Cartilage Choristoma (Soft Tissue Chondroma): A Rare Presentation in the Lower Lip

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Cartilage choristoma (soft tissue chondroma) is an ectopic cartilaginous tissue that is rarely found in oral mucosa. Awareness of such disease entity will guide proper diagnosis and treatment. A case of cartilage choristoma occurring in the lower lip of an 8-year-old child is reported. Potential pathogenetic mechanism and the histologic features of this unusual condition are further discussed.

Keywords: *Cartilage choristoma, soft tissue chondroma, lip lesion*
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INTRODUCTION

Ectopic cartilaginous tissues are rarely found within oral mucosa and submucosa. Both terms, cartilage choristoma and soft tissue chondroma, have been used to designate such unusual occurrences. The term choristoma, as opposed to chondroma, implies that this is a normal piece of tissue in an abnormal location. It also emphasizes the non-neoplastic nature of the cartilage. We present a case of cartilage choristoma occurring in the lower lip of an 8-year-old child. To the best of our knowledge this is the first report of this unusual lesion in the lower lip.

CASE REPORT

An 8-year-old girl, accompanied by her parents, presented with a lower lip exophytic nodule. Her medical history was unremarkable. An intraoral examination revealed a submucosal firm lesion measuring 0.7 by 0.7 by 1.0 cm of the lower lip. The parents confirmed the lesion was present over the past 12 months, while slowly increasing in size over the last three to four months. It was asymptomatic. Upon palpation, the lesion was firm and freely moveable. Our differential diagnosis included sialolithiasis, traumatic fibroma, fibrosed mucocele, inflamed salivary gland (sialadenitis) and benign tumor, NOS. An excisional biopsy was performed under local anesthesia. The lesion was noted to be well circumscribed and partially inferior to the anterior aspect of the mentalis muscle.

Histologic examination revealed well-circumscribed cartilage tissue arranged in a lobular pattern (Fig. 1). The chondrocytes were unremarkable and without evidence of cellular atypia. The cartilage tissue stained positive with an S100 immunohistochemical stain. The histologic differential diagnosis at first included cartilage choristoma, and, benign mixed tumor (a benign salivary gland neoplasm) with prominent chondromatous component. Since serial sectioning revealed no epithelial tumor tissue, benign mixed tumor was eventually excluded as a diagnostic possibility.

DISCUSSION

Cartilage choristomas are uncommon lesions. A small number of cases have been reported in the head and neck region. These lesions have also been referred to as soft tissue chondromas. The initial report of an oral lesion dates back to 1977 (Zegarelli) and it describes a chondroma of the tongue.[1] Additional reports of oral soft tissue chondroma have been reported involving the gingiva,[2] tongue,[3] and buccal mucosa.[4] The designation, cartilage choristoma, first appeared in the dental literature of the 1890s and it describes a lesion of the buccal mucosa.[5] There are other reports of cartilage choristomas occurring in gingiva,[6] maxillary vestibule,[7] and tongue.[8]

Within these sites, cartilage choristomas/soft tissue chondromas have a predilection for the tongue, accounting for 85% of all cases.[2,9] These typically present as a firm nodule ranging between 0.5 to 2.0 cm in size.[7,9] Most patients are asymptomatic. However, those lesions occurring as a dorsal tongue nodule may generate symptoms of dysphagia or nausea.[9] Due to their rarity, minimal assessment of age grouping

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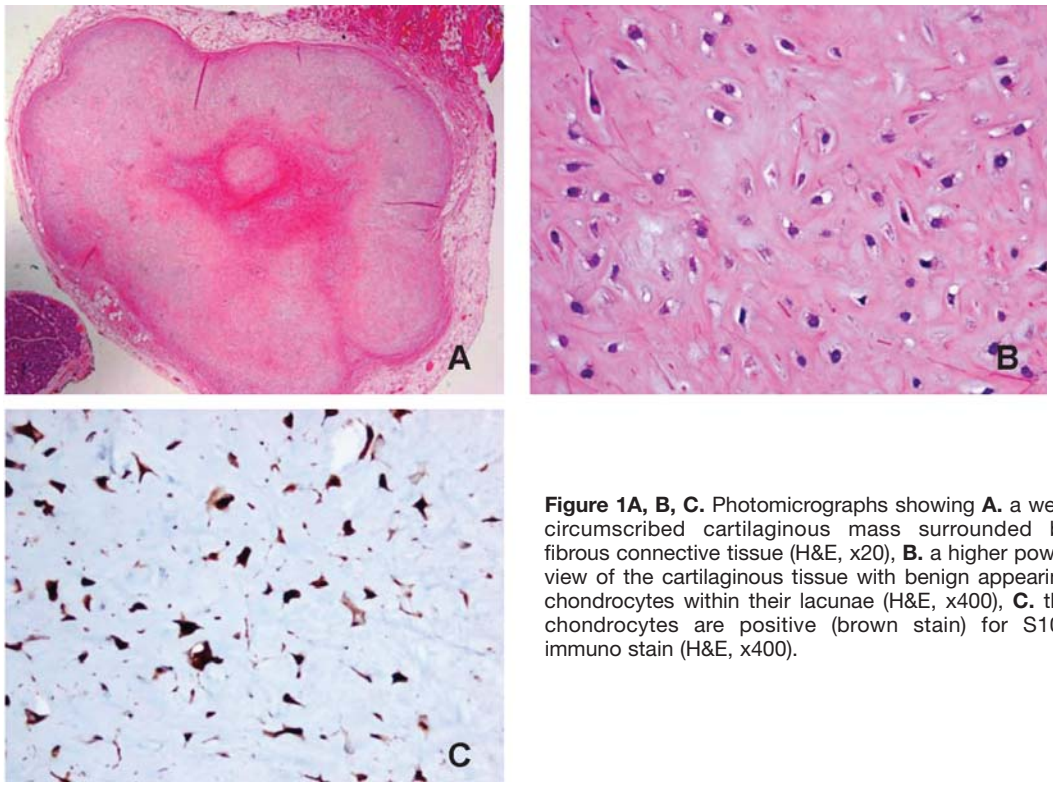

Figure 1A, B, C. Photomicrographs showing **A.** a well-circumscribed cartilaginous mass surrounded by fibrous connective tissue (H&E, x20), **B.** a higher power view of the cartilaginous tissue with benign appearing chondrocytes within their lacunae (H&E, x400), **C.** the chondrocytes are positive (brown stain) for S100 immuno stain (H&E, x400).

and gender predilection is available, and, no strong predilection for either sex or any age group has been reported.[9]

The etiology and pathogenesis for cartilage choristomas are yet to be determined. There are two plausible theories; an embryologic one and a mesenchymal cell one.[6,7] The embryologic premise states that cartilage remnants from embryogenesis may later give rise to cartilage choristomas. In synch with this theory, Matsushita et al.[7] speculated that stimulated heterotopic cartilaginous remnants from the major and minor alar cartilages and nasal septal cartilages are responsible for choristomas discovered in the maxillary vestibule midline. In contrast, the mesenchymal cell theory claims that multipotential mesenchymal cells, when stimulated, will differentiate into chondrocytes and form mature cartilage tissue. Expansion in size may then result from active interstitial and appositional growth. The stimuli for mesenchymal differentiation into chondroblasts and chondrocytes are unknown.

During the microscopic assessment, it is important that the pathologist not mistake a cartilage choristoma / chondroma for a malignant cartilage tumor. While mature cartilage is the predominant histologic finding, some cellular atypia is occasionally observed in cartilage choristomas and chondromas.[2] The features of atypia include binucleation, nuclear hyperchormasia, nuclear pleomorphism and increased mitotic activity. In such cases, a thorough clinical assessment is indicated to rule out a soft tissue chondrosarcoma.[2,7] Because most chondrosarcomas of soft tissues result from direct extension of underlying bony chondrosarcomas, a thorough radiographic survey for intraosseous lesions becomes important.[10] Once a diagnosis of cartilage choristoma / chondroma has been obtained, a conservative local excision is the treatment of choice.[9] There are no known reports of malignant transformation.[9]

In summary, our case is the first one to report a cartilage choristoma / soft tissue chondroma of the lower lip submucosa. Similar lesions may be found in other intraoral locations such as tongue, gingiva, buccal mucosa and vestibules. Simple surgical removal is the treatment of choice.

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