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IDEAS AND TECHNICAL INNOVATIONS

# Combined endoscopic transnasal and transoral approach for extensive upper cervical osteoradionecrosis

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**Abstract**
*Purpose* Osteoradionecrosis (ORN) is a rare yet well-recognized complication following radiotherapy to the head and neck. We illustrate the only case of a spontaneous extrusion of the sequestered C1 arch through the oral cavity and discuss our experience with a combined endoscopic transnasal and transoral approach for cervical ORN.
*Methods* A 56-year-old female presented with a 3-month history of blood-stained nasal discharge. She had been treated with radiotherapy for nasopharyngeal carcinoma 25 years earlier. Flexible nasal endoscopy demonstrated an exposed bone with an edematous posterior nasopharyngeal mass. Computed tomography showed a pre-vertebral mass with destruction of C1 and C2. She underwent occipito-cervical fusion followed by a combined transnasal and transoral endoscopic debridement of non-viable bone in the same perioperative setting. Healing of the raw mucosa was by secondary intention and reconstruction was not performed.
*Results* Histopathological examination reported ulcerated inflamed granulation tissue with no evidence of malignancy. During follow-up, she remained neurologically intact with no recurrence.
*Conclusion* Using both nasal and oral spaces allows placement of the endoscope in the nasal cavity and surgical instruments in the oral cavity without splitting the palate. Hence, the endoscopic transnasal and transoral approach has vast potential to be effective in carefully selected cases of cervical ORN.

**Keywords** Endoscopy · Cervical vertebrae · Osteoradionecrosis · Odontoid process · Nasopharyngeal carcinoma

## Introduction

Osteoradionecrosis (ORN) is a well-recognized complication following radiotherapy to the head and neck. Cervical ORN, first described in 1999 [1], remains a rare occurrence. This paper aims to illustrate an unusual presentation of cervical ORN and discuss our experience with a minimally invasive management. To the best of our knowledge, we present the only case of a spontaneous extrusion of the sequestered C1 arch through the oral cavity and the first report on a combined endoscopic transnasal and transoral approach for cervical ORN.

## Case report

A 56-year-old female presented with a 3-month history of blood-stained nasal discharge. There were no complaints of neck pain or other otolaryngological symptoms. She had been treated with radiotherapy for nasopharyngeal carcinoma 25 years earlier.

Clinically she had restricted movement in her neck in all directions; otherwise, the rest of her neurological examination was unremarkable. Flexible nasal endoscopy demonstrated an exposed bone with an edematous posterior

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nasopharyngeal mass (Fig. 1). Computed tomography (CT) showed a pre-vertebral mass with destruction of C1 and C2 (Fig. 2a, b). She was scheduled for surgery, however, she returned a few days later when she coughed out a fragment of bone which appeared to be the anterior arch of C1 (Fig. 3).

She underwent occipito-cervical fusion followed by a combined transnasal and transoral endoscopic debridement of non-viable bone in the same perioperative setting. Histopathological examination reported ulcerated inflamed granulation tissue with no evidence of malignancy. Her cultures grew normal flora of the naso-oropharynx. Hence, the final diagnosis was ORN of clivus, C1 and C2.

### Surgical technique (anterior approach to the clivus, C1 and C2)

The patient was positioned supine with her head placed on a horse-shoe head rest. The oral endotracheal tube was anchored to the left for easier instrumentation. Her nasal passages were prepared with Moffat's solution (1 ml of 1:1000 adrenaline, 2 ml of 10 % cocaine and 4 ml of 8.4 % sodium bicarbonate). She was registered to the Medtronic AxiEM$^{TM}$ electromagnetic navigation system. Rigid-rod endoscopes 4 mm in diameter and 18 cm in length with lens angles of 0° and 30° were used and mounted on a digital video camera system (Karl Storz). To facilitate the transoral method, the oral Boyle Davis retractor was utilized.

The endoscopic transnasal approach was employed initially where the endoscope and instruments were passed through both nostrils. A unilateral inferior turbinectomy was performed followed by a posterior septectomy to allow more room for instrumentation. In our case, a right inferior turbinectomy was performed. However, a left inferior turbinectomy could also be performed based on the surgeon's preference. The contralateral inferior turbinate and bilateral middle turbinates were preserved. Bilateral sphenoid ostia were enlarged to expose the clivus until a normal superior margin was identified (Fig. 4a). The necrotic anterior arch of C1 was drilled and removed with kerrison rongeurs. Using the transnasal technique alone, we were able to remove the anterior aspect of the odontoid peg almost to the level of its base.

The nasopalatine line (NPL) and nasopalatine angle (NPA) based on the preoperative CT scan (Fig. 2c) can be used to estimate the utmost extent of caudal dissection and window of exposure inferior to the hard palate [2]. As the non-viable bone extended inferiorly, we utilized a combined transnasal and transoral approach in which the 30° endoscope was placed transnasally and pointed downward to visualize the transoral instruments (Fig. 4b). We continued with the high-speed drill transorally in a craniocaudal direction. The anterior part of C2 was drilled until normal bone was reached posteriorly and a shell of healthy odontoid peg remained (Fig. 4c). The surrounding naso-oropharynx granulation was cauterized and debrided.

Healing of the raw mucosa was by secondary intention and reconstruction was not performed. Both nasal cavities were packed with Merocel® nasal tampons.

### Postoperative course

She was extubated after surgery and her recovery was uneventful. There were no complications of cerebrospinal fluid (CSF) leakage or meningitis. The patient was on orogastric feeding during the immediate postoperative period and was allowed oral feeding on post-op day 3. The patient was discharged within 10 days. Postoperative CT imaging revealed the remaining viable C2 and satisfactory fixation and alignment (Fig. 5a). During the 1-year follow-up, there were no concerns regarding dysphagia and our patient remained neurologically intact with no evidence of recurrence, signifying a favorable outcome (Fig. 5b, c).

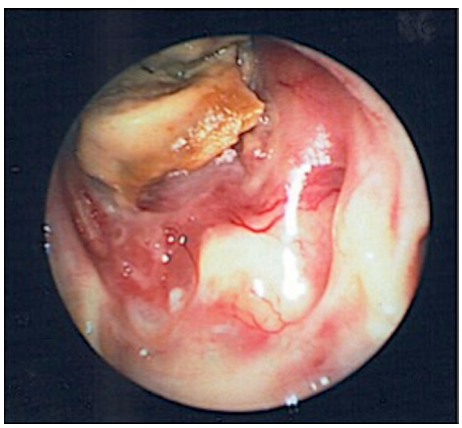

**Fig. 1** Preoperative endoscopic view revealing the exposed, necrotic anterior arch of C1 in the posterior nasopharynx

## Discussion

ORN was originally believed to result from radiation, trauma and infection, wherein trauma permitted bacteria entry with subsequent bone destruction [3]. However, Marx [4] proposed the combination of hypoxia, hypovascularity and hypocellularity to illustrate post-irradiation damage. The irradiated bone is unable to replenish the collagen and cellular components lost, thus resulting in tissue breakdown and necrosis [4].

Patients with cervical ORN often complain of neck pain. Other symptoms include nasopharyngitis, headache, upper extremity pain, dysphagia and kyphosis [5]. Our patient's atypical presentation highlights the importance of a

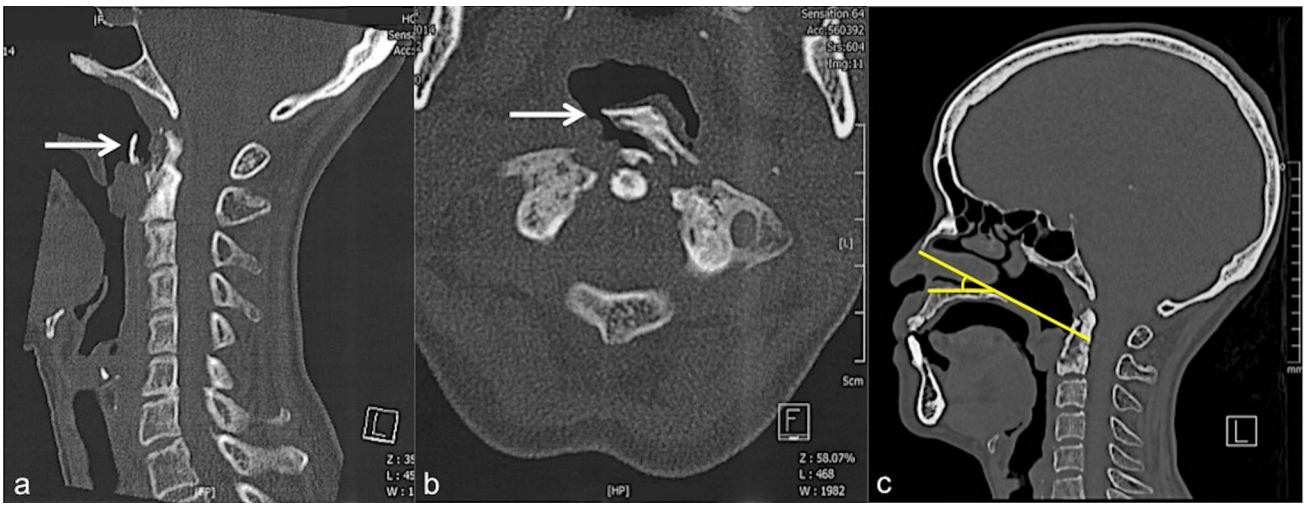


**Fig. 2** **a**, **b** Preoperative sagittal and axial CT scan demonstrating a pre-vertebral soft tissue mass with destruction of C1 and C2 **c** Preoperative sagittal CT scan with the nasopalatine line (NPL) and the nasopalatine angle (NPA). By extrapolating the NPL to the vertebral column, the most inferior limit of dissection with endoscopic instruments can be predicted. The NPA provides the window of exposure caudal to the hard palate

**Fig. 3** Photograph of the sequestered C1 anterior arch that spontaneously extruded

thorough history and examination. There was a case of an expectorated C1 arch secondary to trauma in 1837 [6]. However, to the best of our knowledge, we report the first case of a spontaneous, non-traumatic extrusion of a C1 arch through the oral cavity.

Conservative management of ORN includes antibiotics and hyperbaric oxygen. Necrotic bone is a suitable medium for microorganisms and the risk of infection increases because the exposed bone is constantly surrounded by oral flora. Although hyperbaric oxygen therapy promotes angiogenesis and fibroplasia in irradiated tissue [4], there remains a paucity of evidence supporting its clinical efficacy. Hence, we did not use hyperbaric oxygen therapy for our patient.

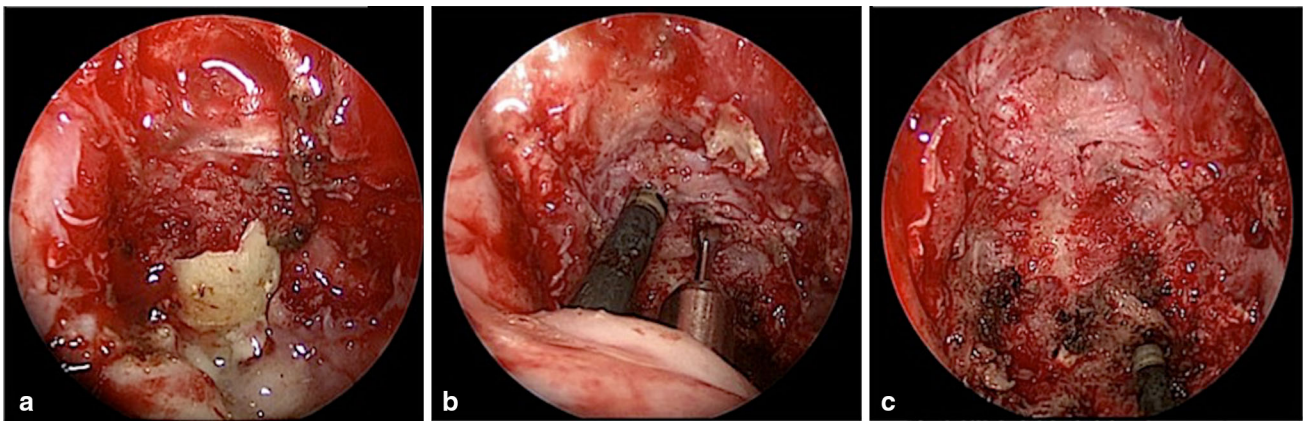


**Fig. 4** Intraoperative endoscopic images. **a** The clivus with a normal superior margin is visualized. **b** A combined transnasal and transoral approach was utilized in which the endoscope was advanced in the nasal cavity and the surgical instruments in the oral cavity. **c** Final view after debridement with normal bone and transverse ligaments visualized

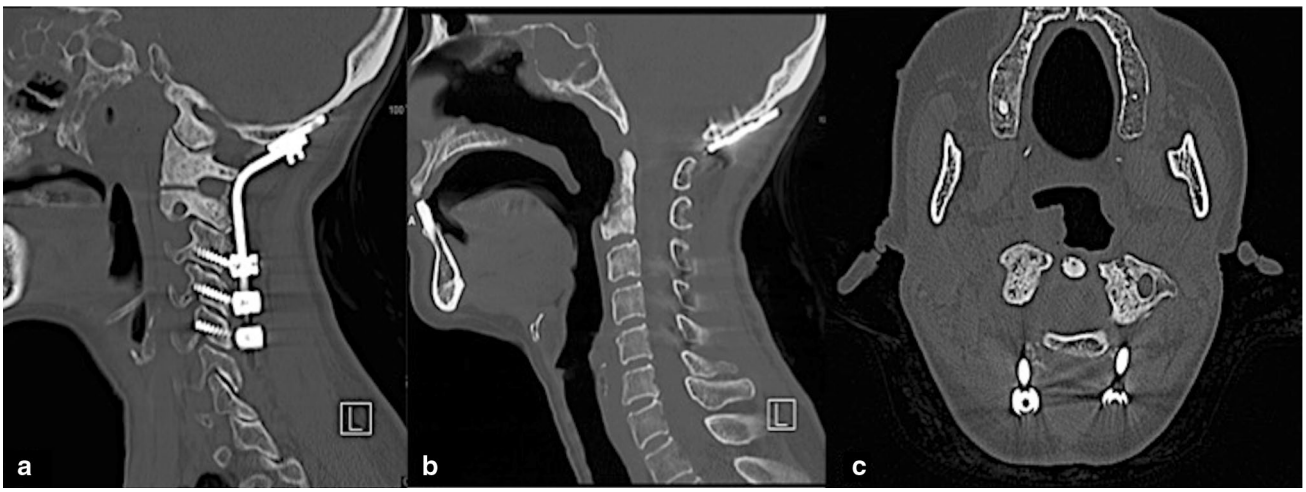


**Fig. 5** **a** Postoperative sagittal CT scan demonstrating satisfactory fixation and alignment after occipito-cervical fusion. **b**, **c** Postoperative sagittal and axial CT scan during the 1-year follow-up revealing the part of C2 that was preserved and no significant tissue damage after removal of the devitalized bone in this minimally invasive procedure

Cervical ORN can lead to devastating neurological and functional deficits, thus advocating more aggressive management. Debridement is imperative to remove necrotic tissue and permit healing by secondary intention. Surgical decompression of neural structures and restoration of spinal stability may be necessary [7]. Inadequate vascularity and hypoxic condition of necrotic bone can cause spinal fusion failure. Nonetheless, our patient had a favorable outcome, which we attributed to the minimally invasive removal of devitalized bone without significant tissue damage.

Management of craniovertebral junction is challenging due to surgical inaccessibility. The main advantage of the endoscope is the ability to provide wide-angled, closed-up views and excellent visualization through a narrow surgical corridor. The endoscopic resection of the odontoid, either transorally or transnasally, has been reported and the technical nuances detailed. However, we would like to describe our experience with a combined endoscopic approach (transnasal and transoral) which has the benefit of offering much greater access to the central cranial base.

The transnasal route allows access from the cribriform plate to the C2 body. Caudal exposure depends on the downward angulation of the endoscope, which is restricted anteriorly by nasal cartilages and posteriorly by the hard palate [8]. Hence, the NPL and NPA are useful to accurately predict the inferior limit of dissection with endoscopic instruments and the window of caudal exposure inferior to the hard palate [2]. The NPL is the line formed by joining the most inferior point of the nasal bone to the posterior part of the hard palate in the midsagittal plane, while the NPA is the angle between this line and the plane of the hard palate [2]. Pathological entities such as cranial migration of the C2 can be managed entirely with a transnasal approach. As for the transoral route, there is adequate access from the lower clivus to the C3 body but occasionally splitting of the palate or mandible is required for greater exposure [8]. This procedure is cosmetically unappealing, prolongs recovery and increases the risk of velopharyngeal insufficiency [9].

However, the problem arises when the pathology lies at the level of the palate such as in our case. We required an approach that can provide optimal exposure of the clivus and maximum room for instrumentation yet minimize morbidity. To overcome our dilemma, we decided to utilize a combined endoscopic transnasal and transoral technique. Combining the approaches (transnasal and transoral) allows placement of the endoscope in the nasal cavity and two or more instruments in the oral cavity. In addition, using both nasal and oral spaces reduces the incidence of blood soiling the endoscope and permits angulation of the endoscope in the cranio-caudal direction. This technique enables effective visualization while work progresses in the inferior aspects of the C2 peg. Hence, we believe the most significant advantage of the combined approach is the substantial improvement in access to the pathology and maneuvring of instruments without compromising safety or efficacy.

Based on our experience, we find that a unilateral inferior turbinectomy rather than simple lateralisation enables optimum exposure for visualization and allows adequate area for bi-nostril instrumentation with minimal invasiveness. This method was also used by Gladi et al. [10]. Despite the resection of the unilateral turbinate, we believe that as long as both the middle turbinates are preserved, we are still able to keep the nasal cavities moist, maintain normal laminar flow of air through the nose and

lessen postoperative crusting [9]. We also widened both sphenoid ostia to expose the clivus and enable identification of vital structures such as the carotid canals, pterygoid canal and vidian nerve. It was imperative that the sphenoid floor was drilled down to identify the sphenoid clivus. The mucosa of sphenoid was normal and thus, was not removed.

Our patient underwent occipito-cervical fusion, as there was concern on the craniovertebral junction stability following evidence of significant destruction of C1 and C2 on preoperative imaging. This was performed before the endoscopic debridement procedure because we believe it was safer to turn the patient to the supine position after posterior stabilization. The benefits of a single-stage procedure include reduced cost, shorter hospital stay and quicker recovery for the patient.

Nevertheless, there are several shortcomings with the endoscopic approach such as a steeper learning curve and difficulty of repair in cases of CSF leak. Persistent CSF leakage predisposes the patient to meningitis and should a durotomy occur, primary closure should be performed. Materials available for reconstruction include synthetic materials (dural substitute) or autologous materials (fat, mucosa or nasal septum, conchal cartilage, temporalis fascia, fascia lata). Fibrin sealants hold the graft in place, and ensure a watertight seal temporarily. The entire repair is usually held in position by a vascularized turbinate flap or nasoseptal flap [11]. The rate of CSF leak is relatively low with 3 cases reported [12–14]. It is not our aim to understate the effect of postoperative CSF leaks but we believe that satisfactory closure of the dura is achievable. There were no complications encountered in our patient.

## Conclusion

This case illustrates the importance of a high index of suspicion for early diagnosis and treatment of cervical ORN, particularly in patients who present atypically. The endoscopic transnasal and transoral approach should be considered as combined technique as opposed to strictly separate routes. This minimally invasive procedure has vast potential to be effective in carefully selected cases of cervical ORN.

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**Compliance with ethical standards**

**Conflict of interest** None declared.

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