Chondroid choristoma of the tongue: A rare case report

ABSTRACT
Choristomas are tumor-like masses consisting of normal cells in an abnormal location. Choristomas of the oral cavity are rare lesions. We report a case of Cartilaginous choristoma on the ventral aspect of the tongue in a 25-year-old female. Clinical features, differential diagnosis, and tumoral origin theories are also discussed along with a meta-analysis of the reported cases in the PubMed database.

Keywords: Benign, cartilage, chondroid choristoma, tongue

INTRODUCTION
The term “Choristoma” can be used to describe tumor-like masses consisting of normal cells in an abnormal location, i.e., a “heterotopic” rest of the cells.[1,2] The occurrence of these entities has been attributed to abnormalities in the embryonic development of the neural tube.[2] It is crucial to distinguish choristomas from hamartomas and teratomas, wherein hamartomas are nonneoplastic, unifocal/multifocal, and developmental malformations, comprising a mixture of cytologically normal mature cells and tissues which are indigenous to the anatomic location, showing disorganized architectural pattern with predominance of one of its components.[3] On the other hand, teratomas are true neoplasms composed of a variety of parenchymal cell types representative of more than one germ layer.[4] Choristomas found in the oral cavity are classified according to the type of tissue, including salivary gland, Cartilaginous, osseous, thyroid, sebaceous, glial, and gastric/respiratory mucosal.[2]

Cartilaginous choristomas in the oral cavity are unusual entities. We present one such rare case of Cartilaginous choristoma on the ventral aspect of the tongue.

CASE REPORT
A 28-year-old female presented with an asymptomatic mass on the ventral aspect of the tongue of 2-year duration. The patient did not give any history of trauma or any other lesion in the oral cavity. The lesion had gradually increased in size in the past 2 years. On intraoral examination, a firm, nontender, well-demarcated, multilobulated submucosal mass measuring approximately 2.3 cm × 1.6 cm was seen on the left ventral aspect of the tongue crossing the midline [Figure 1]. The overlying mucosa was not associated with any inflammation or ulceration but showed areas of whitish discoloration. Tongue movements were normal. A provisional diagnosis of a benign mesenchymal neoplasm was given. Differential diagnosis included salivary gland neoplasm and granular cell tumor. Excisional biopsy of the lesion was done under local anesthesia, and the excised
specimen was sent for histopathological evaluation. The gross specimen measured around 2.6 cm × 1.9 cm, was roughly oval in shape, firm in consistency, and reddish-white in color, and had a lobulated surface. Histopathological examination revealed a nonkeratinized stratified squamous epithelium and an underlying fibrovascular stroma. Deeper part of the stroma showed lobular proliferation of basophilic mature hyaline cartilage surrounded by a fibrotic capsule, with typical chondroblasts arranged in cell nests or in isolation [Figures 2 and 3]. No atypia was evident. A final diagnosis of chondroid choristoma was given. No evidence of recurrence was found in a 6-month follow-up [Figure 4].

DISCUSSION

Choristomas are lesions characterized by the presence of cells not native to the site. Oral choristomas can be composed of various types of tissues, including cartilage, bone, salivary gland, thyroid, sebaceous, glial, respiratory, and gastrointestinal tissue. Oral Cartilaginous choristomas are rare entities with only 32 cases being reported till date, ours being the 33rd case [Table 1 provides a meta-analysis of all the reported cases in the PubMed literature].

These have been reported in patients ranging between 3 and 80 years of age, with tongue being the most common site of occurrence followed by gingiva, buccal mucosa, soft palate, and palatine tonsil. Sixty percent of the tongue lesions are seen on the dorsum of tongue, 32% on the lateral border, and only 8% on the ventral surface. The term choristoma in the oral cavity was introduced in 1971 by Knoll et al. Zegrelli et al. stated that the term “Cartilaginous choristoma of the tongue” should be used to describe only those lesions that show exclusive chondromatous growth. Several theories have been put forward to explain the origin of cartilage in the oral soft tissue; however, the exact etiology of the lesion still remains unclear. According to the embryonal theory, cartilage develops from the heterotropic fetal Cartilaginous remnants. Remnants of Meckel’s cartilage or displacement of Cartilaginous elements from the first four branchial arches to the area of tongue may act as possible sources for the entity to develop. Metaplastic theory states that trauma, irritation, or chronic inflammation could stimulate the pluripotent mesenchymal cells to differentiate into chondrocytes and proliferate to form the lesion, or this transformation could occur de novo. Chou et al. postulated that proper stimulation and active interstitial and appositional growth of multipotent mesenchymal cells could result in cartilage formation. It was also hypothesized that the vestigial rests of cartilage could act as a source of origin of chondroid choristomas. Chromosomal abnormalities involving the 12q13–q15 region...
Table 1: Cases of chondroid choristomas of tongue reported in the English literature since 1890

| Author and year of publication | Age/sex | Duration of lesion (years) | Site of the lesion | Size of the lesion (cm) | Treatment done/any special investigations done |
|-------------------------------|---------|----------------------------|--------------------|------------------------|-----------------------------------------------|
| Berry, 1890[5]                | 49/male | 5                          | Right border of tongue, middle third | -                      | Excision of the lesion. No recurrence         |
| Johns, 1942[6]                | -/male  | 20                         | Right border       | -                      | Excision of the lesion. No recurrence         |
| Bruce and McDonald, 1953[7]   | 52/male | 2                          | Dorsum, anterior third | 0.5                   | Excision of the lesion. No recurrence         |
| Bruce and McDonald, 1953[7]   | 43/female | 1                         | Dorsum, middle third | 0.3                   | Excision of the lesion. No recurrence         |
| Rosen, 1961[8]                | 36/male | >20                        | Left of midline, anterior third | 2×1.5                 | Excision of the lesion. No recurrence         |
| Yoel and Pundyk, 1965[9]      | 36/male | 8                          | Dorsum, middle, and posterior third | 4.5×1                 | Excision of the lesion. No recurrence         |
| Ramachandran and Viswanathan, 1968[10] | 10/female | 2                         | Dorsum, middle third, lateral border | 1                    | Excision of the lesion. No recurrence         |
| Samant and Gupta, 1971[11]    | 16/male | 6                          | Dorsum, right posterior third | 2.5×1                 | Excision of the lesion. No recurrence         |
| Zegarelli, 1977[12]           | 50/female | -                         | Left lateral ventral region | 0.5×0.5               | Excision of the lesion. No recurrence         |
| Del Rio, 1978[13]             | 21/male | -                          | Right posterior ventral surface | 0.5×0.5               | Excision of the lesion. No recurrence         |
| Segal et al., 1984[14]        | 5/male  | 5                          | Left lateral border, anterior third | 2                    | Tumor was excised, no recurrence noticed after 2 years of surgery |
| Segal et al., 1984[14]        | 30/male | 30                         | Left lateral border, anterior third | 1.5                  | Tumor excised using carbon dioxide laser. No recurrence recorded 1.5 years after surgery |
| Yasuoka et al., 1984[15]      | 40/male | 7                          | Dorsum, middle posterior third | -                    | Excision of the lesion. No recurrence         |
| Tohil et al., 1987[16]        | 26/female | -                         | Right anterior and lateral surface | -                    | Excision of tumor. No recurrence               |
| van der Wal and van der Wal, 1987[17] | 61/female | 15                        | Dorsum, left side | 2                    | Excision of lesion. No recurrence             |
| Weitzner et al., 1987[18]     | 61/male | 6 months                   | Dorsum, middle third, right midline | -                   | Lesion was excised. No recurrence             |
| West and Atkins, 1988[19]     | 5/female | Several months              | Vicinity of foramen cecum | 1.5                  | Excision of the lesion. No recurrence         |
| Tani et al., 1989[20]         | 75/female | 2                         | Dorsum, midline in anterior third | 0.7                  | Mass was excised along with a margin of normal tissue and the overlying mucosa No recurrence |
| Trovbridge et al., 1989[21]   | 24/female | >5                        | Left lateral border, middle third | 1×2                  | Excision of lesion, no recurrence after 6 months |
| Moore et al., 1990[22]        | 35/male | 8-0                        | Left dorsum of the tongue | 1×0.8                 | Excisional biopsy under general anesthesia, no recurrence for 5 years |
| Mosqueda-Taylor et al., 1998[23] | 71/female | 2                         | Dorsum, middle third, adjacent to midline | -                    | Surgically excised with a small margin of normal tissue and the covering mucosa, IHC markers - S100, vimentin, EMA, CK Positive cyttoplasmic staining for S-100 in chondroid areas, weak positivity for vimentin in the same area. Negative for EMA, CK No recurrence for 6 months |
| Mosqueda-Taylor et al., 1998[23] | 28/female | 18 months                 | Dorsum, middle third, adjacent to midline | -                    | Surgically excised with a small margin of normal tissue and the covering mucosa, IHC markers - S100, vimentin, EMA, CK Positive cyttoplasmic staining for S-100 in chondroid areas, weak positivity for vimentin in the same area. Negative for EMA, CK No recurrence for 6 months |

Contd...
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| Author and year of publication | Age/sex  | Duration of lesion (years) | Site of the lesion                                                                 | Size of the lesion (cm) | Treatment done/any special investigations done |
|--------------------------------|----------|----------------------------|-----------------------------------------------------------------------------------|--------------------------|-----------------------------------------------|
| Mosqueda-Taylor et al., 1998   | 27/male  | 24                         | Left lateral border, between middle and posterior third                           | -                       | Surgically excised with a small margin of normal tissue and the covering mucosa, IHC markers used - S-100, vimentin, EMA, CK. Positive cytoplasmic staining for S-100 in chondroid areas, weak positivity for vimentin in the same area. Negative for EMA, CK. No recurrence for 6 months |
| Bansal et al., 2005            | 52/male  | -                          | Right lateral border                                                              | 0.5                     | Lesion co-existed with squamous cell carcinoma; right hemiglossectomy done with radical neck dissection |
| Bansal et al., 2005            | 45/male  | -                          | Dorsum, anterior third, left side                                                 | 1.0                     | Excision biopsy of nodule was done, no recurrence in 2 years |
| Weynand and Reychler, 2007     | 66/female| -                          | Right ventral site of tip of tongue                                               | -                       | Lesion with surrounding normal tissue excised under general anesthesia, no recurrence until 25 months of follow-up |
| Yamamoto et al., 2009          | 56/female| 1                          | Right lateral border of tongue                                                    | 1.5                     | Tumor extirpated under local anesthesia, no recurrence in 22 months of follow-up |
| Naik et al., 2009              | 25/female| 5                          | Posterior third of tongue, ventral aspect                                         | 2×3                     | Excision under general anesthesia. No recurrence |
| Pereira et al., 2012           | 64/female| Several years              | Midline region of tongue, on sulcus terminalis                                   | 0.5                     | Excision of lesion (partial glossectomy), IHC markers used - S-100, p63, CK; Strong Immunoreaction for S-100 in chondrocytes, immunoreaction weak for p63 and CK. |
| Kimura et al., 2015            | 34/female| 3                          | Lateral border of tongue                                                          | 2                       | Incisional biopsy for diagnosis confirmation followed by excisional biopsy, no recurrence in a follow-up period of 7 months |
| Semwal et al., 2019            | 55/female| 1                          | Left lateral border of tongue                                                     | -                       | FNAC done, yielded scant myxoid material blocking needle, smears stained with Wright-Giemsa and PAP; smear showed only myxoid stroma with few scattered oval-to-spindle cells, following which excision of specimen was done |
| Present case                   | 28/female| 2                          | Right ventral aspect of tongue crossing midline                                   | 1.3×1                   | Excision of lesion. No evidence of recurrence in 1 year |

FNAC: Fine-needle aspiration cytology, CK: Cytokeratin, IHC: Immunohistochemistry, PAP: Papanicolaou, EMA: Epithelial Membrane Antigen

have been associated with chondroid choristomas of the soft tissues.\(^{[38]}\)

Differential diagnosis of chondroid choristoma includes a variety of benign lesions such as chondroma, pleomorphic adenoma, traumatic chondromatous metaplasia, ectomesenchymal chondromyxoid tumor, and granular cell tumor. It is also important to distinguish it from malignant Cartilaginous neoplasms such as primary chondrosarcoma or metastasis from a primary intraosseous chondrosarcoma.\(^{[28,30,31]}\)

In the present case, the absence of epithelial and mesenchymal components, such as plasmacytoid cells, fusiform cells, cuboidal cells, chondromyxoid stroma along with the absence of morphological patterns of the epithelial cells (trabecular, ductal, cystic, and solid) helped in differentiating it from pleomorphic adenoma.\(^{[39]}\) Absence of lobulated growth pattern, clusters of chondrocytic cells, and surrounding collagenous stroma excluded the diagnosis of chondroma.\(^{[30,40]}\) Traumatic chondromatous metaplasia is seen in edentulous ridges as a result of chronic mechanical irritation from ill-fitting dentures.\(^{[1]}\) In our case, the patient was dentulous, and hence, this differential diagnosis was ruled out. Ectomesenchymal chondromyxoid tumor is histopathologically characterized by the lobular proliferation of ovoid and fusiform cells with occasional foci of atypia in a chondromyxoid background and absence of these features...
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In conclusion, chondroid choristomas are benign developmental lesions which need to be diagnosed correctly for apt management. Free marginal surgical excision is the best treatment of choice. No recurrences have been reported in the literature.

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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