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# Pseudocyst of the auricle in patients with movement disorders: report of two patients with ataxia-associated auricular pseudocysts

Bryce D. Beutler[1], Philip R. Cohen[2]

**1** University of Nevada School of Medicine, Reno, NV, USA
**2** Department of Dermatology, University of California San Diego, San Diego, CA, USA

**Key words:** ataxia, auricle, dyskinesia, ear, Friedreich's, neurological, pinna, pseudocyst, spasticity, spinocerebellar, trauma

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**Corresponding author:** Philip R. Cohen, MD, Department of Dermatology, University of California San Diego, San Diego, CA, 92093, USA. Email: mitehead@gmail.com

## ABSTRACT

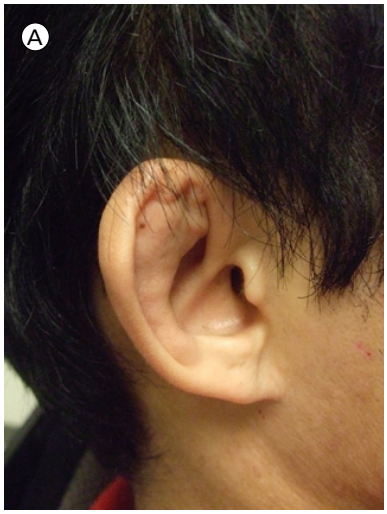
**Background:** Pseudocyst of the auricle is a benign condition of the ear characterized by an asymptomatic, noninflammatory swelling on the lateral or anterior surface of the auricle. It typically presents as a 1 to 5 centimeter cystic lesion located within the scaphoid or triangular fossa. In most patients, the lesion develops spontaneously. However, pseudocyst of the auricle has also been associated with trauma to the ear.

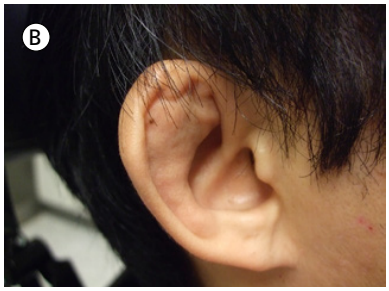
**Purpose:** We describe the clinical findings of two men who developed pseudocyst of the auricle associated with ataxia-induced trauma to their ear. We also summarize the differential diagnosis, the postulated pathogenesis, and the treatment options for this condition.

**Materials and methods:** The features of two men with pseudocyst of the auricle are presented. Using PubMed, the following terms were searched and relevant citations assessed: ataxia, auricle, dyskinesia, ear, Friedreich's, neurological, pinna, pseudocyst, spasticity, spinocerebellar, and trauma. In addition, the literature on pseudocyst of the auricle is reviewed.

**Results:** Pseudocyst of the auricle was observed in two men with neurological disorders: a 33-year-old Asian man with spinocerebellar ataxia and a 47-year-old Caucasian man with Friedreich's ataxia. Each patient had a history of ataxia-induced head and ear trauma. The clinical features of the lesions were sufficient to establish a diagnosis of pseudocyst of the auricle. Neither patient desired treatment.

**Conclusion:** Pseudocyst of the auricle is a benign cystic lesion that is occasionally precipitated by trauma to the affected ear. Patients with neurological disorders, particularly those associated with ataxia and/or dyskinesias, may have an increased risk of developing the traumatic variant of the condition. Diagnosis can usually be established by clinical presentation. However, in some patients, a tissue specimen may be secured for microscopic evaluation to exclude infection or during surgical repair. Various treatment options exist for pseudocyst of the auricle, including: (1) needle aspiration—with or without subsequent injection of an irritant substance—followed by a pressure dressing and (2) surgical deroofing. Alternatively, reassurance of the benign nature of the condition and observation is a reasonable management alternative.



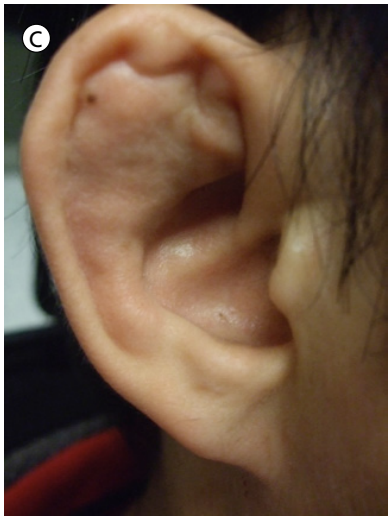




**Figure 1 (a, b, and c).** Distant (a), intermediate (b), and close (c) views of the right ear show a 3.0 x 2.0 centimeter pseudocyst of the auricle in a 33-year-old Asian man with spinocerebellar ataxia. [Copyright: ©2015 Beutler et al.]

## Introduction

Pseudocyst of the auricle is a benign, noninflammatory, asymptomatic cystic lesion of the ear. It most commonly develops progressively over a 4- to 12-week period in the scaphoid or triangular fossa. The lesion may, or may not, be associated with trauma to the affected ear. Patients with neurological disorders that are associated with motor dysfunction, such as ataxias, may be at a heightened risk for developing trauma-related pseudocyst of the auricle.

We describe two men who developed pseudocyst of the auricle after sustaining ataxia-associated head and ear injuries. We also review the differential diagnosis, the postulated pathogenesis, and the treatment options of pseudocyst of the auricle.

## Case reports

***Case 1.*** A 33-year-old Asian man with spinocerebellar ataxia (which clinically became symptomatic 10 years earlier) presented for evaluation of a nontender swelling on his upper right ear that was present for one month. He had poor motor control of his movements and had repeatedly hit his ear against the head support on his motorized wheelchair. Cutaneous examination revealed a firm cystic nodule measuring 3.0 x 2.0 cm on the anterior surface of the auricle of his right ear. The lesion was asymptomatic, flesh-colored, and had progressively enlarged over the previous four weeks (Figure 1). Based on correlation of the clinical presentation and distinct lesion morphology, a diagnosis of pseudocyst of the auricle was established. Since he was at continual risk of repeatedly traumatizing his ear, no intervention was performed to his auricular pseudocyst.

***Case 2.*** A 47-year-old Caucasian man presented for evaluation of a cystic lesion on his left ear. Six months prior, the lesion was interpreted as an abscess with cellulitis and he had undergone an incision and drainage with adjuvant systemic antibiotic therapy. However, the swelling had recently returned. His past medical history was significant for Friedreich's ataxia. He permanently resides in a skilled nursing facility secondary to his ataxia-associated difficulty in motor movements. He has a history of hitting his head on the rails of his bed.

Cutaneous examination revealed a 4.0 x 2.0 cm cystic lesion involving primarily the crura of antihelix of his left ear. It was asymptomatic and noninflammatory (Figure 2). The lesion was initially soft to the touch; however, it had become firm. Based on correlation of the clinical presentation and lesion morphology, a diagnosis of pseudocyst of the auricle was established. Since he was at continual risk of repeatedly traumatizing his ear, no intervention was performed to his auricular pseudocyst.

## Discussion

Pseudocyst of the auricle was first described by the German physician Arthur Hartmann in 1846 [1]. However, the condition did not appear in the English-language literature until 1966, when Engel observed the lesion in Chinese men [2]. One year later, Hansen reported several Caucasian patients who had pseudocyst of the auricle [3].

The initial observations made by Hartmann, Engel, and Hansen remain relevant today. Engel and Hansen both noted that the condition occurs predominantly in men [2,3]. Indeed, in a 1990 world literature review of 114 cases of pseudocyst of the auricle, 93% of auricular pseudocyst patients were men [4]. Their findings were later corroborated by Lim et al., who reviewed the medical records of 41 patients who were treated for pseudocyst of the auricle in Singapore and concluded that 87% were men [5]. The age of onset typically ranges

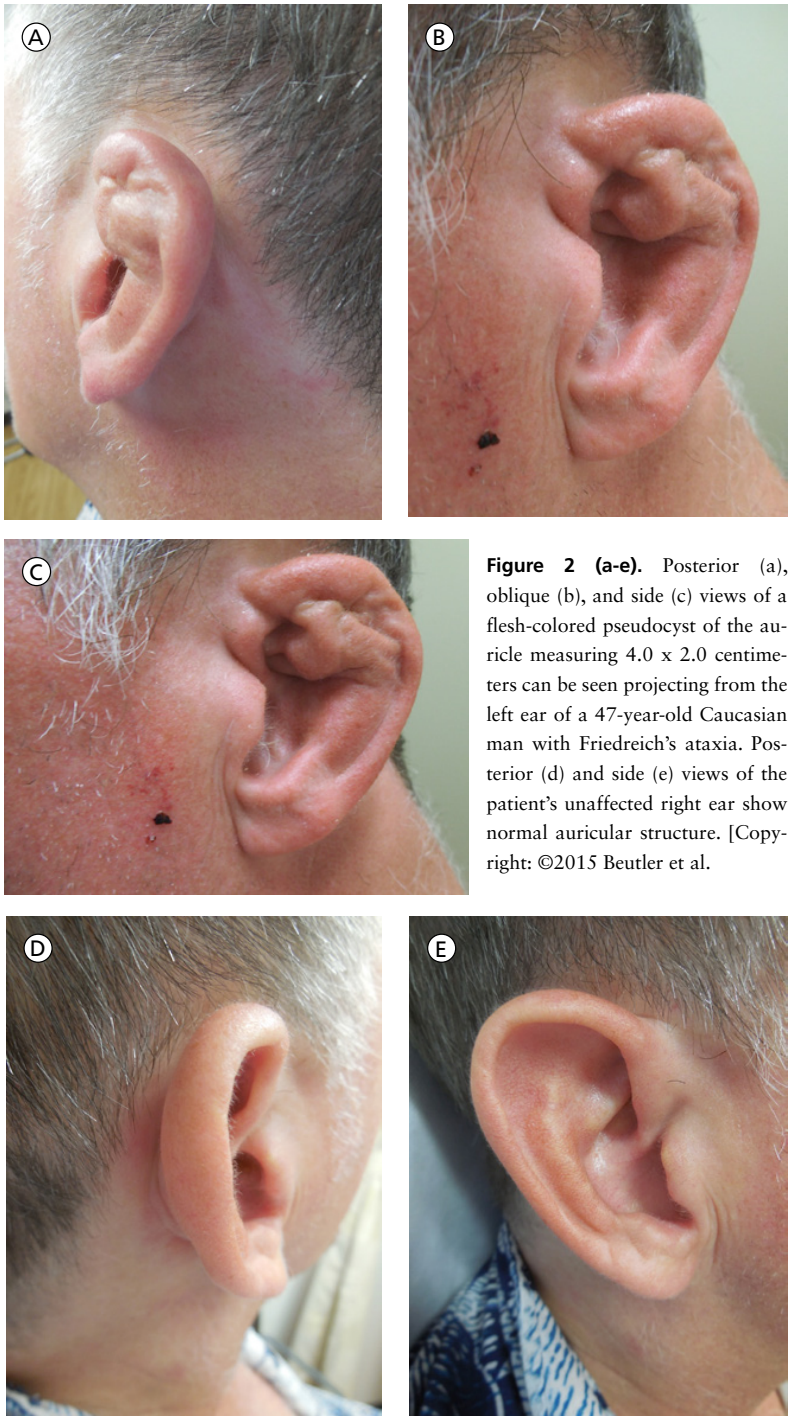



**Figure 2 (a-e).** Posterior (a), oblique (b), and side (c) views of a flesh-colored pseudocyst of the auricle measuring 4.0 x 2.0 centimeters can be seen projecting from the left ear of a 47-year-old Caucasian man with Friedreich's ataxia. Posterior (d) and side (e) views of the patient's unaffected right ear show normal auricular structure. [Copyright: ©2015 Beutler et al.

between 30 and 39 years [4,5]. However, albeit rarely, lesions may appear in either childhood [6] or old age [7]. There is no ethnic predilection.

Pseudocyst of the auricle typically presents as a solitary lesion on the anterior aspect of the ear. In his 1966 report, Engel observed that the pseudocysts "were localized in the upper half of the anterior side of the auricle, usually at the juncture of the crura anthelicis" [2]. Indeed, most patients, including our own, present with lesions that involve the crura of antihelix and scaphoid and triangular fossae. However, pseudocysts may also occur at other sites; in a 2001 review of 17 patients, Supiyaphun et al. observed that lesions often develop in the concha [8].

Regardless of the site of presentation, pseudocysts of the auricle develop progressively over a 4- to 12-week period and present as flesh-colored, nontender, noninflammatory cystic lesions. They range in size from 1 to 5 cm in diameter. The lesions are often soft initially and become increasingly firm over time. They are filled with a viscous fluid that is usually straw-yellow in color, resembling olive oil. However, serous and serosanguinous fluid may be present [4,9]. Pseudocyst of the auricle typically presents unilaterally—most commonly on the right ear—but bilateral presentations have also been reported [4,10-12].

Histologically, pseudocysts are characterized by an intracartilaginous cystic space with surrounding fibrosis. They can be distinguished from cysts by the absence of an epithelial lining on the inner surface of the intracartilaginous cavity. Degeneration of cartilage is common; rarely, calcification may be observed. A perivascular mononuclear infiltrate, predominantly of lymphocytes, is often present [13].

The clinical differential diagnosis of pseudocyst of the auricle is listed in Table 1 [2,4,9,13]. It includes not only local conditions, but also systemic disorders with skin lesions that may mimic pseudocyst of the auricle.

The mechanism of pathogenesis for pseudocyst of the auricle is unknown. In most patients, lesions arise spontaneously. It has been hypothesized that these pseudocysts result from a defect in embryogenesis in which residual planes of tissue are created during the folding of the branchial outgrowths that fuse to form the auricle. The excess tissues could theoretically reopen when subjected to mechanical stress, thus predisposing affected individuals to pseudocyst development [2,14].

It has also been postulated that trauma contributes to pseudocyst formation. Indeed, several patients presenting with pseudocyst of the auricle have reported a recent injury to the affected area of their ear [15,16]. Other patients

**TABLE 1. Clinical differential diagnosis of pseudocyst of the auricle**

| |
|---|
| Angiosarcoma |
| Cauliflower ear |
| Cellulitis |
| Chondrodermatitis nodularis chronica helices |
| Dermal cyst |
| Epidermal inclusion cyst |
| Fibroma |
| Hidrocystoma |
| Perichondritis |
| Relapsing polychondritis |
| Rheumatoid nodule |
| Subperichondral hematoma |
| Tophus |
| Xanthoma |

have engaged in practices that would subject their ears to chronic pressure or friction, such as wearing a tight motorcycle helmet [17] and sleeping on a hard pillow [18].

Two trauma-related etiologies have been proposed. Glamb and Kim hypothesized that constant pressure and/or friction reduces blood flow to the perichondrium, inducing ischemia and subsequent cartilaginous degeneration [17]. Conversely, Choi et al. suggested that repetitive minor traumas lead to the formation of numerous microcysts, which ultimately coalesce to form a large pseudocyst [18]. Both hypotheses are supported by recent work from Miyamoto et al. and Chen et al., who detected elevated lactate dehydrogenase levels within the pseudocyst fluid. Two lactate dehydrogenase isozymes, lactate dehydrogenase-4 and lactate dehydrogenase-5, are present in auricular cartilage and could conceivably be released upon degeneration or tearing of the tissue [19,20].

Patients who have neurological conditions with movement disorders may have an increased risk of developing trauma-induced pseudocyst of the auricle (Table 2) [21, current report]. Each of our patients suffered from neurological conditions characterized by ataxia and sudden, involuntary jerking movements. The spinocerebellar ataxias, also known as autosomal dominant cerebellar ataxias, represent a group of autosomal dominantly inherited progressive neurological conditions. There are over 30 known types of spinocerebellar ataxias; the age of onset, clinical presentation, and prognosis vary between the different types. However, all spinocerebellar ataxias are characterized by ataxia, dysarthria, and ultimately bulbar dysfunction [21]. Friedreich's ataxia is distinct from most other spinocerebellar ataxias in that it is inherited in an autosomal recessive pattern. It is characterized by gait ataxia, dysarthria, dysphagia, spasticity, and loss of muscle tone [22].

Indeed, both of our patients had recent histories of ataxia-related head and ear trauma. We hypothesize that trauma-induced pseudocyst of the auricle may occur more frequently in patients with neurological conditions that have disease-related movement disorders than in the general population. To the best of our knowledge, only one other patient with a movement disorder and pseudocyst of the auricle has been described: a young woman with cerebral palsy who, due to disease-related immobility, developed an auricular pseudocyst after lying in bed with her ear against the pillow for an extended period of time (Table 2) [23, current report]. In contrast to our patients' abnormal motor activity and likely repeated trauma to their affected ear, the auricular pseudocyst in the woman with cerebral palsy was not secondary to trauma.

Treatment of pseudocyst of the auricle should ideally be initiated promptly in order to preserve the normal architecture of the ear. Fortunately, various effective chemical and surgical interventions have been developed in recent years. Therapeutic interventions for successful management of patients' auricular pseudocysts often involves: (1) alteration of the exposed cartilage surfaces and (2) adequate compression of the separated cartilage to enable healing without allowing fluid to accumulate in the prior space between the cartilage [24].

Miyamoto et al. reported excellent results with steroid injection therapy. In a review of 8 patients who were treated with intralesional steroid injections, 3 patients had no recurrences and another 4 were treated successfully after 1 to 3 recurrences; only 1 patient failed to respond to treatment and required surgery [23]. However, in a recent prospective study of 28 patients, Patigaroo et al. concluded that intralesional steroid injections were no more effective than simple observation [25]. Furthermore, corticosteroid injections may result

**TABLE 2. Characteristics of patients with movement disorders and pseudocyst of the auricle**

| Case | Age | Race | Gender | Affected ear | Neurological condition | Treatment | Reference |
|---|---|---|---|---|---|---|---|
| 1 | 16 years | Asian | Female | Not reported | Cerebral palsy | Intralesional corticosteroids | 23 |
| 2 | 33 years | Asian | Male | Right | Spinocerebellar ataxia | Reassurance and observation | Current report |
| 3 | 47 years | Caucasian | Male | Left | Friedreich's ataxia | Reassurance and observation | Current report |

in permanent deformity of the auricle and therefore may not be appropriate for first-line treatment [4,17].

A conservative surgical approach for the management of pseudocyst of the auricle involves induction of fibrosis and scarring on the intracartilaginous cavity of the pseudocyst. Job et al. described a procedure in which incision and drainage of the lesion is followed by curettage of the pseudocyst walls. Bilateral contour pressure dressings are then applied in order to prevent fluid accumulation in the underlying tissue [26].

Two modifications to the aforementioned surgical procedure may help improve patient outcomes. First, several investigators introduced either a 1% iodine tincture or another sclerosant (such as 50% trichloroacetic acid) into the pseudocyst cavity following drainage to induce fibrosis and to reduce the risk of lesion recurrence [2-4,10,27]. Second, button bolsters were used in lieu of pressure dressings [27]; specifically, two sterilized shirt buttons were sutured to the auricle—one to the anterior side and another to the posterior side—following incision and drainage. The buttons not only help maintain localized pressure, but also substantially reduce the risk of recurrence and necrosis of the skin overlying the cartilage [28].

Deroofing the cartilage of the pseudocyst may be the most effective, albeit invasive, treatment. Indeed, multiple researchers have reported outstanding results with this surgical technique [14,25,29]. Deroofing entails curetting and cauterizing the base of the pseudocyst and removing the degenerated cartilage from the roof of the pseudocyst; this procedure eliminates the possibility of any future recurrence. However, it is critically important to excise only the anterior wall of the pseudocyst; accidental removal of the posterior wall can result in deformity of the auricle [18]. Surgical deroofing should be followed by button bolstering for optimal results [27].

Pseudocyst of the auricle is usually asymptomatic. Therefore, observation is a reasonable alternative to management if the patient is unconcerned about the cosmetic appearance of the lesion. In addition, clinical monitoring of the auricular pseudocyst may be appropriate in patients with neurological conditions associated with uncontrolled movements of the head and the potential for repeated trauma to the affected ear.

## Conclusion

Pseudocyst of the auricle is a rare condition of the ear. It typically presents as an asymptomatic, noninflammatory, unilateral cystic swelling affecting the upper auricle. Lesions may develop spontaneously or, less commonly, in response to trauma. Pseudocyst of the auricle occurs predominantly in middle-aged men, but may also occasionally be observed in women, children, and the elderly. Patients with neurological conditions that are associated with movement disorders may be more susceptible to developing trauma-induced auricular pseudocysts.

Pseudocysts of the auricle are typically flesh-colored and range in size from 1 to 5 cm in diameter. They develop progressively over a 4- to 12-week period, becoming increasingly firm over time. Incision and drainage usually reveals a viscous, straw-yellow colored fluid that resembles olive oil. Histologically, pseudocysts appear similar to cysts but lack an epithelial lining on the inner surface of the intracartilaginous cystic space.

Successful treatment for pseudocyst of the auricle initially includes either: (1) aspiration or incision and drainage, followed by injection of an agent to cause fibrosis and adherence of the cartilage or (2) surgical deroofing. Subsequently, adequate and sustained compression—using a button bolster—is important for the intervention to be effective. However, since the lesions are usually asymptomatic, treatment is not always necessary—especially in patients with trauma-related auricular pseudocysts who are at risk for repeat injury to the affected ear.

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