

Cartilaginous choristoma of tongue - A case report and review

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Abstract

Choristoma (aberrant rest or heterotopic tissue) is defined as a histologically normal tissue proliferation, which is not normally found in the anatomic site of proliferation. Choristoma with a proliferation of chondroid tissue is known as cartilaginous choristoma. Clinically, they present as a painless firm nodule and may produce local dysfunction. They occur most frequently in the tongue and less commonly in other sites, such as buccal mucosa, soft palate and gingiva. We report a 35 years female patient with pedunculated globular growth over the left lateral border of the tongue, resembling papilloma but histopathological analysis of the excised specimen revealed to be cartilaginous choristoma.

Keywords: Cartilaginous choristoma, female, tongue

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INTRODUCTION

Choristoma (aberrant rest or heterotopic tissue) is defined as a histologically normal tissue proliferation, which is not normally found in the anatomic site of proliferation.^[1] If the ectopic tissue contains elements from more than one germ layer, they have traditionally been called as teratoma. Choristoma with a proliferation of chondroid tissue is known as cartilaginous choristoma.^[1,2] It was first described by Berry in 1890. It is usually found in the distal extremities and rarely in the soft tissue of the head and neck.^[3,4] This extra skeletal proliferation of bone and cartilage in oral and maxillofacial soft tissue reflects the multipotential nature of primitive mesenchymal cells in this region. Usually, it is developmental in origin; some of these proliferations seem to occur as a result of local trauma. Clinically, they present as a painless firm nodule and may produce local

dysfunction. They occur most frequently in the tongue and less commonly in other sites, such as buccal mucosa, soft palate and gingiva.^[1,2] Here, we report a case of cartilaginous choristoma at the left lateral border of the tongue.

CASE PRESENTATION

A 35 years old female patient visited the non-communicable diseases department of District Hospital for Women in Akola, Maharashtra, India with a slow-growing lump on the left lateral border of the tongue since 2 years [Figure 1]. The growth was initially of small size but slowly grew to a present size of approximately $2.5 \times 2 \times 1$ cm over 2 years. The patient neither had any history of trauma nor crooked tooth on examination. The growth was not painful. The lesion was smoothly

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Figure 1: Clinical photo of pedunculated growth on left lateral border of tongue



Figure 2: post op healed area

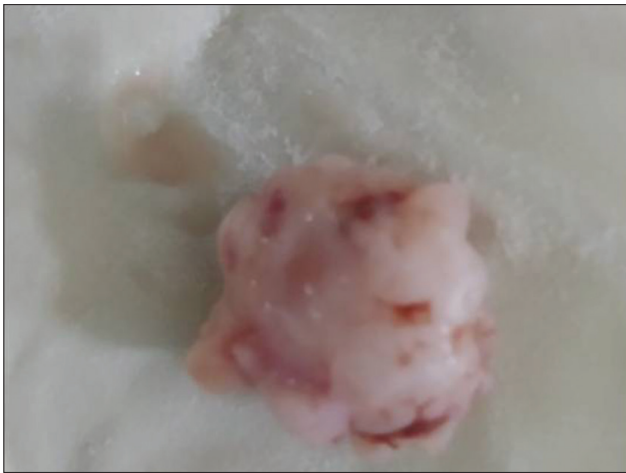


Figure 3: Surgically excised specimen

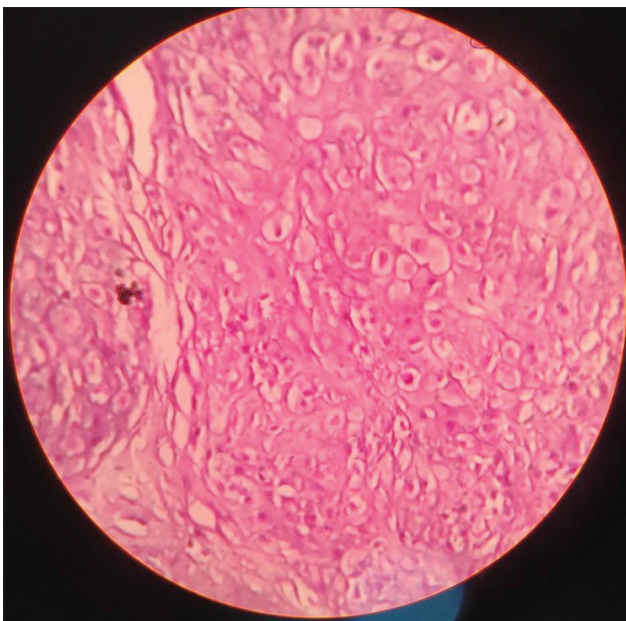


Figure 5: higher magnification view showing mature hyaline cartilage with round chondrocytes trapped inside the lacunae

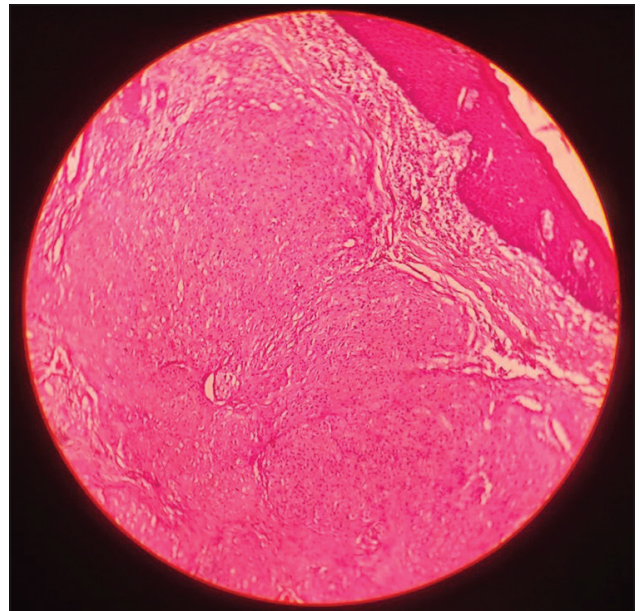


Figure 4: lower magnification view showing well demarcated lobulated myxomatous tissue and hematoxylic cartilaginous tissue in connective tissue stroma.

lobulated, pedunculated and firm to slightly hard on palpation. The overlying mucosa was tensed, pale with no bleeding on provocation. Lymph nodes were not palpable. Clinical diagnosis of papilloma with differential diagnosis of pyogenic granuloma was given. The patient was posted for surgical excision under local anaesthesia. Informed consent was taken before surgical excision. The bilateral lingual block along with local infiltration of 2% lignocaine with adrenaline was given. The lesion was excised in toto with electrical cautery. Wide local excision

of pedunculated growth including surrounding normal tongue tissue (3 mm from all sides) was done. 3.0 vicryl suture were given after approximation of the margins. The sample was fixed in 10% formalin and then sent for histopathological analysis.

The patient was discharged the next day and later had an uneventful healing. Sutures were removed after 7 days. The lesion site was examined repeatedly and after 1 month. The site was completely healed with a smooth lateral border of the tongue [Figure 2].

The excised specimen was firm on palpation, white in colour, cut surface showed whitish areas [Figure 3]. The representative section was taken and embedded in paraffin. Block was cut into thin strips and stained with haematoxylin and eosin stain. Under scanner (4x), the H and E stained section showed stratified squamous surface epithelium with underlying lesional connective tissue stroma. Under low magnification, the surface epithelium showed normal lingual muscle tissue underneath. The tumorous connective tissue stroma was well demarcated and lobulated showing myxomatous tissue and hematoxylic cartilagenous tissue [Figure 4]. Under high magnification, the hematoxylic areas showed mature hyaline cartilage with round chondrocytes trapped inside the lacunae [Figure 5]. So based on the presence of well-demarcated mature hyaline cartilage in a lobulated pattern along with myxoid areas, histopathological diagnosis of cartilagenous choristoma (hamartoma) was made.

DISCUSSION

Choristoma, a congenital anomaly, is better described as a 'heterotopic' rest of cells.^[5,6] Choristoma is a more acknowledged term and it was first described by Krolls *et al.* as tumour-like growths of microscopically common tissue in an unusual location.^[5] In the oral cavity, it is considered as tumour-like growth that has developed from the collection of primordial cells placed at a position far-off from the original tissue or organ. It may consist of a large number of cartilages, bones, fat cells, neural elements, glial tissues, respiratory tissues, thyroid glands and intestinal mucosa.^[5] The presence of mature cartilage is termed as cartilagenous choristoma, Chou *et al.* and Toida *et al.* reviewed the literature. Oral cartilagenous choristomas have been reported in patients with ages ranging from 3 to 80 years with an average age in the third and fourth decade of life. The reported male/female ratio varied between 0.7/1 and 1.2/1, which means that the lesions are almost equally distributed between the sexes.^[4,7,8] The

lesions had been present in the oral cavity for several months up to 30 years and the size ranged from less than 1 cm to several centimetres.^[4,7,8] Different authors have described choristomas at different sites, such as dorsum of tongue,^[1,4,9] lateral border of tongue,^[1,3] ventral surface on tip of tongue,^[8] gingiva,^[5] upper vestibule,^[8] lower lip,^[10] nasopharynx^[11,12] and tonsils.^[13]

The pathogenesis of cartilagenous choristoma is still unclear. Undifferentiated multipotent mesenchymal cells may be considered as a possible origin of cartilage.^[3,13] Another possible origin is from the embryonic remnants. During foetal development, the incomplete resorption of embryonic cartilagenous tissue of the lingual septum, and depending on the persistence after birth, explains a chondromatous proliferation along the tongue midline.^[3,4] However, choristoma of the tonsil appears to be a developmental anomaly associated with the second pharyngeal arch.^[13]

Several hypotheses have been proposed to explain the occurrence of choristoma of the tongue.^[1,2] These include: (1) Origin from cartilagenous embryonic rests, (2) Metaplastic chondroid tissue, (3) Derived from pluripotent cells, (4) Neoplasm or teratoma with a preponderance of cartilage and (5) Mixed salivary gland tumour with a predominance of cartilage. The origin from embryonic rests theory postulates that lesion originates from heterotopic cartilage remnants from any of the first four branchial arches. It is believed that multipotential cells are misplaced during development and sequestered in the tongue.^[1,2] This theory explains the wide distribution of cartilagenous choristoma within the tongue. Another possible embryonic origin is from remnants of Meckel's cartilage.^[1]

The histopathological picture shows a mixture of mature adipocytes or myxoid tissue with islands of cartilage inside a well-defined capsule. Although most of the cases show pure cartilagenous proliferations, few lipocartilagenous and osteocartilagenous lesions also have been documented.^[11] In our case, the lesion tissue is well demarcated and shows areas of mature hyaline cartilage along with myxoid tissue.

Cartilagenous choristoma should be differentiated from cartilagenous metaplasia, which generally occurs in the soft tissue underneath ill-fitting dentures. On the contrary, our patient was a dentate person, without any constant local trauma. Histopathologically, cartilagenous metaplasia is characterised by scattered deposits of cartilagenous cells and calcium that are arranged in a variety of phases of

maturation in solitary or clustered cartilaginous foci.^[12] In our case, there is an absence of calcium and cartilaginous foci.

Differential diagnosis of cartilaginous choristoma includes various benign and malignant conditions. Benign conditions include pleomorphic adenoma, chondroma, neurofibroma, papilloma and ectomesenchymal chondromyxoid tumour.^[3,8] A possibility of a granular cell tumour can be considered as it is a common lesion on the tongue.^[3] Histopathology of our case did not show a wide array of tissue differentiation such as osteoid, adipose, epithelial component, glandular cells as in the case of pleomorphic adenoma. Soft tissue chondroma is microscopically composed of lobules of mature, adult hyaline cartilage, with chondrocytic cells often growing in clusters. One-third of cases may display extensive calcification, particularly in the centre of tumour lobules.^[14] In our case, there is no extensive calcification. Also, neurofibroma, papilloma and ectomesenchymal chondromyxoid tumour (the cells are arranged in chords, strands and sheets within the chondromyxoid matrix) were discarded as diagnosis owing to the characteristic histopathological picture.

It is also important to distinguish it from malignant cartilaginous neoplasms, including primary chondrosarcoma and metastasis from primary intraosseous chondrosarcoma.^[3,8] Our case lacks pleomorphism, mitotic activity and other features of malignancy. Surgical excision is the treatment of choice. No recurrence has been reported in the literature.^[3]

In conclusion, cartilaginous choristoma is a rare lesion in the oral cavity. Clinically, it mimics common lesions such as papilloma, traumatic fibroma, pyogenic granuloma. Hence, a thorough histopathological diagnosis is mandatory.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that

her name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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