

## Bilateral acquired external auditory canal stenosis with squamous papilloma: a case report

Skuamöz papilloma bağlı iki taraflı kazanılmış dış kulak yolu stenozu: Olgu sunumu

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Acquired external auditory canal (EAC) stenosis is described as resulting from a number of different causes such as infection, trauma, neoplasia, inflammation and radiotherapy. Human papilloma virus (HPV) type 6, a deoxyribonucleic acid (DNA) virus, is considered to cause squamous papilloma of the EAC. In this article, we report a case of a 56-year-old male with warty lesions in the left external ear and a totally stenotic right external ear which had similar lesions one year before the involvement of his left ear. On computed tomography of the temporal bone, there was soft tissue obstruction of the right EAC, and thickening in the skin of the left EAC. The middle ear structures were normal on both sides. Biopsy was performed from the lesion in the left ear, and revealed squamous papilloma. We presented this case because squamous papilloma related bilateral acquired EAC stenosis is a rare entity.

Key Words: Acquired stenosis; external auditory canal; squamous papilloma.

Kazanılmış dış kulak yolu (DKY) stenozu enfeksiyon, travma, neoplazi, enflamasyon ve radyoterapi gibi birçok nedenin bir sonucu olarak tanımlanabilir. Bir deoksiribonükleid asit (DNA) virüsü olan human papilloma virüs (HPV) tip 6'nın da DKY skuamöz papillomuna neden olduğu düşünülmektedir. Bu yazıda 56 yaşında sol dış kulağında siğil benzeri lezyonları olan, sağ kulağı bir yıl önce benzer lezyonlar sonrası tama yakın daralmış olan erkek olgu sunuldu. Temporal kemik bilgisayarlı tomografisinde sağ DKY'de yumuşak doku obstrüksiyonu, sol DKY'de cilt kalınlaşması vardı. Her iki taraf orta kulak yapıları normaldi. Sol kulaktaki lezyondan yapılan biyopsi sonucu skuamöz papillom olarak bildirildi. Skuamöz papilloma bağlı iki taraflı kazanılmış DKY stenozu olan hasta nadir bir durum olması nedeniyle sunuldu.

Anahtar Sözcükler: Kazanılmış stenoz; dış kulak yolu; skuamöz papillom.

Acquired external auditory canal (EAC) stenosis is an uncommon entity, and can arise from a number of different causes including infection, trauma, neoplasia, inflammation, and radiotherapy.[1] Its incidence has been estimated at 0.6 cases per 100.000 inhabitants, as reported in the largest series of patients treated for EAC stenosis.[1]

Mechanical cleaning and shaving of the external ear canal with instruments used repeatedly without sterilization are thought to inoculate viral agents. [2,3] Human papilloma virus (HPV) type 6 is considered to cause papilloma of the external ear canal. [2] Aural papillomas are primarily a disease of adult males. [3] We report an additional case of acquired EAC stenosis with squamous papilloma.

## CASE REPORT

A 56-year-old male was admitted to our clinic with warty lesions in his left EAC for one month duration. The chief complaint of the patient was itching and fullness in his left ear. Physical examination revealed warty, fingerlike, keratotic lesions in the left external ear, and the tympanic membrane could not be visualized (Figure 1). He also had aural stenosis with warty lesions in his right ear one year before. Now the right EAC was totally stenotic (Figure 2). The patient and his wife had no lesion in the genital regions.

According to his history, similar lesions had appeared in the right ear one year before the involvement of his left ear. At that time, he had been treated surgically in another clinic with total excision of the EAC skin and with split thickness skin graft reconstruction. Histopatological examination had revealed squamous papilloma. He had been reoperated on due to stenosis of his right EAC. However, the restenosis recurred.

On computed tomography of the temporal bone, there was soft tissue obstruction of the right EAC, and thickening in the skin of the left EAC (Figure 3). The middle ear structures were normal on both sides.

In our clinic, biopsy was performed from the lesion in the left ear, and revealed squamous papilloma and the histopathological evaluation showed hyperkeratosis, acanthosis, papillomatosis, vacuolated cells referred to as "koilocytotic cells" and loss of granular cell layer in the papilla (Figure 4, 5). However, he rejected the surgical treatment advocated for his left ear because of unfortunate experiences he had in the right ear. Medical treatment with topical antibiotics were given to prevent the bacterial infection after biopsy. After three weeks, the warty lesions in the left ear disappeared. The patient was disease free at two years follow-up.

## **DISCUSSION**

Acquired EAC stenosis is described as resulting from a number of different causes such as infection, trauma, neoplasia and inflammation, radiotherapy. Squamous papilloma of the EAC occurs most commonly in Southern China. Such cultural rituals as mechanical cleansing and shaving the ear canal with instruments and using them repeatedly without sterilization are thought to inoculate viral agents.[3] Physical damage to the epithelial cells of the canal from shaving is considered to be the etiology for EAC neoplasm. A DNA virus, HPV type 6 is considered to cause papilloma of the EAC.[2] Although aural papilloma is primarily a disease of adult males,[3] a three-year-old child with aural papillomatosis was also reported.[4] People at risk of condylomata are postulated to develop aural papillomatosis.[2]



Figure 1. Warty, fingerlike, keratotic lesions in the left external

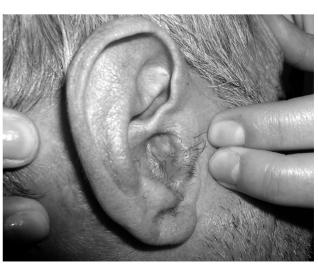
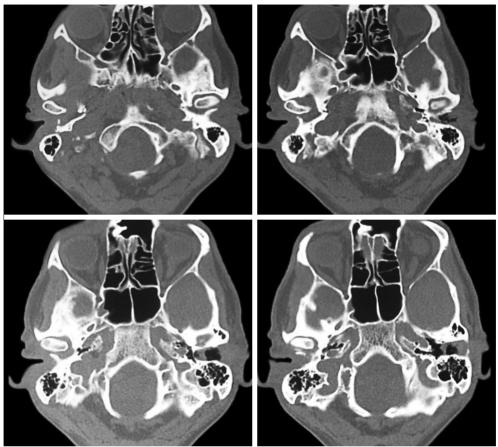


Figure 2. Aural stenosis.

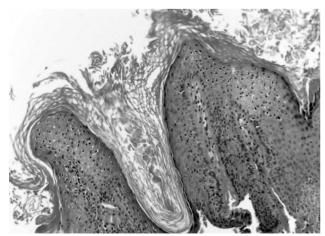


*Figure 3.* Soft tissue obstruction of the right external auditory canal, and thickening in the skin of the left external auditory canal.

There are many forms of papilloma including flat plaques and roughened raised lesions, which are commonly round or oval with a broad base on the skin or mucous membrane.<sup>[2]</sup> They may arise from any site in the EAC.<sup>[3]</sup> Malignant transformation of aural papillomatosis has been reported as

well,<sup>[5]</sup> and HPV types 16 and 18 have been postulated to play an important role in causing malignancy.<sup>[6]</sup> The condition should be differentiated from carcinoma, and hence biopsy is necessary.<sup>[2]</sup>

Histologically, spherical enlargement of the cells, peri-nuclear vacuolation, and absence or a



*Figure 4.* Histopathology showed hyperkeratosis, papillomatosis and acanthosis (H-E x 40).

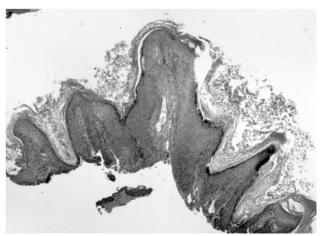


Figure 5. Loss of granular cell layer in the papilla (H-E x 100).

thin layer of cytoplasm can be seen.<sup>[2]</sup> The nucleus is central and often enlarged, angular or wrinkled and it may be associated with moderate degrees of dysplasia.<sup>[6]</sup> The characteristic features of HPV infections are "koilocytotic cells" that distinguish squamous papilloma from other papillomas.<sup>[7]</sup> Koilocytotic cells in histopathological examination of our patient made us to consider HPV in diagnosis but we didn't perform PCR, immunohistochemistry or insitu hybridization.

Several different etiologies can cause EAC stenosis, but a common feature of its pathogenesis is a fibroproliferative inflammatory response. After understanding the strong role of inflammation in the pathogenesis of EAC stenosis, prevention of the formation of granulation tissue and control of underlying infection have been important. For this reason, topical antibiotics and steroids are given in the medical treatment.<sup>[8]</sup>

In survey of the literature, we didn't find any report about the use of alpha interferon or topical therapies for the treatment of squamous papilloma of the external ear. The immune response modulator imiquimod 5% may be a choice for the topical treatment. It has been used for treatment of papillamatosis of the external genitalia and keloid of the skin succesfully.<sup>[9]</sup>

We gave the topical medical treatment to our patient to prevent the bacterial infection after biopsy. We planned to persuade the patient for surgery or CO2 lazer tratment, but in the control examination after three weeks the warty lesions in the left ear disappeared.

Once there is complete obstruction of the external auditory canal, surgery becomes the treatment of choice. The goal of surgery is to remove the fibrous plugs, exposing the tympanic membrane, and to recreate an epithelium lined EAC. The most common postoperative complication is re-stenosis that usually occurs one year after the operation.<sup>[7]</sup>

Evaporation with CO2 laser is suggested to be a good choice for the treatment of squamous papilloma of the EAC as well.<sup>[2]</sup>

In conclusion, several different etiologies can cause EAC stenosis and one of them is squamous papilloma of the external auditory canal. If itchy, warty, keratotic lesions are seen in physical examination we must consider squamous papilloma. The treatment options are excision of the lesion and CO2 laser. Topical antibiotics and steroids are given only to control the inflammation and to control the concurrent bacterial infection in the medical treatment. Once there is complete obstruction of the external auditory canal, surgery becomes the treatment of choice.

## **REFERENCES**

- Becker BC, Tos M. Postinflammatory acquired atresia of the external auditory canal: treatment and results of surgery over 27 years. Laryngoscope 1998; 108:903-7.
- Blair RL, Irani BS, Low C. Aural papillomatosis-treatment with the carbon dioxide laser. J Laryngol Otol 1998;112:565-6.
- 3. Myer CM 3rd, Woodruff SM. Pathologic quiz case 2. Squamous papilloma of the external auditory canal. Arch Otolaryngol 1983;109:200-1.
- 4. Yadav SP, Chanda R, Goyal N, Chanda S. Aural papillomatosis in a 3-year-old child. Int J Pediatr Otorhinolaryngol 2002;66:185-7.
- 5. Friedman I. External ear in Pathology of the ear. 1st ed. Oxford: Blackwell Scientific Publications; 1974.
- 6. Xia MY, Zhu WY, Lu JY, Lu Q, Chen L. Ultrastructure and human papillomavirus DNA in papillomatosis of external auditory canal. Int J Dermatol 1996;35:337-9.
- 7. Elder DE. Disease caused by viruses. In: Elenitas R, Johnson BL Jr, Murphy GF, Xu X, editors. Lever's histopathology of the skin. 10th ed. Philadelphia: Lippincott Williams and Wilkins; 2009. p. 637-66.
- 8. Luong A, Roland PS. Acquired external auditory canal stenosis: assessment and management. Curr Opin Otolaryngol Head Neck Surg 2005;13:273-6.
- 9. Flint PW. Topical therapies of external ear disorders. In: Flint PW, Haughey BH, Lund VJ, Niparko JK, Richardson MA, Robbins KT, et al. editors. Cummings otolaryngology: head and neck surgery. 5th ed. St. Louis: Mosby Inc; 2010. p. 1950-62.