ORIGINAL ARTICLE

Management of Carcinoma of the Temporal Bone

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OBJECTIVES: A retrospective study was performed to evaluate the management of carcinoma of the external auditory canal and the middle ear.

PATIENTS AND METHODS: Thirty patients (14 women, 16 men; mean age 65 years; range 41 to 79 years) underwent treatment for carcinoma of the temporal bone between 1981 and 2002. Histopathological diagnoses were squamous cell carcinoma in 27 cases, adenoid cystic carcinoma in two cases, and melanoma in one case. Seventeen patients received primary treatment while 13 patients presented with residual disease after primary treatment. The patients were classified according to the classification system proposed by the Belgium Consensus Conference in March 2002. The mean follow-up period was five years (range 2 to 276 months).

RESULTS: The mean interval between the appearance of symptoms and first consultation was 22 months (range 1 to 168 months). The first symptoms were otalgia in 16 cases, otorrhea in 16 cases, bleeding from the ear in six cases, hearing impairment in 11 cases, facial paralysis in 11 cases, and neuralgia in two cases. Twelve patients had T_1 and T_2, six patients had T_3, and 12 patients had T_4 tumors. The Kaplan-Meier survival curves showed two-year survival as 82%, 67%, and 32%, and five-year survival as 82%, 67%, and 17% for T_1 or T_2, T_3, and T_4, respectively. At the end of a follow-up period of nine years, the survival rates were 66%, 66%, and 17% for T_1 or T_2, T_3, and T_4, respectively. Overall, complete remission was found in 64.7% and 23.1%, and mortality rates were 35.3% and 76.9% for primary treatment and salvage surgery, respectively.

CONCLUSION: Long-term prognosis of carcinoma of the external auditory canal mainly depends on the stage and primary treatment. Surgery (lateral or subtotal temporal bone resection, in combination with neck dissection and parotidectomy) and adjuvant radiotherapy is the treatment of choice for part of stage T_1 and all T_2 and T_3 tumors. In T_1 tumors, histopathologically confirmed free excision margins may obviate radiotherapy. Additional subclassification based on extension of T_4 tumors may be effective in advanced tumors. Surgery may be considered in T_{4a} tumors, and palliative treatment in most T_{4b} tumors and in all patients with T_{4c} disease.

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Cancer of the external auditory canal and the middle ear is a rare disease with a low incidence of about 1 to 5 per 1,000,000 inhabitants.[1] Chronic inflammatory skin diseases are identified as risk factors. The correct diagnosis is often delayed because the disease may be masked by frequently occurring ear symptoms like chronic otorrhoea. The above-mentioned pathophysiological mechanism has been supported in the literature.[2,3] No uniform internationally accepted classification system exists due to the complicated preoperative assessment of tumor extension. Because of the low incidence and the heterogeneity in staging among various studies, it is difficult to perform a reliable overall statistical analysis. No consensus exists regarding its therapy.

PATIENTS AND METHODS

A retrospective study was performed at the University Hospital Grenoble to evaluate the management of carcinoma of the external auditory canal and the middle ear. Our review included all patients who underwent surgery for a malignancy of the external auditory canal and/or the middle ear between 1981 and 2002. Thirty patients (14 women, 16 men; mean age 65 years; range 41 to 79 years) were found to meet the inclusion criteria. Those with a malignancy involving the external auditory canal, but with the primary tumor being localized at another site (parotid gland, infratemporal fossa) were excluded.

In all the patients, biopsies were obtained, most often under general anesthesia and in some cases under local anesthesia. Histopathological diagnoses were squamous cell carcinoma in 24 cases, of which one was Morbus Bowen, and one was basaloid squamous cell carcinoma; basal cell carcinoma in three cases, adenoid cystic carcinoma in two cases, and melanoma in one case.

Seventeen patients received only primary treatment while 13 patients showed residual disease after primary treatment. The mean follow-up period was five years (range 2 to 276 months).

The patients were classified according to the classification system proposed by the Belgium Consensus Conference in March 2002 (Table 1).[4] This classification system was first proposed by d'Arriaga,[5] it was then modified by the Pittsburgh group in 2000 and 2002.[6,7] The modification made by the Pittsburgh group in January 2002 was related to the facial nerve function, which proposed that disease associated with facial paralysis be classified as a T_4 tumor. The preoperative staging was based on a firm clinical examination and imaging studies. Unlike the Pittsburgh group, we did not use perioperative findings to change the stage.[6]

Treatment modalities consisted of surgery, radiotherapy, and chemotherapy as described below.

Lateral temporal bone resection

This procedure was used in disease limited to the external auditory canal, showing some bone erosion, without the involvement of the middle ear (Fig. 1). An *en bloc* excision of the bony external auditory canal was performed (Fig. 2). The procedure involves a mastoidectomy, an atticoantrotomy, a posterior tympanotomy (preserving the facial nerve), and a division of the incudostapedial joint. This procedure results in the removal of the external auditory canal, the tympanic membrane, the malleus and incus, preserving the epitympanum, hypotympanum, stapes and otic capsule as well as the facial nerve. A superficial parotidectomy for stage T_1 disease and a total parotidectomy for stage T_2 disease is performed, preserving the facial nerve, both in combination with a selective ipsilateral neck dissection of region II and III. Depending on the extent of the disease, and on inva-

Table 1. The staging system according to Pittsburgh group, modified in 2000 and 2002

Stage	Clinical and radiological findings
T_1	Tumor limited to the external auditory canal (EAC) without bony erosion or evidence of soft-tissue involvement.
T_2	Tumor limited to the EAC with bone erosion (not full thickness) or radiographic finding consistent with limited (<0.5 cm) soft tissue involvement.
T_3	Tumor eroding the bony EAC (full thickness) with limited (<0.5 cm) soft tissue involvement, or tumor involving the middle ear and /or the mastoid.
T_4	Tumor eroding the cochlea, petrous apex, medial wall of the middle ear, carotid canal, jugular foramen or dura, or with extensive (>0.5 cm) soft tissue involvement, or facial paralysis.
N	Lymph node involvement is a poor prognostic sign and places the patient in an advanced stage: T_1N_1 to stage III; $T_{2,3,4}$ N_1 to stage IV
M	M_1 disease is T_4 and is considered a very poor prognostic sign.

sion to the tympanic bone and/or superior root of the zygoma, resection of the mandibular condyle and the root of the zygoma may be performed. The dissected area is filled with abdominal fat, or a pedicled musculocutaneous flap is used in cases where the concha cavum or pinna is affected and a partial or total pinna removal is required.

Subtotal temporal bone resection

This approach, described by Lewis and Parson in 1954, was used for tumors of the external auditory canal that extend into the middle ear and/or the mastoid (Fig. 3). A retroauricular incision extending vertically into the neck is combined with a circular conchal incision; if needed, the latter can be adjusted in such a way that the resection may comprise the tragus. The resection results in a lateral temporal bone resection combined with removal of a part of the petrous bone (inner ear, facial nerve canal, sigmoid sinus, and dura); the petrous apex, internal carotid artery, and the internal auditory canal are preserved (Fig. 4). The resection involves the parotid gland, the temporomandibular joint and the mandibular condyle. If necessary, it may be extended to the infratemporal fossa.

In all the cases, a neck dissection of region II and III was performed. During the same procedure, an

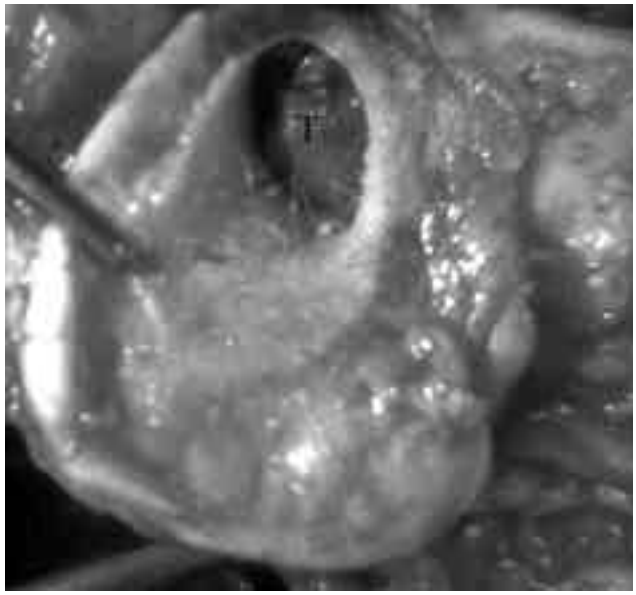

Fig. 1. Intraoperative view of a tumour (T) residing in the external auditory canal.

interposition nerve graft (greater auricular nerve) was used for facial nerve reconstruction. The cavity is obliterated with fat, or a muscle flap may be used (temporal muscle or, even better, a muscular flap of the pectoralis major); in cases in which skin is not available, a musculocutaneous flap may be used (pectoralis major, trapezius).

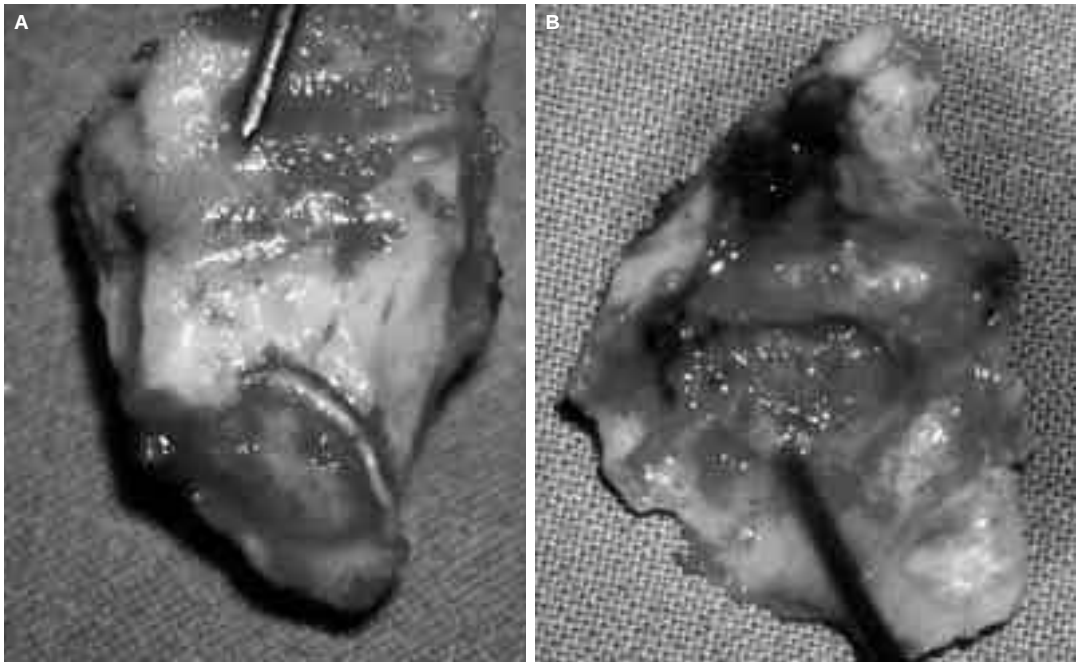

Fig. 2 A. *En bloc* resection specimen of a carcinoma of the external auditory canal. A lateral temporal bone resection was performed. Note that the bone surrounding the tumor and the tympanic membrane is intact. **B.** The lateral view of the same specimen. The tumor filling the external auditory canal is well-visible.

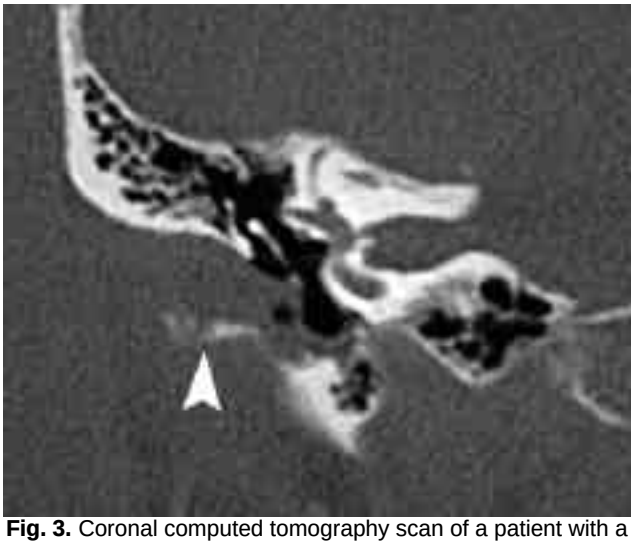

Fig. 3. Coronal computed tomography scan of a patient with a T_3 tumor of the external auditory canal on the right side, showing bone erosion to the tympanic bone (arrowhead) and involvement of the middle ear.

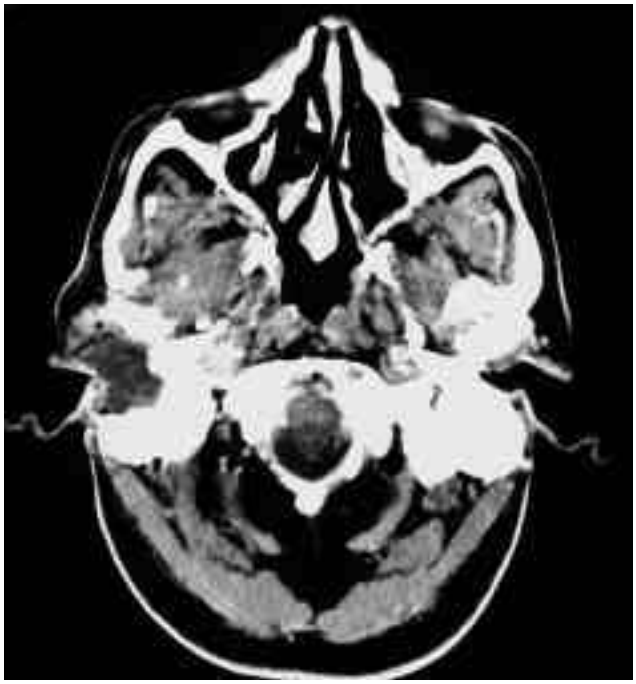

Fig. 4. Axial computed tomography scan showing a recurrent tumor on the right side. Clearly visible is the cavity due to a previous subtotal temporal bone resection.

Total temporal bone resection

This procedure was performed only sporadically and was used for tumors that involved the inner ear and extended to the intrapetrous part of the carotid artery and the petrous apex.

Local resection

This technique was not used in our patients with a curative intention. Patients who may benefit from this procedure are those that need palliative wound cleaning. The technique consists of an extensive cutaneous resection, in combination with bone resection at the level of the tumor. Reconstruction of the bone is achieved with the use of conchal cartilage and the excised skin is replaced by a thin skin graft.

Nonfractionated radiotherapy was administered, at the site of the tumor and/or to the lymph nodes in the neck, in patients for whom surgical treatment was not considered because of poor general health, irresectable tumor, etc. Neoadjuvant nonfractionated radiotherapy was given to some cases of salvage surgery. The remaining patients received postoperative fractionated radiotherapy. Five patients received chemotherapy consisting of 5FU and cisplatin.

RESULTS

The mean interval between the appearance of symptoms and first consultation was 22 months (range 1 to 168 months). The first symptoms were otalgia in 16 cases, otorrhea in 16 cases, bleeding from the ear in six cases, hearing impairment in 11 cases, facial paralysis in 11 cases, and neuralgia in two cases. Five patients had a history of chronic otitis, and four patients were being treated for eczema of the external auditory canal. Three patients had a past history of radiotherapy for a malignancy in the auricular region, dating back to a year for squamous cell carcinoma of the pinna in one patient, to 12 years for squamous cell carcinoma of the external auditory canal in another, and to 20 years for adenoid-cystic carcinoma of the parotid gland in the other.

According to the Pittsburgh classification, modified in 2000 and 2002 (Table 1), 12 patients were classified as having T_1 and T_2 disease, because differentiation between T_1 and T_2 was not obvious. In two cases, computed tomography and/or magnetic resonance imaging were not able to either confirm or exclude bony or cartilaginous erosion at the level of the external auditory canal. Six patients were staged as T_3, all of whom showed invasion to the middle ear, while two patients had erosion to the ossicles. Twelve patients were classified as T_4. Of these, three had invasion to the parotid, three had invasion to the dura, in two cases the jugular foramen was involved (1 with invasion to the carotid artery), three patients

had facial palsy, and 10 patients showed facial paralysis. Initially, none of the patients was diagnosed to have metastasized disease.

Twenty-one patients underwent a lateral temporal bone resection, and four patients underwent a subtotal temporal bone resection. Eleven patients underwent salvage surgery; of these, nine had undergone a partial resection (external auditory canal in 7, and pinna in 2 cases) and two had received chemoradiotherapy before as the only treatment.

Postoperative radiotherapy was used in 16 patients, of whom four patients had already received radiotherapy before surgical treatment. In this population, excision was often combined with a neck dissection (13 patients) and parotidectomy (13 patients).

The Kaplan-Meier survival curves showed two-year survival as 82%, 67%, and 32%, and five-year survival as 82%, 67%, and 17% for T_1 or T_2, T_3, and T_4, respectively. At the end of a follow-up period of 9 years, the survival rates were 66%, 66%, and 17% for T_1 or T_2, T_3, and T_4, respectively.

In our series, of 12 patients with T_1 or T_2, 82% were alive at the end of two and five years. However, long-term follow-up beyond 10 years is essential, because two patients who showed complete remission at 60 months further developed pulmonary metastases and died at 84 months. In this group, only eight patients (66.7%) showed complete remission within a mean follow-up of eight years (range 4 to 23 years). These eight patients underwent lateral temporal bone resection followed by radiotherapy except for one patient whose primary treatment was radiotherapy. Of these eight patients, two developed recurrent/residual disease after surgical treatment followed by radiotherapy, for squamous cell carcinoma of the pinna. In this group, mortality was seen in four patients (33.3%), one due to local recurrence, and three due to pulmonary metastases (1, 1, and 7 years after treatment). Of these, three patients underwent lateral temporal bone resection and one patient local resection of the external auditory canal; all received radiotherapy before (n=2) or after (n=2) surgery; three patients were treated for recurrent/ residual disease.

Of six patients with a T_3 tumor, four (66.7%) showed complete remission within a mean follow-up of five years (range 2 to 9 years). These four patients underwent lateral temporal bone resection followed by radiotherapy except for one patient with a melanoma, who was treated by surgery alone. The remaining two patients (33.3%) died, one at 5 months and the other at 12 months, both due to a local recurrence. One was treated by subtotal temporal bone resection (because of spread to the inferior surface of the temporal bone), the other by lateral temporal bone resection after a previous local resection. Both received radiotherapy postoperatively.

Of 12 patients with a T_4 tumor, only two patients (16.7%) who presented with an extracranial tumor extension (T_{4a}) showed complete remission within three and seven years, respectively. Both underwent lateral temporal bone resection, and resection of the temporomandibular joint, mandibular condyle, and the parotid gland. One received radiotherapy preoperatively, and the other postoperatively. Ten patients (83.3%) died in this group, four of whom had contraindications for surgery including unresectability in two (Fig. 5), and Alzheimer's disease in one. One patient did not accept any treatment. One patient died due to meningitis that developed after surgery performed for pain relieve. The remaining five patients underwent surgery (3 lateral, 2 subtotal temporal bone resections) followed by radiotherapy. The distribution of complete remission and mortality rates according to the type of surgery (primary treatment or salvage surgery) and to tumor stage are summarized in Table 2.

Overall, complete remission was found in 64.7% and 23.1%, and mortality was found in 35.3% and 76.9% for primary treatment and salvage surgery, respectively.

DISCUSSION

Diagnosis

The diagnosis is based on a biopsy which should be taken under general anesthesia in cases in which a malignancy is suspected and a previously taken biopsy under local anesthesia was negative. In our series, there were five cases in which biopsies taken under local anesthesia yielded false negative results. For correct staging, computed tomography and magnetic resonance imaging are indispensable tools because they allow evaluation of the extension to both bony structures and the soft tissue. Magnetic resonance imaging seems to provide important additional information.[8-10] One of the features of the tumor, which is difficult to visualize is spread through the fissure of Santorini.

Table 2. Outcomes following primary treatment and salvage surgery

		Primary treatment (n=17)		Salvage surgery (n=13)	
		%	n	%	n
T_1 or T_2	Complete remission	85.7	6/7	40.0	2/5
	Mortality	14.3	1/7	60.0	3/5
T_3	Complete remission	80.0	4/5	–	
	Mortality	20.0	1/5	100.0	1/1
T_4	Complete remission	20.0	1/5	14.3	1/7
	Mortality	80.0	4/5	85.7	6/7
Overall	Complete remission	64.7	11/17	23.1	3/13
	Mortality	35.3	6/17	76.9	10/13

Staging

Although various staging systems have been used[10-17] the Pittsburgh staging system seems to be the most frequently used system and it allows to compare homogeneously staged patient groups of various centers (Table 1).

The staging system proposed by the Belgium Consensus Conference in March 2002 is the system of the Pittsburgh group which was modified in 2000 an 2002. This system allows an estimation of the prognosis of individual patients. It provides suggestions regarding the therapy; however, no systematic therapy is provided for each stage. Therefore, the treatment may differ for tumors with the same stage. The direction of the spread of the tumor (anterior, posterior, or inferior) does not influence the classification. The presence of facial palsy is a poor prognostic factor and, if present, the patient is staged as having a T_4 tumor. In 1997 Lavieille et al.[18] proposed a new staging system for T_4 tumors, taking into account the direction of the spread of the tumor (Table 3). An overview of survival rates reported in the literature is summarized in Table 4.

Prognosis

In our series, we found that the stage and the anatomical extent of the disease were the primary factors influencing the prognosis. The mean survival rate of patients with metastasized disease was six months. The histology of the tumor was the second most important factor influencing the prognosis. For simi-

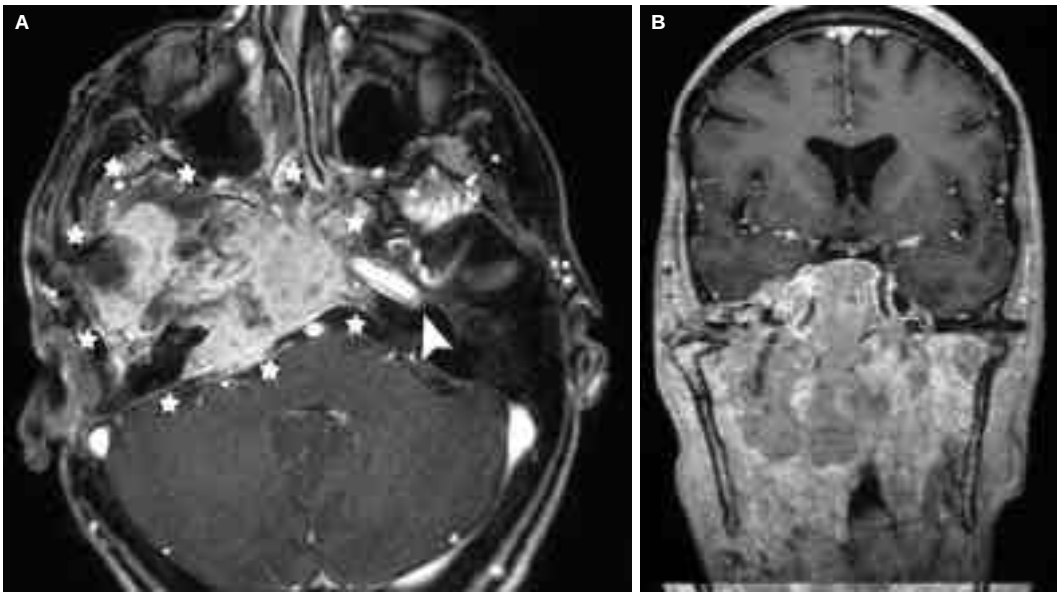

Fig. 5. A. Axial and **B.** coronal magnetic resonance scans with gadolinium, of a patient with an unresectable T_{4c} tumor on the right side. On the axial view, it is visible that the tumour (stars around) extends to the level of the clivus and carotid artery on the contralateral side (arrowhead).

Table 3. Staging on the basis of tumor extension

Stage	Anatomic borders of the tumour
T_1	Skin of the external auditory canal
T_2	Bony or cartilaginous external auditory canal
T_3	Middle ear cavity and external auditory canal
T_4	Intra- or extrapetrous bone extension
• T_{4a}	• Extracranial: infratemporal fossa, skin, parotid
• T_{4b}	• Intrapetrous bone and extradural extension
• T_{4c}	• Meningeal or intradural involvement

lar stages, the prognosis of basal cell carcinoma was better than that of squamous cell carcinoma.[19] The presence of facial nerve paralysis, involvement of lymph nodes and metastatic (M) disease were poor prognostic factors. Incomplete resection of the tumor also results in a poor prognosis.[10]

Therapy

Review of literature shows a large heterogeneity in the management of disease, which may consist of surgery, radiotherapy, and chemotherapy. Two opposing attitudes are observed: noninvasive therapy and maximal invasive radical surgery. Groups from Denmark and the Netherlands support the minimal invasive attitude. While the former[1] performs a local resection in T_1 tumors and may extend surgery on the basis of perioperative findings, the latter[20] uses meticulous tumor debulking followed by local application of 5-fluorouracil ointment, and during follow-up surgical necrotectomy is performed. Their overall survival rates were reported as 92% and 74%, respectively. No effective salvage surgery exists for patients with malignancies of the external auditory canal; therefore, the best therapy remains to be an effective primary therapy.[21]

Surgery

Surgical management according to tumor stage was outlined at the Belgium Consensus Conference in March 2002[4] (Table 5).

For patients with T_1 or T_2 disease, the recommended therapy for T_1 tumors is lateral temporal bone resection, and for T_2 tumors, lateral temporal bone resection followed by radiotherapy. In T_4 disease, intracranial extension (dura, internal carotid artery, jugular foramen, or posterior fossa) is a contraindication for surgery because of the inability to perform a radical excision and the high morbidity related to total temporal bone resection. Surgery for T_4 disease implies transsection of the internal carotid artery, and often the lower cranial nerves, leading to neurological complications and possibly to lethal pulmonary infections especially in the elderly patients.

Table 4. Survival rates in reported series

	Survival (%)			
	T_1	T_2	T_3	T_4
Kinney, 1989[12] (n=30, 2.5 years)	92		72	45
Pfreunder et al., 1999[16] (n=27, 5 years)	86		50	41
Moody et al., 2000[5] (n=32, 2 years)	100	80	50	7
This study (n=30, 2 years)	82		67	32

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 T_1 and T_2 tumors. The Pittsburgh group proposes adjusting the stage on the basis of postoperative histopathology, and using this information in deciding whether or not postoperative radiotherapy is required.[6] The German group of Würzburg[16] supports surgical therapy that may be combined with iridium-192 irradiation depending on the stage of the disease.

Radiotherapy

At present, the treatment protocol of the Belgium Consensus Conference of March 2002[4] consists of radical surgery most of the times followed by radiotherapy (Table 5). Several investigators provided evidence for the beneficial use of radiotherapy following surgery, with improvement in overall survival rates at two years.[6,22] On the other hand, radiotherapy alone for T_2, T_3, and T_4 disease was found to be associated with a poor survival. Moreover, patients whose advanced tumors could not be fully resected showed little benefit from adjuvant radiotherapy in

Table 5. Therapeutic guidelines by stage (Belgium Consensus Conference, March 2002)

Stage	
T_1	Lateral temporal bone resection and neck dissection
T_2	Lateral temporal bone resection and neck dissection followed by radiotherapy
T_3	Subtotal temporal bone resection and neck dissection followed by radiotherapy
T_4	Palliative treatment

terms of survival.[1] Postoperative radiotherapy in T_1 disease is still controversial.[6,10] Curative radiotherapy has been proposed by the Japanese group of Hashi.[23] They suggest that T_1 tumors be irradiated with 65 Gy and in the other stages radiotherapy be followed by a limited excision of the affected bone.

Chemotherapy

In our study five patients received chemotherapy, being as palliative care in three patients, and adjuvant to surgical treatment in one patient, with the latter having a complete remission. Chemotherapy is indicated in cases with residual disease, T_4 tumors, and in metastasized disease (M stage). Residual disease, independent of the stage, is usually detected in the first year after treatment.[13,14] The survival rates after treatment for residual disease are lower than those after complete primary treatment. In our study, chemotherapy did not promote the survival rate, but yielded better pain control. A combination of chemoradiotherapy may be effective in unresectable cases with a two-year survival rate of 75%.[24] In cases in which a single pulmonary metastasis is detected, surgery may be a treatment option; however, in multiple metastases chemotherapy is indicated. Compared with methotrexate, better results have been reported with cisplatin. Combination of cisplatin with 5FU may be the best chemotherapeutical treatment.[4]

In conclusion, current staging systems may provide a prognosis; however, the management options may not always show firm adherence to these staging systems. The long-term prognosis of external auditory canal malignancies is correlated to the tumor stage, biology of the tumor, and the initial treatment. From our point of view, surgical treatment with lateral temporal bone resection or subtotal temporal bone resection followed by radiotherapy appears to be the best treatment for T_1, T_2, and T_3 tumors. In T_1 tumors, histopathologically confirmed free excision margins may obviate radiotherapy. Additional subclassification based on extension of T_4 tumors appears to be effective in the choice of treatment for advanced tumors. Surgery may be considered in T_{4a} tumors, and palliative treatment in most T_{4b} tumors and in all patients with T_{4c} disease.

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