

External auditory canal carcinomas: What does optimal management require?

Les carcinomes du conduit auditif externe : Qu'impose une prise en charge optimale ?

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ABSTRACT

Objective: Carcinomas of the external auditory canal (CEAC) are rare with poorly defined treatment guidelines. The aim of this study is to discuss the therapeutic management of patients with CEAC.

Methods: It was a retrospective study conducted over an 11-year period (2010-2021), including 8 patients with primary CEAC. Cases were recorded in terms of gender, age, symptoms, physical signs, radiological examinations, surgical methods and follow-up.

Results: Median age was 58 years with a sex ratio of 0.6. Main symptoms were hearing loss (n=6), otorrhea (n=4), otalgia (n=4), and peripheral facial palsy (n=2). According to the Pittsburgh classification, CEACs were classified as T1N0M0 in 3 cases, T4aN0M0 in 4 cases, and T4bN0M0 in one case. Surgery consisted of a local resection of the tumor in four cases. A lateral temporal bone resection with total ipsilateral parotidectomy and functional neck dissection was performed in three cases with, respectively, a resection of the ipsilateral pinna (2 cases) and of the temporomandibular joint (2 cases). Adjuvant radiotherapy was indicated in 4 cases. Exclusive radiation therapy was indicated in one case. Histopathological examination confirmed 5 cases of squamous cell carcinoma, 2 cases of basal cell carcinoma, and one case of adenoid cystic carcinoma. Surgical margins were negative in 5 cases. Follow-up was favorable without recurrence in six cases. In one case, a progressive disease with patient death was observed.

Conclusions: Optimal treatment requires early diagnosis and careful imaging assessment of tumor extension to tailor treatment, which often includes surgery and radiotherapy.

Key words: External auditory canal, Carcinoma, Surgery, Radiotherapy, Pronostic

RÉSUMÉ

Objectif : Les carcinomes du canal auditif externe (CCAEE) sont des néoplasies rares dont le traitement n'est pas consensuel. Notre objectif était de discuter les modalités de prise en charge de CCAEs.

Méthodes : Etude rétrospective menée sur une période de 11 ans (2010-2021), incluant 8 patients avec CCAEE primitif. Les facteurs étudiés étaient le sexe, âge, symptômes, les signes physiques, radiologiques les approches chirurgicales et les données du suivi.

Résultats : L'âge médian était de 58 ans avec un sex-ratio de 0,6. Les principaux symptômes étaient la surdité (n=6), l'otorrhée (n=4), l'otalgie (n=4) et la paralysie faciale (n=2). Selon la classification de Pittsburgh, les CCAEE ont été classés : T1N0M0 dans 3 cas, T4aN0M0 dans 4 cas et T4bN0M0 dans un cas. La chirurgie consistait en une résection locale de la tumeur dans quatre cas. Une pérectomie latérale avec parotidectomie totale et un curage fonctionnel sélectif dans trois cas, emportant le pavillon (2 cas) et l'articulation temporo-mandibulaire (2 cas). La radiothérapie était associée dans 4 cas. Elle était exclusive dans un cas. L'examen histopathologique a conclu à un carcinome épidermoïde (5 cas), un basocellulaire (2 cas) et un cas de carcinome kystique adénoïde. Les marges chirurgicales étaient négatives dans 5 cas. Le suivi était favorable sans récurrence dans 6 cas.

Conclusions : Un traitement optimal nécessite un diagnostic précoce et une évaluation minutieuse par imagerie de l'extension tumorale pour adapter le traitement, qui associe souvent la chirurgie et la radiothérapie.

Mots clés : Conduit auditif externe, Carcinome ; Chirurgie ; Radiothérapie ; Pronostic

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INTRODUCTION

Carcinomas of the external auditory canal (CEAC) are extremely rare lesions (1), with an annual incidence of approximately one to six cases per million people, which represents less than 0.2% of all cancers in the head and neck area(2). Squamous cell carcinoma (SCC) is the most common neoplasm at this site(3). Diagnosis is always challenging, and differential diagnosis with numerous other pathologies, mainly malignant otitis externa, is mandatory for precocious management. Surgery with or without adjuvant radiation therapy is advocated by most otorhinolaryngologists as the standard treatment for this disease(2). However, deciding the extent of resection is problematic, because minimal interventions threaten oncologic safety and aggressive surgical intervention is associated with a higher rate of complications(4). This study, compared to the data in the literature, aims to analyze the therapeutic approaches of primary external auditory carcinomas.

METHODS

It was a case series study conducted over an 11-year period from 2010 to 2021 that involved 8 patients diagnosed with histopathologically confirmed primary CEAC. All patients underwent cervical computed tomography (CT) and temporal bone magnetic resonance imaging (MRI). The medical records and radiological results were examined to ascertain the stage of cancer according to the Pittsburgh classification. The minimum follow-up period was 12 months.

RESULTS

Our study included 8 patients. The mean age at diagnosis was 58 years ranging from 49 to 65 years with a gender distribution of 5 women and 3 men. Regarding their personal history, we observed one case of EAC squamous cell carcinoma and one case of EAC basal cell carcinoma already treated with exclusive radiotherapy. The main presenting symptoms were hearing loss (n=6), otorrhea (n=4), otalgia (n=4) and peripheral facial palsy (n=2). The duration of symptoms ranged from 15 days to 3 years. On otological examination, we observed a budding process of the EAC (n=5), a blackish nodule on the anterior wall of the EAC (n=2), and infiltration in the concha (n=2). Two cases had facial palsy, which was grade IV and V, each according to the House–Brackmann scale (HB). Homolateral parotid swelling to the lesion was observed in 5 cases and no cervical lymphadenopathy. The preoperative biopsy of the tumor confirmed the malignancy in all cases. Preoperative audiometry concluded to homolateral conductive hearing loss in 6 cases and a mixed one in the two cases with previous history of radiotherapy. The median hearing threshold in the pathologic ear was 55 dB [35-70]; it was normal in the contralateral side in all cases. The most common histological type was squamous cell carcinoma (SCC) in 5 cases followed by basal cell carcinoma (BCC) in 2 cases

and cribriform variant of adenoid cystic carcinoma (ACC) in one case.

The temporal bone CT scan detected an irregular thickening of the EAC skin causing stenosis in all cases, tympanic bone erosion (n=3), a temporomandibular joint involvement (n=3) and extension into the middle ear (n=1) (Figure1).



Figure 1. CT scan of the temporal bones in axial section showing a total soft tissue filling of the right external auditory canal and the middle ear with lysis of tympanic bone (Patient 5 in table 1).

An MRI of the temporal bone was performed for all patients and revealed a homolateral parotid lymph node with benign cytopuncture in one case and a homolateral parotid involvement in five cases (Figure 2). No regional lymph node metastases or distant metastases were found.



Figure 2. Heterogeneous enhanced Contrast-enhanced T1-weighted brain MRI showing a heterogeneous enhanced soft tissue tumor of the right external auditory canal extending to the middle ear cavity and homolateral temporomandibular joint (Patient 5 in table 1).

According to the Pittsburgh classification, three patients were in an early stage (T1N0M0) and five patients were in advanced stages (T4aN0M0 and T4bN0M0).

All patients except one underwent surgery (figure 3). There were no metastatic lymph nodes in all cases who undergone neck dissection. No parotid extension was found in patients who had parotidectomy. Adjuvant radiotherapy was administered in 4 cases. Exclusive

radiotherapy was indicated for the patient who declined surgical treatment. The follow up period ranged from 1 year to 9 years. No recurrence was noted in six cases (figures 4,5), progressive disease with patient death in one case (Table 1).

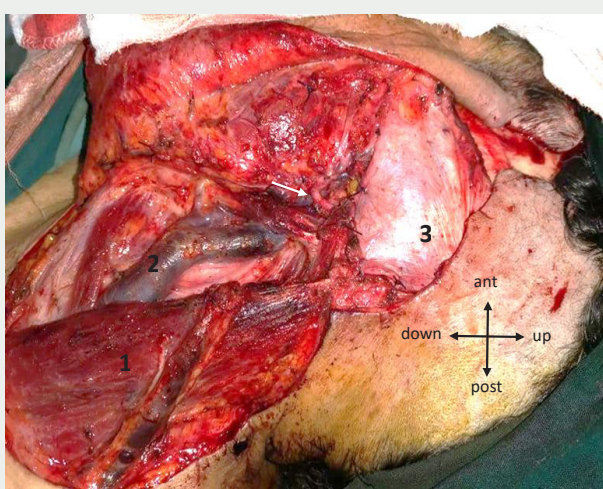


Figure 3. Lateral temporal bone resection with resection of the ipsilateral pinna, total ipsilateral parotidectomy and ipsilateral functional neck dissection. A pediculate temporo-facial muscle with abdominal fat were used to fulfill the petrous cavity (patient 2 in table 1). (1) sternocleidomastoid muscle, (2) internal jugular vein (3): pediculate temporo-facial muscle, (white arrow) left facial nerve.



Figure 4. Postoperative otoendoscopy in a patient operated with sleeve surgery (patient 1 in table I) showing a well epithelialized and permeable right external auditory canal (1: Tympanic membrane, 2: Malleus, 3: Posterior external auditory canal wall)

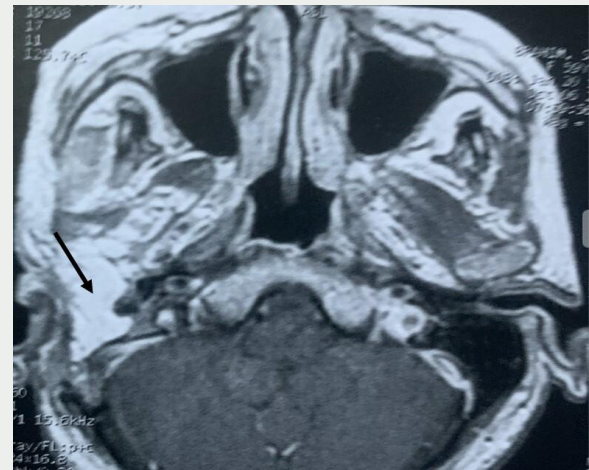


Figure 5. one-year postoperative brain MRI (T1 post gadolinium injection) in a patient operated with a lateral temporal bone resection total parotidectomy and temporomandibular joint resection (patient 5 in table I) : post-operative cavity filled with fat without tumour recurrence.

DISCUSSION

The incidence of CEAC is estimated to be 1-6 people per 1 million population, with a mean age of 61.5 years(5). In our study, mean age at the time of diagnosis was 58 years ranging from 49 to 65. It was similar to data reported in other series of patients with CEAC. Diagnosis is not always easy since the misdiagnosed rate is up to 69%(2). It is frequently misdiagnosed due to its clinical features similar to benign conditions, such as granulation tissue in the external auditory canal (EAC), EAC cholesteatoma, and necrotizing otitis externa(5,6). Many non-specific symptoms were reported, including otorrhea, otalgia, and hearing loss. These symptoms were prevalent in our study. Tinnitus, vertigo, trigeminal nerve involvement, and trismus have been also described in alternative sources from the literature (7–9). Intractable inflammation, a change in discharge with otorrhagia, and severe pain that not respond to treatment must put the chip in the ear and evoke the diagnosis of cancer(9). In addition, a thorough head and neck examination should be performed. Testa et al. identified a regional metastasis rate of 22.7% and a distant metastasis one of 2.3% in 44 cases(10). In our study, no cervical patent lymph nodes nor distant metastases were found. In fact, CEAC are usually associated with local aggressive invasion rather than metastasis.

Hence, face to a budding tissue process of the EAC, if medical interventions prove ineffective or if there is a doubt about the nature of the lesion, a biopsy is warranted. As in our study, the most common histological type of EAC malignancy is SCC, followed by ACC, BCC. Other histopathologic variety were less described such as malignant melanoma, Merkel cell carcinoma, angiosarcoma, adnexal carcinoma, ceruminous adenocarcinoma and lymphoma(9,11).

A preoperative audiogram should be obtained in all patients to evaluate the serviceable hearing in the affected ear and in the opposite one(12). In our study,

Table 1. Table resuming staging, therapeutic approaches, histopathologic data and follow up of patients with external auditory canal carcinoma

Patient	TNM classification	Surgical procedure	Radiotherapy	Histology Margins	Follow up (FU)
Patient 1 (61 years)	T1N0M0	Sleeve surgery with reconstruction of the EAC skin wall using a skin flap from the mastoid region	Adjuvant	SCC Negative	No recurrence on MRI and physical examination (figure 4) FU: 2 years
Patient 2 (65 years)	T4aN0M0	lateral temporal bone resection + total ipsilateral parotidectomy + resection of ipsilateral pinna + ipsilateral functional neck Dissection (Figure 3)	No	SCC Negative	No recurrence on MRI and physical examination; FU: 2 years
Patient 3 (64 years)	T4aN0M0	The patient declined surgical treatment	Exclusive	ACC -	-peripheral facial palsy
Patient 4 (60 years)	T4aN0M0	Lateral temporal bone resection + total ipsilateral parotidectomy + resection of the pinna+ temporomandibular joint + functional neck dissection	Adjuvant	SCC Negative	-peripheral facial palsy No recurrence on MRI and physical examination; FU: 1 year
Patient 5 (59 years)	T4aN0M0 (figure1, 2)	a lateral temporal bone resection + total ipsilateral parotidectomy +resection of the invaded temporomandibular joint + ipsilateral functional neck dissection	Adjuvant	SCC negative	Peripheral facial palsy No recurrence on MRI and physical examination; (figure 5) FU: 2 years
Patient 6 (49 years)	T1N0M0	Sleeve surgery +reconstruction with a pedicled local skin flap	No	BCC negative	No recurrence on MRI and physical examination; FU: 1 year
Patient 7 (57 years)	T4bN0M0	Local surgery + reconstruction with a retro-auricular skin flap with inferior pedicle	no	BCC negative	Progressive disease and patient Death
Patient 8 (51 years)	T1N0M0	Sleeve surgery + reconstruction with a local skin flap rotation	Adjuvant	SCC positive	No recurrence on MRI and physical examination. FU: 3 years

Audiometry concluded to a conductive hearing loss in six affected ears and a mixed one in two cases with normal hearing in the contralateral ones. A temporal bone CT scan is essential to locate and measure bony erosion and evaluate soft tissue tumor extension. Lysis of the bony external auditory canal is crucial to decide the extension of the surgical approach(5). Preoperative MRI is particularly important in the case of advanced CEAC. It offers a better analysis of soft tissue and neurovascular invasion and intracranial spread(5). In clinical practice, the combination of CT and MRI appears as the ultimate preoperative imaging selection, as mentioned by Lee et al(4). Positron emission tomography(PET) scans can also be considered to detect invasions of nearby anatomic structures, such as the parotid gland, as well as metastases to distant organs(5). Tumors that arise from temporal bone, often, invade the parotid gland through the Santorini fissure or the foramen of Huschke(13). If such an extension to the parotid parenchyma is common, CEAC involves intra parotid nodes, only in advanced stages(14). In our study, no cases of parotid gland specimen involvement were detected in patients without preoperative suspicion of invasion. This implies that clinical examination and imaging of the gland are reliable to assess intra glandular extension.

Considering the inaccessibility of the temporal bone to direct examination, a surgically oriented TNM staging system based on radiographic findings was suggested. To date, the most widely used system is the University of Pittsburgh staging system(15) on which our study was based.

The existing issue with EAC carcinomas lies in the absence of a unanimous agreement on treatment guidelines.

Surgery and adjuvant radiotherapy are the treatment of choice(16). In our study two patients had been already treated with exclusive radiotherapy few years before surgery, of which one had a BCC which is known to be radioresistant. Such inadequate therapeutic approach had worsened the outcome with flap necrosis and recurrence despite free margins. We are convinced that therapeutic decision should take in consideration not only the tumor extension but also the histopathologic type. early diagnosis and aggressive primary surgical treatment with postoperative radiotherapy offer the greatest chance of cure. Surgery techniques varied from limited local sleeve excision to total temporal bone resection(1). The first one is indicated for patients with early-stage tumors involving the osseous EAC without extension medial to the tympanic membrane. Subtotal petrosectomy is indicated for advanced stage tumors that extend beyond the EAC such as the middle ear or the skull base. In our study sleeve surgery was performed in three cases of early-stage carcinoma. Surgeons proceed to reconstruction using local skin flaps after tumor removal and lateral temporal bone resection. The appropriate size of the skin flap and blood supply to the graft bed are important to achieve a successful procedure(17). In our study, a flap necrosis was noted in a patient with previous radiotherapy.

Prophylactic neck dissection may be advisable even in cases without patent lymph-nodes because the rate of micro-metastasis to lymphnodes in clinically negative necks is up to 17% in CEAC(18). In our study, all patients with advanced stage underwent lymph node neck dissection, no lymph node extension was identified in all cases. There should be no hesitation in performing

a therapeutic parotidectomy when parotid involvement is apparent, but performing an elective parotidectomy without clinical or radiological evidence of parotid involvement remains controversial(4). Due to the potential for facial nerve injury, it is crucial to refrain from performing unnecessary parotidectomy. In our series, no parotid extension was identified even in advanced cases. Radiotherapy is most commonly indicated in the adjuvant postoperative setting (19). For early-stage (pT1, T2) radically resected EAC squamous cell carcinoma, radiotherapy should only be considered in selected cases with perineural, infiltrative growth or angioinvasive growth, and with a close margin(20).

Regular postoperative follow-up is essential. Physical examination is recommended every 6 months followed by a CT scan every year to detect early recurrence of tumors. If there is suspicion of tumor recurrence on CT scan, MRI is mandatory for a better analysis of lesion extension(21). The poor prognosis of EAC tumors is correlated with extensive tumor invasion, lymph node metastasis, positive surgical margins, and cerebral involvement(21). Our study suggests a favorable survival rate of 100% in earlier stages. However, it decreased in advanced stages. This notable decrease was similarly affirmed by other studies(8,9,22). The overall 5-year survival rate varied in different reports from 51.7 to 61%(8,10,22). The survival rate for SCC is poorer than that for ACC and BCC(5). According to Mazzoni and al, recurrence rate in advanced SCC cases was 41.8% (23). In some series, the estimated 5-year survival rates range from 10 to 15% for advanced cases, and 80–85% for early disease(24).

CONCLUSION

External auditory canal carcinomas are a potentially lethal condition, spreading aggressively into surrounding temporal bone which management is not yet well codified. Early diagnosis is key to ensure the best therapeutic outcomes. Surgery remains the gold standard in the treatment of operable EAC carcinomas. If extensive resection of the temporal bone is usually recommended, extending surgery to the parotid gland, temporal bone, and neck lymph nodes may take in account imaging aspects. Postoperative radiotherapy complements surgery in cases of advanced radiosensitive cancers, positive surgical margins, extracapsular invasion, or lymphovascular/perineural invasion.

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