

Ear lobule reduction using a sub-antitragal groove technique in patients with angiolymphoid hyperplasia with eosinophilia on the earlobe: a case report and literature review

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Angiolymphoid hyperplasia with eosinophilia (ALHE) is a rare condition characterized by nodular lesions predominantly in the head and neck region, often causing discomfort or pain. Treatment remains challenging because of its rarity and the lack of established guidelines. This report presents a case of ALHE affecting the earlobes that was successfully managed using ear lobule reduction surgery and subsequent intralesional steroid injections. A 31-year-old woman with a history of recurrent earlobe masses underwent a partial excision to avoid the loss of the earlobe. Histopathological examination confirmed Kimura disease, a variant of ALHE. Subsequent local methylprednisolone injections effectively controlled the remaining lesions, resulting in significant size reduction without notching. Various treatment modalities have been attempted for this condition; however, recurrence rates remain high. Surgical resection combined with intralesional corticosteroid injections is the preferred approach. In this case, a sub-antitragal groove technique for earlobe reduction was employed to preserve the lateral edge of the ear lobule, minimize the risk of deformity, and achieve a predictable outcome. The sub-antitragal groove technique offers an approach to reduce earlobe size without compromising aesthetics. Further research is required to elucidate the pathogenesis of ALHE and establish standardized treatment protocols for this rare condition.

Abbreviations: ALHE, angiolymphoid hyperplasia with eosinophilia; KD, Kimura disease

Keywords: Angiolymphoid hyperplasia with eosinophilia / Case reports / Ear neoplasms / Kimura disease

INTRODUCTION

Angiolymphoid hyperplasia with eosinophilia (ALHE) is characterized by single or multiple nodular lesions without encapsulation, predominantly in the head and neck region [1-3]. The ear, face, and scalp are the most affected areas, and approximately one-third of patients have lesions on the ear [4]. ALHE can affect different parts of the body, including the extremities, external genitalia, trunk, orbit, oral cavity, and parotid region [2,5]. ALHE often presents as reddish-brown nodules that may bleed, itch, or be painful [6]. However, some patients are asymptomatic [4]. The pathogenesis of ALHE remains unclear, but its etiology has been suggested to be idiopathic or to result from trauma or immunization [7]. These lesions often pose challenges in both diagnosis and management due to their rarity. There is currently no consensus on the treatment of ALHE. This report presents a case of ALHE affecting the ear lobule

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that was successfully managed using surgical ear lobule reduction and subsequent intralesional steroid injection.

CASE REPORT

A 31-year-old woman presented with a 10-year history of recurrent enlarging masses on both earlobes. She had undergone an excision approximately 7 years previously. However, the mass gradually grew over the subsequent years. As a substantial area of overall earlobes was affected, complete resection of the lesion was expected to result in a total earlobe defect with an irreversible earlobe deformity. Therefore, to minimize postoperative earlobe deformity, volume reduction of the lesion with subsequent local steroid injection was planned.

The sub-antitragal groove technique was used for ear lobule reduction (Fig. 1). Two reference lines were drawn. The upper reference line was marked along the sub-antitragal groove, and the lower line depicted the desired earlobe shape after surgery. First, the line connecting the otobasion to the sub-antitragal groove was drawn (line A₁), and then a line following the sub-



Fig. 1. Earlobe reduction using the sub-antitragal groove technique. A 31-year-old woman showing bilateral enlarging mass on both earlobes. First, two reference lines (dashed lines) were drawn along the sub-antitragal groove and desired earlobe shape. Lines A₁, A₂, A₃ and B₁, B₂ were marked to be the same length, respectively. The marked area (white arrows) was excised, and the remaining earlobe flap was rotated.

antitragal groove was drawn (line A₂). Line A₂ extended laterally until it reached the length of line A₁. Line A₃ was marked parallel to the free lateral edge over the same length as line A₂.

The lesion could not be completely removed due to concerns about significant earlobe deformities that might occur following complete resection. Histopathological examination revealed multiple lymphoid follicles and infiltration of lymphocytes and eosinophils, consistent with Kimura disease (KD) (Fig. 2). After

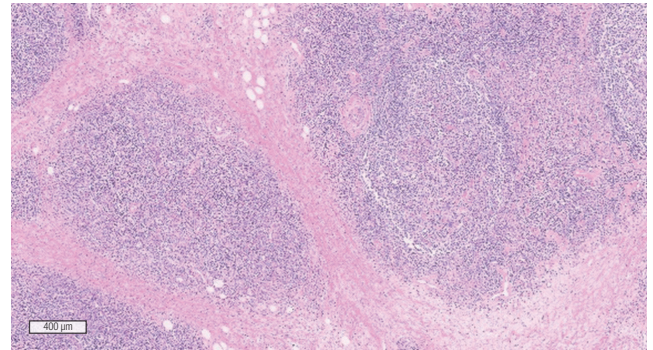


Fig. 2. Pathological features of the resected specimen. Multiple lymphoid follicles and numerous infiltrations of lymphocytes and eosinophils are seen. Increased vascularity with enlarged endothelial cell lining were also seen in the slide (hematoxylin and eosin stain).

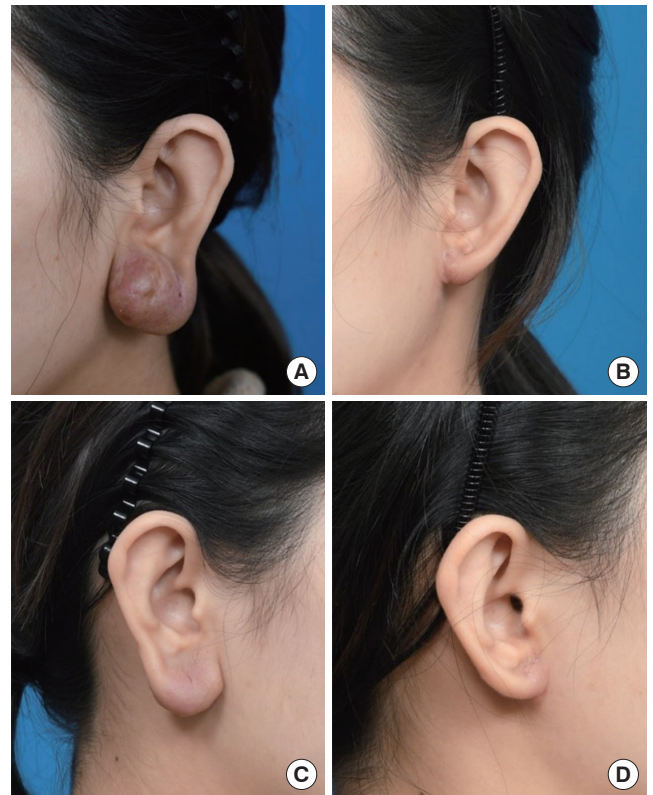


Fig. 3. Surgical outcomes. Preoperative and postoperative photo of 5 months after surgery of the left (A, B) and right ear (C, D) depicts significant postoperative lobular reduction without local recurrence.

2 and 5 months postoperatively, the remaining lesion was treated with a local methylprednisolone injection. The earlobes decreased significantly in size without lateral free-border notching, and the remaining lesion was well controlled with a local steroid injection (Fig. 3).

LITERATURE REVIEW

ALHE is a relatively rare, chronic vascular proliferative disease, primarily affecting the head and neck region. A study examining the frequency of ALHE based on its anatomical location found that the most common sites were the ear, face, and scalp, with frequencies of 36.3%, 28.2%, and 17.3%, respectively. However, reported cases of ALHE have been documented on almost the entire body surface. Notably, cases of ALHE occurring in the lobule constituted 1.0% of the 908 reported cases [6].

The exact cause of ALHE remains unclear, although various hypotheses, including immune-mediated eosinophilia, infection, excess estrogen, atopy, lymphoproliferation, and neoplasia, have been suggested [6,8,9]. Some authors consider ALHE a low-grade neoplasm owing to its high local recurrence rate and progressive nature. Nevertheless, others hold the view that ALHE is an inflammatory reactive condition, which is supported by the findings of immunoglobulin deposition in the lesions [10] and elevation of serum IgE levels [11,12]. Whether ALHE arises as a neoplasm or as a reactive change remains unclear, and further research is needed to confirm the etiology.

ALHE was first described in 1969 and was initially considered to be part of the same disease spectrum as KD, with ALHE being seen as the late stage of KD [13]. Subsequently, histopathological differences between ALHE and KD were reported, leading to the recognition of these diseases as distinct entities [14]. The current consensus now distinguishes KD as a chronic reactive inflammatory disease, while ALHE is characterized as a benign vascular neoplasm [15]. KD typically presents in younger individuals, with a higher prevalence among males, and is characterized by larger and more widely distributed lesions. This condition is often accompanied by peripheral eosinophilia and regional lymphadenopathy [16-18]. However, ALHE is more commonly observed in women and tends to occur between the third and fifth decades of their life. In addition, ALHE does not typically exhibit clinically significant peripheral eosinophilia. Histologically, the main distinction between KD and ALHE is the appearance of an endothelial lining in the newly formed blood vessels. In ALHE, endothelial cells appear plump with large nuclei and abundant eosinophilic cytoplasm, whereas in KD, the endothelial cell lining appears flat [19]. However, standard treatment of both ALHE and KD is unknown; surgical re-

section, use of corticosteroid or radiotherapy has been introduced, but comprehensive therapy has been recommended because of its high recurrence [14].

Various treatment modalities of ALHE have been introduced through literature; however, there is no effective gold standard, and the recurrence rate remains high. Complete surgical resection is recommended, but because of the uncircumscribed nature of the lesions, achieving complete excision is often difficult. Even after complete excision, the recurrence rate remains over 40% [20]. Laser therapy, such as pulsed dye laser, carbon dioxide laser, Nd:YAG laser is reported to be an effective treatment modality [21-23]. Cryotherapy, oral propranolol with or without intralesional or systemic corticosteroids have also been used with complete or partial resolution [24,25]. However, the recurrence rates with these treatments range from 50% to 80% [6]. Due to the high recurrence rate, combination therapy is preferred, often involving surgical resection combined with intralesional corticosteroid injections or adjuvant radiotherapy. In a recent study, it has been reported that Mohs micrographic surgery followed by intralesional corticosteroid therapy was observed to effectively achieve margin-free resection and to maintain the absence of recurrence [26]. Although a definitive gold standard for the treatment of ALHE has not yet been established, due to difficulties in achieving complete resection and a heightened recurrence risk, precise excision and combination therapy to prevent future relapses are preferred. Timely and accurate diagnosis and treatment selection are thus pivotal in ALHE management.

DISCUSSION

In our case, the borders of the lesion were unclear, and complete excision of the ear lobule would have resulted in a significant postsurgical ear deformity. Therefore, instead of complete excision, ear lobule reduction was performed, followed by intralesional corticosteroid administration. After a single corticosteroid injection, the patient's symptoms, including itching and redness, resolved, and no recurrence was observed for up to 6 months after surgery.

Multiple techniques for ear lobule reduction have been reported in the literature. During ear lobule reduction, it is essential to remove excessive tissue without causing significant ear lobule deformity. Traditionally, simple wedge resection, first introduced by Miller [27] in 1924, has been widely used; however, it often leads to distorted or notched free borders. Free borders without notch or distortion with anatomical intertragal notch is crucial in aesthetic and anatomic ear reconstruction [28]. Earlobe reduction techniques involving the medial portion of the

earlobe have also been described. It can avoid secondary earlobe distortion but has limited capabilities for earlobe reduction [29]. The sub-antitragal groove technique was first described by Van Putte and Colpaert in 2017 [30] and has since been used to rejuvenate the ptotic earlobe [30]. This technique aims to preserve the lateral edge of the ear lobule and minimize the risk of free-border notching. Additionally, the scar resulting from the procedure can be effectively concealed within the sub-antitragal groove and anterior tragal crease. In the present study, the sub-antitragal technique was successfully applied to a massive ear lobule. This technique relies on patients' anatomical landmarks and geometric ratios, allowing for predictable ear lobule reduction without the risk of over-resection.

Given the complexities associated with achieving complete surgical resection in ALHE, consideration of combination therapy following excision becomes important. This is particularly important when the condition is known to exhibit a higher prevalence in anatomical regions such as the ear and face [6], where postoperative aesthetic deformity must be considered. Thus, in this case, we considered to treat ALHE on earlobes by ear lobule reduction with sub-antitragal groove technique followed by intralesional triamcinolone injection. When ALHE is clinically assumed on earlobes, earlobe reduction with sub-antitragal groove technique with adjuvant intralesional corticosteroid injection can lead to a favorable outcome without any signs of local recurrence or secondary deformities.

NOTES

Conflict of interest

No potential conflict of interest relevant to this article was reported.

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Ethical approval

This report was approved by the Institutional Review Board of Korea University Anam Hospital (IRB No. 2023AN0305).

Patient consent

The patient provided written informed consent for the publication and use of her images.

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