

CASE REPORT

Bilateral pseudocyst of the auricle

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Pseudocyst of the auricle is characterized by asymptomatic swelling caused by an intracartilaginous fluid accumulation. If left untreated, permanent deformity of the pinna may occur. Many modalities of treatment have been reported. This article summarizes a 2 year retrospective study (2005–2007) performed in the Children Teaching Hospital in Bratislava. This study presents two children who had bilateral pseudocyst of the auricle, and who were treated with different types of surgical treatment and reviews the literature. Purpose of this study is to compare the effectiveness of the two different methods of surgical treatment. The recognition of the ideal way of surgical method of treatment may lead to standardized approach, which can result in successful repair of the auricle with no recurrence (Fig. 4, Ref. 16). Full Text (Free, PDF) www.bmj.sk

Key words: auricular pseudocyst, endochondral pseudocyst, intracartilaginous cyst.

Pseudocyst of the auricle is a benign idiopathic, uncommon condition, which typically presents as a painless swelling over the anterior aspect of the auricle (6). Although it was first reported in Chinese population, persons of all racial groups have been affected, and it is predominantly found in males compared to females (13).

Clinically a pseudocyst manifests as benign, no inflammatory, asymptomatic swelling on the lateral or anterior surface of the auricle, usually in scaphoid or triangularis fossa. It ranges usually in size from 1–5 cm in diameter and contains a clear or yellowish fluid (8).

The etiology of pseudocyst of the auricle is unknown, but two main theories have been reported. The first theory suggests that an auricular pseudocyst often results after repeated minor low-grade trauma, such as rubbing, ear pulling, sleeping on hard pillows, or wearing a motorcycle helmet or earphones. It was suggested that chronic trauma induced cartilage degeneration resulting in progressive dilatation and formation of a cystic space within the auricular cartilage (4). However, analysis of pseudocyst contents revealed a fluid rich in albumin and acid proteoglycans and cytokines, but lacking lysosomal enzymes. Recent analysis of cytokine profile of the fluid indicates markedly elevated levels of interleukin IL-6, which is believed to stimulate chondrocyte proliferation. IL-1, an important mediator of inflammation and cartilage destruction, induces IL-6. IL-1 also stimulates chondrocytes to synthesize proteases and prostaglan-

din E2 while inhibiting the formation of extracellular matrix components (14).

In support of this traumatic etiology, elevated serum lactic dehydrogenase (LDH) values have been reported within the pseudocyst fluid. Two of the elevated isoenzymes, LDH-4 and LDH-5, are proposed as major components of auricular cartilage. These enzymes may be released from auricular cartilage degenerated from repeated minor trauma (10).

The second theory hypothesized that congenital embryologic defect of the auricular cartilage is the predisposing factor in the development of the pseudocyst. The auricle is developed from the first and second branchial arch, and congenital maldevelopment of this branchial arch may result in residual tissue planes within the cartilage which may reopen, giving rise to a pseudocyst (7, 9). Others studies showed, that the auricular pseudocyst might represent an autoimmune disease involving the auricle. The possible mechanism may be an accumulation of reactive exudate

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Fig. 1. Pseudocyst on the right auricle (case 1).



Fig. 3. Pseudocyst on the right auricle (case 2).



Fig. 2. Pseudocyst on the left auricle (case 1).



Fig. 4. Pseudocyst on the left auricle (case 2).

due to immunoreactions of autologous antibody and auricular tissue (2, 3). Other newest study postulates a hypothesis in which an inflammatory response manifesting as perivascular mononuclear infiltrates of predominantly lymphocytes is the cause of the development of this condition. After this triggering inflammatory response on the auricular cartilage, these inflammatory cells release inflammatory cytokines, resulting in a cleavage in the auricle cartilage. An inflammatory fluid slowly accumulates within the cartilage, lifting the anterior segment of the auricular cartilage from its posterior segment. Later on, this inflammatory process slowly settles and is replaced by the histological evidence of perichondrial fibroblastic response and granulation tissue formation (9).

Histology usually reveals an intracartilaginous cyst devoid of epithelial lining containing clear or yellowish fluid, and there are no pathognomic features. In some cases it was found that there is an inflammatory response mediated by perivascular inflammatory response observed in the specimens (9, 15).

The goal of treatment is preservation of anatomical architecture and prevention of recurrence. There are two types of surgical procedure, simple aspiration followed by a pressure dressing applied over the pinna, this procedure has resulted in frequent recurrence (11), and the second successful procedure is surgical incision and drainage of the lesion with excision of affected cartilages and the application of pressure dressing (1, 5, 12, 16).

Case reports

Case 1

A one month baby boy was admitted to our department, presenting with bilateral auricular cystic mass, which was firm, not tender, and the skin over it was normal (Figs 1 and 2). The patient underwent the next day a simple needle aspiration of the cyst under the effect of sedation (midazolam). In the left auricle 2 ml of clear yellowish fluid were aspirated, and in the right about 1 ml of a clear fluid.

On the next day the fluid was re-accumulated and 2 ml thin bloody fluid was re-aspirated and 0.3 ml of 96 % alcohol was injected bilaterally.

After 2 days the patient state has deteriorated and fluid re-accumulation with signs and symptoms of infection (fever, redness of the cyst and pain) developed. Under general anesthesia incision and evacuation were performed, on the left side there was a bloody thick turbid fluid and also there was a granulation tissue which was removed. On the right side incision was done and there was no fluid but there was a granulation tissue which was also removed and pressure dressing was applied. The fluid was sent for microbiological examination; it was negative in the right ear, in the left ear it was positive for *Staphylococcus aureus*. The patient had complete resolution of symptoms with no recurrence.

Case 2

A 9-year old boy was admitted to our department with bilateral cyst of the auricle. The condition started one month prior to admission, the general condition of the child was good, the local examination revealed, in the right ear a cystic mass on the helix particularly in the scaphoid fossa, 1–2 cm in diameter, not tender, skin over it without signs of inflammation (Fig. 3). In the left ear in the crura of the helix there was also cystic mass 1 cm in diameter, not tender (Fig. 4).

The child underwent surgical excision in total anesthesia, with removal of the anterior cartilaginous part of the lesion, and this part was sent to histological examination. A clear yellowish fluid obtained was sent for microbiological examination with negative results of bacterial growth. This procedure was followed by application of pressure dressing. The result was good with no recurrence and with good cosmetic outcome.

Histological examination revealed that there was a nodular lesion consisting of a mature hyaline cartilage. The perichondrial tissues seemed to be thick fibrotic and highly vascularised, there were multiple chondrocytes. Histological conclusion was that the surgically removed tissue represents a mature cartilaginous tissue. The patient had complete resolution of symptoms with no recurrence.

Discussion

Clinically pseudocyst of the auricle manifests as a benign, non-inflammatory, painless swelling on the lateral or anterior surface of the pinna.

In our study, both of our patients presented with a swelling in the anterior aspect of the auricle and none of them had complained of pain, neither a history of acute or chronic trauma of the ears. Others report that bilateral lesions are rare and usually present metachronously. An interesting finding in our study is that both of the patients presented with bilateral lesions and both of them had synchronous appearance. All these findings suggest a systemic pathogenesis in the sense of either congenital or autoimmune etiology of the disease.

The characteristic histological feature of the pseudocyst of the auricle is an intracartilaginous cystic space with no epithelial lining. There is often some granulation tissue and fibroblastic response lining the cystic space and inflammatory response indicated by the presence of perivascular infiltration of leukocytes. In our series one patient had needle aspiration, other patient underwent excision of the cartilage and pressure dressing procedure, histological specimen (a part of cartilage with fluid) was sent, and the findings were mature cartilaginous tissue. The perichondrial tissue seemed to be thickened, fibrotic and highly vascularised, there were multiple chondrocytes – all the findings might correlate with inflammatory theory.

Restoration of the normal architecture of the auricle with no recurrence is the main goal of the treatment. Treatment options reported in the literature include the use of high-dose oral corticosteroids, intralesional corticosteroids, simple aspiration with compression dressing, and surgical excision with compression

dressing (1, 5, 16). Patients involved in our series were small in number (2 patients) and it would be difficult to interpret the effectiveness of all types of treatment. In our study we compare two approaches – a simple aspiration and surgical excision with pressure dressing. Needle aspiration of pseudocyst always results in re-accumulation of the pseudocyst. This observation is evident in one patient who had this procedure with re-accumulation after one day, and when the aspiration was repeated the cyst was also re-accumulated and got infected and the patient general state was deteriorated and he developed signs of infection. The patient finally had an incision and evacuation of the pseudocyst without a removal of anterior part of auricular pseudocyst, and with application of pressure dressing. The outcome was good with complete resolution and with no recurrence.

Definitive treatment option is surgical excision of the anterior segment of the auricular pseudocyst followed by pressure dressing. In our report the patient who had this procedure had a good outcome with no re-accumulation or infection and with cosmetically good results.

Conclusion

Pseudocyst of the auricle is a benign condition of the anterior or lateral wall of the pinna with unknown etiology. It can occur in all races but predominantly in Chinese or white male. Several treatment methods have been reported with variable results. According to our experience, surgical excision of the anterior cartilage with pressure dressing yields excellent results.

References

1. **Chang CH, Kuo WR, Wabg LF, Ho KY, Tsai KB.** Deroofing surgical treatment for pseudocyst of the auricle. *J Otolaryngol* 2004; 33 (3): 177–180.
2. **Chen Q, Fei Y, Zhao T, Luo D, Wu B, Yang X.** Research on the Immunological Cause of Auricular Pseudocyst. *Zhonghua Er Bi Yan Hou Ke Za Zhi* 2001; 15 (7): 304–305.
3. **Chen Q, Zhao T, Yang X.** The Immunological Cause of Auricular Pseudocyst. *Zhonghua Er Bi Yan Hou Ke Za Zhi* 1999; 34 (4): 236–237.
4. **Cohen PR, Grossman ME.** Pseudocyst of the auricle. Case report and world literature review. *Arch Otolaryngol Head Neck Surg* 1990; 116 (10): 1202–1204.
5. **Cohen PR, Katz BE.** Pseudocyst of the auricle: successful treatment with intracartilaginous trichloroacetic acid and button bolsters. *J Dermatol Surg Oncol* 1991; 17 (3): 255–258.
6. **Engel D.** Pseudocysts of the auricle in Chinese. *Arch Otolaryngol* 1966; 83: 197–202.
7. **Hoffmann TJ, Richardson TF, Jacobs RJ et al.** Pseudocyst of the auricle. *J Dermatol Surg Oncol* 1993; 19: 259–262.
8. **Li LJ, Elenitsas R, Bondi E.** Off-center fold. Noninflammatory, fluctuant swelling of the ear. Pseudocyst of auricle. *Arch Dermatol* 2001; 137 (5): 657–662.
9. **Lim L Ming LC, Hong GY, Shuen CS.** Pseudocyst of the auricle: A histological perspective. *Laryngoscope* 2004; 114 (7): 1281–1284.

10. **Miyamoto H, Okajima M, Takahashi I.** Lactate dehydrogenase isozymes in and intralesional steroid injection therapy for pseudocyst of the auricle. *Int J Dermatol* 2001; 40 (6): 380—384.
11. **Ophir D, Marshak G.** Needle aspiration and pressure sutures for auricle pseudocyst. *Plast Reconstr Surg* 1991; 87 (4): 783—784.
12. **Secor CP, Farrell HA, Haydon RC 3rd.** Auricular endochondral pseudocysts: diagnosis and management. *Plast Reconstr Surg* 1999; 103 (5): 1451—1457.
13. **Tan BY, Hsu PP.** Auricular pseudocyst in the tropics: a multi-racial Singapore experience. *J Laryngol Otol* 2004; 118 (3): 185—188.
14. **Yamamoto T, Yokoyama A, Umeda T.** Cytokine profile of bilateral pseudocyst of the auricle. *Acta Derm Venereol* 1996; 76 (1): 92—93.
15. **Zhu L, Wang X.** Histological examination of the auricular cartilage and pseudocyst of the auricle. *J Laryngol Otol* 1992; 106: 103—104.
16. **Zhu LX, Wang XY.** New technique for treating pseudocyst of the auricle. *J Laryngol Otol* 1990; 104 (1): 31—32.

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