# Double osteotomy of mandibula in the treatment of carotid body tumors with skull base extension

**Giorgio Prouse, MD,[a] Daniela Mazzaccaro, MD,[a] Fernanda Settembrini, MD,[b] Michele Carmo, MD,[a] Federico Biglioli, MD,[c]** and **Piergiorgio G. Settembrini, MD,[a]** *Milan, Italy*

**We report two patients with a carotid body paraganglioma that extended to the skull base, a position that is surgically inaccessible by means of a traditional lateral cervical approach. In both patients we were able to remove the lesion by performing a double mandibular osteotomy. Both patients underwent preoperative embolization to reduce the mass. In our experience, this approach has allowed a safe radical excision of exceptionally high lesions with only minor permanent nerve damage. In our opinion this advantage definitely outweighs the consequences of the increased invasiveness of this technique. (J Vasc Surg 2013;58:486-90.)**

Surgical excision of carotid body paragangliomas can represent an important challenge to the most experienced vascular surgeon due to the highly vascularized nature of the tumor and its proximity to vital structures.[1] The incidence of permanent cranial nerve injury and carotid wall damage during the procedure seems to significantly grow with lesions extending cranially, where they are difficult to reach through the traditional lateral cervical approach.[2]

## CASE REPORTS

We describe two patients with carotid body chemodectomas (CBCs) extending to the skull base that required a different surgical approach to ensure a radical and safe excision.

**Patient 1.** The first patient is a 49-year-old woman who came to our attention presenting with a bilateral lesion found after the onset of progressive dysphagia and left ear hypoacusia that led to further investigation. A contrast-enhanced computed tomography scan of the neck showed a 2.1- × 2.6-cm mass originating from the left carotid bifurcation and extending 6.5 cm cranially, involving the jugular foramen and causing the dislocation of vessels and adjacent structures (Fig 1). A 2.4- × 2.7-cm mass without intracranial extension was detected contralaterally. The patient was also found to carry a heterozygous IVS2 mutation of the succinate-ubiquinone oxidoreductase subunit D (*SDHD*) gene.

**Patient 2.** The second patient is a 56-year-old man, a fourth-degree relative of the first patient, who also presented with a bilateral lesion. He had previously undergone surgery elsewhere to attempt the excision of the mass on the left side, which resulted in a major lesion of the internal carotid artery leading to ligation of the vessel and incomplete resection of the tumor. He also sustained a lesion of the left glossopharyngeal nerve and is currently fed by percutaneous endoscopic gastrostomy due to permanent solid food dysphagia. A computed tomography angiography showed a residual 4.2- × 1.7-cm mass on the left and a 5.8- × 2.7-cm mass on the right with skull base extension but without intracranial involvement (Fig 1).

The dimension and skull base extension of both tumors led us to consider a multidisciplinary approach to obtain a wide exposure allowing a radical excision and minimizing the risk of nerve and vessel lesion. We therefore planned preoperative embolization to decrease the risk of bleeding and to potentially reduce the dimension of the mass. A maxillofacial surgeon with expertise in accessing the retromandibular region was consulted.

Both patients underwent preoperative embolization with polyvinyl alcohol (PVA) spheres of 350 to 700 μm diameter. The spheres were injected after a superselective catheterization of the identifiable feeding vessels. The tumor reduced ~30% from its initial diameter.

Surgery in both patients was performed by a team of maxillofacial and vascular surgeons ≤2 days after embolization. Surgical access was achieved by performing an incision along the mandibular inferior margin from the ear to 10 cm distally, following the cutaneous ply. Under the platysma we identified the marginalis mandibulae branch of the facial nerve, which was spared and moved cranially. The incision of the mandibular periosteum was followed by the creeping and cleaning of the mandibula. A vertical ramus osteotomy, combined with parasymphyseal mandibulotomy, is a modified version of the mandibular mobilization previously described by Seward for the excision of parapharyngeal tumors (Fig 2).

The specific position of the parasymphyseal osteotomy was conceived to promote sparing of the inferior alveolar nerve (sensitive branch of trigeminal): osteotomy was performed medially to the mental foramen to guarantee the sensitivity of the inferior lip.[3] The double osteotomy enabled us to overturn the mandibular body and ramus and to anteriorly dislocate the condyle, allowing easy access to the carotid axis and skull base (Fig 3). No intraoral incisions were performed, and tracheostomy was therefore not necessary. Osteosynthesis was accomplished by means of four

From the Division of Vascular Surgery, Ospedale S. Carlo Borromeo, Universitá degli Studi di Milano[a]; the Division of Plastic Surgery, Ospedale di Niguarda Ca' Granda[b]; and Maxillo Facial Surgery, Università degli Studi di Milano, and the Division of Maxillo Facial Surgery, Ospedale S. Paolo.[c]

Author conflict of interest: none.

Reprint requests: Piergiorgio G. Settembrini, MD, Division of Vascular Surgery, Ospedale S. Carlo Borromeo via Pio II 3, 20153, Milan, Italy (e-mail: piergiorgio.settembrini@unimi.it).

The editors and reviewers of this article have no relevant financial relationships to disclose per the JVS policy that requires reviewers to decline review of any manuscript for which they may have a conflict of interest.

0741-5214/$36.00



http://dx.doi.org/10.1016/j.jvs.2012.11.086

**Fig 1.** Three-dimensional volume rendering of patients 1 and 2. **Left,** This image of patient 1 shows a posterior and lateral view of the carotid body tumor and its relation with the mandibula and the skull base. **Right,** This image of patient 2 is a lateral view of the carotid body chemodectoma (CBC). The small axial image in the *lower right corner* shows the presence of bilateral CBCs.

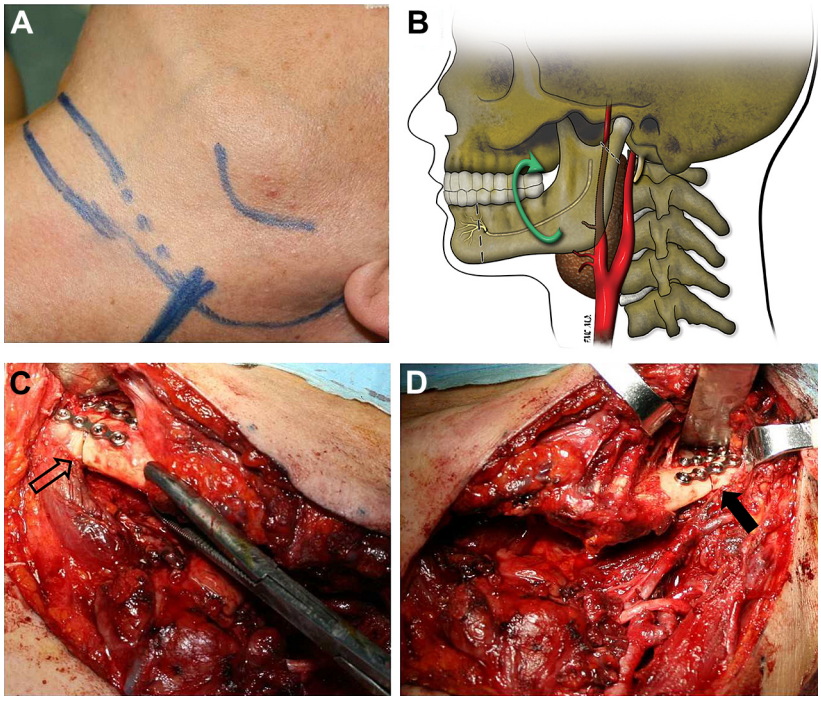


**Fig 2. A,** Relation between the mandibula angle (*blue drawing*) and **(B)** incisions lines drawing of osteotomy subcondylar and mandibular paramediana proximal and distal to inferior alveolar nerve. **C,** Proximal (*clear arrow*) and **(D)** distal mandibular osteotomy (*black arrow*) with titanium plates.

premolded 1-mm-thick titanium plates that had been prepositioned and removed before the osteotomies (Fig 3).

In both patients we were able to gain easy control of the common carotid artery and of the distal segment of the internal carotid artery, providing a surgical field that enabled comfortable recognition of the entire mass and of adjacent vital structures. Near-infrared spectroscopy was used to monitor cerebral perfusion, although carotid clamping was never necessary.

The tumor in both patients was classifiable as a Shamblin group II lesion.[2] At the histologic examination, both lesions presented positive reaction to chromogranin and S-100 protein and were diagnosed as paragangliomas.

Patient 1 presented an immediate postoperative asymptomatic troponin elevation (3.20 ng/mL) with pre-existing negative T waves in the lateral leads. A coronary angiogram was negative for stenotic lesions. Troponin levels normalized on postoperative day 4. She also showed a homolateral vocal cord abduction palsy that was still persistent 26 months after surgery. Intermaxillary fixation to ensure effective dental occlusion was accomplished by the use of rubber bands. Parenteral nutrition was interrupted on

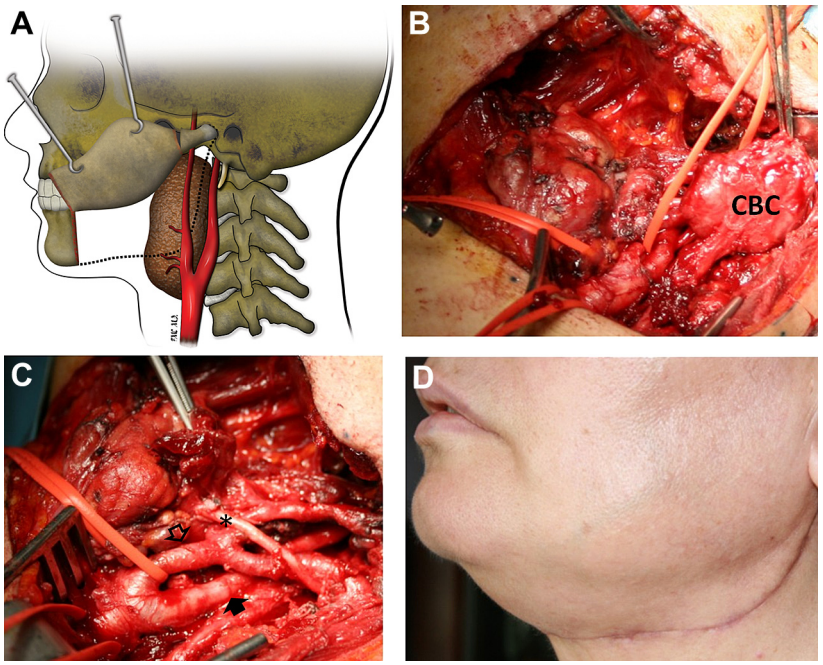


**Fig 3. A,** Drawing with exposure of the internal carotid artery after overturn of mandibula and anterior translation of mandibular condyle. **B,** Intraoperative image of carotid body chemodectoma (*CBC*) during the removal. **C,** Carotid bifurcation without carotid body tumor: the *clear arrow* indicates the external carotid artery, the *black arrow* indicates the internal carotid artery, and the *asterisk* indicates the hypoglossal nerve. **D,** Postoperative photograph shows the result of the intervention with submandibular scar.

postoperative day 7 after complete resumption of oral feeding. No impairment in the temporomandibular joint was noted.

The postoperative course for patient 2, who had the same operation, was uneventful, and he did not sustain any further transient or permanent cranial nerve deficit. Neither patient experienced any difficulty with the temporomandibular joint.

The patients regularly took nonsteroidal anti-inflammatory drugs to control postoperative pain, for 21 and 18 days, respectively; thereafter, they were taken occasionally, with pain gradually disappearing over time. The two patients (who live in a different region of the country) were seen at follow-up visits up to 26 and 3 months, respectively. The second patient was followed up by telephone calls, and radiologic imaging was regularly sent to us for review. Both patients are doing well compared with their preoperative condition. Patient 1 still experiences some minor speech impairment caused by her vocal cord palsy.

## DISCUSSION

CBC is a rare tumor that originates from chemoreceptor cells deriving from mesodermal elements of the third branchial arch.[1] In most patients it is a benign tumor, with a reported metastasis rate of between 2% and 9%.[4] However, CBCs show an inexorable growth pattern with progressively increasing likelihood of cranial nerve damage[5] and airway compression that imposes treatment of all patients with a life expectancy exceeding a few years. The classification proposed by Shamblin et al[2] categorizes the tumor according to its involvement of adjacent vessels and nerves. The Shamblin classification does not, however, encompass the cranial extension of the lesion, although this aspect greatly influences the outcome of surgery.[6] Lesions reaching the skull base and in some cases involving the carotid or the jugular foramen have always challenged the feasibility of surgical treatment.[7-9]

The extreme cranial extension in our patients accounted for an unacceptable risk of surgery-related morbidity if approached from a traditional lateral cervical incision. Preoperative embolization has been used to reduce vascularization and size of CBCs, yielding a dramatically easier excision and thus reducing blood loss and lowering the risk of permanent cranial nerve injury.[10] We do not routinely use embolization unless CBCs are a Shamblin III or II with a high cranial extension.

Two groups, one from Mayo Clinic and one from Guangzhou, China, recently reported their results in 34 and 66 CBCs, respectively, with preoperative embolization with PVA through a superselective catheterization of the feeding vessels in large CBCs. This procedure was safely performed in both series, with no neurologic or hemorrhagic complications; however, one nonflow-limiting dissection without sequelae occurred in the Mayo Clinic series.[11,12]

As an alternative to PVA embolization, spirals and more recently Onyx (ev3 Endovascular, Inc, Plymouth, Minn) have been proposed. Onyx is a relatively new embolic agent that has been used to reduce tumor

vascularization before surgical resection. Elhammady et al[13] reported their favorable experience with preoperative embolization of 18 hypervascular head and neck tumors (including 9 CBCs) using Onyx by direct puncture of the mass under fluoroscopic guidance. In our practice, we have no experience with Onyx because our results with PVA were favorable.

Despite preoperative treatment with embolization, most lesions that extend to the skull base cannot be sufficiently exposed through a lateral cervical access to warrant a radical excision with an acceptable rate of nerve and vessel injury.[14,15] As recently pointed out by the Mayo Clinic group, the distal extension of the CBC plays a very important role in predicting cranial nerve injury; in fact, the projection of the mass extending beyond the C1 vertebra or with a lateral margin of <5 mm from the angle of the jaw on CT or magnetic resonance has an increased risk of these types of complication.[16]

In our opinion, this should warn surgeons against considering the Shamblin classification as the ultimate tool to predict complications from a CBC resection. Therefore, facing two patients with large CBCs reaching the skull base, with one of the two patients having sustained major contralateral nerve and carotid damage, we chose to attempt a novel multidisciplinary approach. A double osteotomy of the mandibula to access the parapharyngeal structures was initially described by Seward[17] and later modified by Brusati et al,[18] who also emphasized the potential complications of this approach. We certainly recognize that this type of incision represents a very invasive approach that carries by itself the potential of permanent damage to nerves and of the generation of temporomandibular disorders. However, weighing out the risk of severe nerve or vessel damage associated with a standard surgical approach and the increased invasiveness of this multidisciplinary approach, we think that the latter was justified.

A wide spectrum of techniques have been described.[19-24] Anterior subluxation of the mandible with nasotracheal intubation improves distal exposure of the extracranial carotid artery and could represent an alternative approach to resect lesions that extend to the skull base.[25] We have seldom used nasotracheal intubation to perform the subluxation of the joint. This maneuver was performed only temporarily with retractors to gain a very distal clamping site. The resection of the styloid process has also been described to access this area; however, this technique seems to be burdened by a high number of complications. Sandmann et al[26] reported cranial nerve injuries in 21 of 29 patients who were treated using the detachment of the styloid process for carotid disease that extended to the skull base. In our division, we have no experience with this technique.

## CONCLUSIONS

The superiority of any of these techniques cannot be determined from a literature analysis; in fact, the small numbers in the reports, the wide spectrum of clinical scenarios, and the lack of a viable classification of the lesions that correlates to their extension and that reliably predicts the occurrence of surgical complications makes a comparison inconclusive. The multidisciplinary approach that we described may not find a widespread application in the treatment of CBCs but certainly offers a further option in order to favor a treatment tailored to each patient's clinical scenario.

The authors thank Dr Fabio Massimo Calliari for providing the drawings for this report.

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Submitted Jun 22, 2012; accepted Nov 18, 2012.