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Case Report

Basal Cell Carcinoma in the Middle Ear: A Case Report and Literature Review

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Although basal cell carcinoma (BCC) often occurs in the head and neck region, BCC in the middle ear is extremely rare. As far as we could determine, only five cases of BCC in the middle ear have been reported previously in the literature. We present the case of a 54-year-old Japanese man with BCC in the middle ear. Preoperative histopathologic examination indicated the possibility of skin appendage tumor. Mastoidectomy and extirpation of the lesion were performed, and the resected tumor was finally diagnosed as BCC. Four months after the initial surgery, the patient had local recurrence and reoperation, and postoperative radiotherapy was given. The patient had repeat local recurrence in the parapharyngeal space 3 years and 2 months following postoperative radiotherapy. The patient did not respond to two courses of chemotherapy with cisplatin and 5-fluorouracil, and eventually died of the primary disease 4 years and 7 months after the initial surgical treatment. We reviewed the five cases previously reported in the literature concerning BCC in the middle ear. More case reports concerning BCC in the middle ear are needed to determine its appropriate management.

KEYWORDS: Basal cell carcinoma, middle ear, recurrence

INTRODUCTION

Neoplastic diseases rarely occur in the middle ear. Although basal cell carcinoma (BCC) often occurs in the head and neck region,[1] BCC in the middle ear is extremely rare. To the best of our knowledge, only five cases concerning BCC in the middle ear have been reported previously[2-5]. The management of BCC in the middle ear has not been fully elucidated. We present a case of a patient with BCC in the middle ear and perform a literature review concerning BCC in the middle ear.

CASE PRESENTATION

A 54-year-old Japanese man had been treated by a local otolaryngologist for left-sided otitis media with effusion for 9 years. The patient presented with a 1-month history of discharge in the right ear. A granulomatous lesion was noted in the patient's left tympanic membrane, and so the patient was referred to our hospital for further evaluation.

At the initial visit, otoscopy revealed a pinkish granulomatous lesion in the posterior part of the tympanic membrane in contact with the eardrum ventilation tube (Figure 1). The right tympanic membrane was normal. No abnormal findings were observed in the head and neck region. The patient had negative T-SPOT®.TB test results. Serum levels of proteinase 3 antineutrophil cytoplasmic antibody and myeloperoxidase-antineutrophil cytoplasmic antibody were 1.0 U/mL (normal range: <3.5 U/mL) and 1.0 U/mL (normal range: <3.5 U/mL), respectively. Audiometry demonstrated a 43-dB conductive hearing loss in the left ear, with no hearing loss in the right ear. The patient did not show any signs of facial palsy or vertigo. Computed tomography targeting the ear revealed a soft tissue density filling the tympanic and mastoid cavities in the left middle ear; bone destruction was not identified (Figure 2). Results of preoperative histopathologic examination suggested the possibility of skin appendage tumor. On laboratory and histopathological examinations, inflammatory diseases such as tuberculosis and anti-neutrophil cytoplasmic antibody-associated disease were ruled out.

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Mastoidectomy and extirpation of the lesion were performed using a transmastoid approach. A tumor was found in the left mastoid sinus and tympanum. On histological examination, dense cells with a high nuclear/cytoplasmic ratio were identified, and the nuclei showed a palisade arrangement on the periphery of cell clumps (Figure 3). The diagnosis was finally confirmed as low-grade BCC in the middle ear. According to Stell's classification,[6] the clinical stage was classified as T2. Positron emission tomography revealed absence of distant metastasis. Four months after the initial treatment, the patient had local recurrence and underwent reoperation and postoperative radiotherapy. A total of 66 Gy of conventional radiotherapy was administered in 33 fractions. In the follow-up, the patient had repeat local recurrence in the parapharyngeal space 3 years and 2 months following postoperative radiotherapy. As the tumor was not resectable because of carotid and intracranial invasion (Figure 4), two courses of chemotherapy with cisplatin and 5-fluorouracil (5-FU) were administered. The treatment was ineffective, and the patient died of the primary disease 4 years and 7 months after the initial surgical treatment.

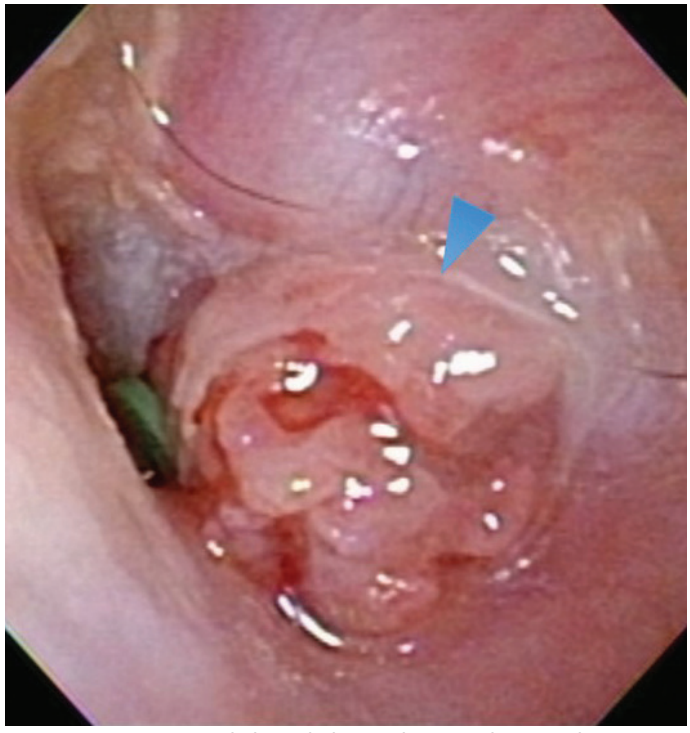

Figure 1. Otoscopy revealed a pinkish granulomatous lesion in the posterior part of the tympanic membrane in contact with the eardrum ventilation tube.

MAIN POINTS

- Basal cell carcinoma in the middle ear is extremely rare. To date, there is no standard management protocol for this condition.
- We reviewed the five cases previously reported in the literature concerning BCC in the middle ear.
- The present case died of the primary disease 4 years and 7 months after the initial surgical treatment. More case reports concerning basal cell carcinoma in the middle ear are necessary to determine its appropriate management.

Literature review including the case reported here in this study

To date, only five patients with BCC in the middle ear have been reported [2-5]. The details, including those of the patient reported in our study, are summarized in Table 1. Patients' ages ranged from 34 to 60 years, with an average of 48.8 years, with equal sex distribution (3 men; 3 women). Hearing loss was reported in five of the six patients. Three patients had otalgia, two had ear discharge, and one had facial palsy and an impaired sense of taste. On local examination, four patients had a granulomatous lesion, one had a pale and bulging eardrum, and one had a small pearl at the pars flaccida. The patient who did not undergo any treatment modality had lung and liver metastasis and eventually died 10 years after the diagnosis. With regard to surgical procedures, tumor resection and mastectomy were carried out in four patients, including our patient, whereas one patient was treated with tumor resection alone. The patient who showed a

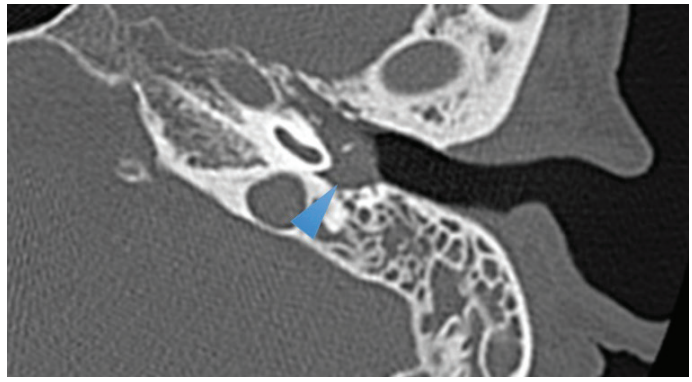

Figure 2. Computed tomography targeting the ear revealed a soft tissue density filling the tympanic and mastoid cavities in the left middle ear, and there was absence of bone destruction.

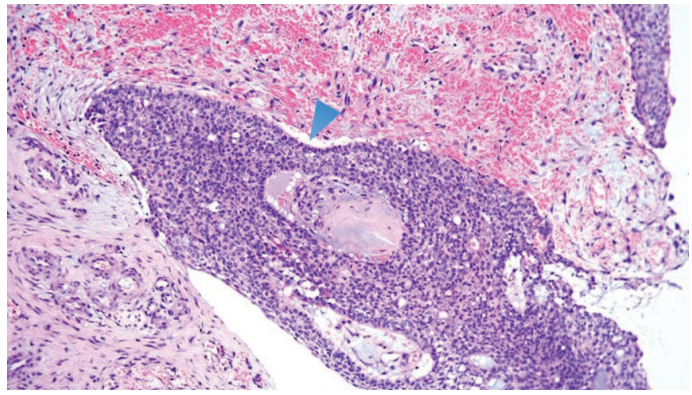

Figure 3. Dense cells with a high nuclear/cytoplasmic ratio were identified, and the nuclei showed a palisade arrangement on the periphery of cell clumps.

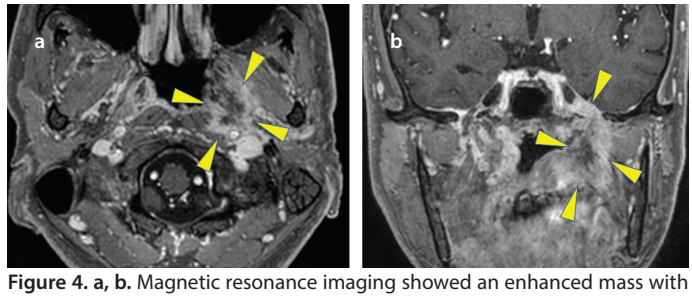

Figure 4. a, b. Magnetic resonance imaging showed an enhanced mass with carotid and intracranial invasion at the parapharyngeal space on axial (a) and coronal (b) T1-weighted imaging (contrast).

Table 1. The list of basal cell carcinoma in the middle ear previously reported

Author	Year	Sex	Age	Symptom	Local findings	Initial treatment	recurrence	2nd treatment	recurrence	3rd treatment	Observation period	Prognosis
Brunner et al. [2]	1953	F	53	O, FP, Taste	brownish granulation	TR and M, RT	local (18 months)	TR, ND	-		4 years	Disease free survival
		F	47	O, HL	grayish-red, firm tissue	TR and M, RT	-				4 years	Disease free survival
Joachims et al. [3]	1988	M	34	O, HL	pale and bulging ear drum	none	-				10 years	Death from illness
Lim et al. [4]	2010	M	60	HL	small pearl	TR	-				unknown	unknown
Shankar et al. [5]	2015	F	45	ED, HL	pinkish proliferative polpoidal mass	TR and M, RT	-				unknown	Disease free survival
Present study		M	54	ED, HL	pinkish granulation	TR and M	local (4 months)	TR, RT	local (3 years and 2 months)	chemotherapy	4 years and 7 months	Death of the primary disease

F: Female; M: Male.
O: otalgia; FP: facial palsy; Taste: impairment of taste; HL: hearing loss; ED: ear dicharge.
TR and M: tumor resection and mastoidectomy; RT: radiotherapy; TR: tumor resection; ND: neck dessection.

small pearl-like BCC underwent surgery alone without radiotherapy because the patient was still able to hear. Two of the previous three patients showed no recurrence following postoperative radiotherapy. The patient who had recurrence following postoperative radiotherapy recovered after undergoing reoperation.

DISCUSSION

Squamous cell carcinoma is the most frequent malignant tumor of the ear, and the auricle is involved in 85% of the cases, the external canal in 14%, and the middle ear in 2% [2]. Although 85%-90% of all BCC lesions occur in the head and neck region,[1] BCC in the middle ear is extremely rare. Although BCC of the external canal originates from the basal layer of the epidermis or the sweat glands in the external canal, the mechanism underlying the occurrence of BCC in the middle ear has not been completely understood [2]. Brunner et al. [2] speculated that BCC in the middle ear originates from the hypotympanum or from the area close to it. Additionally, a primary tumor may run a latent course as long as it is limited to the hypotympanum but may also penetrate the tympanic membrane and grow into the external canal. Moreover, in the present case, the patient was treated for left-sided otitis media with effusion for 9 years. Therefore, there is a possibility that the tumor was localized in the hypotympanum for a long time until it finally invaded the surface of the tympanic membrane.

Stell's classification is generally applied in the staging of middle ear carcinoma [6]. The present case was classified as T2. T2 patients with middle ear carcinoma localized in the temporal bone can be managed successfully with radical mastoidectomy and postoperative radiotherapy [7]. Treatment for BCC mainly consists of surgery, radiotherapy, chemotherapy, and Mohs micrographic surgery [5]. The preferred treatment for BCC is complete resection [8]. The three types of surgical treatments for temporal bone tumors are as follows: lateral temporal bone resection, subtotal petrosectomy, and simple tympanomastoidectomy. The type and amount of surgery for BCC in the middle ear depends on the location of the carcinoma in the ear.[5] When BCCs are located in regions, such as the ear, where aggressive resection would lead to significant functional consequences, the appropriate extent of surgery is controversial.[8] Additionally, it is difficult to maintain a surgical margin during middle ear surgery. In the cases we reviewed, tumor resection and mastectomy were performed. Two of the previous three patients had no recurrence following postoperative radiotherapy. One patient with recurrence following postoperative radiotherapy recovered after undergoing reoperation. In the present case, although the tumor in the middle ear region was controlled with tumor resection, mastoidectomy, and postoperative radiotherapy, recurrence occurred in the parapharyngeal space due to infiltration believed to be through the Eustachian tube. The parapharyngeal space was not included in the scope of surgical resection or radiotherapy. Therefore, BCC that originates in the middle ear may require a subtotal petrosectomy including the Eustachian tube region or extensive irradiation including the parapharyngeal space. Chemotherapy for BCC has been reported to include the administration of cisplatin and adriamycin,[9] or local therapy with 5-FU [5]. In the present case, cisplatin and 5-FU were administered for recurrence in the parapharyngeal space, but both treatments were ineffective. To date, there is no standard management protocol for this condition as the number of patients with BCC in the middle ear is relatively small.

It has been reported that BCC can be locally invasive but can have relatively low metastatic potential.[8] The presence of preoperative risk factors (with a history of malignancy, radiotherapy, scarring, xeroderma pigmentosum, or basal cell nevus syndrome) and the presence of multiple lesions were considered as predictors of recurrence.[1] Our patient had no predictors of recurrence but died of primary disease due to local recurrence. More case reports concerning BCC in the middle ear are necessary to determine its appropriate management.

CONCLUSION

The present case died of the primary disease 4 years and 7 months after the initial surgical treatment. To date, there is no standard management protocol for BCC. More case reports concerning BCC in the middle ear are necessary to determine its appropriate management.

Informed Consent: Written informed consent was obtained from the patient who participated in this study.

Peer-review: Externally peer-reviewed.

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Conflict of Interest: The authors have no conflict of interest to declare.

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