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ORIGINAL RESEARCH

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Treatment outcomes of the external auditory canal and temporal bone malignancy with dura invasion

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Abstract

Objectives: This study aimed to evaluate the characteristics and surgical outcomes of patients with external auditory canal (EAC) and temporal bone (TB) malignancy with dura invasion.

Methods: The medical records of patients with EAC and TB malignancy with dura invasion were retrospectively reviewed. Survival outcomes (overall survival [OS], disease-specific survival [DSS], recurrence-free survival [RFS], and distant metastasis-free survival [DMFS]) were analyzed using the Kaplan–Meier method.

Results: A total of eight patients were included in this study. The median age at diagnosis was 49.5 years (range 12–74 years). The median follow-up periods were 46.5 months. Histologically, four out of eight patients were diagnosed with squamous cell carcinoma (SCC; 50%). The 2-year OS and DSS rates of all patients were 62.5%, and those of EAC SCC patients were 50% and 66.7%, respectively; while the 2-year RFS and DMFS rates of all patients were 37.5%. There was one local recurrence at the resection site (12.5%), two regional neck nodal recurrences (25%), and two distant metastases (25%). Dura resection and duroplasty areas were not involved in the local recurrence case.

Conclusion: In EAC and TB cancer with dura invasion, radical surgery with dura resection may show similar survival outcomes to previous studies without recurrence at the dura resection site.

Level of evidence: IV

KEYWORDS
dura resection, external auditory canal malignancy, squamous cell carcinoma, temporal bone malignancy

1 | INTRODUCTION

Malignant tumors of the external auditory canal (EAC) and temporal bone (TB) are rare, accounting for 0.2% of all head and neck cancers.[1] The incidence is estimated to be one to six cases per million annually.[2,3] The most common histologic type is squamous cell carcinoma (SCC), followed by basal cell carcinoma (BCC) and adenoid cystic carcinoma (ACC).[4]

Treatment options include surgical resection, radiotherapy (RT), chemotherapy, or a combination of these modalities. Regardless of the anatomic limitation and the relationship with surrounding tissues, radical surgery, and combined postoperative RT remain the main forms of treatment.[5] According to previous case–control and retrospective studies,[6–8] treatment strategies depend on the T stage–sleeve resection or RT for T1, lateral temporal bone

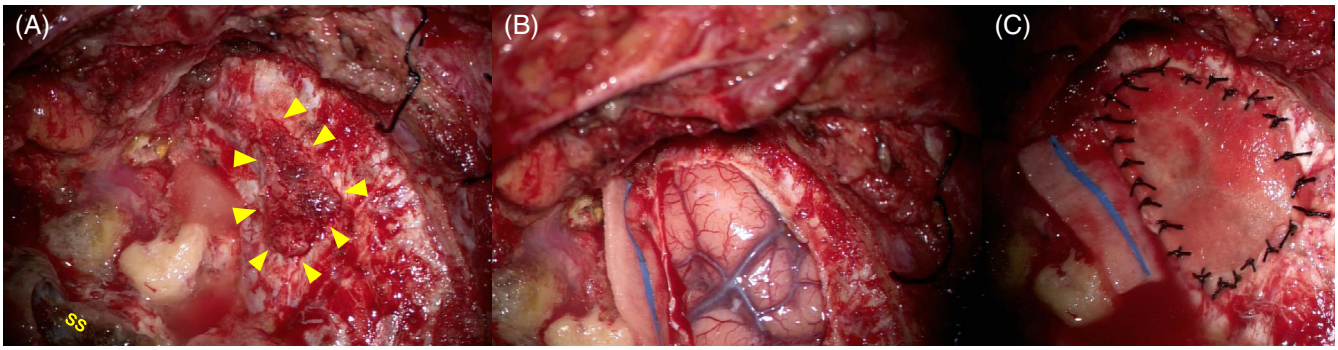

FIGURE 1 Dura resection and duroplasty. Middle fossa is exposed after subtotal temporal bone resection was performed in the left ear (case 7). The area where the tumor invaded the dura is identified (yellow arrowhead). (A) Removal of the dura with securing about 1 cm of safety margin and checking the frozen biopsies for the resection margin. In this case, the long diameter of dural defect is about 3 cm. (B) Duroplasty is done using biocompatible mesh. (C). After this, TachoSil® and surgical glue were used to reinforce. SS, sigmoid sinus.

resection (LTBR) and postoperative RT for T2, and subtotal temporal bone resection (STBR) and postoperative RT for T3 and T4 based on the modified Pittsburgh staging.[1] Recently, some reports had shown relatively good outcomes when definitive concurrent chemoradiation therapy (CCRT) was performed on patients with advanced stages.[9–11]

Advanced T stage, lymph node metastasis, positive resection margin, poorly differentiated histology, facial nerve involvement, and temporomandibular joint involvement were poor prognostic factors in EAC and TB SCC.[1,6,8,12–14] A combined approach with surgical excision and adjuvant RT or CCRT has been tried in the advanced stage of EAC and TB malignancies. However, postoperative complications, such as facial palsy, deafness, cerebrospinal fluid (CSF) leak, meningitis, encephalitis, and brain infarction, can occur.[15]

Dura invasion of the tumor is considered as T4 stage in the modified Pittsburgh staging and is a well-known adverse prognostic factor.[1,6,12,13] After resection of the mass and dura with a safe margin, reconstruction of the dura is sometimes difficult. In these cases, postoperative CSF leakage or wound infection delays the following combined treatment.[15,16]

This study aimed to evaluate the characteristics and surgical outcomes of patients with EAC and TB malignancy involving the dura.

2 | MATERIALS AND METHODS

2.1 | Patients and surgical procedures

We retrospectively reviewed the medical records of patients with histologically proven EAC and TB malignancy with dura invasion. Surgical excision occurred between January 2010 and December 2022 in the Department of Otorhinolaryngology-Head and Neck Surgery of Asan Medical Center. Patients were excluded if they had RT or CCRT without surgery or were referred for recurrence.

STBR and LTBR were performed in six and two cases, respectively. Parotidectomy, selective neck dissection, and surgical defect reconstruction were also performed. Postoperatively, RT or CCRT was applied to all patients. The Institutional Review Board of Asan medical center approved this study (IRB no. 2022-1437).

TABLE 1 Clinical characteristics of all patients (total $N = 8$).

Variable	*N*
Age at diagnosis (years), median (range)	49.5 (12–74)
Sex	
Male	6 (75%)
Female	2 (25%)
Site	
EAC	5 (62.5%)
TB	3 (37.5%)
Histopathology	
SCC	4 (50%)
Others	4 (50%)
Surgical procedure	
Main	
STBR	6 (75%)
LTBR	2 (25%)
Additional	
Parotidectomy	7 (87.5%)
Neck dissection	6 (75%)
Postoperative complication[a]	
Facial palsy	6 (75%)
CSF leakage	1 (12.5%)
Wound infection	2 (25%)
Recurrence[a]	
No recurrence	4 (50%)
Local	1 (12.5%)
Regional	2 (25%)
Distant	2 (25%)

Abbreviations: CSF, cerebrospinal fluid; EAC, external auditory canal; LTBR, lateral temporal bone resection; SCC, squamous cell carcinoma; STBR, subtotal temporal bone resection; TB, temporal bone.
[a]With overlapping in one patient.

TABLE 2 Detailed information of the cases in our study.

Case	Sex	Age	Site	Histopathology	Treatment modalities[a]	Type of TB resection	Parotidectomy	Neck dissection	Facial nerve neurorrhaphy	Reconstruction	Dura resection site	Postoperative complications
1	F	54	EAC	SCC, MD	Surgery + CRT	STBR	Total	SND (I–III, VA)	Cable graft (Sural nerve)	Temporalis muscle, SCM rotation flap	Middle fossa	Facial palsy
2	M	69	EAC	SCC, MD	Surgery + RT	STBR	Total	SND (II)	Cable graft (Sural nerve)	VRAM FF	Middle fossa	Facial palsy, Malocclusion
3	F	35	EAC	SCC, WD	Surgery + RT	STBR	Superficial	SND (IB–III)	Masseteric to facial nerve transfer	SCIP FF	Middle fossa, posterior fossa	Facial palsy, CSF leakage, wound infection
4	M	75	EAC	SCC, WD	Surgery + RT	STBR	Superficial	SND (IIB)	Cable graft (Greater auricular nerve)	SCM rotation flap	Middle fossa	Facial palsy
5	M	19	EAC	Adenoid cystic carcinoma, cribriform	Surgery + RT	LTBR	Total	–	FN preserved	PMMC	Middle fossa	Facial palsy, meningitis
6	M	12	TB	Ewing sarcoma	CTx + Surgery + CTx, proton therapy	LTBR	–	–	FN preserved	TRAM FF	Middle fossa	–
7	M	45	TB	Ceruminous adenocarcinoma, MD	Surgery + CRT	STBR	Superficial	SND (IB–III	Cable graft (Sural nerve)	VRAM FF	Middle fossa	Facial palsy
8	M	74	TB	Angiosarcoma	Surgery + RT	STBR	Superficial	MRND	FN preserved	ALT FF	Middle fossa	Acute infarction, wound infection

Abbreviations: ALT, anterolateral thigh; CRT, chemoradiation therapy; CSF, cerebrospinal fluid; CTx, chemotherapy; EAC, external auditory canal; FF, free flap; FN, facial nerve; LTBR, lateral temporal bone resection; MD, moderate differentiated; MRND, modified radical neck dissection; PMMC, pectoralis major myocutaneous; RT, radiotherapy; SCC, squamous cell carcinoma; SCIP, superficial circumflex iliac artery perforator; SCM, sternocleidomastoid muscle; SND, selective neck dissection; STBR, subtotal temporal bone resection; TB, temporal bone; TRAM, transverse rectus myocutaneous; VRAM, vertical rectus abdominis myocutaneous; WD, well differentiated.

[a]The treatment methods were written in the order in which they were conducted.

TABLE 3 Follow-up results of the cases in our study.

Case	Sex	Age	Histopathology	Recurrence		Expire		2-year status	Final status	Follow-up period (months)
				Site	Time[a] (months)	Cause	Time[a] (months)			
1	F	54	SCC, MD	–	–	Unknown	7.1	DOD	DOD	7.1
2	M	69	SCC, MD	–	–	–	–	NED	NED	50.1
3	F	35	SCC, WD	Regional (neck)	2.4	ICA thrombosis	5.5	DOD	DOD	5.5
4	M	75	SCC, WD	–	–	–	–	NED	NED	42.8
5	M	19	Adenoid cystic carcinoma, cribriform	Distant (lung)	31.9	Lung metastasis	56.7	NED	DOD	56.7
6	M	12	Ewing sarcoma	–	–	–	–	NED	NED	70.0
7	M	45	Ceruminous adenocarcinoma, MD	Regional (neck), distant (lung, bone)	33.0	–	–	NED	AWD	52.8
8	M	74	Angiosarcoma	Local (preauricular)	1.9	Lung metastasis	5.7	DOD	DOD	5.7

Abbreviations: AWD, alive with disease; DOD, dead of disease; MD, moderate differentiated; NED, no evidence of disease; SCC, squamous cell carcinoma; WD, well differentiated.

[a]Duration after the initiation of treatment.

2.2 | Dura resection and duroplasty

After histological confirmation of the mass involving the dura, the dura was resected and reconstructed (duroplasty). Several layers of surgical materials were stacked for dura reconstruction, including the temporalis fascia or biocompatible mesh (Lyoplant® [B. Braun Aesculap], Surgisis® [Cook]), a topical fibrin-based hemostat (TachoSil® [Baxter Healthcare Corp.]), and fibrin glues (Greenplast® [Greencross] and Tisseel® [Baxter Healthcare Corp.]; Figure 1).

2.3 | Statistical analysis

The primary outcomes were overall survival (OS) and disease-specific survival (DSS). The secondary outcomes were recurrence-free survival (RFS), distant metastasis-free survival (DMFS), and postoperative complications. Statistical analyses were performed using the SPSS Statistics for Windows, version 22.0 (IBM Corp.). Descriptive statistics were used to analyze the demographics and characteristics, and survivals were estimated using the Kaplan–Meier method; p value < .05 was considered statistically significant.

3 | RESULTS

A total of eight patients were included in this study. The median age at diagnosis was 49.5 years (range 12–74 years). The clinical characteristics of the patients are shown in Table 1. There were six males and two females. The median follow-up periods were 46.5 months. As the primary tumor site, five cases originated from EAC (62.5%) and three from TB (37.5%). The histologic results showed four patients with SCC (50%) and four patients with other types: one each with ACC, Ewing sarcoma, ceruminous adenocarcinoma, and angiosarcoma. In all SCC patients, the primary site was EAC. According to the modified Pittsburgh staging, all patients were T4 stage with suspected dura invasion at the time of diagnosis, and there was no patient with distant metastasis. Lymph node metastasis was clinically suspected in two SCC patients and one angiosarcoma patient.

Detailed information on patients with surgical procedures is presented in Table 2. All patients underwent STBR or LTBR with dura resection and duroplasty. All patients received a surgical excision followed by postoperative RT. Pre- or postoperative chemotherapy was combined in three patients. The main surgical procedure was STBR (75%), and parotidectomy (87.5%) or neck dissection (75%) was also performed. After the facial nerve resection, facial nerve anastomosis was done with a cable graft in four cases. The dura invasion was identified in the middle fossa dura in all patients. In one case, the posterior fossa dura was also invaded, and a wide range of resections, including the middle and posterior fossa dura, was required to remove the tumor. The area of dural resection was larger than 4 cm^2 in all patients. A vascularized musculocutaneous free flap or muscular rotation flap reconstructed the surgical defect. Facial palsy (75%) was the most common postoperative complication, followed by wound infection (25%), CSF leakage (12.5%), and meningitis (12.5%). In addition, malocclusion and acute cerebral infarction occurred in one case (12.5%).

The follow-up results of each case are summarized in Table 3. Three patients (37.5%) showed no recurrence or metastasis until the last follow-up. Two patients had regional neck nodal recurrences (25%), and two had distant metastasis (25%). Local recurrence was identified in one patient with angiosarcoma at the preauricular area (case 8). At the 2-year follow-up, three patients had died (cases 1, 3,

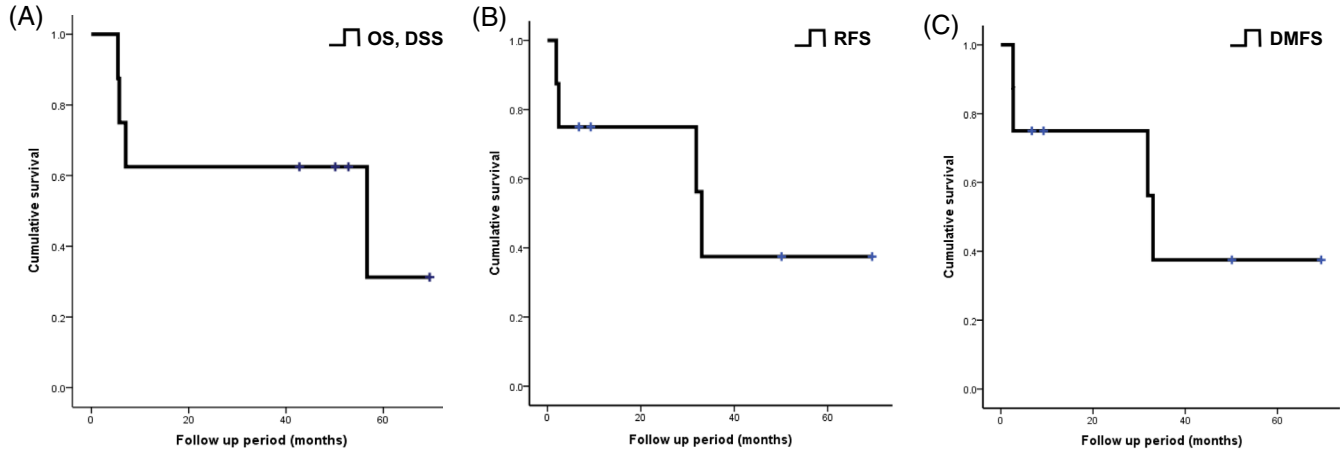

FIGURE 2 Kaplan–Meier curve for survival outcomes of EAC and TB cancer: overall survival and disease-specific survival (A), recurrence-free survival (B), and distant metastasis-free survival (C). EAC, external auditory canal; TB, temporal bone.

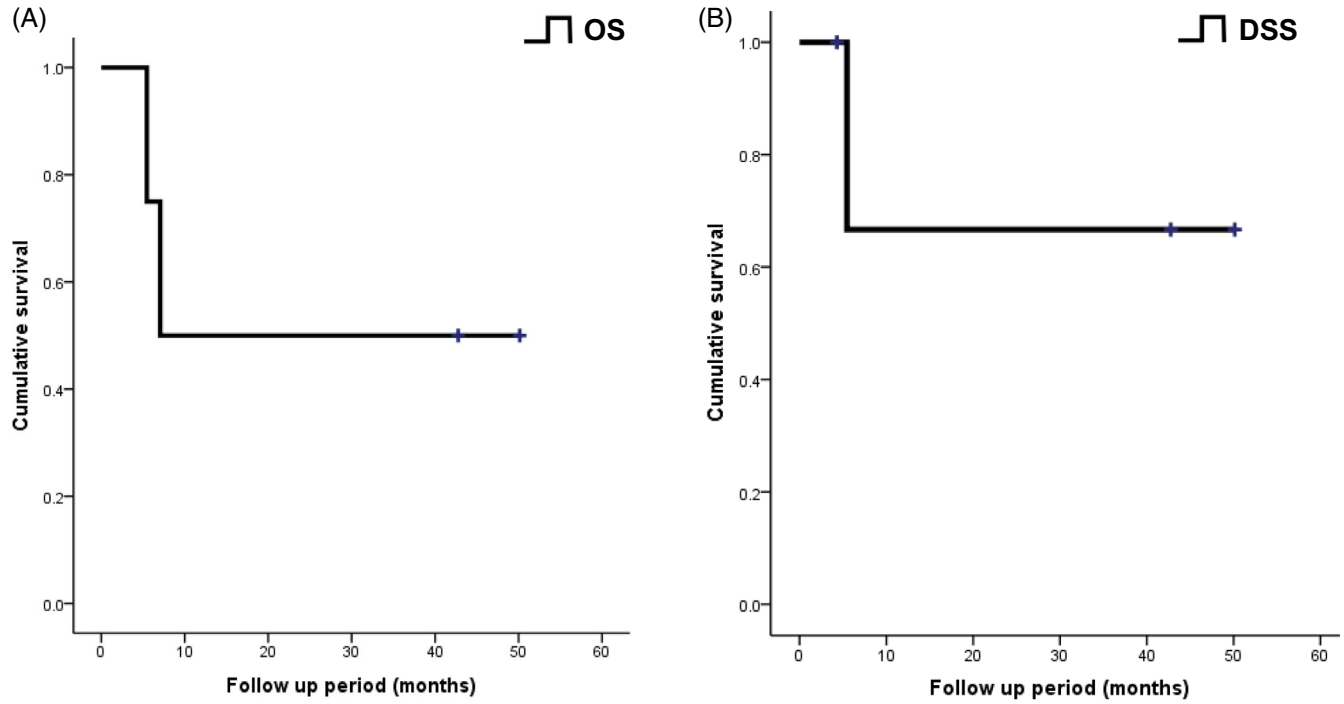

FIGURE 3 Kaplan–Meier curve for survival outcomes of EAC SCC: overall survival (A), disease-specific survival (B). EAC, external auditory canal; SCC, squamous cell carcinoma.

and 8), while five had no evidence of disease. Among these, one patient with ACC had a distant metastasis in the lung and died 56.7 months after the treatment (case 5). In another patient with ceruminous adenocarcinoma (case 7), regional recurrence and metastasis were observed at 33 months, but the patient was still alive after 56.7 months of follow-up.

The Kaplan–Meier curves analyzing the 2-year survival outcomes are schematized in Figures 2 and 3. The 2-year OS and DSS rates of all patients were 62.5%, and those of EAC SCC patients were 50% and 66.7%, respectively. The median OS time of all patients was 56.7 months, and that of EAC SCC patients was 7.1 months. The 2-year RFS and DMFS rates of all patients were 37.5%. The median RFS time of all patients was 33 months.

4 | DISCUSSION

When the dura is invaded, the EAC cancer is evaluated as a highly advanced state. According to a recent large-scale study on T4 EAC cancer conducted in Japan, the most critical anatomical areas influencing the patient outcome were the facial nerve and dura.[17] This result that the dura involvement significantly impacts the prognosis is consistent with that of

TABLE 4 Summary of surgical outcomes of EAC and TB cancer in previous other studies.

Study	Total *N*	Staging, mPSS (*N*)	Treatment modalities (*N*)	Surgical procedures (*N*)	Survival outcomes
Moody et al.[1] TB ca (SCC)	32	T1 (7) T2 (5) T3 (6) T4 (14)	Surgery (9) Surgery + RT (23)	LTBR (21) STBR (6) TTBR (5)	2-year OS T1: 100% T2: 80% T3: 50% T4: 7%
Gillespie et al.[26] TB ca (SCC)	15	T1 (5) T2 (3) T3 (3) T4 (4)	Surgery (6) Surgery + RT (9)	LR (3) LTBR (8) STBR (4)	2-year OS T1–2: 100% T3: 50% T4: 15%
Kunst et al.[27] TB ca (SCC)	28	T1 (12) T2 (2) T3 (4) T4 (10)	Surgery (5) Surgery + RT (23)	LR (12) LTBR (11) STBR (2)	2-year OS T1–2: 85% T3–4: 64% 10-year OS T1–2: 85 T3–4: 46%
Lassig et al.[28] TB ca (SCC)	30	T1 (4) T2 (3) T3 (6) T4 (14)	Surgery (10) Surgery + RT (16) Surgery + CRT (2)	LTBR (28)	2-year DFS T1–2: 100% T3: 67% T4: 56%
Bacciu et al.[6] TB ca (SCC)	45	Stage I (5) Stage II (6) Stage III (15) Stage IV (19)	Surgery (18) Surgery + RT (27)	LTBR (21) STBR (24)	5-year OS: 67.6% 5-year DSS Stages I–II: 100% Stages III–IV: 65.1% 5-year RFS Stages I–II: 100% Stages III–IV: 59.6%
Essig et al.[24] TB ca (SCC)	35	T1 (3) T2 (9) T3 (5) T4 (18)	Surgery (8) Surgery + RT (27)	LTBR (35)	2-year OS: 72% 5-year OS: 49% 2-year DFS: 68% 5-year DFS: 59% 2-year DSS: 79% T1–2: 100% T3–4: 70% 5-year DSS: 62% T1–2: 100% T3–4: 45%
Leong et al.[16] TB ca (SCC)	35	Stage I (4) Stage II (1) Stage III (1) Stage IV (29)	Surgery + RT (35)	LTBR (10) ETBR (25)	2-year OS Stages I–III: 100% Stage IV: 48.6% 5-year OS: 48.6%
Mazzoni et al.[25] EAC ca (SCC)	41	T1 (6) T2 (6) T3 (8) T4 (21)	Surgery (18) Surgery + RT (23)	LTBR (30) STBR (11)	5-year DFS T1–2: 67% T3–4: 41% 5-year DSS T1–2: 92% T3–4: 48%
Nam et al.[14] EAC ca (SCC)	26	T1 (10) T2 (8) T3 (4) T4 (4)	Surgery (11) Surgery + RT (12) Surgery + CTx (3)	LR (3) LTBR (17) ETBR (4) STBR (2)	5-year OS: 70.4% 5-year RFS: 61.8%

Abbreviations: CRT, chemoradiation; CTx, chemotherapy; DFS, disease-free survival; DSS, disease-specific survival; EAC, external auditory canal; ETBR, extended temporal bone resection; LTBR, lateral temporal bone resection; LR, local resection; mPSS, modified Pittsburgh staging system; OS, overall survival; RFS, recurrence-free survival; RT, radiotherapy; SCC, squamous cell carcinoma; STBR, subtotal temporal bone resection; TB, temporal bone; TTBR, total temporal bone resection.

previously reported studies.[6,12,13,18] The sufficient excision of the dura is anatomically challenging, making it difficult for clinicians to determine the best treatment strategy for patients with suspected dura invasions.

Several clinical staging systems have been proposed for the malignancy of EAC SCC. In this study, the modified Pittsburgh system was used, which was first proposed by Arriaga et al.[19] and modified by

Moody et al.[1] This staging system is specifically applied for the SCC of EAC and is most widely used. The eighth edition of the American Joint Committee on Cancer (AJCC) staging system has also provided the staging guidelines of cutaneous SCC of the head and neck, which can also be applied to the SCC of EAC and auricle. Previous studies have shown that the Pittsburgh system is more correlated with the survival outcome of EAC cancer than the AJCC system.[6,8,20] In addition to the SCC of the middle ear and EAC, a staging system for other histological types (BCC, ACC, and adenocarcinoma) was also presented by Stell and McCormic[21] and subsequently modified by Clark et al.[22,23]

In general, postoperative RT has been performed after radical surgery such as STBR or total temporal bone resection (TTBR) for stage T4 EAC cancer. In previous studies that analyzed the prognosis of T3 and T4 stages of TB SCC, a 5-year DSS was 45%–65%.[6,24,25] The 2-year OS was 50%–64% at T3 stage and 7%–49% at T4 stage.[1,16,26,27] Two-year disease-free survival (DFS) of T4 stage patients was 56%.[28] In a recently published large-scale review article, the 5-year survival rate for T4 disease was 14%–49%.[29] In our results, the 2-year OS and DSS of EAC SCC patients were 50% and 66.7%, respectively, consistent with the reported results (Table 4).

TTBR, which was implemented in the past, is rarely conducted because of its high surgical morbidity and no proven survival benefit.[6,30,31] Instead, at T4 stages, STBR and parotidectomy are mainly performed with dura resection, if necessary.[32] In our cases, all EAC SCC patients underwent STBR and parotidectomy with dura resection. In five cases, when direct invasion of the facial nerve was suspected, facial nerve neurorrhaphy was performed after facial nerve resection. Surgical reconstruction was performed by the plastic surgery team using vascularized musculocutaneous free flap or muscular flap.

Since dural invasion suggests a poor prognostic factor, it makes it difficult to decide whether to conduct surgical resection, though it is not a contraindication to surgery. During the follow-up, there was no recurrence at the dura resection site in all our cases. Local recurrence of the primary site occurred only in one case, at the preauricular area, which was not related to the dura resection site. According to the previous literature, surgical excision was only recommended when the size of the isolated dural involvement was less than 1 cm in preoperative MRI.[33] In all our cases, the area of the dura defect was 4 cm^2 or more after sufficient resection with a safety margin. Our results suggest trying curative surgery rather than palliative treatment even if the size of the dura invasion is greater than 1 cm, thus changing the previous principle.

When the extent of dura reconstruction is wide, another factor that makes it difficult to decide whether to perform surgery is the postoperative complication. According to our findings, only in one case, where a wide dural resection was performed, including the middle and posterior fossae, postoperative CSF leak occurred, and surgical exploration was done. Postoperative meningitis and acute brain infarction were suspected in one case each; however, the severity was mild and was controlled with conservative treatment. These are similar to that reported previously.[15] In addition, flap necrosis, osteoradionecrosis, and trigeminal neuralgia have also been reported.[16] According to our results, even if the area of dura resection exceeds 4 cm^2 or more, successful surgical results can be guaranteed without critical complications when a complete dura repair occurs.

RT after radical surgery is the mainstream treatment in advanced stages of EAC cancer. It has not been established whether neoadjuvant or adjuvant chemotherapy has an impact on prognosis. In our cases, preoperative chemotherapy was performed only in patients with Ewing sarcoma. Postoperative RT was performed in all patients, and chemotherapy was performed in three patients. Since the histopathological diagnosis of patients was heterogeneous and the number of cases was limited, it was difficult to statistically analyze the effect of postoperative chemotherapy on prognosis. This should be analyzed and discussed in a large-scale study.

CCRT was suggested to be an alternative treatment to surgery, showing 67% of the 5-year DSS in T4 patients.[9] In addition, for patients with unresectable disease due to invasion of internal carotid artery or extensive dura, CCRT was applied.[10] In T3–4 patients, the prognoses of the group that underwent conventional surgery and postoperative RT and the group that underwent definitive CCRT were 52.1% and 55.6%, respectively.[11] This was similarly observed in a recent large-scale study.[15] Based on these results, CCRT could be considered another treatment option for non-surgical cases. In this series, postoperative CCRT was applied to the patients.

This study had some limitations: a single institute, a retrospective study, and a small number of cases. Sufficient large-scale, long-term research is still needed to provide clear guidelines for this disease. Despite these limitations, the authors suggest that radical surgery with dura resection and repair may be helpful in patients with EAC and TB cancer with dura invasion. Since radical surgery of TBR had a potential risk of postoperative morbidities, CCRT could be used as an initial or adjuvant treatment.

5 | CONCLUSION

In advanced-stage cancer of EAC or TB, radical surgery with dura resection and postoperative RT may show similar survival outcomes to previous studies without recurrence at the dura resection site.

CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest.

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