Case Report

Chondroind Syringioma of the Lip in a Pediatric Patient

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KEY WORDS

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ABSTRACT

Chondroid syringoma (CS), also known as mixed tumor of the skin, is a rare, benign adnexal tumor composed of both epithelial and mesenchymal components. It primarily affects middle-aged to elderly individuals, typically presenting as a solitary, slow-growing nodule on the head and neck, most commonly the scalp, nose, or cheeks. Its occurrence in the pediatric population is exceedingly rare, and presentation on the lip is even less common. We present a unique case of a 9-year-old female child who developed a CS on her left lower lip. The patient presented with a painless, firm nodule that had been slowly growing for approximately six months. Clinical differential diagnoses included mucocele, fibroma, and other benign salivary gland tumors. Complete surgical excision was performed, and histopathological examination confirmed the diagnosis of chondroid syringoma. This case highlights the unusual presentation of CS in terms of both age and anatomical location, emphasizing the importance of considering rare entities in the differential diagnosis of pediatric cutaneous masses and the necessity of histopathological confirmation for definitive diagnosis and appropriate management.

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Introduction

Chondroid syringoma (CS), or mixed tumor of the skin, is an uncommon benign cutaneous adnexal tumor first described by Virchow in 1863 and later coined by Hirsch and Helwig in 1961 [1]. CS has a very low incidence of only 0.1% - 0.98 % [2].

It is characterized by the presence of epithelial elements, typically resembling sweat gland ducts or tubules, and mesenchymal components, predominantly chondroid (cartilaginous) and myxoid stromal tissue [2].

CS most commonly affects adults, particularly those in their fifth to seventh decades of life, with a slight male predominance [3]. The most frequent anatomical sites include the head and neck region, with the scalp, nose, and cheeks being common locations. Less frequently, it can occur on the trunk or extremities [4].

The occurrence of CS in children is exceptionally rare, with only a handful of cases reported in the literature [5]. Furthermore, its presentation on the labial mucosa, especially the lower lip, is also uncommonly reported, as lesions in this area are more typically mucoceles, fibromas, or minor salivary gland tumors. This report details a unique case of CS arising on the left lower lip of a 9-year-old female, highlighting its unusual presentation in a pediatric patient and the diagnostic challenges it poses. An exhaustive literature review revealed that the present case is the first presentation of CS occurring on the lower lip in a paediatric patient [2].

Case Presentation

A 9-year-old female child presented to the outpatient clinic with a chief complaint of a slowly growing, painless mass on her left lower lip. The lesion had been present for approximately six months, with no history of trauma, spontaneous bleeding, discharge, or associated pain. Her medical history was unremarkable, and there

Figure 1: Clinical picture of the lesion

was no family history of similar lesions or genetic syndromes. There were no history of drug reaction, trauma and cheek biting.

On physical examination, a solitary, well-circumscribed, firm, and non-tender nodule was observed on the vermilion border of the left lower lip. The lesion measured approximately 1.5cm×1.0cm, was yellowish-colored, and the overlying skin appeared intact with no signs of ulceration or inflammation (Figure 1). It was mobile on palpation and did not appear to be attached to deeper structures. Regional lymph nodes were not palpable.

Based on the clinical presentation and location, the initial differential diagnoses included a mucocele, fibroma, lipoma, hemangioma, and a benign minor salivary gland tumor such as a pleomorphic adenoma. Due to the firm consistency and persistent nature of the lesion, an excisional biopsy was planned for definitive diagnosis and therapeutic management.

Under local anesthesia, the lesion was completely excised with a narrow margin. The excised specimen was sent to the Department of Oral Pathology and Microbiology for histopathological examination. Grossly, the specimen was a firm, well-defined, encapsulated nodule, grayish-white on cut section (Figure 2).

Figure 2: The excised tissue

Histopathological examination revealed a well-circumscribed, partially encapsulated tumor composed of both epithelial and mesenchymal components. The epithelial component consisted of nests, cords, and ductal structures, some of which were cystic, lined by single or double layers of cuboidal to columnar epithelial cells. (Figure 3a) These structures were interspersed within a prominent and diverse mesenchymal stroma. The stromal component predominantly displayed a chondromyxoid matrix, characterized by a basophilic, mucinous background with embedded chondrocyte-like cells (Figure 3b). There was no evidence of cellular atypia, pleomorphism, increased mitotic activity, or necrosis, confirming the benign nature of the lesion. Based on these pathognomonic histopathological features, a final diagnosis of CS was made.

The post-operative course was uneventful. The patient made a full recovery, and the surgical site healed well. At a 12-month follow-up, there was no sign of recurrence.

Discussion

CS is a rare benign adnexal tumor of controversy regarding its exact histogenesis, though it is widely believed to originate from eccrine or apocrine sweat glands [2]. Its rarity is amplified when considering pediatric cases. A comprehensive review of the literature

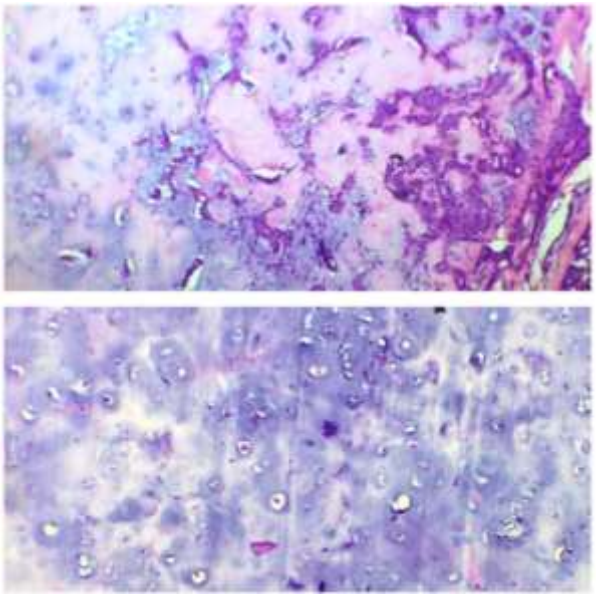

Figure 3: a: Photomicrograph reveals nests of cuboidal to columnar epithelial cells in chondro- myxoid connective tissue stroma. (Hematoxylin and Eosin 20×), **b:** High power view of chondromyxoid mesenchymal element with few chondrocytes (Hematoxylin and Eosin 40×)

reveals only a limited number of CSs reported in individuals under 18 years of age [5,6]. Furthermore, while the head and neck region is a common site, the specific localization on the lip is unusual, particularly as the lip is rich in minor salivary glands, making tumours of salivary gland origin more probable [7]. CS is rare on the labial mucosa of the lips; it is extremely rare on the lower lip. A review of the literature revealed 43 case reports of CS of lip with 38 cases reported in upper lip and 5 cases reported in the lower lip. The present case report is the first report of CS in a paediatric patient (Table 1).

The clinical presentation of CS is typically a solitary, firm, non-tender nodule that grows slowly over months to years. This aligns with the presentation in our 9-year-old patient. However, the non-specific clinical appearance often leads to a broad range of differential diagnoses. For a lip lesion in a child, common considerations include: mucocele, lipoma, fibroma, hemangioma and minor salivary gland tumor [8].

The definitive diagnosis of CS relies entirely on histopathological examination. The hallmark feature is the admixture of epithelial elements (ductal, tubular, or cystic structures) and a prominent mesenchymal stroma, which can be chondroid, myxoid, fibrocytic, or adipose [2]. The presence of a significant chondroid component is critical for the diagnosis of chondroid syringoma. It is important to differentiate CS from pleomorphic adenoma (PA) of minor salivary glands, given their close histological resemblance and similar composition of epithelial and mesenchymal elements. While CS traditionally refers to tumors of skin adnexal origin and PA to salivary gland origin, some authors consider them to exist on a spectrum, particularly when arising in areas with mixed adnexal and salivary gland components like the lip [9]. However, the predominance of sweat gland-like epithelial components in our case favored the diagnosis of CS. Additionally, CS shows epithelial differentiation towards adnexal structures while adnexal structures are usually absent in PA, serous and mucous cells are quite evident in PA but not in CS and PA shows epithelial cells arrangement in sheets, strands and ducts while in CS the prominent arrangement of epithelial cells is tubulocystic.

The diagnosis of CS is generally based on histopathology the hallmark features include nests of cuboidal and polygonal cells, intercommunicating tubule-alveolar structure lined with two or more rows of cuboidal cells, ductal structure, and matrix of varying structure [3, 10]. Malignant transformation of CS is exceedingly rare but has been reported. Features suggestive of malignancy include rapid growth, ulceration, infiltrative borders, and microscopically, cellular atypia, high mitotic activity, necrosis, and infiltrative growth [10-12]. Our patient's lesion showed none of these malignant features, and its benign nature was confirmed.

Table 1: A review of the published cases of chondroid syringioma (CS) in the lip

S.NO	Authors	Number of cases	Site	Patient details
1	Stout and Gorman (1959) [4]	16	Upper lip	N/A
		2	Lower lip	N/A
2	Triantafyllou and Rapidis (1986) [6]	1	Upper lip	38M
3	Adalam and Wood. 1986 [2]	1	Upper lip	N/A
4	Bekerecioglu *et al*. 2002 [8]	3	Upper lip	24F, 27M, 28 F.
5	Shimizu *et al.* (2003) [7]	1	Upper lip	68 M
6	Arikan *et al.* (2004) [20]	1	Upper lip	73 M
7	Dubb and Michelow (2010) [11]	1	Upper lip	58 F
8	Girgis *et al*. (2015) [14]	1	Upper lip	23 M
9	Shilpa *et al.* (2016) [5]	1	Upper lip	48 M
10	Kundu *et al.* (2016) [12]	1	Upper lip	46 M
11	Min *et al.* (2016) [9]	5	Upper lip	39M, 44M, 47M, 64M, 65F
12	Reddy *et al.* (2017) [3]	1	Upper lip	35M
13	Syed *et al.* (2019) [13]	1	Upper lip	44 M
14	Goel *et al.* (2020) [18]	1	Upper lip	23 M
15	Rodrigues *et al.* (2021) [1]	1	Lower lip	43 M
16	Vázquez Hernández *et al.* (2021) [17]	1	Upper lip	65M
17	Palit *et al.* (2021) [15]	1	Lower lip	18 M
18	Gotoh S *et al.* (2021) [16]	1	Lower lip	58 M
19	Chon J *et al.* (2024) [10]	1	Upper lip	40 M
20	Oussalem A *et al*. (2025) [19]	1	Upper lip	63 M

Abbreviations: S.NO: Serial number, NA: Not available, M: Male, F: Female

The treatment of choice for CS is complete surgical excision with clear margins. Recurrence is rare after complete removal but can occur if excision is incomplete [8, 11, 13 -15]. The prognosis for benign CS is excellent following surgical intervention, as demonstrated by the uneventful post-operative course and no recurrence in our patient.

This case report is significant as it contributes to the limited literature regarding pediatric CS, particularly its occurrence on the lower lip. It highlights that while rare, CS should be considered in the differential diagnosis of any firm, slowly growing, painless cutaneous nodule in children, even in atypical locations. Early biopsy and histopathological examination remain crucial for accurate diagnosis and appropriate management. [21] To conclude, this research has few limitations including no use of immunohistochemistry and limited follow up time.

Patient Consent

Informed consent was obtained from the patient's legal guardian for the publication of this case report and accompanying images.

Conclusion

CS is a rare benign adnexal tumor that typically affects adults. This case report describes an unusual presentation of CS on the left lower lip of a 9-year-old female child, underscoring its rarity in the pediatric population and this specific anatomical site. Clinical diagnosis of such lesions can be challenging due to their non-specific appearance. Complete surgical excision is the treatment of choice, leading to an excellent prognosis. This report emphasizes the critical role of histopathological examination for definitive diagnosis and underscores the importance for clinicians to consider uncommon benign entities when evaluating pediatric cutaneous masses, even in atypical locations.

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Conflicts of Interest

The authors declare no conflicts of interest.

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