

## Pure cartilaginous choristoma on the ventral surface of tongue: A double rarity

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Received - 09 June 2019

Initial Review - 26 June 2019

Accepted - 30 July 2019

### ABSTRACT

Choristomas are tumor like mass that are aggregates of microscopically normal tissue in an abnormal location. Commonly reported choristomas are of Osseous, Cartilaginous, Glandular and Glial types. The oral cavity is an unusual site of presentation where the most common site is dorsum of the tongue. Ventral aspect is an extremely rare site with only four cases reported till date. We report here a case of a 24-year-old male who presented with a small swelling on the ventral surface of the tongue. The histopathological examination revealed features of a well-circumscribed mass composed of exclusively mature cartilage encased within a dense fibrous connective tissue. The case is presented for its double rarity of site which is a ventral surface of the tongue and the histopathology which revealed a pure form of cartilaginous choristoma.

**Keywords:** *Chondroma, Heterotopia, Oral cavity, Undersurface.*

Choristomas (aberrant rest or heterotopic tissue) first described by Krolls et al are defined as histologically normal tissue proliferation in the anatomic sites where they are not normally found [1]. It may consist of a large number of cartilage, bone, fat cells, neural elements, glial tissues, respiratory tissues, thyroid glands, and intestinal mucosa [2,3,4]. Choristoma with a predominant proliferation of chondroid tissue is known as cartilaginous choristoma.

The oral cavity is a rare site of presentation where they occur most frequently in the tongue, the most common site being the dorsum followed by a lateral border. Only four cases till date have been reported worldwide on the ventral surface of the tongue. This paper reports the fifth case of a cartilaginous choristoma on the ventral aspect of tongue in a young male which was of a pure type on microscopy; thus, making it a case with double rarity.



Figure 1: Small firm swelling on ventral surface of tongue.

### CASE REPORT

A 24-year-old male reported to the ENT OPD with a chief complaint of swelling on the ventral surface of the tongue since childhood. It was a slow-growing, painless mass with no associated complaints. There was no significant past history or family history.

General examination and vitals of the patient were within normal limits. Local examination revealed a well-defined mass, pink to brown in colour measuring 1 x 1 cm in size with a firm consistency (Fig. 1). A complete hemogram was advised which was unremarkable, following which an excision biopsy was done and the tissue was sent to the Department of Pathology for histopathological examination.

The gross specimen measured approximately 1 cm × 1 cm in size, oval with a pinkish brown colour, and a soft to firm consistency. On cutting, the cut surface was homogenous, grey-brown (Fig. 2). The histopathological examination showed

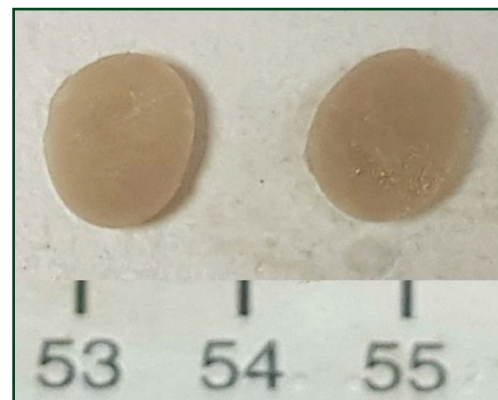
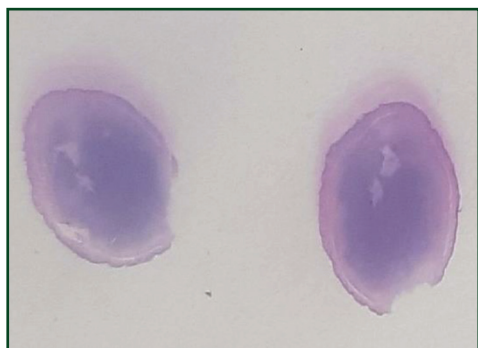


Figure 2: Grossly swelling was of 1 x 1 cm with a homogenous shiny brown cut surface.

**Table 1: Cases of cartilaginous choristomas of the tongue reported till date.**

S. No.	Age (years)	Sex	Location (On Tongue)
1	49	M	Right Lateral border
2	-	M	Right border
3	52	M	Dorsum
4	43	F	Dorsum
5	36	M	Left of midline
6	36	M	Dorsum
7	10	F	Dorsum and lateral border
8	16	M	Dorsum
9	50	F	Ventral surface
10	21	M	<i>Ventral surface</i>
11	5	M	Left lateral border
12	30	M	Left lateral border
13	40	M	Dorsum
14	26	F	Right anterior and lateral surface
15	61	F	Dorsum
16	61	M	Dorsum
17	5	F	Base (near foramen cecum)
18	75	F	Dorsum
19	24	F	Left lateral border
20	35	M	Dorsum
21	71	F	Dorsum
22	28	F	Dorsum
23	27	M	Left lateral border
24	3	F	Dorsum
25	52	M	Right lateral border
26	45	M	Dorsum
27	66	F	Tip of tongue
28	73	F	<i>Ventral surface</i>
29	22	F	<i>Ventral surface</i>
30	64	F	Dorsum
31	60	F	Dorsum
32	54	M	Left lateral border
<b>[Present case]</b>			
33	24	M	<i>Ventral surface</i>

**Figure 3: Photomicrograph showing the scanner view of the H & E stained section.**

keratinized stratified squamous epithelium lined tissue that showed a well-circumscribed mass composed exclusively of basophilic mature cartilage separated from the overlying epithelium by compressed fibro-collagenous tissue. No other tissue was seen admixed (Fig. 3 and 4).

Based on these microscopic findings, a final diagnosis of cartilaginous choristoma of pure type was given. The follow-up period was uneventful until the time of submission of the article.

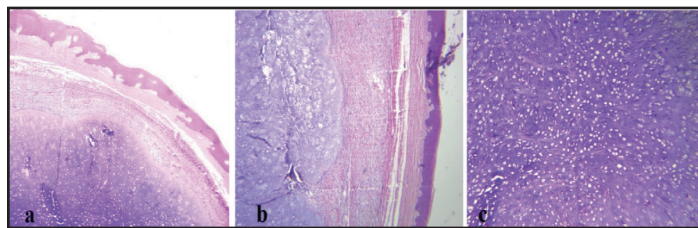
## DISCUSSION

Choristomas are defined as a proliferation of any histologically normal tissue in an abnormal location. Choristomas of the oral cavity are rare lesions [4]. Cartilage, salivary gland, bone, thyroid, sebaceous gland, brain tissue and gastric mucosa have been identified as the sources of intraoral choristomas [2,4]. The most common site in the oral cavity is the dorsum of the tongue, followed by lateral border [4]. Ventral surface of the tongue is a rare site and only four cases have been reported in the literature till date.

Cartilaginous choristoma was initially explained by Berry et al in 1890 and the first case reported in the oral cavity was in 1913 [3,4]. Several hypotheses have been proposed to explain the occurrence of choristoma of tongue. These include: (a) Origin from cartilaginous embryonic rests, (b) Metaplastic chondroid tissue, (c) Derivatives from pluripotent cells, (d) Neoplasm or teratoma with a preponderance of cartilage and (e) Mixed salivary gland tumor with a predominance of cartilage [5].

This lesion has been reported in patients ranging from 10 to 80 years and occurs more commonly in females. Clinically it presents as a slow-growing, painless, firm swelling [4]. Pure cartilaginous choristomas are composed of only mature hyaline cartilage and are an extremely rare occurrence. According to Zegarelli et al, the term cartilaginous choristoma of the tongue should be used only for those lesions that show an exclusive chondromatous growth [6]. Keeping this in view, till date, only thirty-two cases of cartilaginous choristomas on the tongue have been reported [Table 1] [1,4,5,7,8,9,10].

Histopathologically, cartilaginous choristomas of the tongue often show the predominant cartilaginous component admixed of other type(s) of tissue, such as bone and adipose

**Figure 4: Photomicrograph showing the (a) stratified squamous epithelium with well-circumscribed cartilaginous tissue [H & E x 40]; (b) same area in higher magnification [H & E x 100]; (c) exclusive mature hyaline cartilage [H & E x 400].**

tissue. It is important to differentiate cartilaginous choristomas with other benign and malignant conditions that may clinically mimic it. Benign conditions include pleomorphic adenomas with extensive chondroid degeneration, chondromas, and ectomesenchymal chondromyxoid tumors [1,4]. A few malignant cartilaginous neoplasms, including primary chondrosarcomas and metastasis from a primary intraosseous chondrosarcoma may also sometimes enter in the differential diagnosis considerations [9].

These differential diagnoses can most of the times be ruled out by histopathological examination alone, but sometimes ancillary techniques such as immunohistochemistry and cytogenetics may be required to do so. High positivity for S100 protein, a marker of myoepithelial cell suggests chondromatous tissue and has been applied in some previously reported cases of cartilaginous choristomas [9,10]. In our case, the diagnosis was evident on histopathology, therefore, Immunohistochemistry was not required for confirmation. Surgical excision is the treatment of choice. No recurrence has been reported so far in any of the cases mentioned in the literature [2,3,4].

## CONCLUSION

Cartilaginous choristomas have been reported in the dorsum of the tongue quite a few times, but only four cases have been reported on the ventral surface. Thus it is necessary that both the clinician and the pathologist must be aware of this benign but rare entity as it may clinically mimic other oral pathologies. It must be included in the differential diagnosis of all oral cavity

lesions with a predominant chondromatous morphology as it is completely curable by surgical excision.

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*Funding: None; Conflict of Interest: None Stated.*

**How to cite this article:** Bhargava M, Singh A, Singh V, Misra V. Pure cartilaginous choristoma on the ventral surface of tongue: A double rarity. *Indian J Case Reports*. 2019;5(4):366-368.

Doi: 10.32677/IJCR.2019.v05.i04.024