

이개연골에 발생한 가성낭종 1예

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A Rare Case of Auricular Endochondral Pseudocyst

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= Abstract =

Auricular endochondral pseudocyst is a very rare, benign intracartilaginous cystic lesion which most commonly presents as a cystic mass in the anterior plane of the auricle. We present a case report of a 48-year-old man with a fluctuating lesion of 3 week's duration on the left auricle, with no specific history of trauma or disease. Initial incisional drainage revealed an abundance of serous fluid, which quickly recurred. Surgical removal of the hypertrophic perichondrium forming the pseudocyst anterior wall and ear cartilage curettage was carried out with intraoperative absolute alcohol sclerotherapy, followed by compression dressings. The auricle healed uneventfully, with a good final cosmetic result and no recurrence within a 6-month follow-up period. We report this unusual case as the first in Korean plastic surgery with a review of the literature.

Key Words : Auricular endochondral pseudocyst, Benign idiopathic cystic chondromalacia

INTRODUCTION

Endochondral pseudocyst of the auricle, first reported in 1966, is a very rare, benign cystic lesion of unknown etiology that presents as spontaneous, asymptomatic swelling over the anterior aspect of the auricle, commonly in the scaphoid or triangular fossa.¹⁾ Usually unilateral, the lesions are normally 1~5 cm in diameter, with intracartilaginous accumulation of a clear or yellowish sterile viscous fluid. This condition has a tendency to occur in young, healthy Asian males, especially of Chinese ethnicity.^{1,2)} Diagnosis

is confirmed by correlation of clinical findings and histologic features, which appear as a lack of epithelial lining of the cyst and no other distinct pathognomonic findings.³⁾ Several treatments have been suggested in the literature, including simple aspiration, intralesional sclerotherapy and incision with drainage.^{3,4)} However, because recurrences are frequent and usually leave unsatisfactory results, improper therapies may cause poor outcomes. We present a case of endochondral pseudocyst of the auricle, which was successfully treated by surgical excision, sclerotherapy and compression dressings.

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CASE REPORT

The patient, a 49 year-old male, presented with a painless lesion on the left auricle of approximately 3 weeks' duration. He had no history of previous trauma or disease on the left ear, and was in good general condition. The

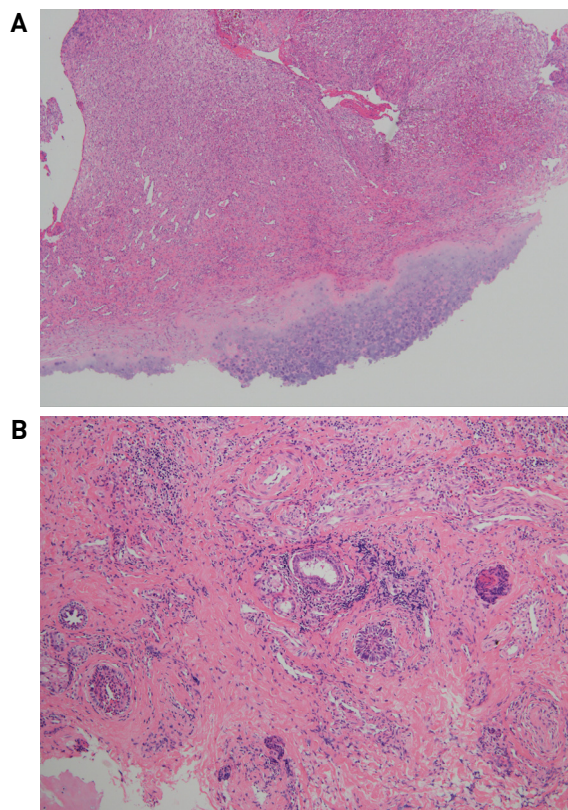


Fig. 3. Cross section of pseudocyst (A) (H&E, X 40) (B) (H&E, X 100) Microscopic histopathology demonstrated no epithelial cell layer lining the inner surface, with chronic inflammatory cell infiltration and neovascularization.

lesion appeared as a 2.4 cm swollen, fluctuating mass on the scaphoid fossa of the left auricle. Initial incisional drainage performed as an outpatient procedure revealed an abundance of serous fluid inside a collapsible cavity with egg-shell-like walls of a crispy consistency. Notwithstanding the compression dressing applied, the lesion was once again bulging with fluid 2 days later (Fig. 1).; a review of the literature led to suspicion of auricular endochondral pseudocyst and a decision to operate. Under local anesthesia, surgical exploration was performed by extending the previous incision. This exposed the anterior wall of the pseudocyst including hypertrophic perichondrium and the posterior wall formed of ear cartilage, with some granulation tissue inside the cavity (Fig. 2-A, 2-B). The anterior wall was separated from the overlying skin and surgically excised, and the posterior cartilage wall was curetted. To further decrease the risk of recurrence, the posterior wall of the surgical site was flushed with 2 cc of absolute alcohol for intraoperative sclerotherapy (for the purposes of additional posterior wall curettage and cavity obliteration), after which the alcohol was removed and the site thoroughly irrigated with normal



Fig. 1. Preoperative clinical view Initial incisional drainage led to fluid re-accumulation and recurrence in just 2 days.

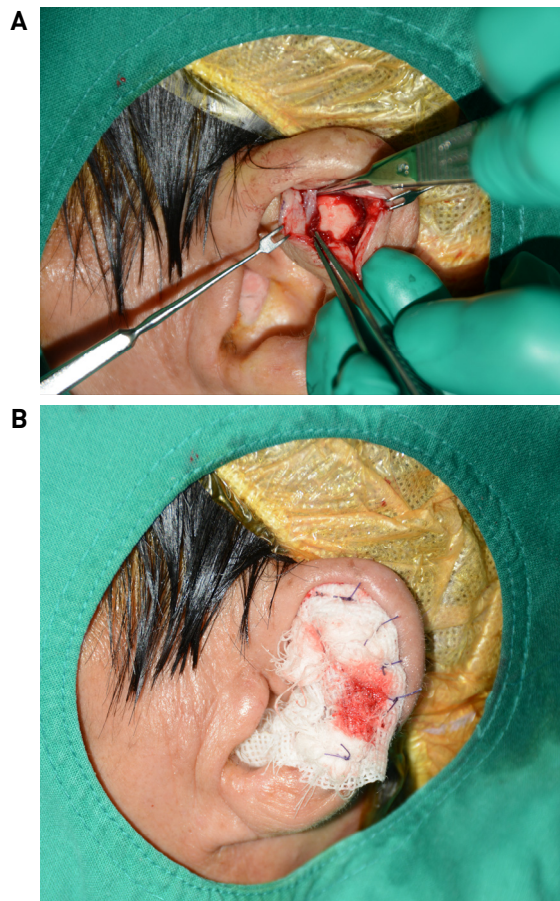


Fig. 2. Intraoperative view (A) Surgical exploration exposed hypertrophic perichondrium of the anterior wall (held in upper forceps) and posterior wall ear cartilage with granulation tissue (indicated by lower forceps) in between. (B) Simple skin repair was followed by a tie-over compression dressing using pull-out sutures.

saline. After repairing the skin incision, a tie-over compression dressing was applied with pull-out sutures and



Fig. 4. Postoperative clinical view after 6 months. Surgical wounds healed uneventfully, with a good final cosmetic result. No recurrence appeared within a 6 month follow-up period.

maintained for 5 days followed by conventional gauze compression dressing for a week. Histologic examination showed no epithelial cell lining of the cavity walls, chronic inflammatory cell infiltration and neovascularization, confirming the diagnosis (Fig. 3-A, 3-B). Surgical wounds healed uneventfully with a good final cosmetic result and no recurrence within a 6-month follow-up (Fig. 4).

DISCUSSION

Pseudocyst of the auricle, a cystic condition of the auricle lacking epithelial lining, was first histologically examined and coined by Engel among Chinese living in Hong Kong.¹⁾ This condition has been especially well-documented among Chinese males, with several authors such as Cho, Zhu and Tan emphasizing the relatively high occurrence in that ethnicity group.⁵⁻⁷⁾ First reported in Korea in 1996 by dermatologists presenting 3 cases and subsequently in 2007 by otolaryngologists, this lesion has been named as endochondral pseudocyst, intracartilaginous cyst and benign idiopathic cystic chondromalacia.^{4,8)} Because of its rare incidence, clinicians may easily be unaware of this condition and misdiagnose it, delaying proper management with disfiguring consequences.

The precise etiology is unknown, but several pathogenic hypotheses have been suggested. Engel hypothesized that the release of lysosomal enzymes causes damage to the car-

tilage, resulting in autolysis and degeneration.¹⁾ However, later analysis of cyst fluid failed to isolate such lysosomal enzymes.⁵⁾ Another hypothesis postulates a congenital embryologic dysplasia of the ear cartilage as the subjacent disorder in the formation of an intracartilaginous cystic lesion. The cadaveric dissection of normal ears done by Zhu and Wang supports this theory.⁶⁾ But no anatomic examination of affected ears has assured this pathogenic relationship. Chronic repetitive minor trauma, such as sleeping on hard pillows or wearing headphones or helmets, which unbeknownst to the patient may cause perichondrial ischemia and cartilage deformation, could have been closely associated with pseudocyst formation considering a few reports related to acute events.³⁾ Lim et al also proposed an inflammatory reaction etiology, based on findings of lymphocyte and monocyte infiltration in 16 cases.²⁾

Diagnosis of endochondral auricular pseudocyst is mainly based on clinical features and routine histology. The fluid composition is nondiagnostic, therefore, analysis of the aspirate is not required. The differential diagnosis may include inflammatory cartilage disorders such as chondrodermatitis helices, subperichondrial hematoma and relapsing perichondritis, but they can be discerned from clinical characteristics. Unlike other subperichondrial auricular diseases, the histological characteristics of auricular endochondral pseudocyst are thinned cartilage, an intracartilaginous cystic lesion without epithelial lining, hyalinizing degeneration along the cystic space border and granulation tissue suggesting a reparative or regenerative process.

Treatment should achieve not only successful resolution without recurrence but also preservation of normal structure and aesthetic appearance of the auricle.^{3,8)} If left untreated, it can damage and deform the auricle. Prior studies have suggested various methods of treatment, with needle aspiration or incision and curettage both commonly used; compressive dressings with buttoning or bolstering should be applied to lower the high recurrence rate of such simple treatments and prevent cartilage hypertrophy. The efficacy of systemic steroid treatment is uncertain and carries the risk of complications.^{8,9)} Intralesional sclerotherapy with corticosteroids or iodine has also been used after aspiration, but with very high recurrence rates. Moreover, steroid injection can result in skin pigmentation and cartilage deformation.^{3,4,9,10)} Absolute ethanol, widely used in cavitary

diseases, was chosen as a post-excision sclerosant in this case for its efficacy in tissue conglutination and cavity obliteration with little side effects.¹¹⁾ Deroofing with removal of the anterior cartilaginous plate of the pseudocyst while preserving the posterior wall with curettage is probably the most effective treatment method with minimal recurrence. By maintaining the posterior aspect of the pseudocyst, the structural frame of the auricle is retained with the intact remnant wall preventing the “floppy ear” deformity.¹²⁾

We hereby report a case of auricular endochondral pseudocyst, the first recognized by Korean plastic surgeons, with a review of the literature. This is a very rare lesion and is not easily diagnosed without clinical experience, especially as it has only been reported among Otolaryngologists and Dermatologists, whereas other specialists or clinicians such as General Surgeons and Plastic Surgeons also commonly see patients with ear lesions. With various treatment options and a notorious tendency to recur, we achieved an excellent outcome by combining surgical deroofing, sclerotherapy and compressive dressing. Notwithstanding the rare prevalence, clinicians should take into account the possibility of auricular pseudocyst while treating ear lesions to accomplish satisfying results.

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