Hematogenous Renal Cell Carcinoma Metastasis in the Postoperative Temporal Bone

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Abstract
Metastatic renal cell carcinoma (RCC) involving the temporal bone is a rare entity. It is usually asymptomatic and misdiagnosis as acute otitis media, mastoiditis, and Ramsay-Hunt syndrome in early onset is not uncommon. We report a case of RCC metastasis to the postoperative temporal bone in the middle of molecular targeted therapy. A 60-year-old man presented left facial palsy with severe retro-auricular pain and he also underwent left middle ear surgery for cholesteatoma more than 30 years before and had been aware of discontinuous otorrhea; therefore, initially we speculated that facial palsy was derived from recurrent cholesteatoma or Ramsay-Hunt syndrome. Exploratory tympanotomy revealed RCC metastasis and postoperative MR indicated hematogenous metastasis. To the best of our knowledge, no report was obtained on temporal bone metastasis in the middle of chemotherapy or hematogenous metastasis in the postoperative middle ear. Metastasis in the temporal bone is still a possible pathological condition despite the development of present cancer therapy. Besides, this case indicates that hematogenous metastasis can occur in the postoperative state of the temporal bone.
Case Presentation

A 60-year-old man presented with rapidly progressing left facial palsy and severe retroauricular pain. He had not experienced vestibular symptoms; however, the patient reported hearing loss and tinnitus on the left ear. Before coming to our center, he had consulted a private ENT (Ear-Nose-Throat) clinic. Despite the unusually severe pain, an initial diagnosis of symptoms was Ramsay-Hunt syndrome, and oral steroid therapy was initiated. He had also undergone left-sided nephrectomy for renal cell carcinoma (RCC) 2 years earlier, and metastasis to a hilar lymph node had been detected 1 year postoperatively. No local recurrence had been observed, and molecular targeted therapy (sunitinib) had been performed for the RCC metastasis at the first consultation in our center. Past otologic history included left-sided middle ear surgery for cholesteatoma more than 30 years earlier, which had presented as the ipsilateral conductive hearing loss on pure tone audiogram and awareness of discontinuous otorrhea. Ear endoscopy revealed a reddened region around the posterior-superior ear canal (Fig. 1), but no mucosal otorrhea or perforation of the tympanic membrane or posterior canal wall was observed. High-resolution CT revealed an intratympanic and mastoid mass involving the facial nerve in the mastoid and tympanic segments (Fig. 2a). Due to the postoperative state, distinguishing whether the pathology represented recurrent cholesteatoma, viral infection, or tumor invasion was difficult. Otalgia was not able to be controlled using oral analgesics, so exploratory tympanotomy and mastoidectomy were planned for biopsy at the perifacial nerve on the day following the first visit, to avoid any delays in diagnosis of a potentially malignant tumor. The patient and his family consented to biopsy of the facial nerve envelope. No cholesteatoma was detected on biopsy, and granulomatous tissue was seen to be filling the tympanic and mastoid cavities. Beneath the granulomatous tissue, a white mass was observed involving the facial nerve. A biopsy specimen was obtained from around the mastoid segment of the facial nerve, and histologic appearance of the specimen demonstrated patterns of clear cell type-RCC compatible with metastasis of the original tumor (Fig. 3). Otalgia and retro-auricular pain were relieved for a few days after surgery, but severe pain recurred, and postoperative MRI showed a mass occupying the sigmoid sinus, indicating hematogenous metastasis (Fig. 2b). Consultation with experts in urology led to palliative care.

Discussion

Metastasis can occur when cells break away from a cancerous tumor and travel through the bloodstream or lymph to other areas of the body. Determination of how a metastasis arose in a region with little in the way of blood supply or lymph networks is thus difficult. Metastatic cancer cells in the temporal bone are more likely to have traveled through the blood from distant organs or tissues, due to the relative absence of lymphatics. However, the postoperative state commonly leads to scar formation and reductions in blood supply to the surrounding area, as usually seen on re-operation. Though in addition to the rarity of RCC metastasis in the temporal bone, with metastasis occurring in the middle of treatment of original cancer and a region of reduced blood supply, a major problem in this case was that both the decisions of the urologist and ENT doctors brought about delayed diagnosis and the patient was troubled by severe pain and unable to receive sufficient terminal care. Clinical application of molecular targeted therapy has been the first-line option for metastatic RCC since the beginning of this century [1]. Although median progression-free survival is report-
edly 6–11 months with molecular targeted therapy, compared with 5–6 months with the previously dominant immunotherapy, molecular targeted therapy remains palliative [2]. Metastatic RCC cells can reach the head and neck area via normal hematogenous flow. The most important route of metastasis is tumor embolization via the Baston plexus, the anastomosis of the valveless vertebral epidural venous system [3]. In our case, the sigmoid sinus was completely occupied by tumor, and tumor extension had reached the temporal bone. These findings imply that the metastasis had occurred through the Baston plexus. RCC metastasis in the temporal bone is quite uncommon [4–6], and this is the first reported case of not only RCC metastasis to the temporal bone during molecular targeted therapy, but also metastasis to the temporal bone.

Conclusion

It is understandable that metastasis occurs in a higher vascularized region, and it arises as a result of uncontrollable advanced cancers. Besides, it is no doubt that the introduction of chemotherapy leads to the less rapid progression of cancer compared with the past. However, the possibility of the disease derived from a malignancy or metastasis to the temporal bone must be kept in mind for patients presenting with hemifacial palsy accompanied by severe pain, even if the first disease was not only considered to be cured or under treatment, but also in a postoperative state.

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Statement of Ethics

The Ethics Committee of Kansai Medical University approved this case for presentation and publication. Informed consents were obtained from the patient for publication of this case report and accompanying images.

Disclosure Statement

The authors declare no conflicts of interest associated with this case report.

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**Author Contributions**

Masaya Konishi carried out the surgery and participated in the sequence alignment and drafted the manuscript. Hiroshi Iwai also carried out the surgery and participated in the clinical management. Kensuke Suzuki suggested this case for publication and helped to draft the manuscript. All authors read and approved the final manuscript.

**References**


**Fig. 1.** Endoscopic view of the left ear. Not only reddened and swelling tympanic membrane, but also a partial defect of the posterior ear canal wall can be observed.
Fig. 2. a Preoperative coronal CT scan shows a large jugular bulb on the right, and the facial nerve (arrow-head) and a boundary to the facial nerve in the mastoid segment is obscure. Similar findings are also indicated in the axial view (arrow). Tumor invasion to the facial nerve could not be conclusive and distinguish from cholesteatoma recurrence and viral infection through this image. b Postoperative MR image showing a mass occupying the jugular bulb on the right (arrowhead) and its continuity extend to the sigmoid sinus (arrow).

Fig. 3. Photomicrograph of the biopsy specimen shows large neoplastic cells with clear cell type-cytoplasm (arrow), which suggests metastasis of RCC (HE stain, ×200). RCC, renal cell carcinoma.