

Recurrent Pseudocyst Pinna: A Rational Approach to Treatment

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Abstract- Objective: The objectives of this study were to evaluate a rational approach in the management of recurrent pseudocyst pinna, so as to ascertain the definitive treatment of frequently recurring condition.

Materials and Methods: Forty eight (48) patients were diagnosed with pseudocyst of pinna who were unresponsive to aspiration and intralesional steroid injection at our tertiary care referral centre (ENT deptt. of SKIMS Medical college) between December 2009 to April 2012. Forty one (41) patients showed recurrence within 10 days after needle aspiration and intralesional steroid with contour dressing and were randomly divided into two groups. Group I comprising of 26 patients, underwent incision and drainage with curettage followed by buttoning and group II comprising of 15 patients, underwent surgical deroofting of the cyst along with buttoning.

Results: 39 males and 9 females with mean age of 32.6 ± 4.5 years were taken in this study. 29 (60.417%) patients had left sided lesion, whereas 19 (39.563%) patients had right sided lesion. Most common site of involvement was scaphoid fossa in 29 (60.417%) followed by concha in 16 (33.33%) and triangular fossa in 3 (6.25%) patients. 41 (85.41%) patients showed recurrence within 10 days after needle aspiration and intralesional steroid injection and contour dressing. 7 (26.923%) of the 26 patients showed recurrence after incision and drainage with buttoning. These 7 patients that recurred along with the 15 patients underwent surgical deroofting with buttoning showed no recurrence. In our study one patient developed perichondritis initially followed by cauliflower ear, otherwise the results were cosmetically acceptable. Biochemical study of the aspirated fluid revealed markedly elevated activity of Lactate dehydrogenase (LDH) levels in all cases.

Conclusion: Pseudocyst of pinna is a rare, benign, idiopathic, asymptomatic, cystic swelling over the anterior aspect of auricle and can be a source of disappointment for the surgeon because of frequent recurrence after its treatment. Optimal treatment for recurrent pseudocyst of the pinna involves removal of anterior cartilaginous leaflet of the pseudocyst with repositioning of the overlying flap of skin followed by buttoning which accomplishes the twin objectives of complete resolution of the condition without any recurrence. This results in a normal appearing auricle with minimal scarring.

I. INTRODUCTION

Pseudocyst of pinna is a rare, benign, idiopathic, asymptomatic, cystic swelling over the anterior aspect of auricle and can be a source of disappointment for the surgeon because of frequent recurrence after its treatment. Engel[2] in 1966, first reported the pseudocyst of auricle in the Chinese population. It is predominantly seen in young male adults, being uncommon before the age of 20 years and after 60 years. This rare disorder results from spontaneous accumulation of a sterile, oily, yellowish fluid and presents clinically as a painless, solitary, fluctuant, non-inflammatory, dome shaped cystic swelling on the anterior surface of pinna. Majority of cysts are located in the scaphoid [1] and the triangular fossae. It ranges usually from 1-5 cm in diameter. The etiology of pseudocyst of the auricle is unknown, but the 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[2] or wearing a motorcycle helmet or headphones[8]. It was suggested that chronic trauma induced cartilage degeneration resulting in progressive dilatation and formation of a cystic space within the auricular cartilage[6]. 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 arches, and congenital maldevelopment of this branchial arch may result in residual tissue planes within the cartilage which may reopen, giving rise to a pseudocyst[4]. Other studies showed that the auricular pseudocyst might represent an autoimmune disease involving the auricle [11]. The possible mechanism may be an accumulation of reactive exudates due to immunoreactions of autologous antibody and auricular tissue. 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 auricular 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

Index Terms- Pseudocyst, Cystic swelling, cartilage, pinna

process slowly settles and is replaced by the histological evidence of perichondrial fibroblastic response and granulation tissue formation.

Histologically, pseudocyst of the auricle is characterized by an intercartilagenous cavity lacking an epithelial lining, hence the name pseudocyst. Irregular thinning and hyalinization of the cartilage peripheral to the cavity is also seen. Later stage of pseudocyst may also show intracartilagenous fibrosis and granulation tissue[4]. Lim et al[3] in their histological review of 16 cases of pseudocysts, found a perivascular mononuclear infiltrate of predominantly lymphocytes in all the cases.

Despite the large number of treatment modalities that have been described for this condition, the definitive treatment still remains controversial because of high recurrence rate.

II. MATERIALS AND METHODS

Forty eight (48) patients were diagnosed with pseudocyst of pinna who presented with asymptomatic painless cystic swelling over anterior aspect of auricle and were managed at our tertiary care referral centre (ENT deptt. of SKIMS Medical college) between December 2009 to April 2012. All the patients were initially subjected to needle aspiration and intralesional steroid injection with contour dressing. The biochemical analysis of the aspirated fluid was done. Forty one out of the forty eight patients showed recurrence within 10 days after needle aspiration and intralesional steroid with contour dressing. These 41 patients were randomly divided into group I comprising of 26 patients who underwent incision and drainage with curettage followed by buttoning and group II comprising of 15 patients who underwent surgical deroofting of the cyst along with buttoning under local anaesthesia. All patients gave informed written consent and all the procedures were performed in an outpatient setting.

III. RESULTS

39 males and 9 females with asymptomatic pseudocyst of pinna were treated at SKIMS MC Hospital between Dec. 2009 to April 2012. The age of patients ranged from 28 to 40 years, with the mean age of 32.6 ± 4.5 years. 29 (60.417%) patients had left sided lesion whereas 19 (39.583%) patients had right sided lesion. All the patients had painless swelling of pinna as presenting symptom, with an average duration of symptoms of 3 weeks ranging from 15 to 28 days. The scaphoid fossa was the most common site of involvement. 29 (60.417%) patients presented with lesion in the scaphoid fossa followed by concha in 16 (33.33%) and triangular fossa in 3 (6.25%) patients. 41 (85.41%) patients showed recurrence within 10 days after needle aspiration and intralesional steroid injection and contour dressing. 7 (26.925%) of the 26 patients in group I had recurrence after incision and drainage with buttoning within 10 days after removal of buttons. The 7 patients that recurred then underwent deroofting with buttoning alongwith 15 patients of group II. Thus a total of 22 patients underwent surgical deroofting with buttoning. None of the patients had recurrence following this technique. The patients

were followed for 6-8 months. In our study, one patient developed perichondritis initially followed by cauliflower ear 2 months after the surgery. The patient was diabetic, underlying comorbidity may have contributed to the unfavourable outcome, otherwise the results were cosmetically acceptable. Biochemical study of the aspirated fluid revealed markedly elevated activity of Lactate dehydrogenase (LDH) and a preponderance of LDH-4 and LDH-5 in all patients.

IV. DISCUSSION

Pseudocyst of the auricle is characterized by a unilateral, asymptomatic, cystic swelling of the helix or the antihelix, most often located in the scaphoid fossa[2]. Engel[2] in 1966 first reported the pseudocyst of the auricle in the Chinese. This rare disorder results from spontaneous accumulation of a sterile, oily yellowish fluid, resembling olive oil[1]. It is mostly observed in young males and presents clinically as a benign, non inflammatory, painless swelling on the lateral or the anterior surface of pinna[1],[2]. The etiology of pseudocyst is still incompletely understood, but several mechanisms for its pathogenesis have been put forth. Originally, Engel[2] hypothesized that abnormal release of lysosomal enzymes from local chondrocytes caused progressive dilatation and formation of an intracartilagenous cavity. Another theory hypothesized that congenital embryonic dysplasia of the auricular cartilage is a factor in the development of pseudocyst. The auricle develops from 6 tubercles on the first and second branchial arches. It has been suggested that during the complex fusion and folding that form the auricle, residual tissue planes may sometimes be formed within the mesenchyme that gives rise to the auricular cartilage. It is the reopening of these tissue planes later on that result in the formation of pseudocyst[4]. Another factor that has been postulated to play some part in the pathogenesis of pseudocyst is chronic low grade trauma. Engel[2] suggested that the Chinese habit of sleeping on a hard pillow might be a causative factor in the pseudocyst formation. Other types of minor trauma that have been cited include the carriage of large hampers and sacks on shoulders[6], motorcycle helmets and stereo headphones[8] and even the Italian habit of pulling the pinna on birthday[9]. Choi[6] suggested that repeated minor trauma led to an overproduction of glycosaminoglycans, which starting as microcysts within the cartilage, coalesce to form a larger lesion or pseudocyst. However, trauma has not been documented in most cases of pseudocyst. Tan[7] found a positive history of trauma in only 10% of patients, whereas Cohen[1],[23], reported a history of trauma to the lesional site in 30% of patients. In our study, 24 (50%) patients gave a history of trauma, whereas 5 (10.4%) patients were motorcycle helmet wearers. This gives evidence to the fact that minor repeated trauma may be critical to the pathogenesis of this condition. Elevated levels of LDH in the pseudocyst fluid also points towards a traumatic basis of pseudocyst formation. Two of the elevated isoenzymes, LDH-4 and LDH-5, are proposed as major components of human auricular cartilage. These enzymes may be released from the auricular cartilage degenerated from repeated minor trauma[10]. The autoimmune basis of pseudocyst formation was analysed by

Chen et al[11], who measured in the cystic fluid and blood samples of 44 cases the content of IgG, IgA, IgM and complement C3, the content of immune complex, DNA of cytomegalovirus, antinuclear antibody and reactions between samples and frozen sections of healthy white rats. In view of the marked male preponderance of this condition, we propose a hormonal basis of pseudocyst formation. Cohen[1] reported that 93% of the cases occur in males. Other studies have reported a similar male preponderance (Tan[7], 87.5%; Lim[3], 87.8%). In our study 39(81.25%) patients were males. This striking male prevalence can be explained on the differential action of estrogen and testosterone in inducing cytokines. Interleukin 1 is a cytokine that plays a major role in inflammatory response in the context of infections and immune mediated diseases. Monocytes/macrophages are the primary source of IL-1. Interleukin 1 stimulates the production of mediators such as prostaglandin E2, nitric oxide, cytokines, chemokines, and adhesion molecules that are involved in articular inflammation. Furthermore, IL-1 stimulates the synthesis and activity of matrix metalloproteinases (MMP) and other enzymes involved in cartilage destruction[24]. Morishita et al studied the effect of sex hormones on the production of IL-1 by the human peripheral monocytes and concluded that the estradiol and progesterone inhibited the production of IL-1 from human peripheral monocytes. Similar results were obtained by Richette[13] et al and Posma et al. The authors propose that the hormones modify the inflammatory response induced by chronic low grade trauma in susceptible individuals having a certain autoimmune defect or congenital dysplasia of the auricular cartilage that invariably leads to pseudocyst formation.

There is much controversy regarding the management of pseudocyst pinna. Restoration of the normal architecture of the auricle with no recurrence is the main goal of the treatment. Various treatments reported in literature include simple aspiration, intralesional injection of corticosteroids and aspiration in combination with bolstered pressure sutures or plaster of paris cast[9],[15],[16],[22]. More invasive techniques like incision and drainage of the cavity followed by its obliteration by curettage, sclerosing agent and pressure dressing; open deroofting that involves removal of the anterior cartilaginous leaflet of pseudocyst with repositioning of the overlying flap of skin have also been recommended. Both needle aspiration and incision drainage of the pseudocyst result in prompt accumulation of the fluid in the lesion[1],[2]. Aspiration or incision and drainage followed by compression decreases the recurrence rate. Compression has been achieved using traditional contour dressing, bolstered pressure sutures[15], plaster of paris casts[16] and recently using buttoning. The role of corticosteroids either systemic or intralesional is controversial with varying success rates. Minocycline is thought to work as a sclerosant through its anti-inflammatory and immunomodulatory mechanisms. Recently, the use of fibrin glue as a sealer between the 2 leaves of the cartilage has been described by Tuncer[17] with positive results in one patient of recurrent pseudocyst. However, it is difficult to interpret the significance of these observations as both involved few patients. Various surgical techniques have been

proposed for treating this condition. Incision and drainage was the earliest to be described but has shown to have a high recurrence rate[2]. Zhu and Wang[18] have reported a method of inserting a small drainage tube in to the cavity of the pseudocyst with a guidewire. Paul et al[19] described in a single patient the successful use of a 3-mm punch biopsy of the inferior border of the pseudocyst followed by application of a bolster.

Surgical deroofting of the pseudocyst was first described by Choi[6] in 31 patients with no recurrence and cosmetically good results in 90% of the patients. He had used contour dressings for compression. Lim[3] modified this technique by using buttoning as a compression method, in 41 patients and reported no recurrence with good cosmetic outcome in all the patients. Chang[20] reported similar results in 10 patients of pseudocyst in whom deroofting was done. The use of buttons was first described in otolaryngology literature as a method to maintain adequate localized pressure in the ear in patients with auricular hematoma. Almost all the reported studies[2,3,6,7,20] on this mysterious condition have been described in the Chinese population. This study is among non-Chinese (Indian) population. Most of the studies report a high male preponderance[1,2,7]. The mean age of presentation was 32.6 ± 4.5 years, similar to those reported by Tan[7] (38.2 years) and Choi[6] (42.8 years) showing it to be a disease primarily of young adults. In our study, scaphoid fossa was the most common site of involvement in 29 (60.417%) patients. Choi[6] described scaphoid fossa (80.6%) as being the most common site of pseudocyst.

On comparing the various modalities, we found that aspiration followed by pressure dressing is an ineffective method of treating pseudocyst as only 7 patients responded to this modality. Incision and drainage followed by buttoning considerably decreased the recurrence rate. This could be explained on the basis that buttons offer a better and a more constant compression as compared to contour dressings. However, surgical deroofting of the cavity followed by buttoning was associated with 0% recurrence and offered a cosmetically acceptable result in all cases. Buttoning provides the best form of compression as it offers a constant pressure, is easy to apply, is aesthetically appealing and is patient friendly as it avoids the need for large bandages.

V. 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. The epidemiological profile of this condition is similar in Chinese and non-Chinese (Indian) population. A hormonal influence modulating the inflammatory process explains the marked male preponderance of this condition. Several treatment methods have been reported with variable results. Optimal treatment for recurrent pseudocyst of the pinna involves removal of anterior cartilaginous leaflet of the pseudocyst with repositioning of the overlying flap of skin followed by buttoning which accomplishes the twin objectives of complete resolution of

the condition without any recurrence. This results in a normal appearing auricle with minimal scarring.

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PREOPERATIVE



AFTER ASPIRATION



INTRAOPERATIVE



POSTOPERATIVE

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