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ORIGINAL ARTICLE

A unique procedure of joined transoral and retropharyngeal high cervical approach (JTRC) without mandibulectomy for treating upper cervical neoplasm involving both C2 and C3

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Abstract
Purpose To investigate a unique procedure of joined transoral and retropharyngeal high cervical approach (JTRC) without mandibulectomy for treating upper cervical neoplasm involving both C2 and C3.
Method A 23-year-old male patient had neoplasma involving C2 and C3 that caused pathologic fracture of C2 and bony destruction of C3. The neoplasm excision and cervical spine reconstruction were performed through JTRC approach without splitting up the mandible. In this approach, there were two surgery windows that could be applied in turns by closing or opening the mouth to gain an ideal exposure. By rotating the mandible, we could gain maximized exposure for either surgery windows. In addition, there was an overlay zone between transoral approach and high cervical retropharyngeal approach that could eliminate the blind area behind the mandible.
Results All the procedures were successfully performed; the surgical incisions healed without infection. By operating alternatively between the two surgery windows, we have successfully performed neoplasm excision and cervical spine reconstruction involving both C2 and C3. Pathologic results showed metastatic renal cell cancer to the resected cervical tumor and confirmed the patient's diagnosis of tuberous sclerosis (Bourneville disease). A 3-month postoperative cervical spine radiography and CT scan demonstrated a favorable placement of the bone implant as well as a favorable instrumentation.
Conclusion The JTRC approach could provide good surgical exposure for treating disorders involving both upper and lower cervical spine without splitting up the mandible.

Keywords Standard transoral approach (STOA) · Transmandible approach by mandible splitting (TMA) · Retropharyngeal high cervical approach (RHC) · Joined transoral and retropharyngeal high cervical approach (JTRC)

Background

In patients with disorders involving the craniocervical junction or upper cervical spine, standard transoral approach (STOA) could well expose the lesions from the lower part of clivus to the C2/3 disk [1, 2], providing a direct access for tumor resection or screw insertion. However, in cases with involvement of C3 or distal cervical vertebrae, the common transoral approach fails to offer enough exposure for surgery due to anatomic limitations from the tongue and mandible. An expanded transoral approach, transmandible combined with anterior cervical approach by mandible splitting (TMA), could provide more extensive exposure from clivus to lower cervical spine compared with standard transoral approach, and be used to surgically treat patients with osseous metastases involving both upper cervical and lower cervical spine [3–6]. However, this technique-demanding approach usually had associated postoperative complications including mandibular osteotomy nonunion, occlusal disturbance, sensitivity disturbance and carotid artery thrombosis. Additionally, the high

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morbidity due to the huge surgical wound has limited its wide clinical use.

In this report, we introduced a new method to deal with pathologies involving both upper and lower cervical spine without the need of splitting the mandible. We named it as "joined transoral and retropharyngeal high cervical (JTRC) approach", where the mandible was kept intact and did not require surgical splitting during osseous metastases resection and reconstruction.

Case report

A 23-year-old male patient presented with severe neck pain. Neurological examination revealed no paresis, normal deep tendon reflexes and full power in both upper and lower extremities. Neck CT (computer tomography) scan (Fig. 1) revealed destructive osseous lytic lesions in C2 and C3: a pathologic fracture crossed the base of odontoid and extended into the C2 vertebra; another small round lytic lesion located in the posterior C3 vertebra body. The posterior cortex of C2 vertebra had been destroyed. Additionally, there were sclerotic metastases in C4 vertebra body and spinal processes of C2, C3 and C4. T2-weighted magnetic resonance images (MRI) of cervical spine in the sagittal plane (Fig. 2) demonstrated neoplasm involving both C2 and C3 vertebrae bodies, while the C2 collapsed from pathologic fracture and expanded around, with a long T2 signal in the front of C2–4 indicating hemorrhage. Axial T2 image showed a mass lesion destroying the body of axis, and invading into the spine canal, touching the thecal sac without compression. Bilateral vertebrae arteries were not invaded by the metastatic lesion, and passed through the groove normally.

Additional MRI of abdomen revealed a retroperitoneal tumor, which appeared to be originated from the left kidney. Fine-needle biopsy of the cervical spine lesion and retroperitoneal mass both demonstrated vascular leiomyoma with low-grade malignant. Thus, the patient was diagnosed as tuberous sclerosis (Bourneville disease) associated with pathologic fracture of C2.

In view of the low-grade malignancy possibility of the cervical lesions, and anatomical complexity in the upper cervical region, en bloc resection was not our choice. We decided to use piecemeal excision and anterior autograft bone implant with plate fixation combined with posterior instrumentation and fusion for treating method.

Surgical procedures

Our surgical procedures include: (1) removal of the ventral lesion and reconstruction of cervical spine stability via anterior approach; (2) occipitocervical fusion via posterior approach.

The patient was placed in supine position. A nasal intubation was performed after general anesthesia induction. Surgical field was prepared with 10 % povidone iodine and sterile drapes. A Codman retractor was used to expose the oral cavity and a fiberoptic light to ensure favorable visualization of the posterior pharyngeal wall. A straight incision was made along the midline of pharyngeal wall. After that, a monopolar electrocautery was used to strip the bilateral longus colli for sufficient exposure of the anterior arch of C1 and anterior part of C2. The axis was invaded by tumor and bulge into the surrounding soft tissue. A pathologic fracture could be seen at the base of odontoid. A rongeur was used to remove the tumor piece by piece and special attention was paid to avoid damaging the vertebral artery. The anterior arch of atlas was keep intact, while axis body, most part of dens and C2/3 disk were resected thoroughly by piecemeal under the transoral surgery window. However, the C3 was hindered by the tongue and mandible, which was out of reach via this approach. Subsequently, the Codman retractor was taken off and the patient's mouth was closed. Another high

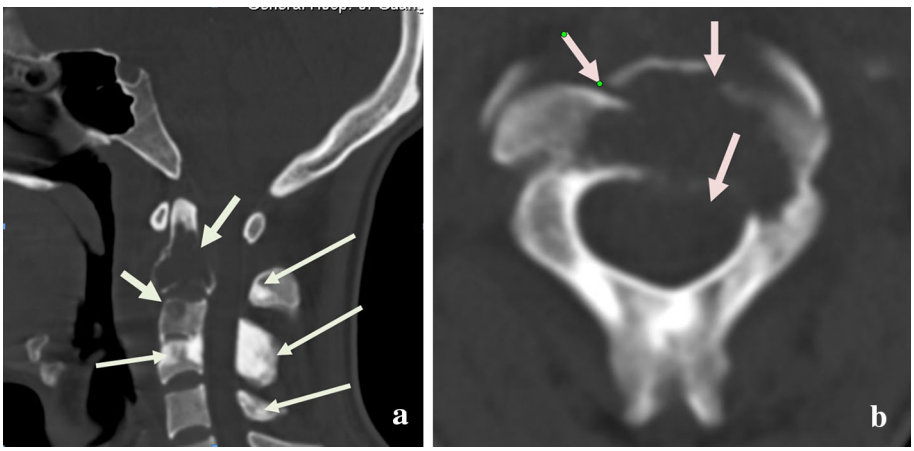

Fig. 1 **a** Sagittal neck CT in bone window showing lytic lesions in C2 and C3 (*short arrows*), noting that the posterior cortex of C2 was destroyed by the metastasis, and sclerotic metastases in C4 vertebra and spinal processes of C2–5 (*long arrows*). **b** Axial image of C2 vertebra showing pathologic fractures in the metastatic lytic lesion in C2 (*arrows*)

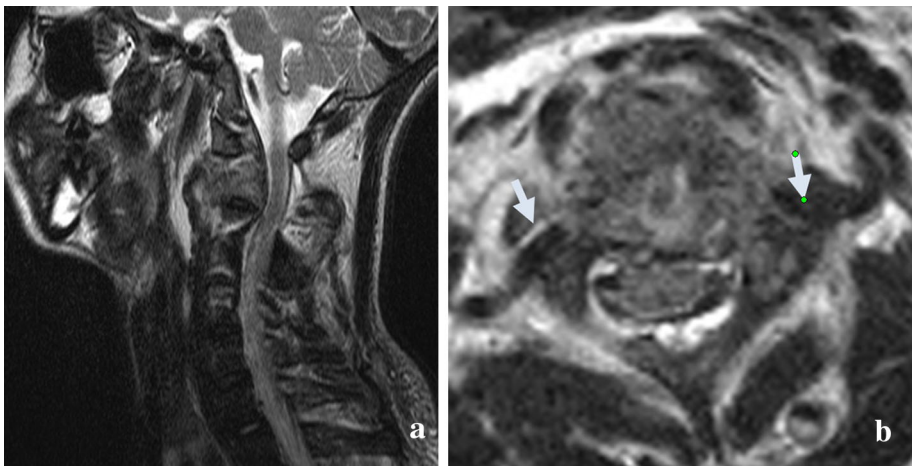

Fig. 2 **a** T2-weighted MRI of cervical spine in the sagittal plane demonstrated neoplasm involving both C2 and C3 vertebrae body, the C2 collapse for pathologic fracture and expand around, with a long T2 signal in the front of C2–4 indicating bleeding. **b** The T2 axial image shows a mass lesion destroying the body of axis, and expand a little into the spine canal, which touches with the thecal sac without compression. Both sides, vertebrae arteries could be seen passing through the groove without being invaded (*white arrow*)

cervical retropharyngeal incision was made at right side neck to achieve another operation window for exposure of C2–5 vertebrae.

A long straight incision along the sternocleidomastoid muscle was made, and then the platysma muscle was dissected. A gap between the carotid artery sheath and esophagus bronchi sheath was detected and retracted outward, allowing the access of C3 and C4 vertebrae. After ligation of the thyroid upper artery and lingual artery, the circum-tongue soft tissue was retracted upward as far as possible with a retractor or periosteal detacher to gain access to C2/3 gap. The hypoglossal nerve, lingual nerve, esophagus and carotid artery were carefully protected. Then, the remaining tumor in C3 was removed thoroughly (Fig. 3).

After that, a perfect shape and size bone bulk from the ilium was harvested and implanted into the tumor excision cavity carefully (Fig. 4), striding from anterior arch of C1 to C4 body; then a proper size titanium plate was chosen for fixation. After the anterior plate was applied and secured into place through the high cervical window, two locking screws were inserted into the anterior arch of C1 and body of C4 by different surgery windows, respectively (Fig. 5). All the procedures were performed smoothly as we successfully detour the mandible for surgical procedures from C1 to C4 and avoided otherwise a huge wound brought by mandible splitting (TMA).

The incision at the pharyngeal wall was closed in two layers, and then the high cervical incision was also sutured

Fig. 3 The neoplasm of C2 and C3 has been removed through JTRC approach

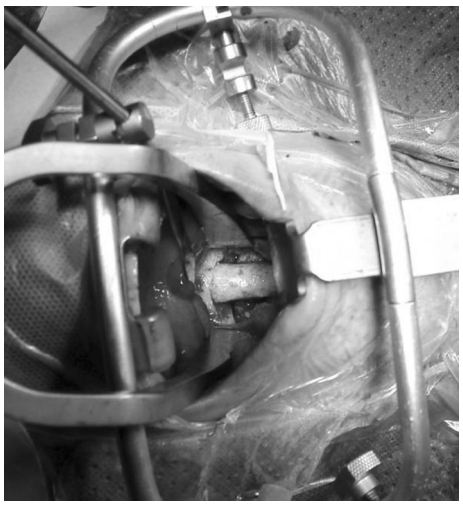

Fig. 4 A perfect shape and size bone bulk from the ilium was harvested and implanted into the tumor excision cavity carefully, striding from anterior arch of C1 to C4 body

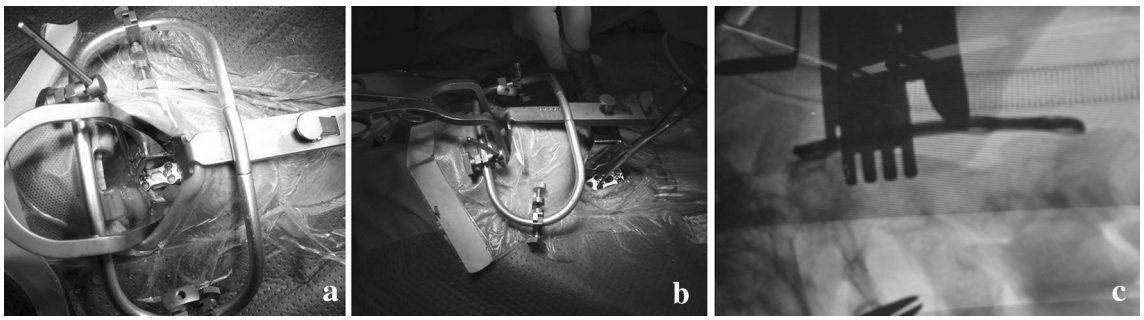

Fig. 5 **a**, **b**, **c** Using fluoroscopy monitor, a suitable plate was placed in the front side of cervical from C1 to C4. Then the screws were inserted through the transoral window or retropharyngeal window, respectively

after a drainage tube was placed after vertebrectomy of both C2 and C3, we worried about mechanical insufficiency with anterior instrumentation. Then the patient was turned to prone position. A posterior instrumentation and fusion were added.

Results

All the procedures were successfully performed, then the patient was sent to intensive care for 24 h. After the intubation was taken off, the patient was transferred to ordinary unit. The patient received 7 days of nasogastric tube feeding without complications. The surgical incisions healed with no infection. At 10th days after surgery, the patient started a rehabilitation program. Pathologic results of the resected cervical tumor showed metastatic renal cancer and confirmed the patient's diagnosis of tuberous sclerosis (Bourneville disease). Then patient received rapamycin for further treatment. He was discharged on postoperative day 14 with a rigid cervical collar. A postoperative cervical spine radiography and CT scan demonstrated a favorable placement of the bone implant as well as a favorable instrumentation. At 3-months follow-up, there was no displacement and loosening of the instrumentation on CT and radiography (Fig. 6), and the patient recovered to work.

Discussion

When we deal with lesions in the upper cervical region, there are three kinds of anterior surgical approaches: (1) the standard transoral approach (STOA), which is commonly chosen for ventral lesions located between the clivus and the upper cervical vertebra. This approach can provide direct access to the lesions, allowing excellent decompression of the ventral medulla and upper cervical spinal cord. Via this approach, we can also perform resection of neoplasms, release and/or reduction for atlantoaxial facet joint, and even atlantoaxial fixation with plate [7–9]. However, for the cases with lesions extending into lower cervical spine, the standard transoral approach fails to provide adequate exposure for surgery due to constrain by the mandible. (2) The retropharyngeal high cervical approach [10, 11] (RHC), which has been recognized as an extension of common anterior cervical approach. This approach could also be used for removal of lesions involving the atlas and axis. Compared to the transoral approach, the surgeries via this approach are less likely to be complicated by infection. However, it has limitations of awkward trajectory, restricted depth of exposure and indirect midline access to the upper cervical spine. Especially, when the mandible is small or in low position, the mandible would hinder the complex procedures involving the upper cervical spine. (3) Transmandible combined with anterior cervical approach by mandible splitting (TMA), which is an enlarged transoral approach by splitting up the mandible to further expose of lower cervical spine. This approach is commonly applied to treat ventral lesions involving both upper and lower cervical spine [12–15]. Although a good exposure can be achieved via this approach, it is not widely accepted due to high incidence of complications, including dysphagia, dysphonia, dysmasesia, non-union of the split mandible, and cosmic problems. After surgery via this approach, the patients' quality of life is poor; they have to receive nutrition through nasogastric tube for a long time until fusion of the split mandible bone.

Thus, we set to explore a novel method to achieve an extensive exposure for both upper and lower cervical spine without splitting up the mandible, thus avoiding the notorious complications.

In this report, we introduced a new method to deal with pathologies involving both the upper and lower cervical spine without splitting up the mandible. We named it as "joined transoral and retropharyngeal high cervical (JTRC) approach", as it means, by this approach, the mandible is kept intact and does not require surgical splitting while performing neoplasm excision and reconstruction just like that in TMA.

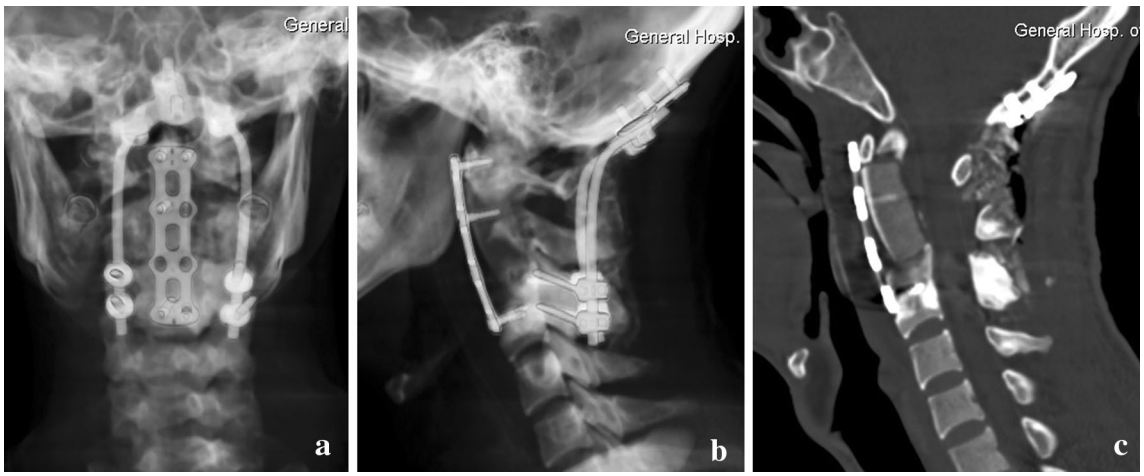

Fig. 6 **a**, **b** The postoperation cervical radiography shows perfect instrumentation. **c** The postoperation cervical reconstruction CT image shows the neoplasm has been removed and replaced with the bone implant

In our surgical procedures, two surgery windows were set up: a transoral operation window and a high cervical retropharyngeal operation window. Surgical procedures can be shifted in either window as needed to get an ideal trajectory and work path without splitting up the mandible.

There seems to be a relative blind area under the mandible. But actually, we could avoid it by switching the two windows in turns. When we were operating within the mouth, the mandible opened downward, and when the works switched into the high cervical windows, the mouth was closed. The mandible could be push upward as far as possible, which worked like a door that sways around the mandibular joints. This is a very important idea to understand the advantage of this union approach. Through this mechanism, we could gain maximized exposure for either surgery windows. We could find an overlap zone (Fig. 7) that could be accessed by both the upper and down surgery windows, thus leaving no blind area between two windows. Although the overlap zone may look a little like a tunnel, it was adequate for procedures.

When we worked in the high cervical windows, the thyroid upper artery and lingual artery needed ligation. The hypoglossal nerve, lingual nerve, esophagus and carotid arteriy are important organs that needed careful protection during the procedures. Actually, there was no need to expose the nerves deliberately; the carotid artery and esophagus could be retracted softly to the side to provide enough room for exposure.

In the overlap zone, the work trajectory was still a little awkward compared with the mandible splitting approach. When the neoplasm invades the surrounding soft tissue extensively, this JTRC approach seems not optimal for performing en bloc resection, or the pharyngeal wall needs to be closed with a free flap, which is its limitation.

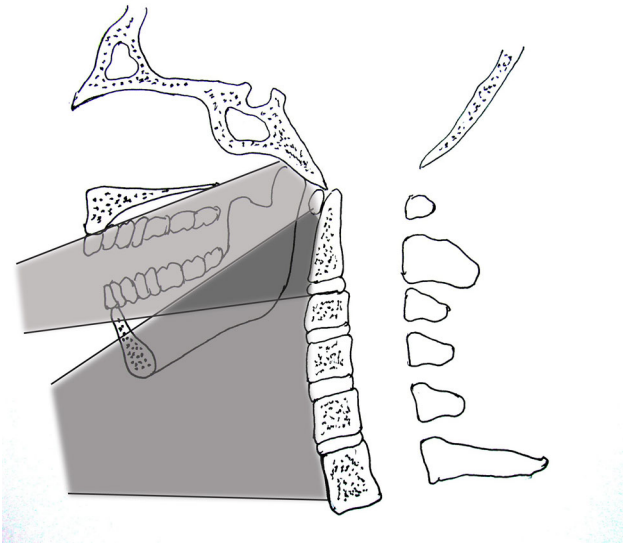

Fig. 7 The principle of joined transoral and retropharyngeal high cervical (JTRC) approach. There are two operation windows that could be achieved by this method (the transoral and retropharyngeal high cervical window). They could be used freely in turn, and the *black area* is the overlapping zone of the two operation windows. Therefore, we could keep away the mandible to perform tumor resection and reconstruction with instrumentation just like splitting the mandible

In summary, the advantages of JTRC include (1) smaller incision and less invasion than TMA. (2) Good cosmic results, as no incision or scar at the face. (3) Better life quality for postoperation than TMA. The patient could recover to normal eating rapidly. The outcomes of the patient indicated this new method is favorable in improving the life quality, decrease incidence of complications and gain a better cosmic result than MSTO. However, contraindications of this surgical approach may include mouth-opening disability, restricted neck extension, poor

condition of the mandible with small size or hypoplasia, and oral infection.

Our preliminary experience with this patient suggests that the JTRC approach may provide good surgical exposure for treating disorders involving both upper and lower cervical vertebrae without splitting up the mandible.

Since the development of endoscopic techniques and the design of the "fibroscope", endoscopy has been routinely performed in neurosurgery. Recently, endoscopic endonasal, transoral and transcervical approaches have been developed as promising alternatives to the traditional transoral microsurgery to the craniocervical junction (CVJ) [16, 17]. I suppose that, if combined with endoscopy assistant, our JTRC would be a more minimally invasive and convenient method for treating a variety of pathological conditions that affect the craniovertebral junction.

Compliance with ethical standards

Conflict of interest All the authors declare no conflict of interests.

Ethical statement All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards. No identifiable patient information was included in this case report. Informed consent was waived as per IRB policy on this retrospective case report. The manuscript has not been published before or is not under consideration for publication anywhere else and has been approved by all co-authors.

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