Intra-temporal facial nerve abscess due to basal cell carcinoma of external auditory canal: A unique presentation

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ABSTRACT
Infranuclear facial palsy following facial nerve abscess in intra-temporal course has not been described in literature as per our knowledge. After extensive search of literature, only one case of facial nerve sheath abscess was found in a case of leprosy, but the abscess was in the intra-parotid course of the nerve. Basal cell carcinoma (BCC) is a rare malignant tumor of external auditory canal (EAC). Facial nerve palsy in BCC occurs only in advance stages when the tumor erodes the floor of EAC to involve the nerve. Contrast enhanced computed tomography of the temporal bone is useful in diagnosing the entity pre-operatively and in planning the management in such cases. This case is being reported as facial nerve sheath abscess in intra-temporal course due to malignancy of the EAC has not been reported as yet.

Key words: Basal cell carcinoma, external auditory canal, facial nerve abscess

INTRODUCTION
Non-melanoma skin cancer (NMSC) is the most common cancer in the United States with an incidence 18-20 times greater than that of malignant melanoma.[1] The lifetime risk of developing NMSC were estimated to be 29-55% for basal cell carcinoma (BCC) and 7-11% for squamous cell carcinoma (SCC).[2] About 80% of NMSC are BCC and 20% are SCC.[3]

BCC represents approximately 11% of tumors of the external auditory canal (EAC) and temporal bone.[4-6] Its actual incidence in population is reported to be one to six patients per million per year.[7,8] Because of its high frequency and low mortality, BCCs are not routinely registered and incidence estimates have largely come from ad hoc studies.[9] Due to its rarity, it is difficult to develop a proper staging system and treatment for BCC. The American Joint Committee on cancer includes auricular malignancy in their staging system for non-melanomatous lesions of the skin.[10] Facial nerve sheath abscess leading to facial nerve paralysis is an extremely unique presentation in a case of BCC of EAC.

BCC incidence has a strong inverse relationship with latitude.[11] In general, chronically sun exposed parts of the body develop a larger proportion of BCCs than those with less or infrequent exposure.[12] BCC is the predominant skin cancer in Caucasians[13] and its treatment imposes a substantial economic burden.[14,15] Its incidence in the Indian population is very rare. Our patient was a farm laborer and this segment of population is exposed to excessive sunlight and that can be one of the risk factors in our patient.

CASE REPORT
A 45-year-old female presented to neuro-otology out-patient department of our tertiary care institute with complains of right ear discharge, which was occasionally blood tinged, for about 9 months. Subsequently, she developed right-sided facial weakness which was slowly progressive for last 5 months. Hearing loss was also present but it was not accompanied by any history of tinnitus or giddiness. On examination, her right external ear was filled with pale...
reddish fleshy polypoidal mass and tympanic membrane landmarks were not visible. Pure tone audiogram showed moderate conductive hearing loss. With the above findings, differential diagnosis of chronic suppurative otitis media, glomus tympanicum or jugulare and SCC of EAC was kept. Biopsy from the EAC was done and sent for histopathology. Meanwhile, contrast enhanced computed tomography (CECT) of temporal bone was also got done. CECT temporal bone showed right-sided facial nerve sheath ballooning with edema of the nerve present. The tympanic membrane was involved by the mass but the middle ear was free of disease in the CECT temporal bone scan [Figures 1 and 2].

The patient was then planned for surgery of the right ear canal wall-down mastoidectomy with facial nerve decompression. Intra-operatively, canal wall-down mastoidectomy was done and facial nerve was exposed from geniculate ganglion to stylomastoid foramen. While clearing the disease, we encountered dehiscence of the facial bony canal in its vertical course and the nerve sheath was highly edematous with twice the caliber of the normal size. When facial nerve sheath was incised in its vertical portion, pus was encountered with yellowish-cheesy content coming out of the sheath [Figure 3]. Cheesy content was immediately sent for frozen section along with a portion of sheath of nerve and it was reported to be free of malignant cells. Even though facial nerve sheath was not involved by tumor cells, we resected the edematous portion of the nerve taking 5 mm of margin on both sides and primary grafting of resected facial nerve was done with greater auricular nerve. Malleus and incus were removed from the attic region to ensure complete disease removal from middle ear. Temporalsis fascia grafting was done to cover the middle ear and mastoid cavity.

Histopathology of the sent tissue showed a tumor composed of infiltrating solid nest and lobules with peripheral palisading of tumor cells. The tumor cells are small to medium sized with high nucleo-cytoplasmic ratio having hyperchromatic nuclei and small amount of cytoplasm. Mitotic figures are also evident and the tumor is also infiltrating surrounding fibrocollagenous tissue. A bit of tissue lined by squamous epithelium along with subepithelial tumor is also seen, suggestive of BCC. Post-operative CECT at 3 months follow-up did not show any recurrence of the disease in the middle ear and EAC, although facial palsy improved from House-Brackmann grade 5 to grade 4.

**DISCUSSION**

Intra-temporal facial nerve sheath abscess due to malignancy of EAC and middle ear has not been reported in literature so far. Only one case of facial nerve sheath abscess has been reported in a case of leprosy, but it was in the intra-parotid course of facial nerve. Although facial nerve palsy in BCC due to erosion of floor of EAC is a known complication, but development of intra-temporal facial nerve sheath abscess is not documented or reported in the literature till date and hence we are reporting this unique presentation.

Malignant tumors of the EAC and temporal bone are rare in occurrence. They account for less than 0.2% of all head
and neck tumors. Out of these, SCC is most common variant followed by BCC. BCCs are slow-growing tumors which spread locally and rarely metastasize. Excessive sun exposure is a risk factor for their occurrence. Temporal bone involvement most often occurs as tumor extends along the EAC toward the tympanic membrane and then it involves the middle ear. However, in our case, the tumor was in deeper EAC, also involving the tympanic membrane, but it was not involving the middle ear. The facial nerve involvement in our case was through the erosion of the floor of EAC.

The incidence of persons affected by BCC is likely to substantially underestimate the true incidence of this cancer due to the common occurrence of multiple primary tumors within individuals synchronously or at different times. It has been observed previously that 43% of people affected by BCC developed a subsequent BCC within 4.5 years of active surveillance in the same population and meta-analysis of seven independent studies showed that mean 3 year risk of BCC is around 44% after an initial diagnosis of BCC in North America.

A study by Buettner and Raasch that included only single lesions per person reported BCC incidence was highest on the combined subsites of lip, orbit, nasolabial, ear, nose, and cheek. Our patient also had BCC of the EAC, which is included in the common sites for BCC. The top five subsites (nose, cheek, eye area, forehead, and ears) correspond closely to the subsites that receive the greatest UV exposure, apart from the forehead which receives the second highest level of UV exposure but had the fourth highest incidence of BCCs. This difference could be explained by the sun protection of the forehead offered by hair cover (which can decrease solar UV exposure by up to 80%). The anatomic site distribution of BCCs on non-facial regions was less predictable and not as directly associated with general UV exposure.

Diagnosis of BCC is mainly based on histopathological examination of the mass either from EAC or from middle ear. Nodular BCC is the most typical, usually arising in head and neck region. It is a slow-growing, fleshy or pearly colored nodule, which is locally invasive. Other varieties like superficial, pigmented and morphiec types are not common in head and neck region. CECT should be done pre-operatively to see extension of disease, to know the stage of the disease and to plan management accordingly.

Extension of BCC from EAC to adjacent surrounding tissues can occur either through natural anatomical fissures or through erosion of bone. Anterior spread occurs through fissures of Santorini, petrosquamous sutures, and foramen of Lushka to temporo-mandibular joint and parotid area causing trismus and pain. Inferior spread through erosion of floor of EAC into the stylomastoid foramen and fallopian canal can leads to facial weakness or palsy. Medial spread can involve tympanic membrane and middle ear cleft and can result in facial nerve palsy and conductive deafness. Medial extension with extension into hypotympanum can also lead to jugular foramen syndrome. Superior spread to epitympanic space along with erosion of tegmen tympani causing intracranial extension can be seen in advance cases only.

Due to the rarity of these tumors, there is a lack of a universally accepted staging system, thus making it difficult to analyze data, formulate, and evaluate a treatment strategy. Pre-operative clinical staging is difficult in these tumors because large parts of the temporal bone are not amenable to clinical examination. Treatment of choice for BCC is radical surgery taking a healthy margin of 1-1.5 cm to prevent recurrence followed by risk adapted post-operative radiotherapy. The suggested surgery for complete resection of tumor of the EAC, middle ear, and temporal bone is a lateral temporal bone resection (resection of the bony EAC, the tympanic membrane, the malleus and the incus with the medial limit of the incudostapedial joint), stage dependent, combined with a neck dissection. The most important survival factor is removal of the primary tumor with histologically clear margins. Risk factors for incomplete excision and recurrence includes sensitive locations like ears, eyes, and nose. In these cases, it may be better to perform a more radical excision taking 2 cm healthy margin. Incomplete excision leads to tumor recurrence with 50% occurring in first 2 years and 66% in first 3 years. So, complete resection with a healthy margin during primary surgery is very important, though it is challenging in ear cases due to its complicated anatomy as compared with other skin bearing areas. Moh’s microsurgery, which is the gold standard for BCC of the skin, is not applicable in tumors of EAC and temporal bone. The main advantage of Moh’s microsurgery (MMS) is the ability to examine 100% of the surgical margins, making incomplete excision less likely. The cure rates reported for primary tumors treated with MMS were 93.3% and 94.8%, respectively.

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