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A RARE OF CHONDROGENIC CHORISTOMA OF THE CHEEK, MIMICKING SOFT TISSUE CHONDROMA

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Abstract

Background: Chondrogenic choristoma, also known as cartilaginous choristoma, is a rare benign lesion characterized by the presence of mature cartilage in an anatomically inappropriate location. Although it has been reported in several sites of the head and neck, involvement of the cheek is uncommon and may present a diagnostic challenge because of its morphological similarity to soft-tissue chondroma. Case Presentation: I report a rare case of chondrogenic choristoma of the cheek in a 5-year-old patient evaluated at Bhayangkara Hospital, Palembang, Indonesia. The patient presented with a cheek mass clinically suspected to represent a soft-tissue tumor. The excised specimen measured 1.2 × 0.5 × 0.4 cm. Histopathological examination demonstrated encapsulated, a well-circumscribed lesion composed of lobules of mature hyaline cartilage. The cartilaginous tissue was predominantly hypocellular, with mature chondrocytes located within lacunae and embedded in abundant hyaline extracellular matrix. The chondrocytes showed bland cytological features without significant nuclear atypia or mitotic activity. No necrosis, destructive infiltration, or other malignant features were identified. There was no evidence of osseous involvement in the examined specimen. Conclusion: Chondrogenic choristoma should be considered in the differential diagnosis of a well-circumscribed cartilaginous mass in the cheek, particularly in pediatric patients. The distinction from soft-tissue chondroma may be challenging on histopathological examination alone and requires correlation with the clinical and radiological findings. Recognition of this rare entity is important to avoid misclassification as a true cartilaginous neoplasm.

Keyword: *chondrogenic choristoma, cheek, soft-tissue chondroma*

A. INTRODUCTION

Choristomas are uncommon, tumor-like lesions composed of mature, histologically normal tissue occurring in an anatomically inappropriate location. When the ectopic tissue is composed predominantly of mature cartilage, the lesion is termed a chondrogenic or cartilaginous choristoma. (Sakrikar et al, 2022)These lesions are considered non-neoplastic and are thought to result from developmental heterotopia, although other mechanisms, including aberrant differentiation of mesenchymal tissue, have also been proposed. These

lesions are generally regarded as benign, non-neoplastic developmental lesions and are considered to represent heterotopic tissue resulting from developmental or embryologic abnormalities. However, their exact pathogenesis remains uncertain, and alternative mechanisms, including aberrant differentiation or metaplastic transformation of mesenchymal tissue, have also been proposed (Fu et al, 2024)

Cartilaginous choristomas of the oral cavity are particularly rare. The tongue is the most frequently reported intraoral site, whereas involvement of other locations, including the buccal mucosa and other soft tissues of the oral and maxillofacial region, is considerably less common. (Al-Ali & Hantzakos, 2024). Because of its rarity and the absence of large population-based studies, its true incidence and prevalence remain unknown. Most available epidemiological information is derived from individual case reports, small case series, and literature reviews rather than from prospective epidemiological studies. Recent literature continues to describe oral cartilaginous choristoma as a rare entity, particularly when occurring outside the tongue. A 2025 literature review of cartilaginous choristoma of the hard palate noted that fewer than 50 cases had been reported for this unusual site, emphasizing the limited number of documented cases in the oral cavity.(Ueno S et al,2023). Clinically, these lesions typically present as slow-growing, firm, and generally painless or asymptomatic masses, and their nonspecific appearance may lead to consideration of other benign soft-tissue lesions (Gonçalo et al, 2023). Histopathologically, they are characterized by mature cartilage in an anatomically inappropriate location, with the diagnosis established through microscopic examination (Sakrikar et al, 2022)

The pathogenesis of chondrogenic choristoma remains incompletely understood. Unlike true cartilage neoplasms, a choristoma represents mature, histologically normal tissue occurring in an anatomically inappropriate location. Recent reports continue to support a developmental origin, although the precise mechanism responsible for the formation of ectopic cartilage has not been definitively established (Al-Ali & Hantzakos, 2024; Fu et al., 2024). One of the principal hypotheses is the embryonic rest theory. According to this concept, multipotential mesenchymal cells or cartilaginous embryonic remnants may become displaced or incompletely resorbed during craniofacial development. These ectopic cells subsequently remain within the developing soft tissues and may retain the capacity to differentiate into mature chondrocytes. The resulting mature hyaline cartilage may persist as a localized, tumor-like mass despite being unrelated to the normal cartilage-bearing structures. Recent literature has continued to cite embryonic dysplasia or misplaced developmental tissue as a possible

mechanism for oral cartilaginous choristoma (Al-Ali & Hantzakos, 2024; Fu et al., 2024). Another proposed mechanism is chondroid metaplasia, in which pluripotent or multipotent mesenchymal cells within the soft tissue undergo chondrogenic differentiation following local stimuli. Trauma, chronic mechanical irritation, and chronic inflammation have been proposed as potential stimuli for this process. However, the available evidence remains limited, and the absence of a history of trauma or chronic irritation in many reported cases argues against a universal reactive mechanism. Recent reports therefore continue to regard metaplastic transformation as one of several possible pathogenetic mechanisms, alongside the developmental theory involving heterotopic embryonic cartilage remnants or misplaced pluripotent cells (Nezam et al., 2022

An important diagnostic consideration is the distinction between cartilaginous choristoma and benign cartilaginous neoplasms, particularly soft-tissue chondroma. Recent literature has emphasized that this distinction may be challenging because both lesions may demonstrate mature, well-organized cartilage and relatively bland chondrocytes. Clinical presentation, anatomical location, relationship to adjacent structures, and histopathological findings should therefore be considered collectively (Carvalho et al, 2025). Soft-tissue chondroma itself may occur in unusual extraosseous locations of the head and neck, including the masseteric space, further illustrating the potential diagnostic overlap between these entities (Yadav et al, 2023). The distinction between chondrogenic choristoma and soft-tissue chondroma can therefore be challenging. Lobular architecture, hypocellularity, mature chondrocytes, and abundant hyaline matrix may be encountered in both lesions and should not be regarded as independently diagnostic. Clinical and radiological correlation, including assessment of the relationship between the lesion and adjacent bone, joint structures, and other potential sources of cartilage, is important when establishing the final diagnosis. Recent reports of oral cartilaginous choristoma have similarly emphasized the importance of integrating clinical and histopathological findings to avoid misclassification as a neoplastic lesion. (Carvalho et al, 2025)

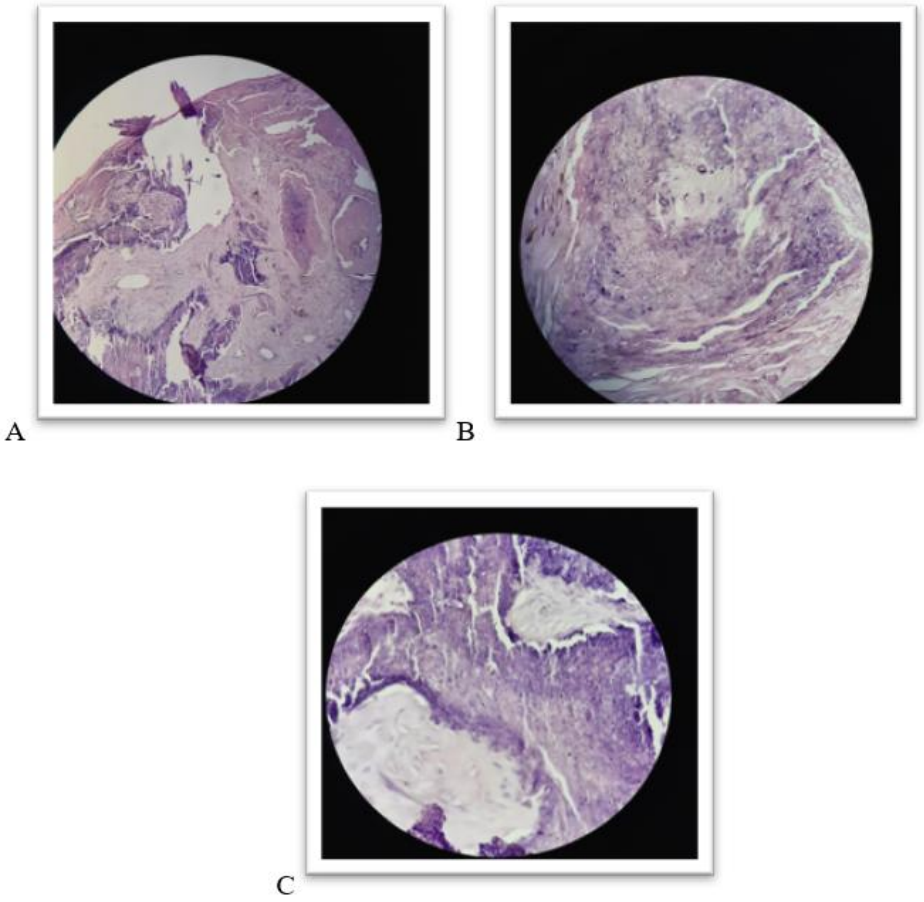
The occurrence of a cartilaginous lesion within the cheek of a 5-year-old child is particularly unusual. In the present case, the lesion was initially clinically suspected to represent a soft-tissue tumor and measured only 1.2 × 0.5 × 0.4 cm on excision. Histopathological examination demonstrated lobules of predominantly hypocellular mature hyaline cartilage without significant cytological atypia or mitotic activity. The combination of the patient's young age, unusual anatomical location, mature heterotopic cartilage, and absence

of malignant features prompted consideration of chondrogenic choristoma as the most appropriate diagnosis, while soft-tissue chondroma remained the principal differential diagnosis. Therefore, this case is presented to highlight an unusual occurrence of chondrogenic choristoma of the cheek in a pediatric patient and to discuss the diagnostic challenges in distinguishing this rare heterotopic lesion from soft-tissue chondroma.

CASE PRESENTATION

A 5-year-old child presented to Bhayangkara Hospital, Palembang, Indonesia, with a mass in the cheek region. The lesion was clinically suspected to be a soft-tissue tumor. The available clinical evaluation and radiological examination, did not indicate a definite relationship between the lesion and the underlying bone. The patient subsequently underwent excisional biopsy for histopathological evaluation.

Gross examination revealed a well-circumscribed tissue fragment measuring 1.2 × 0.5 × 0.4 cm. The specimen was submitted for routine histopathological examination. Microscopically, the lesion was well circumscribed and composed predominantly of lobules of mature hyaline cartilage. The cartilage was predominantly hypocellular and contained mature chondrocytes located within lacunae. The chondrocytes exhibited bland cytological features without significant nuclear atypia or pleomorphism. The intervening cartilage matrix was abundant and homogeneous.No significant mitotic activity, necrosis, or destructive infiltrative growth was identified. There was no evidence of malignant transformation. No osseous component or apparent involvement of adjacent bone was identified in the examined specimen.



From picture above A. Histopathological examination of the chondrogenic choristoma showing a well-circumscribed and encapsulated lesion composed predominantly of mature hyaline cartilage within fibrous connective tissue.(H&E, 4x10) B. Intermediate-power view of the chondrogenic choristoma showing well-formed mature hyaline cartilage arranged in lobules and surrounded by fibrous connective tissue. The cartilage contains relatively uniform chondrocytes located within lacunae and embedded in an abundant hyaline extracellular matrix. (H&E, 10x10) C. High-power view showing mature hyaline cartilage composed of relatively uniform chondrocytes located individually or in small groups within well-defined lacunae. The chondrocytes display bland nuclei without significant pleomorphism or cytological atypia. The surrounding extracellular matrix is homogeneous and hyaline in appearance. No increased mitotic activity, necrosis, or malignant cytological features are identified (H&E, ×400).

Based on the patient's young age, the unusual cheek location, the presence of mature hyaline cartilage in an anatomically inappropriate soft-tissue site, and the absence of cytological atypia or destructive growth, the findings were considered most consistent with a chondrogenic (cartilaginous) choristoma. Soft-tissue chondroma was considered the principal

histopathological differential diagnosis. The final diagnosis was established by integrating the clinical and anatomical findings with the histopathological features.

B. RESULTS AND DISCUSSION

Chondrogenic, or cartilaginous, choristoma is a rare benign lesion characterized by the presence of mature cartilage in an anatomically inappropriate location. Within the oral cavity, these lesions are uncommon and have been reported predominantly in the tongue, with other locations occurring considerably less frequently. Recent literature continues to emphasize the rarity of oral cartilaginous choristomas and their potential to clinically mimic other soft-tissue lesions (Sakrikar et al, 2022) .

The present case is notable because the lesion occurred in the cheek of a 5-year-old child. The cheek is an unusual location for a cartilaginous choristoma, and the young age of the patient further contributes to the uncommon presentation. Most reported oral cartilaginous choristomas have involved the tongue and have occurred in older patients, although the lesion may be encountered at other ages and anatomical sites. The unusual location in the present case highlights the importance of considering choristoma when mature cartilage is identified in a soft-tissue lesion at an unexpected anatomical site (Sakrikar et al., 2022; Fu et al., 2024).

Clinically, oral choristomas are generally described as slow-growing, firm, and relatively indolent masses. Because their clinical appearance is nonspecific, they may initially be diagnosed as other benign soft-tissue lesions. In the present case, the cheek lesion was clinically suspected to represent a soft-tissue tumor, illustrating the difficulty of establishing the diagnosis based solely on clinical findings. Fu et al. (2024) similarly emphasized that the diagnosis of oral choristoma primarily relies on histopathological examination because of its tumor-like clinical presentation.

Radiological assessment in the present case showed no significant osseous abnormality, bone destruction, or calcified lesion. The absence of an apparent relationship with the underlying bone supports the interpretation of the lesion as an extraosseous soft-tissue process. (Crompton et al., 2025; Usuki et al., 2025) This finding is relevant because the anatomical relationship between a cartilaginous lesion and adjacent bone may contribute to the differential diagnosis. Soft-tissue chondroma may also occur in unusual extraosseous locations of the head and neck; Yadav et al. (2023), for example, reported an extraskeletal soft-tissue chondroma involving the masseteric space.

Histopathologically, the present lesion consisted of well-circumscribed lobules of

mature hyaline cartilage with predominantly hypocellular areas, mature chondrocytes within lacunae, and abundant hyaline matrix. The chondrocytes showed bland cytological features without significant atypia or pleomorphism. No significant mitotic activity, necrosis, or destructive infiltrative growth was identified. These findings are consistent with the benign histological appearance described in recent reports of cartilaginous choristoma, in which mature hyaline cartilage is identified in an ectopic location without features of malignancy (Al-Ali & Hantzakos, 2024).

Histopathologically, the present lesion consisted of well-circumscribed lobules of mature hyaline cartilage with predominantly hypocellular areas, mature chondrocytes within lacunae, and abundant hyaline matrix. The chondrocytes showed bland cytological features without significant atypia or pleomorphism. No significant mitotic activity, necrosis, or destructive infiltrative growth was identified. These findings are consistent with the benign histological appearance described in recent reports of cartilaginous choristoma, in which mature hyaline cartilage is identified in an ectopic location without features of malignancy (Sakrikar et al, 2022; Al-Ali & Hantzakos, 2024).

The principal differential diagnosis in this case was soft-tissue chondroma. Both chondrogenic choristoma and soft-tissue chondroma may contain mature hyaline cartilage and relatively bland chondrocytes, creating potential histopathological overlap. Castano B, et al. 2024). A recent report by Carvalho 2025, specifically addressing the distinction between chondroma and cartilaginous choristoma, described a lobulated proliferation of well-organized cartilage containing mature chondrocytes and highlighted the diagnostic difficulty between these entities.

The distinction between the two entities should therefore be based on an integrated assessment rather than on a single microscopic feature. In the present case, the combination of the patient's young age, unusual cheek location, mature and predominantly hypocellular hyaline cartilage, absence of cytological atypia or destructive growth, and lack of radiographic evidence of bony involvement favored chondrogenic choristoma. This approach is consistent with recent literature emphasizing the importance of correlating the clinical and anatomical context with histopathological findings when evaluating oral choristomas and distinguishing them from neoplastic lesions (Fu et al., 2024; Carvalho et al., 2025).

Another consideration is cartilaginous metaplasia, although this was less favored in the present case. Cartilaginous metaplasia generally consists of smaller or scattered foci of cartilage associated with an identifiable local stimulus or tissue alteration, whereas the present lesion

demonstrated a well-circumscribed, organized proliferation of mature hyaline cartilage. Sakrikar 2022, also emphasized the importance of distinguishing cartilaginous choristoma from cartilaginous metaplasia based on the pattern and organization of the cartilage and the clinical context.

Complete surgical excision is generally considered the treatment of choice for oral cartilaginous choristoma. Recent case reports have described favorable outcomes following excision, with no recurrence during the reported follow-up periods (Al-Ali & Hantzakos, 2024). In the present case, complete excision provided both definitive treatment and adequate tissue for histopathological diagnosis. Continued clinical follow-up is appropriate because of the rarity of the lesion and the limited number of reported cases.

Overall, this case illustrates an unusual presentation of chondrogenic choristoma arising in the cheek of a young child. The combination of mature hyaline cartilage, bland chondrocytes, hypocellularity, absence of malignant features, and lack of radiographic evidence of osseous involvement supported the diagnosis. Awareness of this rare entity is important because its unusual In the present case, the absence of an apparent osseous connection and the well-circumscribed nature of the lesion support a localized extraosseous process. Therefore, wide oncological resection is generally unnecessary when the histopathological findings confirm a benign chondrogenic choristoma. Conservative complete excision with preservation of adjacent structures is appropriate for a localized lesion without evidence of malignant transformation or infiltrative growth.

The status of the surgical margins should nevertheless be considered during pathological assessment. Although recurrence appears to be uncommon, complete removal is preferable to minimize the possibility of residual ectopic cartilage. Some literature has suggested that removal of the associated perichondrial tissue may be considered because residual perichondrium could theoretically provide a source for further cartilaginous growth. However, evidence supporting a substantial recurrence risk in oral cartilaginous choristoma remains limited because of the rarity of the lesion. (Sethi H et al, 2024)

Postoperative management is generally straightforward and depends primarily on the anatomical site and extent of the surgical procedure. For a cheek lesion, routine wound care, assessment of mucosal healing, and evaluation of facial function are appropriate. Histopathological confirmation should be communicated to the treating clinician so that unnecessary additional treatment can be avoided in the absence of malignant features.

Clinical follow-up is recommended, although there is no established standardized

surveillance protocol specifically for chondrogenic choristoma because of the small number of reported cases. Periodic clinical examination is reasonable to assess wound healing and detect any local recurrence. Recent reports have documented favorable outcomes without recurrence following complete excision, including a 2024 cartilaginous choristoma of the nasopharyngeal region and a 2025 oral cartilaginous/osteocartilaginous choristoma (Al-Ali & Hantzakos, 2024; Usuki et al., 2025).

Overall, the management of the present case is consistent with the approach reported in the recent literature: complete local excision followed by histopathological confirmation and clinical follow-up. Because the lesion demonstrates mature hyaline cartilage without atypia or destructive growth and lacks an apparent connection with bone, additional chemotherapy, radiotherapy, or radical surgery is not indicated.

C. CONCLUSION

Chondrogenic choristoma of the cheek is an exceptionally uncommon benign lesion, particularly in a young child, and represents an important diagnostic consideration when mature cartilage is identified within an otherwise nonspecific soft-tissue mass. In the present case, the combination of the patient's young age, unusual cheek location, absence of radiographic evidence of osseous involvement, and histopathological demonstration of well-circumscribed, predominantly hypocellular mature hyaline cartilage without atypia, significant mitotic activity, necrosis, or destructive growth supported the diagnosis of chondrogenic choristoma. The principal clinical significance of this case lies in its potential to mimic a soft-tissue neoplasm, particularly soft-tissue chondroma, despite its benign and heterotopic nature. Recognition of this entity and careful integration of clinical, radiological, anatomical, and histopathological findings are essential for accurate diagnosis, appropriate surgical management, and avoidance of unnecessary concern regarding neoplastic transformation. This case also expands the limited spectrum of reported oral cartilaginous choristomas by documenting an unusual presentation in the cheek of a pediatric patient.

CONFLICT OF INTEREST

The authors declare that they have no conflict of interest related to the publication of this case report.

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