Case Report

Cartilaginous Choristoma of the External Auditory Canal

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Abstract

Choristoma, a subtype of heterotopia, represents the presence of histologically normal tissue at an embryologically aberrant site. The finding of a heterotopic cartilaginous mass within the bony external auditory canal is an uncommon occurrence. We report a case of cartilaginous choristoma in a 5-year-old male patient. Otoscopic examination demonstrated a round, white, firm mass located on the anterior wall of the external auditory canal, covered by normal-appearing epithelium. Palpation with a ball probe revealed firm, non-tender lesions, raising suspicion of osseous or cartilaginous masses rather than soft-tissue tumors. The mass was surgically excised through a transcanal approach. Histologically, the mass was composed of mature cartilage without any neoplastic characteristics. This case emphasizes the need to consider choristoma in the differential diagnosis of a white mass lateral to an intact tympanic membrane.
Keywords: *cartilaginous choristoma, external ear canal, heterotopic.*

Cartilaginous Choristoma pada Liang Telinga

Abstrak

Choristoma merupakan salah satu bentuk heterotopia, adalah suatu jaringan yang secara histologi normal tetapi secara embriologi berada pada lokasi yang abnormal. Keberadaan suatu massa kartilago heterotopik pada liang telinga pars osea adalah jarang. Kami melaporkan sebuah kasus cartilaginous choristoma pada anak laki-laki usia 5 tahun. Pada pemeriksaan otoskopik didapatkan massa bulat berwarna putih pada dinding anterior liang telinga. Massa padat, putih yang dilapisi epitelium yang normal. Palpasi menggunakan ball-probe menunjukan bahwa lesi bersifat keras dan tidak nyeri tekan, mengarahkan bahwa massa cenderung oseous maupun cartilaginous dan bukan tumor jaringan lunak. Pembedahan dilakukan melalui pendekatan transkanal. Secara histologi, lesi terdiri dari jaringan kartilago matur tanpa fitur neoplastik. Kasus ini menekankan pentingnya mempertimbangkan choristoma pada kasus lesi berwarna putih yang berada di lateral membran timpani yang intak.
Kata kunci: *cartilaginous choristoma, external ear canal, heterotopik***.**

Introduction

Choristoma is a tumor-like developmental growth of normal microscopic tissue in an abnormal site.[1] Choristomas most commonly contain bone, cartilage, or both. Choristomas with chondroid tissue proliferation are known as cartilaginous choristomas.[2] Cartilaginous choristoma (CC) is typically an incidental finding in asymptomatic individuals.[1] Many reports describe different types of choristomas in the head and neck, such as gastric mucosa in the tongue, salivary gland tissue in the middle ear, and osseous or cartilaginous masses in the intraoral soft tissues.[3] Lee[4] reported 36 cases of cartilaginous choristoma in his research conducted between 1990 and 2004. Furthermore, Yamahara's[3] review of the literature identified only 53 reported cases of cartilaginous masses on the bony EAC. The age distribution of patients with choristoma has been reported to range from 3 to 52 years, with a preference for the left side.[5] These data originate from case series and have not been established through a systematic review. This report discusses the management of cartilaginous choristoma in the bony external auditory canal (EAC). Although cartilaginous choristoma is generally considered a benign and incidental finding, its rarity and similarity to other bony or cartilaginous lesions can pose diagnostic challenges. They may lead to unnecessary, extensive surgical procedures. Reporting this case highlights the importance of accurate clinical assessment, careful intraoperative evaluation, and appropriate pathological interpretation to guide optimal management.

Case Description

In July 2022, a 5-year-old otherwise healthy boy was brought by his mother to the Otorhinolaryngology–Head and Neck Surgery outpatient clinic of Ngesti Waluyo Christian Hospital, Temanggung, Central Java, Indonesia. The chief concern was a white mass in the left external auditory canal, incidentally noticed by the mother one month earlier while bathing the child. The boy himself had never reported any symptoms, including ear pain, itching, discharge, bleeding, hearing difficulty, tinnitus, vertigo, or facial weakness. There was no history of ear trauma, foreign body insertion, recurrent otitis externa, or previous ear surgery. The mother stated that the child had never allowed ear cleaning with cotton buds, and therefore, the exact time of onset of the lesion could not be determined.

Otoendoscopic examination revealed a solitary, round, pearly-white, firm mass measuring approximately 3 × 3 mm arising from the anterior wall of the left external auditory canal, located 7–8 mm from the tympanic annulus (Figure 1). The lesion was covered entirely by intact, normal-appearing canal skin, non-tender, and immobile on gentle palpation with a ball-probe, with no bleeding on manipulation and no cerumen impaction. The tympanic membrane was fully visualized and normal in both ears. Examination of the right ear, nose, nasopharynx, oral cavity, and neck was unremarkable.

Under general anesthesia, endoscopic transcanal excision was performed. The mass was situated between the squamous epithelium and the periosteum, displayed a clear plane of cleavage, and was removed entirely without curettage or drilling of the underlying bone. The excised specimen measured 3 × 4 × 3 mm. Histopathological examination revealed that the mass consists of cartilage surrounded by squamous epithelium and keratin, without evidence of malignant transformation, and is consistent with a cartilaginous choristoma (Figure 2).

At the 12-month follow-up visit, the left external auditory canal remained widely patent, with no stenosis or recurrence, and the child remained asymptomatic. Thus the final diagnosis was cartilaginous choristoma of the left external auditory canal.

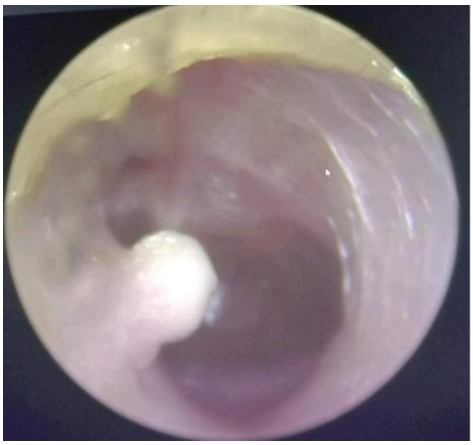

Figure 1. Left Ear Endoscopy

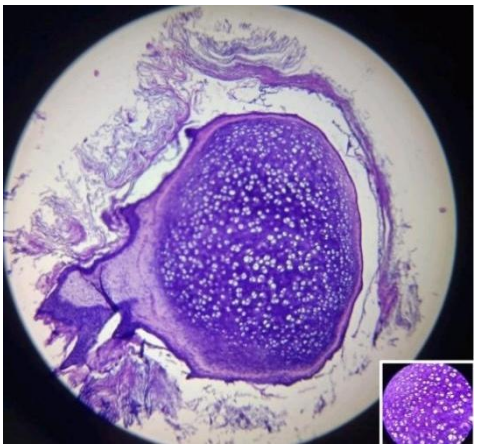

Figure 2. Microscopic Finding of the Lesion

Discussion

The presence of a heterotopic cartilaginous mass within the bony external auditory canal represents an infrequent and noteworthy clinical observation. Knowledge of this entity remains comparatively limited. Historically, cartilaginous lesions of the EAC were initially designated as chondromas. However, the revised nomenclature, cartilaginous choristoma, is now preferred. This updated terminology more accurately characterizes the lesion as a tumor-like proliferation of histologically normal tissue occurring in an aberrant location.[1] Lee's[4] findings indicated that histopathological examination of these lesions revealed a tumor-like mass composed of normal chondrocytes. It is pertinent to note that cartilaginous tissue is not a constituent of the normal bony external auditory canal. Consequently, the presence of a cartilaginous mass in this site, arising from tissue where cartilage is not typically found, aligns with the definition of a cartilaginous choristoma.[4]

Cartilaginous choristomas are predominantly located on the medial aspect of the anterior wall of the external auditory canal, anterior to the manubrium and the short process of the malleus. These lesions typically range in size from 1 to 4 mm and are often asymptomatic. In some instances, they may manifest as otitis externa. Asymptomatic lesions or those exhibiting no growth may be managed with observation.[5] When a lesion is identified in a typical location for a cartilaginous choristoma, it is crucial to differentiate it from exostosis and keratosis. Similarly, when a lesion occurs in an atypical location, the differential diagnosis should also include keratosis, fibroma, and osteoma. Definitive diagnosis and differential diagnosis of a cartilaginous choristoma are established through biopsy, and the treatment of choice is complete surgical resection. Unlike a chondroma, a cartilaginous choristoma is composed solely of normal cells, facilitating relatively straightforward surgical removal. Notably, no cases of recurrence following complete resection or malignant transformation have been reported, suggesting a favorable prognosis.[6]

While Kim[6] has described cartilaginous choristomas as largely asymptomatic, Raghavan[7] reported a case of a choristoma of the EAC associated with atresia, situs inversus totalis, and congenital hearing loss. Histopathological analysis in that instance revealed a salivary gland choristoma, and the patient experienced postoperative facial nerve palsy.

In the present case, the lesion appeared to be in contact solely with the periosteum but did not seem to originate from it. The ease with which the lesion was removed using an elevator suggests its location between the squamous epithelium and periosteum of the bony EAC. One possible consideration is that this lesion represents an early stage of a periosteal chondroma. However, periosteal chondromas have the potential to continue growing after skeletal maturation, with some lesions reaching up to 6 cm in size.[4] In contrast, the lesion in this case measured only 3 mm in diameter.

The precise etiology of cartilaginous choristomas remains undetermined. However, it is hypothesized that aberrations during the differentiation of the first and second branchial clefts during the fetal period may predispose to choristoma formation.[8] The pathogenesis is

thought to involve the aberrant migration of embryological cartilage cells, possibly originating from heterotopic cartilaginous embryonic rests of Meckel's or Reichert's cartilage of the first or second branchial arch. These cartilage precursor cells may mistakenly migrate into the primitive external ear canal during the invagination of the first pharyngeal cleft and proliferate within the deeper tissues of the canal. Nevertheless, their definitive origin remains elusive.[3] An alternative theory posits that undifferentiated multipotent mesenchymal cells may represent a potential cellular origin for cartilage formation.[9]

A variety of other lesions can present with similar characteristics and should be included in the differential diagnosis of a solitary mass of the external auditory canal. Exostoses may be an incidental finding in the EAC; however, they are typically bilateral in patients with a history of frequent aquatic exposure and exhibit a broad base with complete tympanic bone involvement.[10] Osteomas can also occur in the EAC and are characteristically attached to the tympanosquamous suture, lateral to the isthmus. Cholesteatoma, while less common in the EAC, is another diagnostic consideration. Although keratin horns typically arise in sun-exposed areas, they have been reported as incidental findings in the EAC. Malignancies can also occur in this location. Meticulous otoscopic examination, a thorough review of patient risk factors, and appropriate imaging modalities can aid in diagnosing choristomas and differentiating this relatively benign lesion from more serious masses.[1]

Hamartomas are characterized by excessive proliferation of mature tissues normally found in a specific area of the body but arranged in a disorganized manner and often dominated by a single tissue type[11]. In contrast, choristomas consist of histologically normal tissue located in an anatomical site where that tissue does not typically exist, representing a heterotopic or ectopic presence. The terms choristoma and heterotopia are frequently used interchangeably in literature. However, the definitions of hamartoma and choristoma can vary, with some malformations classified as hamartomas, while many ectopic tissues fit the definition of choristomas. Both lesions can occur in various parts of the body.[11]

The ability to distinguish between cartilaginous choristoma and periosteal chondroma, both preoperatively and intraoperatively, is of clinical significance. This distinction extends beyond mere terminology, as the diagnosis dictates the extent of surgical excision. In cases of cartilaginous choristoma, simple excision of the lesion is typically successful. Conversely, periosteal chondromas necessitate complete excision with curettement of the underlying bone to minimize the risk of recurrence. Given that cartilaginous choristoma is a less familiar entity and lacks distinct histological differences from chondroma, pathologists may frequently diagnose cartilaginous masses of the bony EAC as simple chondromas if provided with limited clinical or intraoperative information. Therefore, to facilitate more accurate pathological diagnosis, surgeons should provide detailed information to the pathologist regarding the lesion's relationship to the periosteum, specifically noting whether the mass was in contact with the periosteum or whether there was evidence of underlying periosteal erosion.[3]

This case provides several important clinical lessons. First, a small, firm, white mass located lateral to an intact tympanic membrane should prompt consideration of cartilaginous choristoma in the differential diagnosis, particularly when palpation suggests a non–soft tissue origin. Second, careful intraoperative evaluation of the lesion's relationship to the periosteum is essential, as this information has direct implications for surgical planning and pathological interpretation. Providing detailed intraoperative findings to the pathologist may prevent misdiagnosis as chondroma and avoid unnecessary aggressive surgical intervention. Finally, simple excision without curettement appears to be sufficient for cartilaginous choristoma, supporting a conservative surgical approach when this diagnosis is suspected.

Conclusion

The presence of a heterotopic cartilaginous mass in the bony external EAC is relatively rare. The term "Cartilaginous Choristoma" rather than "chondroma" or "hamartoma" may be more appropriate for these lesions. The differential diagnoses are exostoses, osteomas, and cholesteatomas. If removal of CC is needed, simple excision can be done via endoscopic transcanal guidance.

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Authors' Contributions

Angga Kusuma served as the primary surgeon, responsible for the patient's clinical management and surgical treatment, as well as data acquisition. Theresia Hening D.A., performed the histopathological examination and contributed to the interpretation of the pathological findings. Fiane de Fretes was responsible for drafting the manuscript and organizing the literature review. All authors contributed to the critical revision of the manuscript, read, and approved the final version of the manuscript.

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Data and Materials Availability

The data and materials supporting the findings of this study are available from the corresponding author upon reasonable request.

Consent Form

The authors certify that they have obtained all appropriate patient written consent forms. In the form, the patient's parent has given their consent for images and other clinical information to be reported in the journal. The patient's parent understands that the name and initial will not be published, and due efforts will be made to conceal the identity.

Competing Interests

The authors declare that they have no competing interests.

AI Usage Declaration

Artificial intelligence tools (ChatGPT) were used solely to assist with language editing and text refinement. All content generated by AI has been reviewed, verified, and approved by the authors, who take full responsibility for the manuscript.

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