Auriculotemporal Carcinoma - A Retrospective Case Series

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Abstract

Rationale: Eleven cases with auriculotemporal cancer were reviewed for prognostic analysis. **Patient Concerns:** Follow-up ranged from 1.2 to 12 years (median 5.01 years). **Diagnosis, Treatment and Outcome:** Three patients with parotid gland carcinoma, out of those, two had chemoradiotherapy, died in the first 2 years of treatment. They were at stage T4 and tumour progressed with distant metastasis. Otorrhoea was the most common symptom in patients with primary temporal bone carcinoma. One patient with auricular carcinoma had a recurrence at the primary site 13 months after surgery. One patient with T1, two patients with T2 and one patient with T3 have completed a 5-year survival period. One patient with T1 and another one with T2 are still at a 2-year follow-up period with no recurrence. **Take-Away Lessons:** Complete resection is the treatment of choice. Post-operative radiotherapy is highly recommended. The most decisive prognostic indicator is the advanced stage. Early diagnosis has great importance.

Keywords: Chemotherapy, ear, myocutaneous flaps, radiotherapy, temporal bone carcinoma

Introduction

Temporal bone cancer represents <0.2% of all tumours. It is usually associated with chronic inflammation.[1] The most common location is the external auditory canal but rapidly proceeds to middle ear involvement. Otalgia, hearing loss, facial palsy and purulent discharge are the main signs. Persistent discharge which fails to improve with treatment, growing granulomatous tissue and bony erosion should raise a suspicion of malignant progress. It may also develop from a regional extension. Metastatic tumours are reported following haematogenous and lymphatic spread from distant primary sites.[2]

Treatment alternatives range from primary chemoradiotherapy to surgical resection. It has low survival rates and follows a fatal course with high recurrences. The overall 5-year survival rates vary with different reported series from 43.2% to 77% which decreases to 7% for advanced tumours.[1,3] A survival analysis of the case series is required to update tumour management.

Case Report

Eleven cases were included (2008-2021). Follow-up ranged from 1.2 to 12 years (median 5.01 years). There were one female patient and 10 male patients with ages ranging between 44 and 72 (mean; 59). Eight patients had surgery as an initial treatment. One patient had metastatic spread of primary stage-III stomach carcinoma to the internal auditory canal. He received radiochemotherapy and died in a year. Three patients were at stage T1, two patients were at T2, one patient was at T3 and four patients were at T4.[1] The tumour was originating from the auricle in three patients (one basal cell carcinoma), from the parotid in four patients (three adenocarcinomas and one adenoid cystic carcinoma) and from the ear canal in three patients (all squamous cell carcinoma). Otorrhoea was the most common complaint. Two patients with primary parotid cancer with temporal bone invasion had radiochemotherapy [Figure 1a-c]. They died in the first 2 years of treatment. They were at stage T4 and the tumour progressed with metastasis [Table 1]. Four patients with primary parotid cancer had facial paralysis. One patient had a history of removal of pleomorphic adenoma of the parotid gland and developed carcinoma several years later. He had parotidectomy and neck dissection which recurred in a year. He had temporal bone resection with abdominal free flap repair. However,

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he died due to several metastatic lesions [Figure 2a-d]. The facial nerve was resected in three cases and reconstructed in two of them. One patient with primary parotid gland cancer and House–Brackmann grading-IV facial paralysis had parotidectomy with facial nerve resection.

Two patients with auricular carcinoma had primary resection with auriculoplasty [Figure 3a-c]. One patient with auricular cancer and nodal involvement in the neck had resection of the temporal bone with auricular removal, neck dissection including sternocleidomastoid muscle and total parotidectomy. The defect was reconstructed with the pectoralis regional flap [Figure 4a-c]. He had a recurrence at the primary site 13 months after surgery. One patient with T1 carcinoma of the ear canal had sleeve resection and split-thickness grafting of the ear canal without bony drilling. Another patient with T1 primary carcinoma of the temporal bone had a canal wall down tympanoplasty. One patient with T4 temporal bone carcinoma had a resection of the temporal bone with parotidectomy and facial neurectomy [Figure 5a-e]. The facial nerve was reconstructed with hypoglossal-facial anastomosis. One patient with T1, two patients with T2 and one patient with T3 have completed a 5-year survival period. One patient with T1 and another one with T2 are in the 2-year follow-up period with no recurrence. Six patients (50.4%) had disease-free survival during the follow-up period.

Discussion

Temporal bone cancer has a poor prognosis despite extensive measures. Early identification is important since advanced cases are indicative of worse outcomes. The extent of temporal bone surgery is dependent on the invasion of the tumour and should be treated aggressively. The involvement of dura with intracranial extension is not the limit of operability but has an important impact on the survival rate. Advanced cases also have a high rate of distant metastasis. It is noteworthy to consider parotidectomy when the tumour is particularly located at the anterior part of the auditory canal.[4] On the other hand, occult metastasis at the very early stage of carcinoma of the auditory canal is possible.[5] The histopathological finding of undifferentiated epithelial cancer and nodal/extranodal spread is associated with poor outcomes.[3] Perineural invasion is not as significant as positive surgical margins which decrease survival.[1] Surgery of malignancy involving the petrous temporal bone rather than the external ear only clearly presents a challenge to provide safe resection margins. Facial nerve involvement at the time of presentation could be indicative of advancement and poor prognosis.[4,6] However, it can be resected and simultaneously reconstructed with a favourable outcome.

The primary site was the auricle in four cases. They are usually diagnosed early and present a better prognosis. However, the

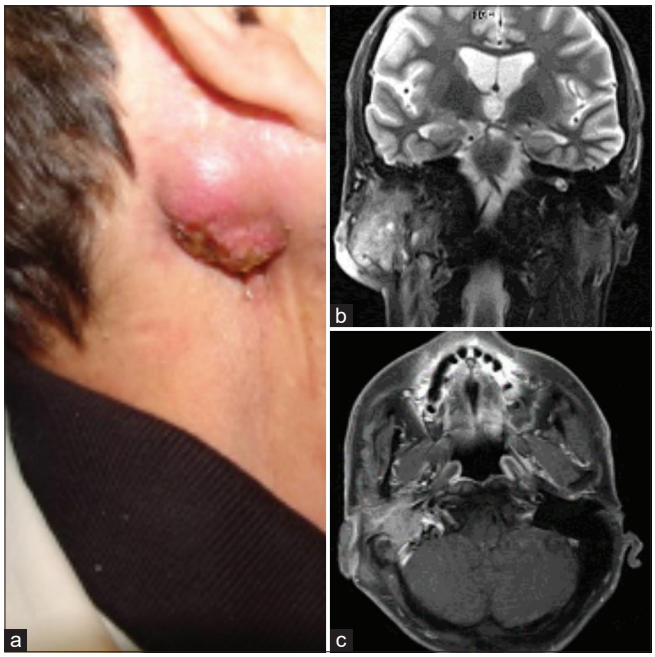

Figure 1: (a) Right parotid carcinoma with temporal bone infiltration. T2-weighted MRI on (b) coronal and (c) axial view shows the extensive location of the tumour. Tumour infiltrates the jugular foramen medially. The posterior fossa dura seems to be invaded by tumour growth

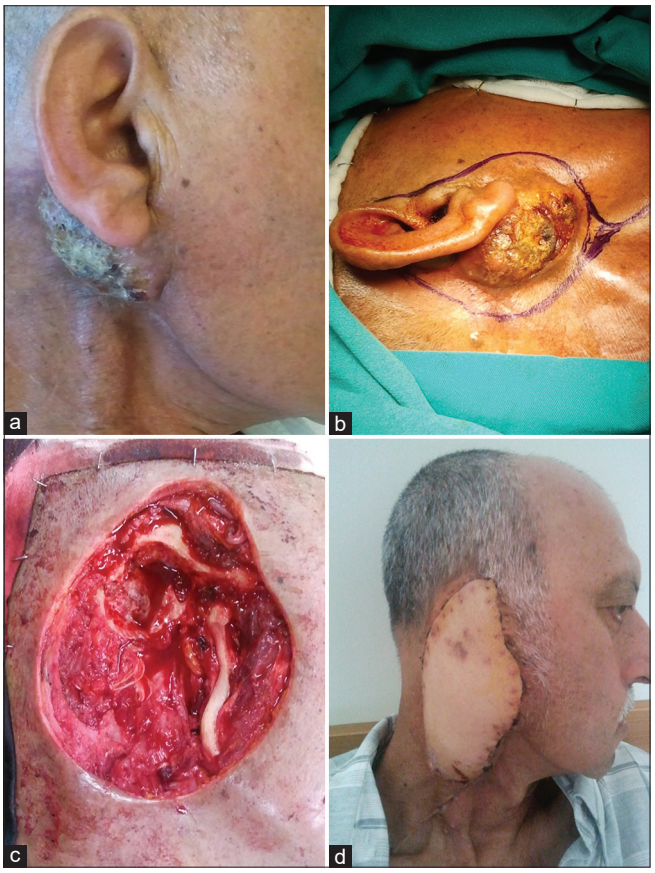

Figure 2: (a) Recurrent parotid carcinoma in a patient who had parotidectomy and neck dissection before. (b and c) Mandibular periosteum, lateral aspect of fibrocartilage articulation capsule of the temporomandibular joint, remnant of the parotid gland and facial nerve were included in the lateral temporal bone surgery with total auricular resection. (d) An abdominal free flap was prepared to reconstruct the defect

Table 1: The profile of the patients with auriculotemporal carcinoma

Case	Age	Sex	Primary location	Histopathology	Chief complaints	Tumour size (cm)	TNM	Medical story	Treatment	Follow-up and outcome
1	57	Male	Auricle	SCC	Tumour	4	T2NoMo	-	Class-II defect Primary resection + RT	11 years
2	67	Male	Auricle	SCC	Tumour	5	T3N2aMo	-	Class-III defect Temporal bone resection (lateral Type-II) + parotidectomy + neck dissection + pectoralis major flap + RT-CT	HBG-I 13 months The patient had local recurrence
3	72	Female	Auricle	BCC	Tumour	1	T1N0M0	-	Class-II defect Primary resection	2 years
4	44	Male	Stomach	AC	Hearing loss, vertigo	0.5	Metastatic	Surgery for stomach CA + RT-CT	CT	6 months Died
5	54	Male	Parotid	ACC	FP HBG-III	5	T4N1M1	-	RT + CT	Lung metastasis 2 years Died
6	64	Male	Parotid	AC	FP HBG-III	3	T4N1M1	-	RT + CT	Multiple metastases 6 months Died
7	62	Male	Parotid	AC	Otalgia FP HBG-IV	2	T2N0M0	-	No auricular defect Parotidectomy + facial nerve resection + partial mandibular resection + mastoid apex resection (lateral Type-IV) + cable graft reconstruction of facial nerve + RT-CT	HBG-VI 2 years
8	67	Male	Parotid	SCC ex pleomorphic adenoma	Otalgia FP HBG-III	6	T4NxM0	Parotidectomy + neck dissection + RT	Class-III defect Temporal bone resection (lateral Type-III)+ partial mandibular resection + rectus abdominis free flap + CT	HBG-VI Lung metastases 7 months Died
9	54	Male	EAC	SCC	Otorrhoea Hearing loss	1.5	T1N0M0	-	Class-I defect Sleeve resection of EAC (lateral Type-I)	14 years
10	52	Male	EAC	SCC	Otorrhoea Otalgia Hearing loss	2	T2N0M0	-	Class-I defect Canal wall down Mastoidectomy (lateral Type-I) + RT	HBG-I 12 years
11	56	Male	Middle ear	SCC	Otalgia Hearing loss	3	T3N1M0	Tympanoplasty	Class-III defect Temporal bone resection (lateral Type-III) + parotidectomy + 7-12 anastomosis + pectoralis major flap + RT	HBG-II 9 years

CA: Carcinoma, SCC: Squamous cell CA, BCC: Basal cell CA, AC: Adenocarcinoma, ACC: Adenoid cystic CA, FP: Facial paralysis, HBG: House-Brackmann grading, RT: Radiotherapy, CT: Chemotherapy, EAC: External auditory canal, TNM: Tumour-node-metastasis

auricle has a good blood supply. The overlying skin is thin and the tumour may rapidly invade the cartilage and the bone which makes precise resection and reconstruction difficult to achieve. Delayed treatment endangers the survival rate which falls considerably when auricular cancer invades the temporal bone.[1] Three patients had squamous cell carcinoma and one elder female had basal cell carcinoma of the auricle. Basal cell cancer is a slow-growing, invasive tumour and it rarely metastasises but it may exhibit deep extension. Wide resection is required. Otherwise, the risk of recurrence is high.[1] Reconstruction of large defects when total auricular resection is needed should be planned before surgery. Multistaged procedures can be complicated due to infection and tissue contraction. Inflammatory tissue response and infection are major concerns for synthetic or alloplastic implants. It is practical to classify temporal bone cancers according to the site of origin since the histopathologic pattern may change. Epithelial and basal cell carcinoma are frequently seen in the auricle and external ear. Primary adenocarcinoma and mucoepidermoid carcinoma frequently involve the external and middle ear.[7] Adenocarcinoma of the endolymphatic sac is defined in a distinct category since it is oftentimes associated

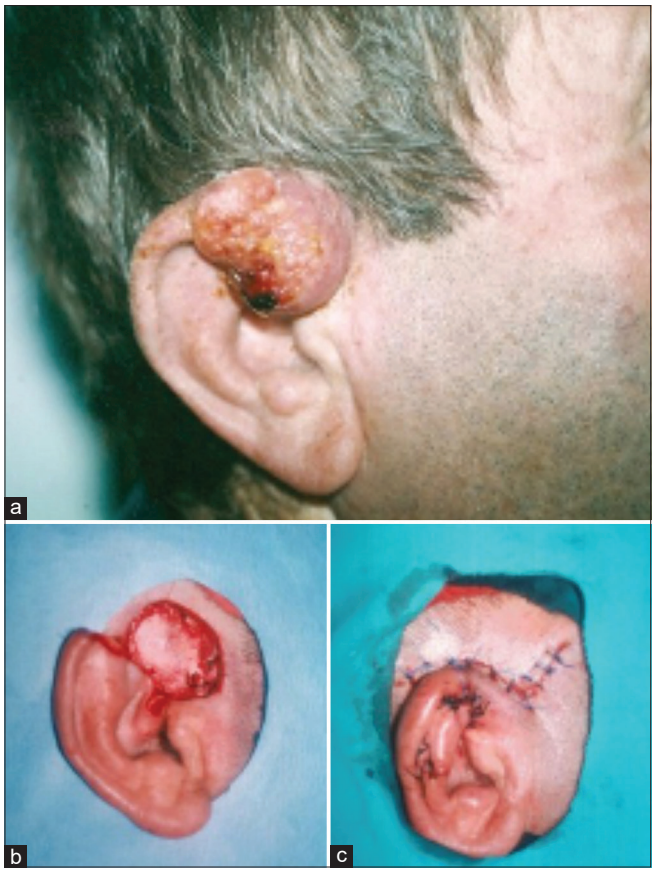

Figure 3: (a) Auricular carcinoma, (b) Primary resection, (c) Reconstruction of the tumour

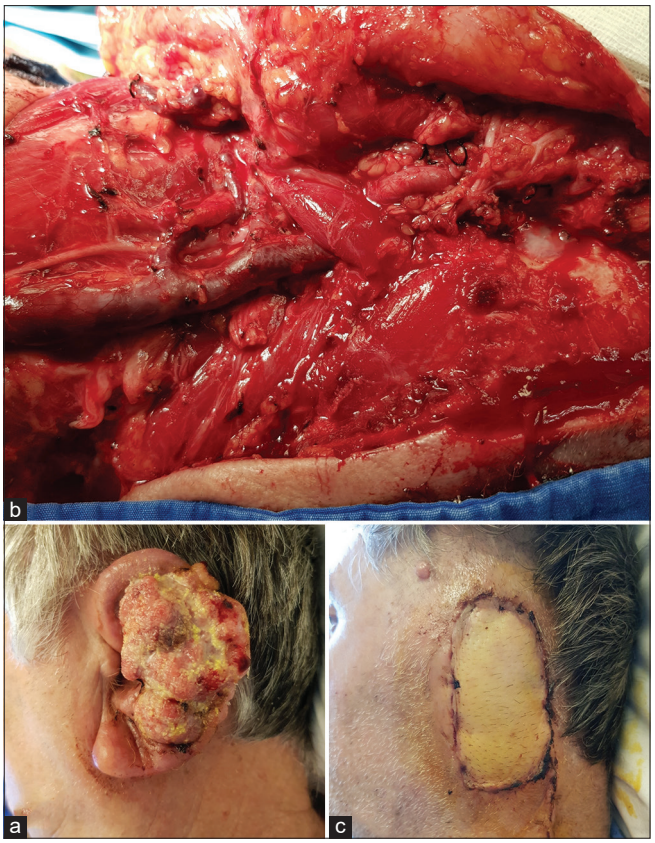

Figure 4: (a) Auricular carcinoma with ipsilateral nodal involvement. (b) The patient had total parotidectomy with temporal bone resection and radical neck dissection. (c) The defect was reconstructed with pectoralis regional flap

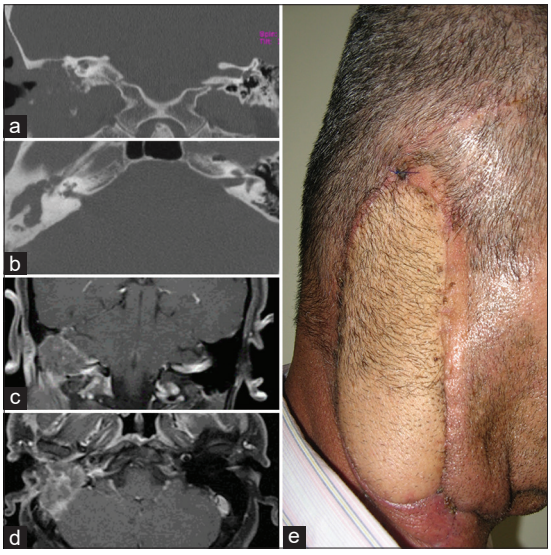

Figure 5: The patient with primary carcinoma of the right temporal bone. He had tympanoplasty before due to chronic otitis media. Temporal bone tomography on coronal (a) and axial (b) demonstrates irregular destruction of the mastoid bone. MRI on coronal (c) and axial (d) view confirm the mass occupying lesion in the right mastoid. The patient had lateral temporal bone resection with the removal of the parotid gland and the facial nerve. The nerve was reanimated with hypoglossal-facial anastomosis. Pectoralis regional flap was used to cover the defect (e)

with Von Hippel–Lindau disease, a genetic disorder. The bilateral presentation can occur but the metastatic progression is less likely.[8] Sarcomas involving temporal bone are more frequent in adolescent/paediatric ages.[9]

Complementary radiotherapy following surgery has shown increased survival for advanced cases.[1,3] Studies indicate that radiotherapy following surgery even for early-stage tumours has proven to be effective for survival.[10] Chemotherapy has a promising role in advanced cases. Comparative studies indicate that the overall survival rate for definitive chemoradiotherapy is as good as surgery followed by radiotherapy (52.1% vs. 55.6%). When high morbidity is inevitable due to surgery or the unresectability of advanced cancer, definitive chemoradiotherapy might be appropriate. A treatment plan based on starting chemoradiation for all advanced cases and proceeding with salvage surgery when indicated is worth consideration. However, bony erosion is correlated with a worse prognosis compared to widespread soft-tissue involvement with the tumour. Survival rates are negatively influenced by the extent of bony lesions and radiotherapy does not improve the prognosis.

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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