# Cartilaginous Choristoma of the Bony External Auditory Canal: A Study of 36 Cases

Fei-Peng Lee

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ORIGINAL RESEARCH

# Cartilaginous Choristoma of the Bony External Auditory Canal: A Study of 36 Cases

**Fei-Peng Lee, MD,** Taipei, Taiwan

**OBJECTIVE:** The study goal is to present the clinical data of 36 cases of heterotopic cartilaginous mass in the bony external auditory canal and to clarify the terminology used to describe this clinical entity.

**STUDY DESIGN AND SETTING:** We conducted a medical record review of 36 consecutive patients with lesions (26 of which were excised) seen at two Departments of Otolaryngology in Taiwan.

**RESULTS:** Histopathological examination revealed that all 26 excised lesions were composed of mature hyaline cartilage, and on this basis, cartilaginous choristoma was diagnosed.

**CONCLUSION:** The presence of a heterotopic cartilaginous mass in the bony external ear canal is not as rare as it was once thought to be. The descriptive term "cartilaginous choristoma" rather than "chondroma" may be more appropriate for these lesions.

**EBM RATING:** C.



The presence of a heterotopic cartilaginous mass in the bony external auditory canal is an unusual and interesting finding. Relatively little information about this entity has accumulated. Only 17 cases, including our previously reported 6 cases, have been well documented in the literature.[1-5] Because histopathological examination revealed these lesions were composed of mature hyaline cartilage, we and other authors had designated this lesion "chondroma" of the external auditory canal in previous reports.[1-5] With heightened awareness, an increasing number of such cases were seen in our department in recent years. It appeared that this lesion was not as rare as it was once thought to be. Consequently, we now favor the term "cartilaginous choristoma" rather than "chondroma" since these lesions represent tumor-like growths of histologically normal tissue in an abnormal location.[6]

The purpose of this article is to assess the clinical and histologic features of 36 consecutive cases of such lesions and to discuss their pathogenesis and the terminology used to identify these lesions.

## MATERIAL AND METHODS

From 1990 to 2004, 36 cases of cartilaginous choristoma of the bony external auditory canal were clinically diagnosed by the author in the Department of Otolaryngology, Taipei Medical University Hospital and Chang Gung Memorial Hospital. There were 19 males and 17 females. Patients' age ranged from 6 to 70 years, with a mean of 38 years. The right ear was affected in 18 cases and the left in 18 cases. These lesions usually presented as a single, white, hard, round, horn- or club-shaped mass with a smooth surface, and the size was 1 to 3 mm in diameter. Of the 36 lesions, 35 were photographed using a rigid otoscope or oto-microscope and photo-documented with color slides or videotapes, and one was well drawn and the drawing included in the chart. The external ear was also photographed if concurrent congenital abnormalities were found. The lesion appeared to be asymptomatic and incidentally found during a routine otoscopic examination. Twenty-six patients agreed to undergo surgical excision and received a pathological diagnosis. After local anesthesia with xylocaine-epinephrine solution injection was provided, excision under otomicroscopic guidance was performed in the operating room in 16 patients, and excision under video-otoscopic guidance was performed in the outpatient department in 10. All these lesions were beneath the squamous epithelium and found to

From the Department of Otolaryngology, Taipei Medical University Hospital, Taipei Medical University.

Reprint requests: Fei-Peng Lee, MD, Associate Professor, Department of Otolaryngology, Taipei Medical University Hospital, 252 Wu-Hsing Street, Taipei, 110, Taiwan;

E-mail, fplee@tmu.edu.tw.

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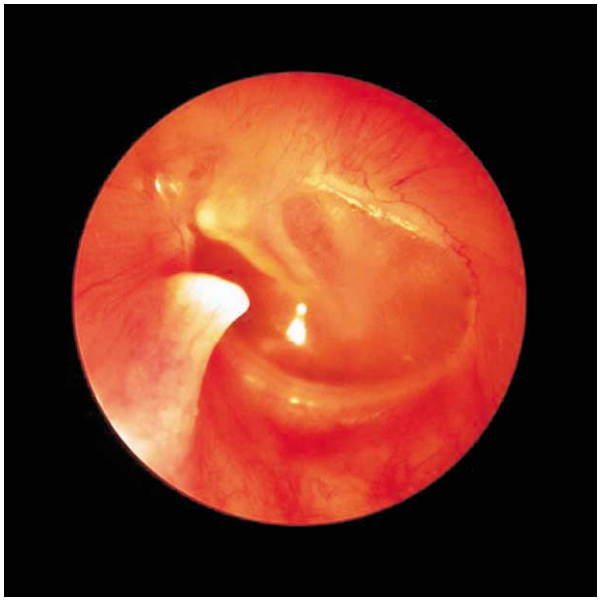

**Fig 1** A surgically and histopathologically proved horn-shaped cartilaginous choristoma in front of the handle of the malleus (case 21).

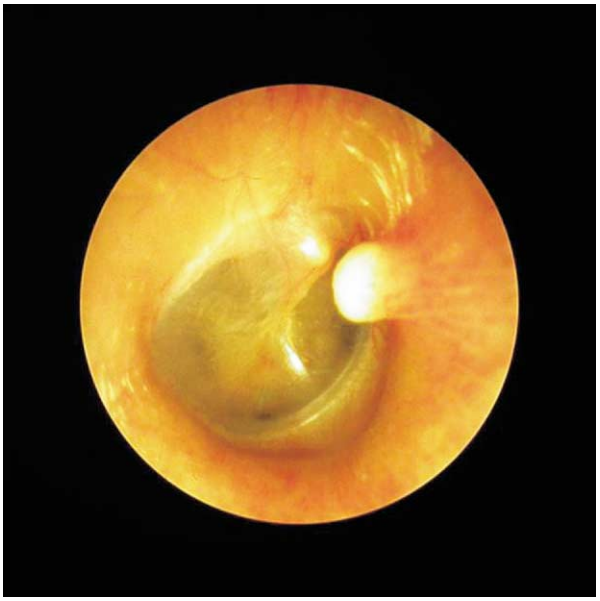

**Fig 2** A surgically and histopathologically proved club-shaped cartilaginous choristoma in front of the handle of the malleus (case 5).

be in loose contact with the periosteum. These lesions could be easily removed with a pick, needle, or elevator. The lesions in 10 cases were observed only and not excised.

In 6 cases, the small whitish pearl-like lesion in the external auditory canal, which is not a typical location for cartilaginous choristomas, was biopsied, and the biopsy revealed cartilaginous choristoma at the superior wall of the lateral part of the bony external ear canal in 1 case and epidermal cysts in the other 5.

## RESULTS

Of 36 lesions, 35 were located typically in the medial portion of the anterior wall of the bony external ear canal, just in front of the short process and handle of the malleus (Figs 1 and 2); only one lesion was found at the superior wall of the lateral part of the bony external ear canal (Fig 3). In 1 patient, the tympanic membrane and handle of the malleus of the ipsilateral ear were abnormally shaped (Fig 4). A congenital accessory lobe in the front of the tragus of the ipsilateral ear occurred in 6 patients (Fig 5) and double accessory lobes occurred in 1. The size of the excised lesions ranged from 1 to 3 mm in diameter.

Microscopically, all 26 excised lesions were composed exclusively of differentiated hyaline cartilage (Figs 6 and 7). None of these lesions had histological features of fibrosis, calcification, ossification, or malignant transformation. The lesions were initially recognized as benign chondromas; however, the diagnosis of cartilaginous choristoma was finally made.

All patients had an uneventful postoperative course with no postoperative recurrence after a mean follow-up of 2 years and 10 months (range, 3 months to 9 years).

## DISCUSSION

Chondromas can arise from skeletal or extra-skeletal soft tissue. Chondromas from skeletal structures can be classified into enchondromas and periosteal chondromas. An enchondroma is a benign hyaline cartilage neoplasm located within the medullary cavity of a bone. This lesion is most commonly reported in the small bones of the hands and feet.[7] Periosteal chondroma (ecchondroma) is a benign hy-

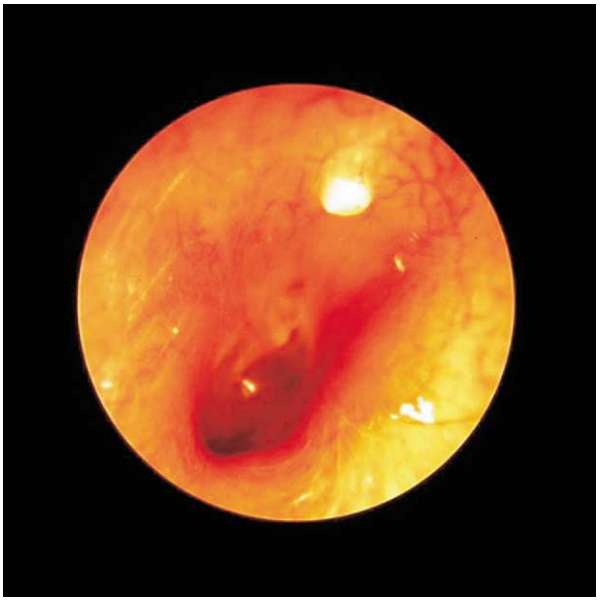

**Fig 3** A surgically and histopathologically proved cartilaginous choristoma at the superior wall of lateral part of the bony external auditory canal (case 20).

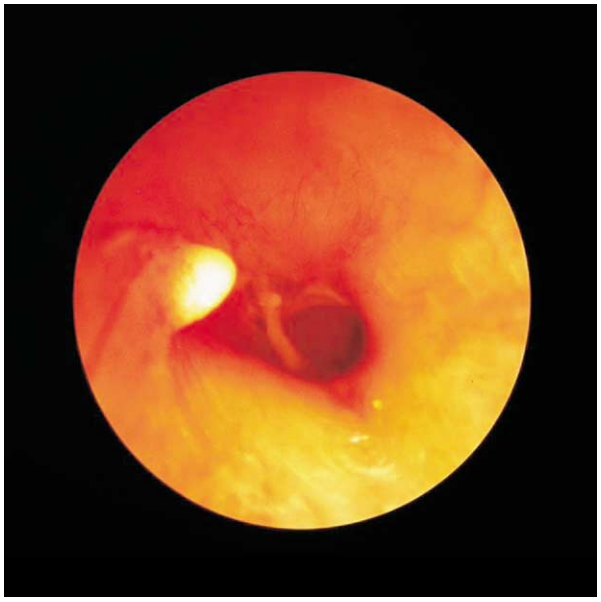

**Fig 4** A surgically and histopathologically proved cartilaginous choristoma of the bony external auditory canal concurrently associated with deformed shape of the malleus (case 14).

aline cartilage neoplasm of bone surface that arises from periosteum.[7] It always arises from the diaphyseal plate region, and preferred sites include the proximal humerus, femur, tibia, and the phalanges of the hand. Initially, the lesion is small and is located on the surface of the bone. As it enlarges, it erodes the underlying cortex in a saucer-like fashion, displaying a sclerotic rim and producing a buttress of periosteal new bone formation.[8]

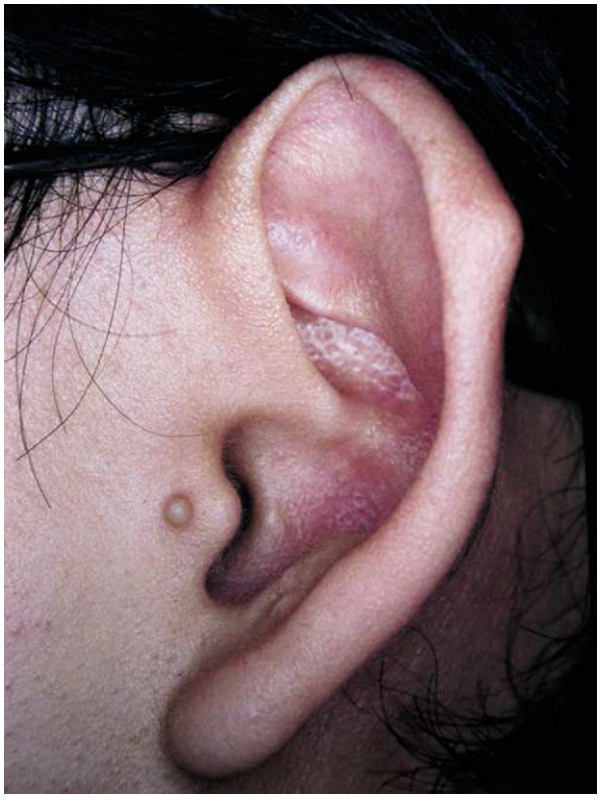

**Fig 5** A small accessory lobe was concurrently found in front of the ipsilateral tragus (case 16).

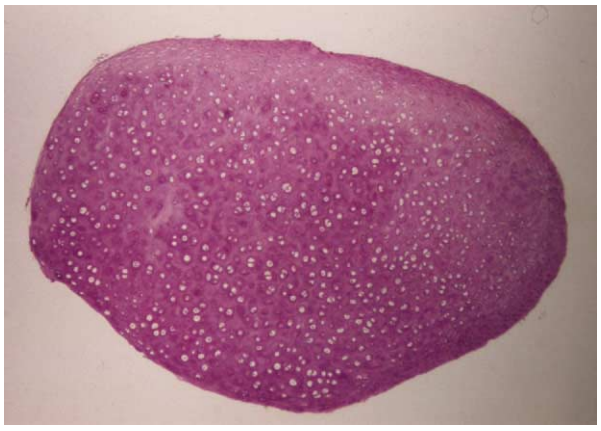

**Fig 6** Histopathology of the cartilaginous choristoma of the bony external auditory canal (case 16). The tumor consists of mature hyaline cartilage. (Hematoxylin-eosin stain, 40×.)

Soft-tissue (extra-skeletal) chondromas are benign soft-tissue tumors occurring in extra-osseous and extra-synovial locations. By definition, they do not arise in nor are they attached to these bony structures.[9] This lesion is predominantly composed of adult-type hyaline cartilage and devoid of other differentiated elements, except osseous, fibrous, and/or myxoid stroma. The tumor affects chiefly the soft tissues of the hands and feet.[7,9-11]

In the head and neck, cartilaginous tumors are seen most commonly in the larynx and in the ethmoido-sphenoid region.[12] Otherwise, chondromas may arise from tissue in which cartilage is normally not found, such as skin of the nasal bridge,[13] tonsils, thyroid, neck, oral mucosa, tongue, nasopharynx, and cerebral falx.[6,12,14,15] Doubts exist about the true nature of these lesions (ie, whether they are true chondromas, choristomas, or harmatomas).[6]

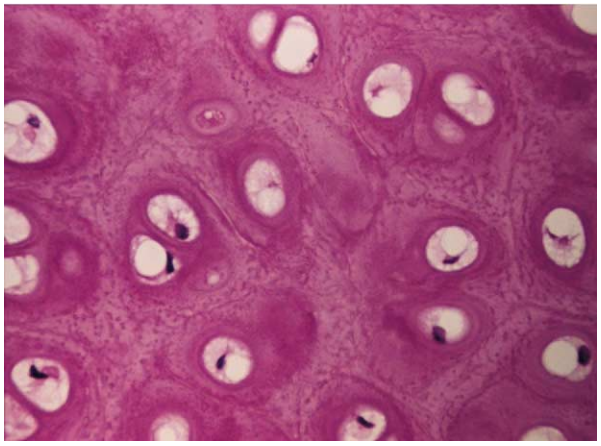

**Fig 7** Histopathology of the cartilaginous choristoma of the bony external auditory canal (case 16). The tumor consists of mature hyaline cartilage. (Hematoxylin-eosin stain, 200×.)

Choristoma is defined as a tumor-like growth of normal tissue occurring in an abnormal location.[6] The author assumed that such cartilaginous lesions in the bony external auditory canal were, for a variety of reasons, cartilaginous choristomas rather than chondromas. These reasons were:

1. Histopathological examination of the lesion revealed a tumor-like mass of normal chondrocytes. Cartilaginous tissue is not normally found in the bony external auditory canal.[16] This cartilaginous mass arises from tissue in which cartilage is normally not found, and thus it fits the definition of a cartilaginous choristoma.
2. All the excised lesions were located between the squamous epithelium and periosteum of the bony auditory canal. They could be easily removed with a pick, needle, or elevator. The lesion appeared to be in contact only with the periosteum but appeared not to arise from the periosteum. Possibly, the lesion is an early stage of periosteal chondroma. However, periosteal chondroma may continue to grow after skeletal maturation, and some lesions may attain a large size of up to 6 cm.[8] In this study, the size of the lesion generally ranged from 1 to 3 mm in diameter and no progressive enlargement of tumors was observed in nonoperated cases.[1] Also, no erosion of the periosteum of the bony external ear canal was found. Since the lesion has very limited growth potential and seldom exceeds 4 mm in diameter,[1] its neoplastic nature is doubtful and therefore, the name cartilaginous choristoma rather than chondroma is more appropriate.[17]
3. Most of the lesions were asymptomatic. This is a characteristic of choristomas.
4. Seven (19%) of these 36 cases had similar congenital external ear malformation of the accessory lobe, which typically is located in front of the tragus of the ipsilateral ear. This feature suggests the possibility that the lesion originates from similar embryological developmental faults.

Some authors support the opinion that a chondroma is a harmatoma. However, in this series the term "harmatoma" is not recommended since it is limited to simple and spontaneous growths composed exclusively of components derived from local tissue.[15]

Embryologically the external ear canal develops from the first branchial groove, and the tragus is formed by contributions from the first branchial arch.[18] In this series, 7 of these 36 (19%) patients had a similar congenital external ear malformation of the accessory lobe located in front of the tragus of the ipsilateral ear. One patient had an ipsilateral congenitally malformed handle of the malleus. Embryologically, the handle of the malleus is formed by contributions from Reichert's cartilage of the second branchial arch.[18] Moreover, most of the cartilage of branchial origin is hyaline. These lesions may have developed from heterotopic cartilaginous cell nests in Meckel's cartilage of the first branchial arch or Reichert's cartilage of the second branchial arch. But, since it has been impossible to follow the embryological development of these lesions, their true origin remains speculative.

Although 19% of these patients also had an accessory ear lobe, these lesions probably do not represent ectopic external ear cartilage. This conclusion is based largely on the finding that this lesion is composed of easily recognizable hyaline cartilage, and that the cartilaginous components of the external helix and accessory auricles are composed of elastic cartilage.[13]

Among 47 cases of cartilaginous choristoma of the bony auditory canal, including 11 published cases by other authors[1-5] and the 36 cases in this study, the patients' ages ranged from 5 to 70 years with average of 34 years. Most cases had been diagnosed in patients between the ages of 20 and 49. The right ear was affected in 25 patients and left ear in 22 patients. Twenty-six were males and 21 were females. Forty-five (95%) occurred typically in the medial portion of the anterior wall of the bony external ear canal, just in front of the short process and handle of the malleus. Only 2 occurred at the superior and posterior wall of the bony external ear canal, but none were found in the inferior wall. These lesions have been reported to be 1 to 4 mm in diameter and located 1 to 3 mm lateral to the drum.[1] The mechanism of the exclusive occurrence in front of the short process and handle of the malleus is not well understood. Generally, there were no clinical manifestations, and the lesion was found incidental to routine otoscopic examination, but it occasionally presented with repetitive external otitis.[2]

When the lesion occurs at the typical site (ie, the medial portion of the anterior wall of the bony external auditory canal just in front of the short process of the malleus), the differential diagnosis of exostosis and keratoma should be included. Diffuse exostoses are broad-based hyperostotic lesions, usually bilateral and symmetric, and found in patients with a history of frequent participation in aquatic activity.[19]When the lesion is not located at the typical site, the differential diagnosis of keratoma, fibroma, and osteoma should be included. Excisional biopsy and pathological examination are required for definitive diagnosis. Because it is small and asymptomatic, the lesion is easily misdiagnosed as an early-stage exostosis and a patient with this lesion is generally treated with observation alone instead of surgical excision. Its true incidence may be underestimated. Histologically, the cartilaginous choriostoma of the bony external ear canal consists of a well-circumscribed mass of mature hyaline cartilage.

Of 36 lesions in this series, 26 (72%) were excised and histopathologically examined to verify this new clinical entity. However, these lesions are benign and most cases do not cause symptoms. Moreover, no progressive enlargement of tumors was found in nonoperated cases.[1] These lesions can remain under observation until they cause symptoms, when excision is indicated. Cartilaginous choristoma can be successfully excised under oto-microscopic guidance with-

out traumatic injury to the tympanic membrane. In our experience, use of a video telescope for excision of such lesions is also an effective, safe, and convenient alternative guidance strategy for this surgical technique.

No recurrence of adequately excised lesions has been reported in the literature. Notably, chondrosarcomas can appear in the temporal bone.[20] To our knowledge, malignant changes in cartilaginous choristomas of the bony external auditory canal have never been reported.

## CONCLUSION

The presence of a heterotopic cartilaginous mass in the bony external ear canal is not as rare as it once was thought to be. The descriptive term "cartilaginous choristoma" rather than "chondroma" may be more appropriate for these lesions.

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