

Verrucous carcinoma of the temporal bone

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Abstract

Verrucous carcinoma is a highly differentiated variant of squamous cell carcinoma. In the literature, 11 cases of primary verrucous carcinoma of the temporal bone have been reported. We present a 48-year-old woman who had undergone radical mastoidectomy because of chronic otitis media 20 years ago; consequently, verrucous carcinoma occurred in the mastoid cavity. We discuss verrucous carcinoma of the temporal bone with the review of literature.

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1. Introduction

Verrucous carcinoma is a slowly and locally growing neoplasm that rarely metastasizes. Ackerman [1] first described this neoplasm as a low-grade tumor and also a clinicopathologic variant of squamous cell carcinoma. In the head and neck region, this lesion is principally seen in the oral cavity and larynx, and rarely in the ear [2]. The literature review revealed only 11 cases of primary verrucous carcinoma of the temporal bone [3–10].

2. Case reports

A 48-year-old woman who had a bilateral sensorineural hearing loss and bleeding in the left ear with left peripheral facial paralysis was admitted to Çukurova University Medical School Hospital. She had a history of left radical mastoidectomy in her left ear 20 years earlier, and had a revision mastoidectomy 8 years ago in different centers because of chronic otitis media. Moreover, she has complained of left peripheral facial paralysis for 20 days, and has received oral steroid. In the otoscopic examination, the left ear cavity was filled with granulation tissue and a whitish warty tissue. Left peripheral facial paralysis was also present. Her oropharyngeal and systemic examination was unremarkable with no cranial nerve deficit or palpable lymphadenopathy.

The audiogram showed total sensorineural hearing loss in the left ear. Her computed tomography showed a defect

consistent with previous radical mastoid surgery in the left mastoid. Left mastoid cavity filled with a soft tissue that had contacted with the facial canal's horizontal and vertical segments (Fig. 1A, B).

The biopsy taken from the granulation tissue in the left ear cavity has been reported as chronic inflammation. Although a definitive histologic diagnosis was not obtained, we performed a revision radical mastoidectomy. During the surgery, keratinized tissue, granulation tissue, and cholesteroloma was encountered in the mastoid cavity and removed. While this tissue was being removed, facial bony canal was found to be dehiscence. Great care was taken during the removal of this tissue over the dehiscence part of the facial canal. A large meatoplasty was made after the removal of all of these tissues. Histopathology of the specimen was reported as verrucous carcinoma in the left ear. Histopathologic examination revealed marked hyperkeratosis, acanthosis, papillomatosis, and well-differentiated tongues of squamous epithelium extending into the underlying stroma. There was no cytologic atypia (Fig. 2). During the 26-month follow-up period after surgery, the mastoid cavity remained dry and free of disease, and the facial functions of the patient were deemed acceptable.

3. Discussion

Although verrucous carcinoma has a histologically benign appearance, it behaves like a locally aggressive tumor. The structures near the primary area are attacked due to persistent local growth. If there is any lymphadenopathy, it is often reactive. The metastatic potential of this tumor is very rare.

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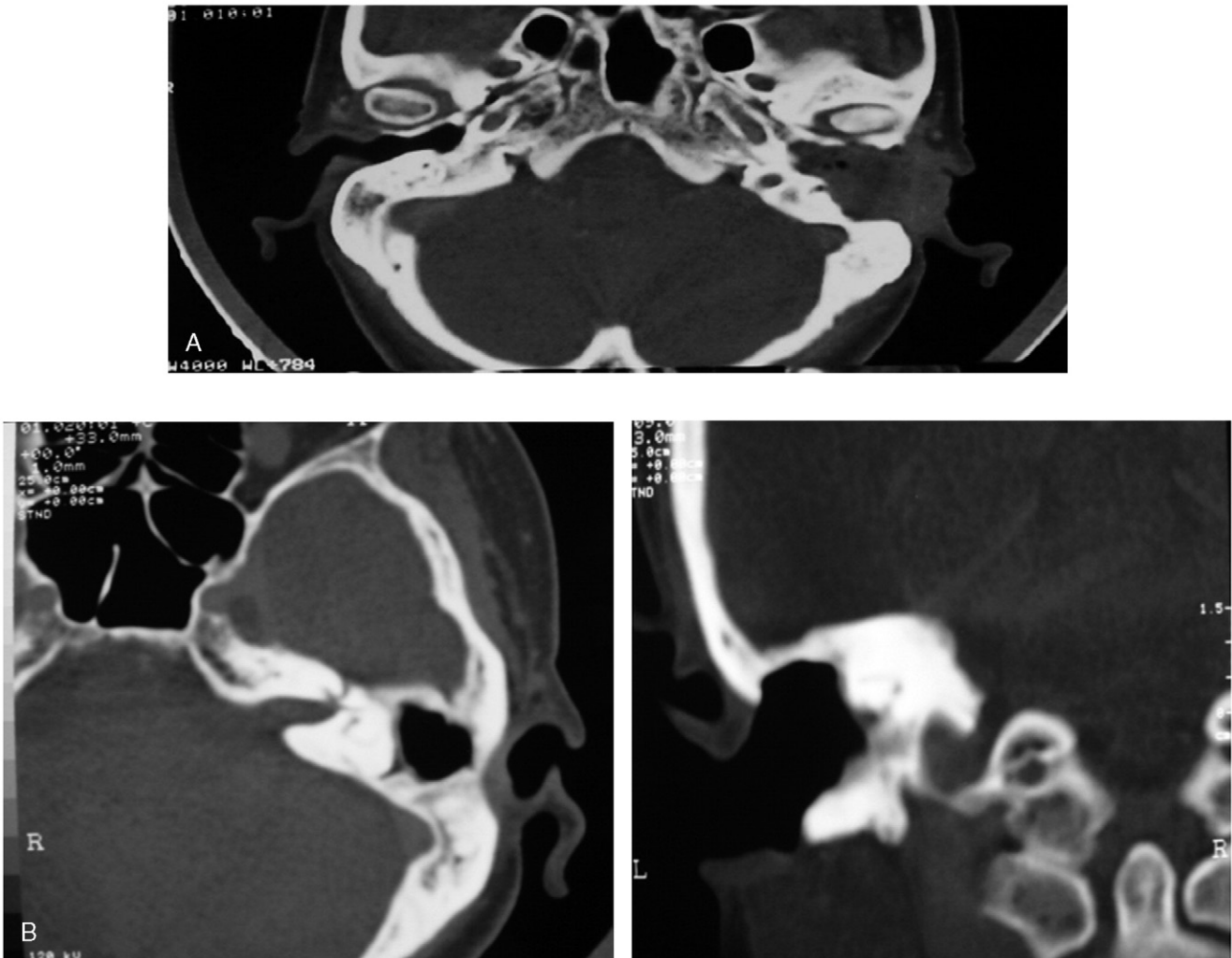


Fig. 1. (A) Preoperative temporal bone computed tomography of the patient demonstrating soft tissue filling the left mastoid cavity. (B) Postoperative temporal bone computed tomography of the patient demonstrating the tumor-free mastoid cavity.

Because the diagnosis of this tumor requires multiple and deep biopsies, the pathologic diagnosis of this neoplasm is often difficult. Small or superficial biopsy specimen often reveals only hyperkeratosis, acanthosis, and apparently benign papillomatosis [11]. Perhaps the only sign of malignancy is increased keratinocytes and nuclear area [12]. The biopsy specimen taken from our patient was not diagnostic. The revision mastoidectomy was planned to remove all the pathologic tissues and have them sent for histologic examination. Verrucous carcinoma was reported as the final diagnosis of our patient after the examination of the surgical specimen with multiple and deep sections.

Chronic local irritation plays a role in the etiology of verrucous carcinoma. When we review the literature, including our present case, 8 of 12 reported cases had a history of chronic otitis media, and 6 of these patients had previously undergone ear surgery [3–10]. Also, 2 of them had worn hearing aids in the affected ears for many years and 1 patient was a chronic ear picker [7]. Regarding chronic irritation, Ackerman [1] emphasized the role of tobacco

chewing in the verrucous cancer of the oral cavity. The occurrence of most larynx verrucous carcinoma cases in smokers supports this theory [13]. In addition, the temporal bone squamous cell carcinoma is often observed in patients who had chronic otitis media with lifelong otorrhoea [13–15].

The localization of the primary lesions of verrucous carcinoma in the temporal bone in these 12 patients were in external auditory canal (5 cases), in postoperative defect (6 cases), and middle ear (1 case) [3–10]. The lesion in our patient was localized in the radical cavity without any further extension.

In the treatment of the verrucous carcinoma, radiotherapy remains controversial. According to some authors, it carries a risk of anaplastic transformation to very aggressive squamous cell carcinoma and is definitely contraindicated in verrucous carcinoma [16–18]. However, some authors claim that it is an alternative treatment of this neoplasm [19,20]. Although Tharp and Shidnia [19] have reported no anaplastic transformation after the radiotherapy for head and neck verrucous carcinoma cases, according to their literature

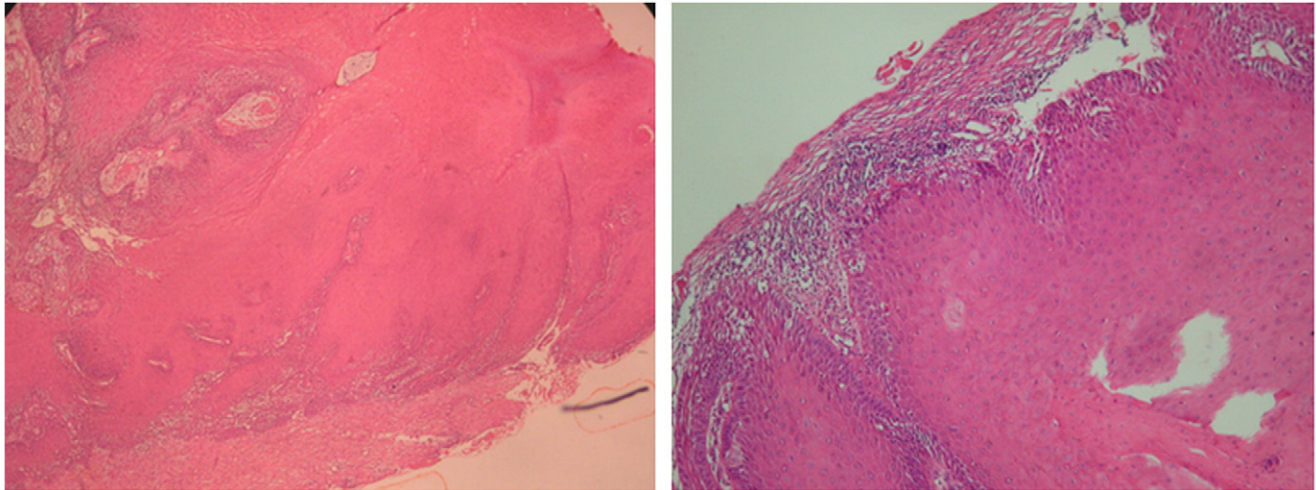


Fig. 2. Well-differentiated tongues of squamous epithelium pushing into the underlying stroma (left: hematoxylin and eosin stain, magnification $\times 40$; right: high-power view, hematoxylin and eosin stain, magnification $\times 100$).

review the risk of anaplastic transformation has been reported as 7%. Nevertheless, they suggest that the local control of the verrucous carcinoma with radiotherapy is less than 50%, but is between 74% and 86% with surgery [19].

Surgery seems to be the best therapeutic alternative in the treatment of this tumor. If the tumor is irresectable, radiotherapy may be suitable. We have operated on our patient without knowing that the diagnosis was verrucous carcinoma, but revision surgery was enough in the management of our patient without any further plan of postoperative radiotherapy. However, we have closely followed up our patient in the postoperative period.

The prognosis of the verrucous carcinoma is related to the spread of tumor. Lymph node metastases are surprisingly low in number and have been reported in only 9% of patients [21]. Five of 7 reported cases had extratemporal infiltration and subjects died within 20 months after the diagnosis. On the other hand, 3 of 4 cases without extratemporal survived for more than 4 years. The prognosis of the fourth case is unknown. Our case also had no extratemporal invasion and after our revision mastoidectomy operation, she has been tumor-free for 26 months, and her facial function has improved reasonably. In the follow-up period, we will be checking her postoperative temporal bone computed tomography for every 6 months. However, we would like to emphasize that a close follow-up of such patients is very important to detect any sign of recurrence as early as possible.

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