesion is reported [2–9].