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RESEARCH ARTICLE
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BILATERAL EXTERNAL AUDITORY CANAL CARCINOMA

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Abstract- Carcinoma of External Auditory Canal is rare and Carcinoma of Bilateral External Auditory Canal is even rarer. We are presenting a case of Carcinoma of Bilateral External Auditory Canal and how it was managed.

Mesh Terms- Lateral Temporal Bone Resection, Carcinoma of External Auditory Canal

Introduction- Carcinoma of External Auditory Canal (EAC) is a rare tumor with prevalence of 1-6 per million populations[1]. Accordingly Carcinoma of Bilateral EAC is exceeding rare with just 5 reported cases so far. Carcinoma of EAC often mimics benign and infective conditions such as Chronic Suppurative Otitis Media (CSOM) and Otitis Externa thus making it challenging to diagnose. Patients often present with symptoms of otorrhea, hearing loss and occasional ear bleed. Such patients are treated conservatively initially, leading to delay in proper treatment. Late presentation includes mass in the ear, facial palsy, vertigo and tinnitus and indicates involvement of middle ear and inner ear by the tumor.
We present a case of Bilateral EAC Carcinoma, in a 32 years old dwarf, which presented 5 months apart.

Case Report

A 32 years old dwarf presented to us with history of Left sided otorrhea since past 3 years and Left sided deafness and ear fullness since past 3 months. He also gives history of occasional ear bleed from left side. He was diagnosed as Left CSOM and was being treated conservatively for past 3 years by a physician. The patient denies history of tobacco and alcohol abuse and there no history of him being exposed to radiation therapy.
Upon otoendoscopy, an exophytic mass could be seen in Left EAC completely occluding the EAC. Tympanic Membrane and most of the EAC wall skin was not seen. [Figure 1]The mass bled on touch but was painless. The mass was biopsied and histopathological examination revealed it to be Poorly Diffentitated Adenocarcinoma. Audiological Examination revealed moderate to severe mixed hearing loss on left side. HRCT Temporal Bone [Figure 2] showed polypoidal soft tissue mass in Left EAC with subtle erosions of bony EAC and non-visualization of Left Tympanic Membrane. Middle ear and inner appeared to be uninvolved. Anatomy was found to be unfavorable with low placed dura and forward sigmoid sinus. Due to such anatomical variations, the possibility of performing a Posterior Tympanotomy appeared difficult and a modified Lateral Temporal Resection was planned. FDG-PET Scan [Figure 3] showed hyper metabolic lesion in Left EAC and

bilateral low grade avid cervical lymph nodes at levels IA and IB. Based on clinical and radiological examination, the tumor was staged as T2. The patient was then posted for Lateral Temporal Bone Resection with Superficial Parotidectomy and Sampling of the cervical lymph nodes followed by Bilateral Modified Neck Dissection with Obliteration of Left Mastoid Cavity and Postoperative Radiotherapy. An Intraoperative Nerve Integrity Monitor was used for identification of facial nerve.

All the surgical margins were negative for malignancy and the unilateral Lymph Nodes were found to be metastatic with no extracapsular spread. The patient underwent 30 cycles of radiotherapy postoperative.

In 5th month of post-operative period, a routine otological examination revealed a polypoidal mass in the Right EAC on the posterior canal skin.[Figure 4] The mass was biopsied and histopathological examination revealed it to be Squamous Cell Carcinoma-in-situ. HRCT Temporal Bone[Figure 5] revealed as soft tissue in Right EAC confined to the lower part of Posterior Canal Wall Skin with no evidence of involvement of bone, middle ear or inner ear. Interestingly, the anatomy on right side was favorable with no anatomical variations. Based on the clinical examination and radiological investigations the tumor was staged as T1. The patient was then posted for Lateral Temporal Bone Resection with Superficial Parotidectomy. All the margins were found to be negative for tumor and middle ear reconstruction was done.

DISCUSSION

Squamous cell carcinoma of the temporal bone and external auditory canal is a rare tumor with a reported incidence of between 1 to 6 cases per million population per year [1] and it accounts for less than 0.2% of all tumors of the head and neck [2].To our knowledge there have been only 5 cases of Bilateral Carcinoma of EAC have been reported so far. In general, tobacco and alcohol use are the two most important risk factor associated with squamous cell cancers of the head and neck. Squamous cell carcinoma of the temporal bone and external auditory canal is often associated with chronic otitis media and exposure to radiation therapy [1].

Otorrhea and otalgia are the most common symptoms of temporal bone tumors [3]. Because these symptoms are similar to those of otitis externa and chronic suppurative otitis media, EAC cancer is easily misdiagnosed and treated as those common otologic diseases. This causes delay in the proper management of the tumors. The diagnosis is based on biopsy of the mass. The biopsies taken under local anesthesia often yield false negative results, necessitating repeat or deeper biopsy.

So it's prudent to consider deeper biopsy under general anesthesia. Because of the rarity of squamous cell carcinoma of the temporal bone and auditory canal, developing an accurate tumor staging system and treatment has been challenging[4]. The University of Pittsburgh staging system for primary squamous cell cancer of the external auditory canal was proposed by Arriaga et al. in 1990 to attempt to classify the disease prior to treatment[5].Although it has its limitations for determining soft tissue extension of the tumor, the development of a TNM staging system using preoperative high-resolution CT scans of the temporal bone has been confirmed by other studies to accurately reflect the extend of the disease and planning of tumor resection [6]. The staging system does not include the direction of the spread of the tumor (anterior, posterior, or inferior). This explains the diversity of approaches employed in some patients in this series. Another confounding issue is the difficulty in differentiating between T1 and T2 tumors. Gillespie et al. in 2001 published a retrospective chart review of 15 patient treated for squamous cell carcinoma and reported that the University of Pittsburgh staging system correlated with patient outcome and was more sensitive than preoperative radiological staging[7].

Modified Pittsburg staging system (Morris et al., 2012; Moody et al., 2000; Moffat and Wagstaff, 2003).

Tumor	Description
T1	Tumor limited to the EAC without bony erosion or evidence of soft tissue involvement.
T2	Tumor limited to the EAC with bone erosion (not full thickness) or limited soft tissue involvement (G0.5 mm).
T3	Tumor eroding the osseous EAC (full thickness) with limited soft tissue involvement (G5 mm) or tumor involving the middle ear and/or mastoid.
T4	Tumor eroding the cochlea, petrous apex, medial wall of the middle ear, carotid canal, jugular foramen, or dura; or tumor with extensive soft tissue involvement (95 mm), such as involvement of temporomandibular joint or styloid process; or with evidence of facial paresis.
Lymph nodes	
N0	No regional nodes identified
N1	Single ipsilateral regional node <3 cm in size
N2a	Single ipsilateral regional node 3–6 cm in size.
N2b	Multiple ipsilateral nodes
N2c	Bilateral or contralateral nodes
N3	Node >6 cm
Tumor stage	
Stage I	T1 N0
Stage II	T2 N0
Stage III	T3 N0
Stage IV	T4 N0 and T1-T4 N+

The treatment of carcinoma of EAC is usually surgical and followed by postoperative radiotherapy in some cases. In March 2002, Belgium Consensus Conference was held Catholic University of Leuven, St. Lucas University Hospital, Leuven. At this conference evidence-based guidelines for the management of patients with external auditory canal carcinomas were laid down. The recommended therapy for T1 tumors is lateral temporal bone resection with neck dissection, and for T2 tumors, lateral temporal bone resection with neck dissection followed by radiotherapy. Treatment for T3 lesions is Subtotal Temporal Bone Resection with neck dissection followed by radiotherapy whereas treatment for T4 lesions is Palliative Chemo-radiotherapy.

In all the previous five reported cases of Carcinoma of Bilateral EAC, there has been an interval of few months to few years between presentation of disease in the opposite side. And all the patients have had undergone postoperative radiotherapy. Only one case, reported by Çağdaş Elsürer et[9] all did the carcinoma present itself bilaterally at the same time because the patient had undergone Radiotherapy for Nasopharyngeal Carcinoma. So based on this evidence, it might not be farfetched to assume that Radiotherapy does play a role in initiating carcinomatous changes on the previously normal side, in a few individuals. Further research is warrantied to reach a definitive conclusion.

SUMMARY

- Carcinoma of External Auditory is a rare entity and Carcinoma of Bilateral External Auditory Canal is even rarer with only 5 reported cases worldwide.
- Carcinoma of EAC mimics benign conditions like Chronic Suppurative Otitis Media and Otitis Externa, leading to delay in diagnosis.
- HRCT and MRI Temporal bone play a vital role in staging the disease and planning the management.
- Lateral Temporal Bone Resection with Parotidectomy and Neck Dissection with or without postoperative radiotherapy is the treatment of choice.

- The rest 5 reported cases show formation of cancer after exposure to radiation suggesting an adverse effect of radiotherapy in susceptible individuals.
- Further research is warranted for any conclusive correlation.

Figure 1

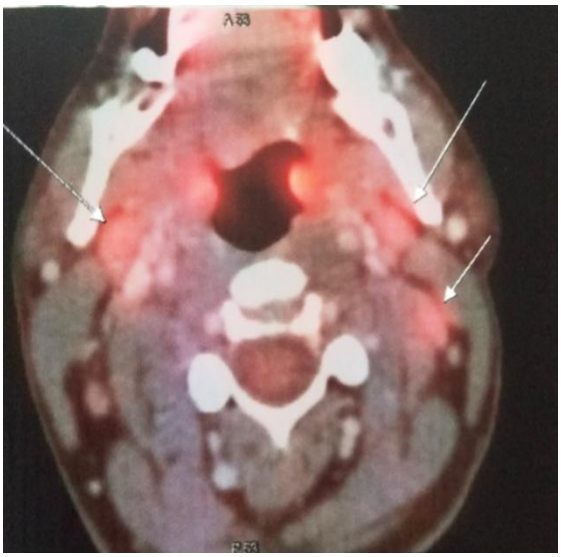

Figure 2

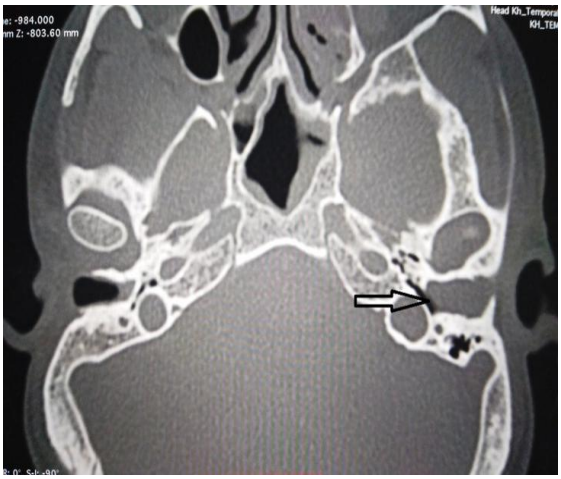

Figure 3

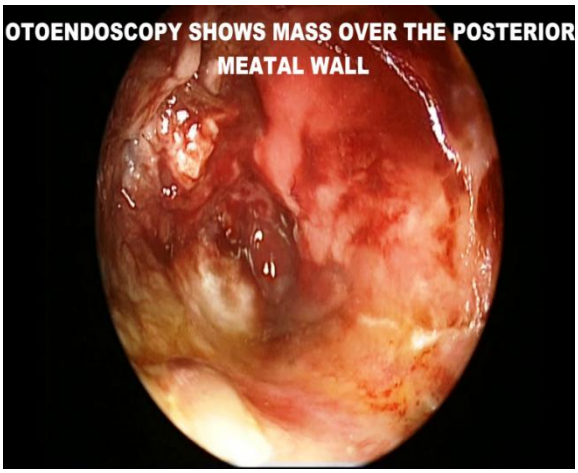

Figure 4

Figure 5

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