

# Transoral Mandibular Tongue-Splitting Approach in Upper Cervical Epidural Abscess: A Case Report and Review of the Literature

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## Abstract:

**Introduction:** The transoral mandibular tongue-splitting approach is typically performed for the treatment of upper cervical tumor and instability but has not been performed for the treatment of upper cervical epidural abscess (UCEA). We report the first case of UCEA successfully treated with a transoral mandibular tongue-splitting approach.

**Technical Note:** A 62-year-old man who had medical histories of tracheotomy with intubation and dermatopathy due to radiation therapy for the treatment of nasopharyngeal carcinoma presented with neck pain and limb weakness. The imaging examination revealed bone erosion of C2-C4 vertebrae and abscess at the level of C2-C4, supporting a diagnosis of UCEA. The transcervical approach could not be used for treatment; therefore, the transoral mandibular tongue-splitting approach was used successfully to perform decompression, debridement, and iliac bone grafting. Subsequently, we reviewed the literature pertaining to the use of the transoral mandibular tongue-splitting approach. The approach can be invasive and cause some complications. However, no fatal complications have been reported, and all patients demonstrated a favorable neurological outcome with reduced neurological deficits.

**Conclusions:** This case and subsequent literature review suggest that the transoral mandibular tongue-splitting approach may be effective for the improvement of neurological outcomes without fatal complications in patients with UCEA. There may be an increasing number of patients with UCEA requiring the transoral mandibular tongue-splitting approach due to the increasing prevalence of immunocompromized status and the aging population.

## Keywords:

upper cervical epidural abscess (UCEA), transoral approach, mandibular tongue-splitting, transcervical approach, neurological outcome, complication

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## Introduction

The transoral mandibular tongue-splitting approach has been performed mainly for the treatment of upper cervical tumor<sup>1-6</sup>. However, the approach is technically demanding and invasive with many possible complications<sup>1-6</sup>. Thus, the indication may be limited to patients requiring surgical treatment with a wide operative field and anterior stabilization. An upper cervical epidural abscess (UCEA) can cause fatal respiratory failure and/or tetraplegia, and emergent surgical intervention is required in patients with progressive limb paralysis<sup>7</sup>. Although the transoral approach is frequently used for the treatment of UCEA<sup>8-17</sup>, it may be inappropriate if a wide operative field and anterior stabilization are re-

quired<sup>18-27</sup>. For such patients, the transoral mandibular tongue-splitting approach may be useful, but it has not been used for the treatment of UCEA due to its invasiveness and possible complications<sup>18-27</sup>. Here, we describe a patient with UCEA who was successfully treated *via* transoral mandibular tongue-splitting approach.

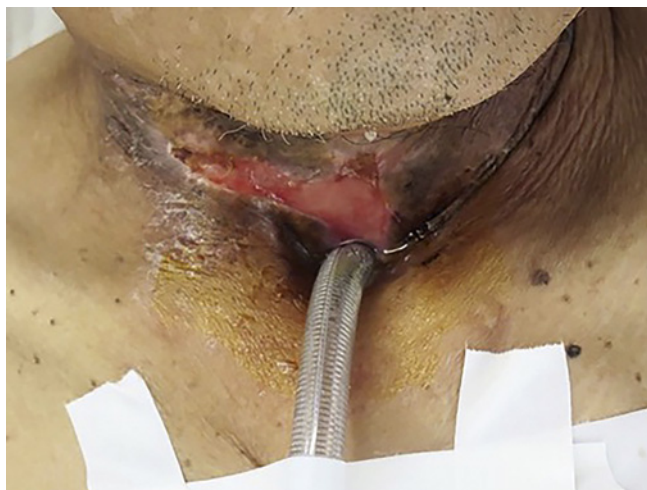
## Technical Note

A 62-year-old man presented with neck pain and limb weakness. He had medical histories of tracheotomy with intubation and dermatopathy due to radiation therapy for the treatment of nasopharyngeal carcinoma (Fig. 1). Laboratory tests revealed an elevated inflammatory response, and com-

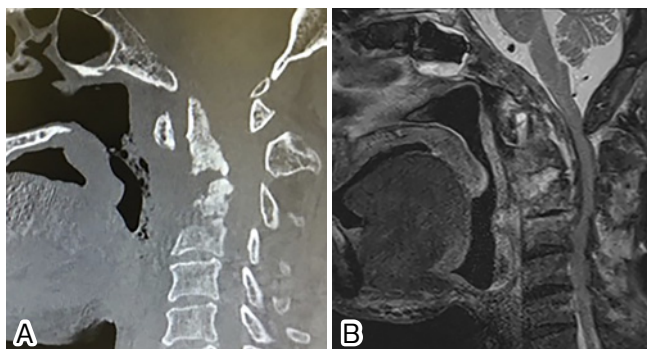
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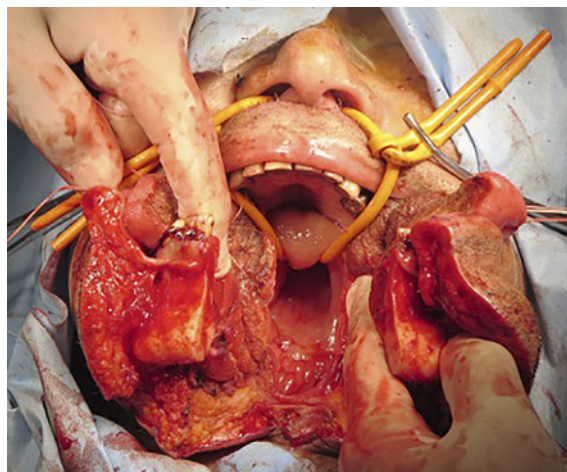
**Figure 1.** Post-tracheotomy, post-intubation, and post-dermatopathy due to radiation therapy for the treatment of nasopharyngeal carcinoma.



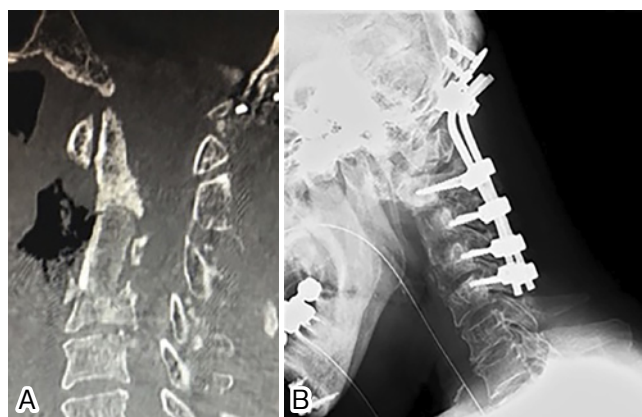
**Figure 2.** Sagittal plane of CT scan before treatment (A). Sagittal T2-weighted MRI before treatment (B). These images show bone erosion of C2-C3 vertebrae (A) and cervical epidural abscess at the C2-C3 levels (B).

puted tomography (CT) of the cervical spine demonstrated bone erosion in C2-C4 vertebrae (Fig. 2A). Subsequent T2-weighted magnetic resonance imaging revealed a high signal intensity lesion (Fig. 2B), indicating an abscess in C2-C4. Bacterial cultures were negative. The patient was diagnosed with UCEA, and intravenous antibiotics were administered (tazobactam/piperacillin). However, the patient exhibited deterioration of limb weakness, and repeat CT revealed progression of the erosion in C2-C4 vertebrae, as well as UCEA-related spinal cord compression. To prevent progressive neurological deficits due to the abscess, the patient underwent decompression, debridement, and iliac bone grafting by the transoral mandibular tongue-splitting approach.

A Nelaton tube was inserted into the nasal cavity and advanced to the oral cavity to elevate the soft palate. Transoral mandibular tongue splitting (Fig. 3) was achieved by an otolaryngologist and an oral surgeon. After debridement of the UCEA and infected granulation tissue, C2 vertebrae were excavated. Iliac bone grafting was performed to stabilize the



**Figure 3.** Transoral mandibular tongue-splitting approach. Median mandibular splitting, median longitudinal incision of the tongue, partial resection of eroded lesion of C2-C3 vertebrae, debridement of the abscess, suturing tongue, and titanium plate fixation of the mandible were performed.



**Figure 4.** Postoperative images. (A) Sagittal plane of CT scan. (B) Lateral radiograph.

excavated C2 vertebrae. Soft tissues and tongue were sutured, followed by mandibular fixation using a titanium miniplate. A halo-vest was placed to prevent postoperative dislocation of the bone graft.

Operative culture revealed *Pseudomonas aeruginosa*, and high-dose intravenous empirical antibiotic therapy was continued. One week after the first operation (Fig. 4A), the patient underwent posterior occipitocervical arthrodesis (Oc-C4) with instrumentation (Synthes GmbH., Eimattstrasse, Oberdorf, Switzerland) (Fig. 4B).

His symptoms, such as severe cervical pain and neurological deficit, rapidly improved following surgery. Perioperative incidental dural tear was detected, which may have been influenced by radiation therapy<sup>28</sup>. There were some minor complications, such as limited jaw mobility and superficial mucosal infections. However, meningitis, malocclusion, and a reduction in the occlusal force were not

**Table 1.** Characteristics of Reported Cases of Transoral Mandibular Tongue-Splitting Approach.

Reference	Year	Age (years)/ Sex	Primary disease	Level	Tracheotomy	Surgical procedure	Blood loss (mL)	Surgical time (h)	Complication (n)
Vishteh et al <sup>1</sup>	1999	11/M	Occipitocervical instability	Oc-C2	Done	Single-stage procedure: transoral bilateral sagittal split mandibular osteotomy combined with soft palate-splitting approach (unspecified)	Not available	Not available	Limited jaw mobility (4) Superficial mucosal infection (2)
		20/F	Klippel-Feil anomaly	Oc-C2	Done		Not available	Not available	
		49/F	Rheumatoid arthritis/basilar invagination	Oc-C2	Done		Not available	Not available	Macroglossia (1) Micrognathia (1)
		68/F	Rheumatoid arthritis/basilar invagination	Oc-C2	Done		Not available	Not available	Retrognathia (1)
Hiromasa et al <sup>2</sup>	2012	23/F	Upper cervical tumor	C2-C3	Done	Two-stage procedure: first stage, posterior debridement and fusion; second stage, transoral mandibular tongue-splitting extirpation (unspecified)	Not available	Not available	None
		65/M	Upper cervical tumor	C1-C2	Done	Two-stage procedure: first stage, posterior debridement and fusion; second stage, transoral mandibular soft palate-splitting extirpation (unspecified)	Not available	Not available	
		66/F	Upper cervical tumor	C2-C3	Done	Two-stage procedure: first stage, posterior debridement and fusion; second stage, transoral mandibular tongue-splitting extirpation (unspecified)	Not available	Not available	
Ortega-Porcayo et al <sup>3</sup>	2014	43/M	Upper cervical tumor	C2-C4	Done	Two-stage procedure: first stage, posterior bilateral laminectomies and facetectomies of C2-C4 and instrumentation (Occiput-C5-C6-C7); second stage, transoral mandibular tongue-splitting approach with anterior screw-plate fixation (C2-C4)	2100	9	None
		23/F	Upper cervical tumor	C2-C3	Done	Two-stage procedure: first stage, posterior bilateral laminectomies and facetectomies of C2-C4 and instrumentation (C1-C3-C4); second stage, transoral mandibular tongue-splitting approach using anterior titanium cage with bone matrix fixation (C1-C4)	900	5	

**Table 1.** continued

Reference	Year	Age (years)/ Sex	Primary disease	Level	Tracheotomy	Surgical procedure	Blood loss (mL)	Surgical time (h)	Complication (n)
Logroschino et al <sup>4</sup>	2004	59/M	Upper cervical tumor	C2	Done	Two-stage procedure: first stage, transoral mandibular tongue-splitting approach with anterior screw-plate fixation (C2); second stage, posterior instrumentation (Occiput-C2-C3-C4)	Not available	Not available	None
		63/F	Upper cervical tumor	C2	Done		Not available	Not available	None
Stulík et al <sup>5</sup>	2007	27/M	Upper cervical tumor	C2	Done	Two-stage procedure: first stage, posterior bilateral laminectomies and facetectomies of C2 and instrumentation (C1-C3-C4); second stage, transoral mandibular tongue-splitting approach using anterior cage with iliac bone graft fixation (C1-C3)	300	8	Superficial mucosal infection (1) Liquorrhea (1)
Menon et al <sup>6</sup>	2019	35/M	Occipitocervical instability	C2-C3	Done	Single-stage procedure: transoral mandibular tongue-splitting approach with anterior screw and/or plate fixation (C2), with/without posterior instrumentation (unspecified)	Not available	Not available	Superficial mucosal infection (1)
		26/M	Occipitocervical instability	C2-C3	Done		Not available	Not available	Superficial mucosal infection (1)
		46/F	Upper cervical tumor	C2	Done		Not available	Not available	Limited jaw mobility (1)
		67/M	Upper cervical tumor	C2	Done		Not available	Not available	Superficial mucosal infection (1)
		38/M	Upper cervical tumor	C2	Done		Not available	Not available	Superficial mucosal infection (1)
51/M	Upper cervical tumor	C2	Done	Done	Not available	Not available	Superficial mucosal infection (1)		
Current case		62/M	Upper cervical epidural abscess	C2-C4	Done	Two-stage procedure: first stage, transoral mandibular tongue-splitting approach with iliac bone graft (C3); second stage, posterior instrumentation (Occiput-C2-C3-C4-C5)	284	7.5	Limited jaw mobility (1) Superficial mucosal infection (1)



noted. Intravenous antibiotics were switched to oral administration of ciprofloxacin for a total of 6 months, with normalization of the inflammatory response and imaging confirmation of abscess disappearance. Although a bridging callus was not noted due to radiation therapy, the dislocation of the bone graft and implant was not detected at 2 months after surgery. Despite this progress, the patient died of progressive nasopharyngeal carcinoma 6 months postoperatively.

## Discussion

The transoral mandibular tongue-splitting approach is used mainly for tumor resection and stabilization of the upper cervical spine<sup>1-6</sup>. Despite its wide operative field, it is technically demanding and invasive with many complications<sup>1-6</sup>.

We reviewed the literature pertaining to the transoral mandibular tongue-splitting approach (Table 1). In all reported cases, preoperative tracheotomy was performed to prevent respiratory failure<sup>1-6</sup>. Cervical instability was treated with single-stage or two-stage posterior instrumentation<sup>1-6</sup>. Considering the blood loss and surgical time, this approach could be invasive. However, the neurological outcome was favorable in all patients<sup>1-6</sup>. Four of 19 patients died of progressive cancer at >4 months postoperatively; all patients without a history of cancer achieved full recovery.

Some authors have reported that UCEA was successfully treated by posterior stabilization and decompression, which provides a wide operative field and is a common approach for orthopedic surgeon<sup>13,29</sup>. However, we selected transoral mandibular tongue-splitting approach for five reasons in this case: first, anterior debridement and stabilization were needed, considering the abscess location and C2-C3 vertebral erosion. Second, posterior stabilization and decompression risked promoting implant infection. Third, the transcranial approach was contraindicated due to neck dermatopathy after radiation therapy. Fourth, postoperative management was relatively simple in terms of tracheotomy and intubation, which was placed for the treatment of nasopharyngeal carcinoma. Fifth, cooperation between otolaryngologists and oral surgeons is necessary in our hospital to successfully perform the technique.

The incidence of spinal epidural abscesses has increased due to the increasing prevalence of immunocompromized status and the aging population<sup>30,31</sup>. Moreover, the main source of UCEA is contiguous spread, such as that in the case of otorhinolaryngologic disease and tooth extraction<sup>8,27</sup>. Thus, there may be an increasing number of patients with UCEA requiring anterior debridement and stabilization.

This is the first report of the successful treatment of UCEA with transoral mandibular tongue-splitting approach, which may prevent neurological deficits without fatal complications in patients with UCEA.

**Conflicts of Interest:** The authors declare that there are no relevant conflicts of interest.

**Author Contributions:** Takaomi Kobayashi, Tadatsugu Morimoto, and Kazumasa Maeda wrote and prepared the manuscript, and all of the authors participated in the study design. All authors have read, reviewed, and approved the article.

**Informed Consent:** The patient and his family provided consent for submission of the case for publication.

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