Case Report

Incipient Warthin tumor of intraparotid lymph node in a patient with squamous cell carcinoma of the external auditory canal: An incidental finding

ABSTRACT

The occurrence of intranodal salivary gland neoplasm is uncommon; squamous cell carcinoma (SCC) of the external auditory canal (EAC) is another rare occurrence. Clinically, SCC of EAC presents with symptoms similar to other benign otologic conditions. A case of Stage I SCC in EAC region is presented here in a 60-year-old male patient with incidental intranodal Warthin tumor along with the histological differential diagnosis. The patient is being followed up. There is no evidence of recurrence 1 year and 11 months after surgery.

Keywords: External auditory canal, squamous cell carcinoma, Stage I, Warthin tumor

INTRODUCTION

Literature infers carcinoma of the external auditory canal (EAC) as an uncommon entity.[1,2] Zhen *et al*. quoted that malignancy of EAC is so rare that an otolaryngologist may encounter only a few or even no EAC tumors in their career.[1] The annual incidence of squamous cell carcinoma (SCC) of EAC is about 1 case per million people.[3] For progression of the tumor, periauricular soft tissues, the parotid gland, the temporomandibular joint , and mastoid are the most common sites.[2] The previous radiotherapy has been considered to be the most important factor in association with tumors of EAC; although, chronic suppurative otitis media and exposure to chemicals have also been stated.[4] Usually, the patient presents with symptoms quite similar to other benign otologic conditions, resulting in inadvertent delayed diagnosis.[5] Late diagnosis is common which worsens the prognosis.[2]

As aforementioned, the tumor may progress to preauricular soft tissue and parotid gland, superficial parotidectomy is usually done to obtain anterior tumor-free margin in carcinomas of EAC. Salivary gland tumors have been reported to arise inside the lymph nodes ranging from benign tumors such as Warthin tumor (WT) and pleomorphic adenoma to malignant neoplasms such as acinic cell carcinoma and mucoepidermoid carcinoma.[6] WT is considered to be the most common salivary gland neoplasm to originate in ectopic salivary tissue.[6]

Here, we present an incidental finding of inchoate WT of size 3.5 mm × 3 mm in a patient treated for Stage I SCC of EAC who presented early with a complaint of swelling inside ear.

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CASE REPORT

A 60-year-old South Indian male patient presented to the Head and Neck Surgical Oncology OPD with a chief complaint of swelling and pain inside the left ear of 3 months duration. Medical history revealed that the patient was hypertensive and diabetic with a history of angioplasty. Patient also recalled recurrent ear infection in the past. He is a chronic smoker (40 years), with a habit of tobacco chewing (for the past 1 year) and was taking alcohol in the past, which he quit around 12 years ago. Past family history revealed that patient's mother died of tongue SCC. On examination, a polypoidal growth was noticed involving the posterior and inferior walls almost filling the canal, with no clinically enlarged intra- or peri-parotid gland, pre/postauricular or neck nodes.

He had already consulted a surgeon who had taken biopsy of the tumor located in the left EAC, for histopathological diagnosis. The hematoxylin and eosin slides and paraffin blocks of tissue were submitted for review at onco-pathology division of our institution. The slide was reviewed and reported as well-differentiated SCC.

Radiological examination displayed an irregular enhancing lesion in the left EAC without bony invasion. There was no middle ear extension. The right EAC was normal [Figure 1]. Since the tumor was confined to left EAC without bony erosion or evidence of soft-tissue extension, the patient was categorized in Pittsburg stage I. After discussion in the multi-speciality board, lateral temporal bone resection (LTBR) with superficial parotidectomy, and selective neck dissection was done with preservation of facial nerve.

Postauricular incision was placed extending anteriorly and approaching into the neck. Posterior-based flap was raised followed by cortical mastoidectomy. Antrum, facial nerve canal, and incus were identified. Using drill lateral temporal bone was delineated all around till ear ossicles, and the malleus and incus were removed. LTBR was completed using chisel and hammer. The facial flap was used to fill in the middle ear. Superficial parotidectomy was done with the preservation of branches of the facial nerve. Level II and III lymph nodes were dissected out, and the wound was primarily closed.

Radical specimen received in our department constituted of left LTBR with left superficial parotidectomy and left level II to III cervical lymph nodes. The specimen was cut open, which revealed an ulceroproliferative growth of size 1.4 cm × 1.3 cm × 0.4 cm situated 1.1 cm from superior resected margin, 0.7 cm from posterior resected margin, 0.8 cm inferior resected margin, and 0.6 cm from the anterior margin. Sections from EAC lesion showed an invasive epithelial neoplasm composed of islands of tumor cells showing increased nucleo-cytoplasmic ratio, moderate nuclear pleomorphism, vesicular nuclei with prominent nucleoli, and abundant keratin pearl formation. Stroma showed dense inflammatory reaction [Figure 2a]. The temporal bone was free of tumor microscopically. All resected soft tissue, and bony margins and parotid gland were free of tumor. No perineural or lymphovascular invasion was seen. Two intraparotid lymph nodes were identified, larger node measured 9 mm. Smaller node showed reactive changes, while the larger node showed double-layered oncocytic epithelium arranged in complex papillary architecture and thin core composed of lymphoid tissue [Figure 2b-d]. Adjacent nodal tissue was unremarkable. Eight neck nodes showed reactive changes. A final diagnosis of

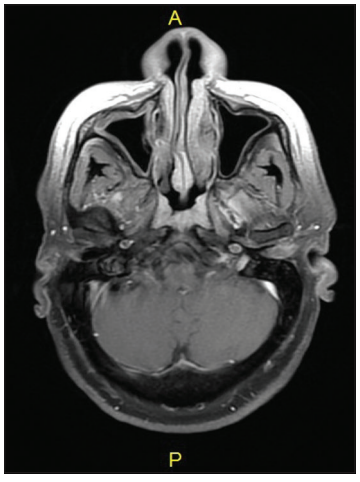

Figure 1: Contrast-enhanced T1-weighted image showing enhancing lesion of the left external auditory canal. No bony erosion was seen

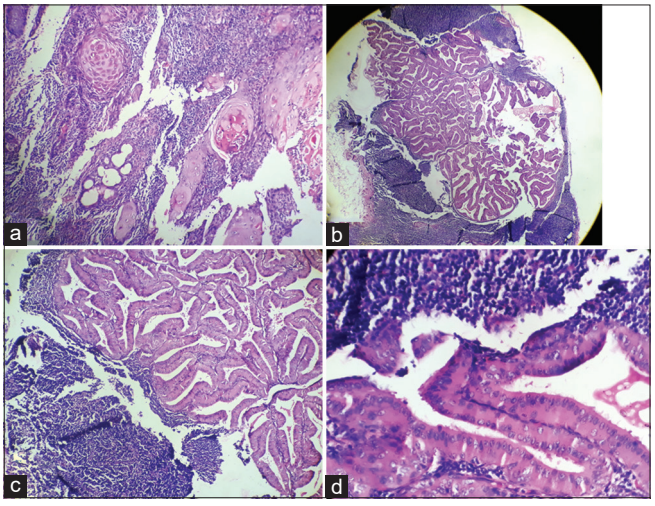

Figure 2: (a) Photomicrograph showing well-differentiated squamous cell carcinoma of external auditory canal; (b-d) Photomicrograph showing Warthin Tumor intra-parotid lymph node composed of double-layered oncocytic epithelium in papillary architecture with a thin core containing lymphocytic infiltrate. Normal appearing lymph node structure is also seen

pT1 N0 well-differentiated SCC of the left EAC (AJCC 8th edition) with incipient intraparotid lymphnodal WT was made. Figure 3a shows the gross specimen of LTBR specimen. The operated site healed uneventfully with no evidence of recurrence 1 year and 11 months after surgery [Figure 3b]. The patient is under close follow-up.

DISCUSSION

SCC of EAC and intranodal WT may not be related to each other, nor do they affect the clinical course. However, the presence of two uncommon entities is of pathological and academic interest. We hereby report a case of SCC of EAC in a 60-year-old male patient with 3.5 mm incipient WT in the intraparotid lymph node. Regarding carcinoma of the EAC, a rare disease, chronic inflammation, and radiation are the most common etiologies postulated in the literature.[4] The patient reported with swelling inside the left ear since 3 months, leading to an early diagnosis and prompt treatment. Moreover, the patient gave a history of recurrent otic infection, which could be considered as a possible etiological cause. Unlike the present case, these tumors show a female preponderance.[3] The patient's age was 60 years, which is in concordance with the usual age group of 55–65 years.[3] While SCC is reported as the most common temporal bone tumor among most of the studies,[7,8] Zhen reported adenoid cystic carcinoma as the most common malignancy followed by SCC.[1]

Relative to EAC SCCs, a few entities should be considered in the differential diagnosis. Differentiation of inflamed seborrheic keratoses (SK) from SCC may be challenging, especially when SK shows inflammatory cytological atypia once irritated. Limited tissue sampling may add to the diagnostic difficulty. The key differentiating feature is the "stromal invasion" seen in SCC. In doubt, the identification of lesion extent by radiological imaging might be helpful with a repeat biopsy, including deeper tissues.[9] Other entities to be given utmost importance are keratoacanthoma and reactive pseudoepitheliomatous hyperplasia. Ulceration, abundant mitoses with marked anaplasia favor SCC while epithelial lip (the marginal buttress of the epithelium) and sharp outline between tumor and stroma, if found, favors keratoacanthoma.[10] Pseudoepitheliomatous hyperplasia can result from chronic infections, chronic ulcers, granular cell tumor, halodermasis, and pyoderma gangrenosum. Gacek *et al*. advocated that immediate generous biopsy of the lesion should be performed when the clinical presentation is typical of malignancy such as >2 months history of pain and bloody otorrhea along with an ulcerated soft-tissue mass in the ear. Antibiotics with an observation period 2–3 weeks are sufficient to assess a smooth soft-tissue mass of <2 months duration without pain.[11] Other differentials include chronic otitis media, benign papilloma, aural or otic polyps, verruca vulgaris, and actinic keratoses.

Treatment for invasive SCC of the canal is temporal bone resection (partial, total, or piecemeal), depending on the extent of the tumor followed by covering the defect with skin or muscle flap. Postsurgical radiation theray is given in advanced stages. Five-year survival rate of 50% is reported in various series using this protocol.[12,13] Surgery alone has been advocated as the sole treatment for very early T1 stages like the present case.[5] The present case was managed by LTBR with left superficial parotidectomy and neck dissection.

Histological examination of the left superficial parotid gland showed an intranodal small WT. The radiological images were retrospectively analyzed by the radiologist. An oval hypertense lesion was noticed in the superficial lobe [Figure 4], probably corresponding to the node examined histopathologically. Rest of the parotid gland parenchyma was unremarkable. WT is

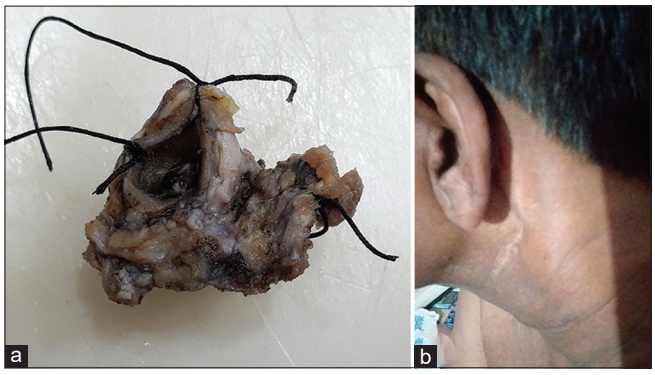

Figure 3: (a) Gross specimen of lateral temporal bone resection specimen; (b) postoperative picture of the patient 1 year and 11 months after surgery

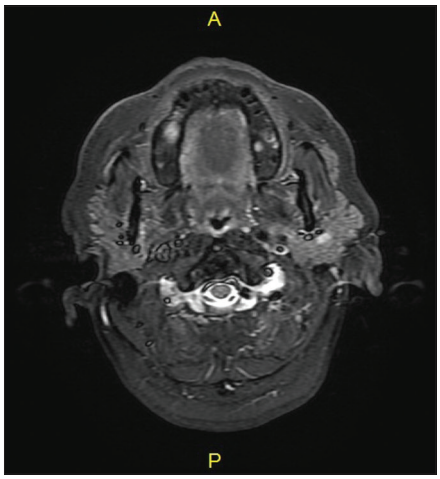

Figure 4: STIR image showing hyperintense oval lesion in the superficial lobe of the left parotid gland probably corresponding to intranodal Warthin tumor diagnosed histologically

regarded as a tumor-like growth. The inclusions of the salivary gland ducts and acini in the medullary regions of the parotid and occasionally extraparotid lymph nodes may give rise to a variety of both benign and malignant salivary gland tumors.[6] The predominant hypothesis being that these tumors arise from a proliferation of oncocytic epithelium and lymphoid tissue-derived from entrapped salivary ductal tissue within parotid lymph nodes.[13] EBV infection, usage of tobacco, and steroid hormone (progesterone) have been implicated in the development of WT. Secondary accumulation of lymphoid elements as an inflammatory lymphoid cell response to an epithelial neoplasm is another hypothesis in the pathogenesis of Warthin's tumor. However, Teymoortash *et al*. based on their immunohistochemical study demonstrated sinus endothelial/virgultar cells in Warthin tumor as a specific component of lymph nodes. They further opined that WT arises within intra-parotid lymph nodes, most probably from heterotopic salivary gland tissue and suggested that WT should not be regarded as adenoma.[14] WT, however, may be considered as a true neoplasm. The tumor has been reported to transform to epithelial and lymphoid malignancies in extreme rare cases.[15-17]

The patient is under regular follow-up and is doing well.

CONCLUSION

SCC of EAC if diagnosed at early stage results in better patient outcome. Most of the time, symptoms are nonspecific and thus, in case of proliferative growth in EAC, SCC although rare, should be kept in the differential diagnosis. The present case is a rare synchronous presentation of two uncommon tumors.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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