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Clinical study of preoperative angiography and embolization of hypervascular neoplasms in the oral and maxillofacial region

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Objective. The purpose of this study was to investigate the angiographic features of hypervascular head and neck neoplasms and to evaluate the effects of embolization on these lesions.

Methods. Angiograms and operation records of 25 patients with hypervascular neoplasms (23 neck paragangliomas, 1 hemangiopericytoma, and 1 hemangioendothelioma) were retrospectively analyzed, and the effects of 8 embolization procedures were estimated.

Results. Angiograms demonstrated that 23 neck paragangliomas (NPs) were manifested as richly vascularized lesions, and were divided into 3 types. Type I NPs (n = 2) were located cranial-laterally to the carotid bifurcation, and were removed with an intact carotid artery and injured vagus nerve. Type II (n = 17) lesions widened the bifurcations in 16 of 17 cases. During the operation, the continuity of the internal carotid arteries was preserved in 15 of 17 cases. Type III (n = 4) lesions enveloped the carotid bifurcations, and were removed together with the bifurcations in 3 of 4 cases. In 6 type II paraganglioma cases intra-arterial embolization was employed and the paragangliomas were removed with less blood loss (238 mL) than the nonembolized group (600 mL). Additionally, the infratemporal hemangiopericytoma and the parotid hemangioendothelioma were embolized and removed uneventfully.

Conclusions. Angiographic studies are highly valuable for the diagnosis and preoperative analysis of hypervascular head and neck neoplasms. Also, embolization therapy may minimize intraoperative blood loss. Both methods should be employed in a team approach to lesion treatment.

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Hypervascular neoplasms, including paraganglioma, angiofibroma, and other vascularized tumors, are rarely encountered by oral and maxillofacial radiologists or surgeons. Thus, accurate diagnosis and treatment may be problematic. These neoplasms are typically slow-growing, painless tumors that occur in the midcervical, nasopharyngeal, or other deep extracranial areas, and are closely associated with the carotid vessels and cranial nerves. The mainstay of treatment is complete resection; this, however, may induce severe complications such as uncontrollable bleeding, cranial nerve deficits, or cerebral ischemia. Several methods, such as neck ultrasonography (US), computed tomography (CT), and magnetic resonance imaging (MRI), are used to define the size, extent, and vasculature of the tumor. Despite mild invasion, superselective angiography provides an accurate arterial "map" and identifies the tumor's blood supply and flow dynamics. Additionally, selective arterial embolization may be performed concomitantly to minimize intraoperative blood loss.[1-3] All of these procedures afford surgeons the advantage of easier tumor removal with a decreased incidence of complications. In the present study, the angiograms and operation records of 25 patients with hypervascular neoplasms were reviewed retrospectively, and the effects of 9 embolization procedures were assessed.

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CLINICAL MATERIALS AND METHODS

From March 1987 to December 2004, 25 patients with hypervascular neoplasms were admitted for angiography and surgery at Peking University School of Stomatology. The group consisted of 23 patients with histologically proven neck paragangliomas (NP), 1 patient with hemangiopericytoma, and 1 patient with hemangioendothelioma. Of the 23 NP patients, 11 were male and 12 were female with a mean age of 38.8 years (range = 14-64 years). The NPs were clinically slow-growing and pulsatile cervical masses, with occasional pain; 7 of them were present with bruits and 1 evidenced Horner's syndrome and dysphasia. Thirteen patients underwent CT scanning. The patient with hemangiopericytoma presented a parapharyngeal mass with nasal

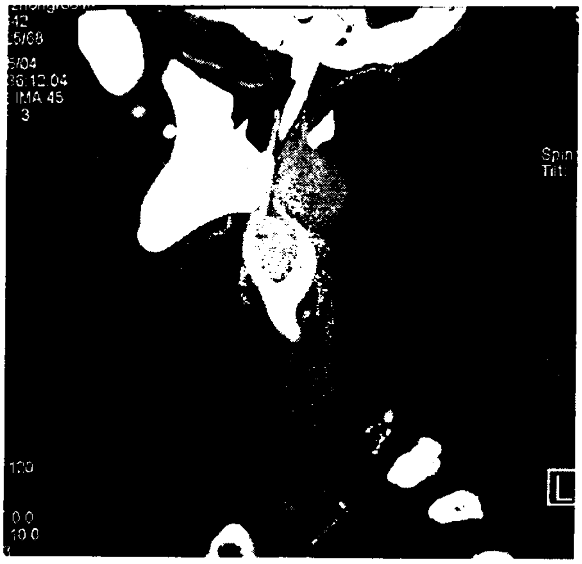

Fig. 1. Sagittal CT of a 36-year-old man with a midcervical mass showed 2 tumors, 1 splaying the carotid bifurcation and the other locating cranially to the bifurcation.

Fig. 2. Axial CT of a 50-year-old woman exhibited an irregular mass in the left deep parotid lobe, as well as a smaller mass on the right neck.

obstruction and decreased hearing. The patient with hemangioendothelioma was referred to our hospital for a large parotid mass. This patient had recently undergone surgery, however the operation was unsuccessful due to severe bleeding.

All patients underwent bilateral carotid arteriography via a transfemoral route under local anesthesia. Tolerance to a possible interruption of internal carotid blood flow was assessed by the angiographic demonstration of excessive cross-filling in the Circle of Willis. Eleven patients with NP, 1 with hemangiopericytoma, and 1 with hemangioendothelioma had undergone superselective external carotid arteriography, and the angiograms were carefully analyzed to detect extra-to-intracranial anastomosis. Nine of these patients experienced selective embolization with Ivalon (polyvinyl alcohol [PVA], sized at 250-350 μm) and Gelfoam particles, via 4 French catheters. The following results of the angiography were reviewed by 2 experienced oral and maxillofacial radiologists: tumor vasculature and feeding branches, the correlation of tumor stains to the carotid bifurcation, enlargement of the bifurcation, displacement of the external carotid artery (ECA) or internal carotid artery (ICA), invasion of the ICA, and the effects and side effects of embolization.

Before surgery, the Matas Test was performed on all patients, and this was verified to last at least 30 minutes. Twenty-three patients with NPs were operated on using a transverse cervical skin incision. The common carotid artery (CCA) was first dissected free and was surrounded by a vessel loop; next, the distal part of the ICA was similarly isolated, if possible. After identifying and preserving the adjacent cranial nerves and separating the tumor from the surrounding tissues, the tumor was removed from the carotid system periadventitially. The main trunks of the carotid arteries, as well as the ECA branches, were preserved whenever possible. The following results were reviewed: preservation of the ICA, ECA, and bifurcations, repair of the ICA or bifurcations if injured, cranial nerve or cerebral deficits, and intraoperative blood loss. Student *t* test was performed to compare the blood loss between embolized and nonembolized cases. The hemangiopericytoma was resected via a submandibular approach, thereafter the mandible was cut off to allow full exposure. The hemangioendothelioma was removed through an enlarged "S" incision routinely used for a parotid mass. The intraoperative blood loss and possible complications were registered.

RESULTS

Appearance of CT images

CT results for 11 of 13 NPs showed well-defined masses beneath the sternocleidomastoid muscle with a size range of 3 to 8 cm (average = 5.2 cm). The masses were evenly enhanced after intravenous injection of contrast media. One of 13 NPs exhibited enhanced double masses within the left cervical sheath. In this case, the relationship between the tumor and the carotid artery was clear, specifically, the bifurcation was enlarged, with the ICA displaced posterior-laterally and the ECA anteriorly (Fig. 1). However the arterial feeders of the carotid artery were not identified.

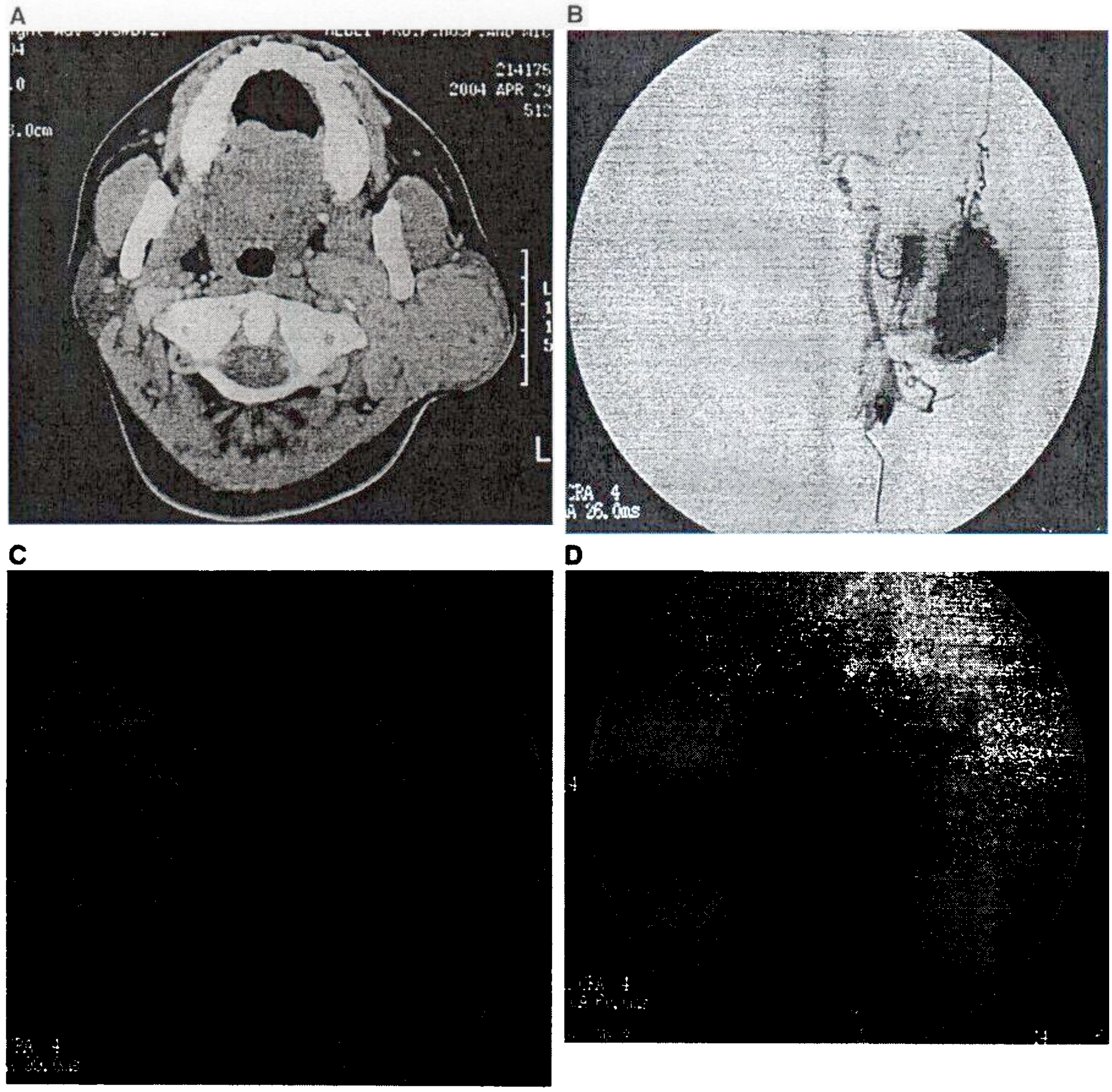

Fig. 3. **A**, Axial CT of a 40-year-old woman showed an irregular large mass involving the parotid and parapharyngeal space. **B**, Left common carotid arteriography, frontal view, showing the large tumor blush at the parotid area. **C**, Lateral view of the same arteriography. **D**, The tumor disappeared after PVA and Gelfoam embolization through the posterior auditory artery and small feeders of the ECA trunk.

Enhanced CT scanning demonstrated that the other NP case had an irregular mass of the left deep parotid lobe. Additionally, a smaller mass was concomitantly observed on the right neck, allowing consideration of bilateral NPs (Fig. 2). The hemangiopericytoma was an enhancing well-defined mass that involved the parapharyngeal and infratemporal space, and the hemangioendothelioma exhibited an unevenly enhanced irregular mass that involved the parotid and parapharyngeal space (Fig. 3).

Appearance of Angiograms and effects of embolization

Twenty-three NPs exhibited dense tumor stains of the upper neck, which was divided into 3 types.

- Type I (n = 2): the tumor stains located cranial-laterally to the carotid bifurcation, and both ICA and ECA were displaced anterior-medially. The feeders were solely from ECA. An embolization attempt was abandoned in 1 NP case due to identification of occipital to vertebral anastomosis.

Table I. Results of angiography and surgery of 17 patients with type II paragangliomas

		Angiography						
No.	*Sex/age*	*Bif.*	*ICA*	*ECA*	*Embolized vessels*	*Records of operation*	*Blood loss (mL)*	*CN deficits*
1	F/52	E	PL	A	No	ICA preserved, ECA dissected, Bif. repaired	800	X, XII
2	F/46	E	PL	A	No	ICA preserved, ECA ligated	500	No
3	M/42	E	PL	A	APA	ICA/ECA preserved	300	No
4	F/42	N	PL	En.	STA	ICA preserved, ECA ligated	150	No
5	F/50	E	PL	En.	No	ICA preserved, ECA dissected	900	XII
6	F/42	E	PL	A	APA	ICA preserved, ECA dissected, Bif. repaired	250	No
7	M/44	E	PL	A	APA	ICA/ECA preserved	250	No
8	M/44	E	PL	A	No	ICA/ECA preserved	350	No
9	M/33	E	PL	En.	No	ICA preserved, ECA dissected, Bif. repaired	900	No
10	F/23	E	PL	En.	No	ICA preserved, ECA dissected, Bif. repaired	1000	No
11	M/13	E	PL	A	No	ICA dissection and anastomasis to CCA	400	XII
12	F/53	E	PL	A	No	ICA/ECA preserved	100	No
13	M/23	E	PL	En.	No	ICA dissection and anastomasis to CCA	1000	X, XII
14	M/59	E	PL	A	No	ICA preserved, ECA dissected	150	No
15	M/36	E	PL	A	No	ICA preserved, ECA dissected, Bif. repaired	500	No
16	M/26	E	PL	A	APA, FA, STA	ICA/ECA preserved	400	X, XII
17	F/42	E	PL	A	OA	ICA/ECA preserved	80	No

CN, Cranial nerve; *Bif.,* bifurcation; *ICA,* internal carotid artery; *ECA,* external carotid artery; *F,* female; *M,* male; *E,* enlargement; *N,* not enlarged; *PL,* posterior-lateral displacement; *A,* anterior displacement; *APA,* ascending pharyngeal artery; *En.,* enveloped; *STA,* superior thyroid artery; *CCA,* common carotid artery; *FA,* facial artery; *OA,* occipital artery.

- Type II (n = 17, Table I): the tumors located at the bifurcations and enlarged the bifurcations in 16 of 17 cases. The ICA didn't have feeding branches and was displaced posterior-laterally in all cases. One of the tumors displaced the ECA anteriorly and was fed mainly by a branch originating from the bifurcation An embolization attempt was stopped because the 4F catheter could not be advanced far enough to avoid reflux into the internal carotid artery. Five of the 17 cases enveloped the ECA and were fed by ECA branches. Eleven displaced the ECA anteriorly and were fed by multiple ECA branches. Six of the 17 cases underwent embolization: 3 by the ascending pharyngeal artery (APA), 1 by the superior thyroid artery (STA), 1 by the occipital artery and the other by the APA, STA, and facial arteries. Despite mild pain, the embolization procedures were uneventful and tumor blushes nearly disappeared in all cases (Fig. 4).
- Type III (n = 4): with the average size of 7.5 cm (range = 6 to 8 cm), the tumors encapsulated the ICA, ECA, and carotid bifurcations in all cases (Fig. 5), and were fed by ECA branches as well as feeders from the ICA trunk. One patient developed blindness soon after angiography, and a follow-up angiography confirmed occlusion of the ICA at the narrow segment. One patient experienced decreased vision during embolization through the ECA trunk, and angiography revealed that the ophthalmic artery was partially occluded, probably from anastomosis to the middle meningeal artery. Following treatment, the vision of the 2 patients was not improved.

In all 23 patients, the Circle of Willis was patent and excessive cross-filling was observed via angiography.

The hemangiopericytoma exhibited dense tumor blush in the infratemporal space and was successfully embolized through feeders of the internal maxillary and ascending pharyngeal artery. The hemangioendothelioma, which was highly vascularized on angiogram, disappeared after embolization of the posterior auditory artery and small feeders of the ECA trunks (Fig. 3, *B-D*).

Treatment outcomes

In the 2 cases with type I NPs, the tumor was removed completely with preservation of the carotid arteries; however, the vagus nerve was encapsulated by the tumor and injured. The intraoperative blood loss was 200 mL and 800 mL, respectively. At discharge, 1 patient had dysfunction of cranial nerve X and XII, and the other had a vagus nerve deficit. Six of 17 type II NPs (Table I) were removed with intact ICA and ECA. Nine of the 17 NPs were removed with preservation of the ICA, however the ECA was ligated (n = 2) or dissected (n = 7). Additionally, 5 of the 9 NPs underwent suture repairs of the involved bifurcations. Two of the 17 NPs were removed together with the involved bifurcations, and an end-to-end anastomosis between the ICA and CCA was performed. At discharge, 3 patients developed deficits of cranial nerve X and XII and 2 had deficits of cranial nerve XII. In the embolized group, the surgeons had an impression that the tumor was shrunken and easily removed from adjacent structures. The average blood loss of the embolized cases (n = 6) was 238 mL, which

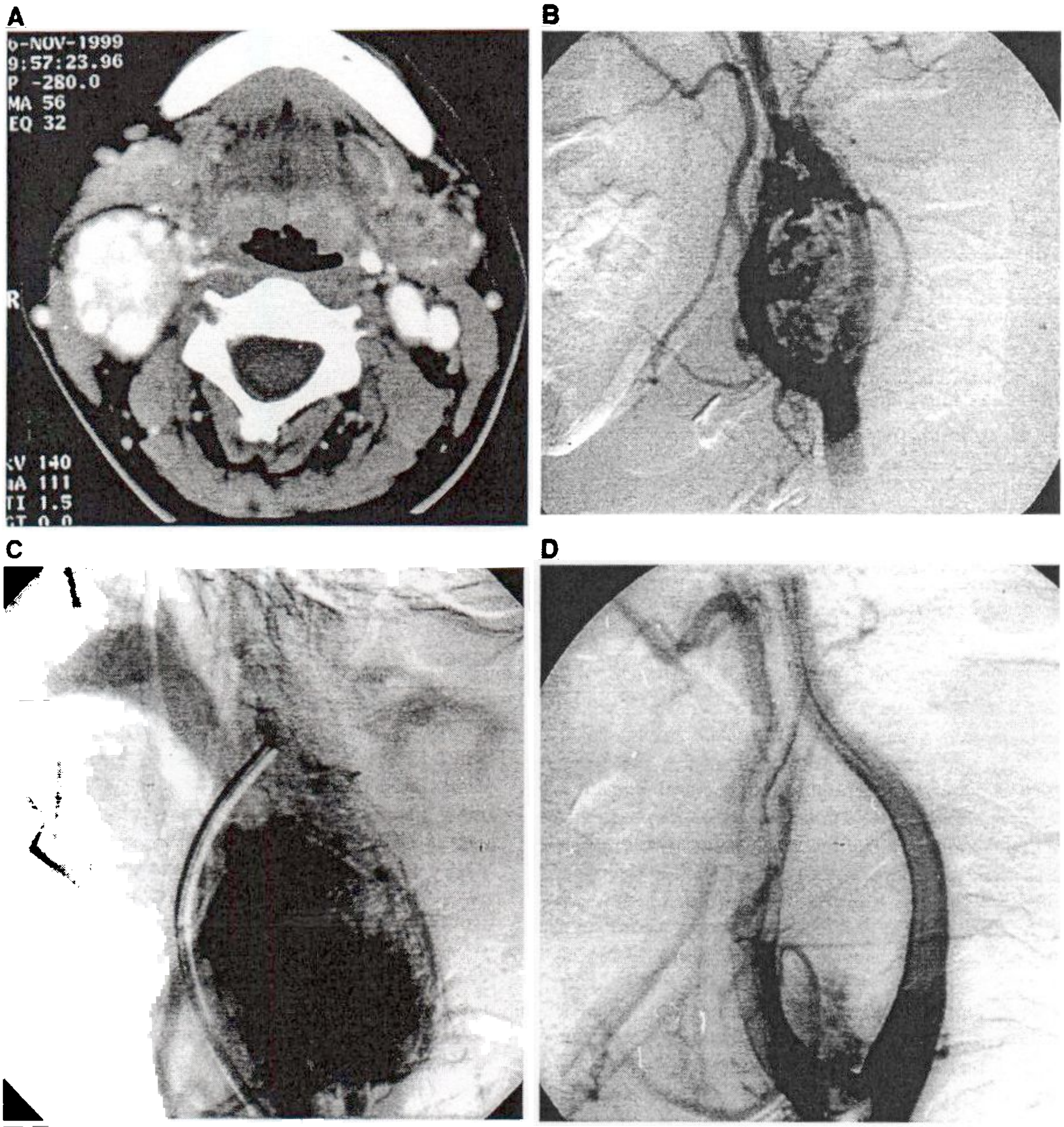

Fig. 4. **A**, Axial CT showed a highly enhanced tumor in the right neck sheath of a 42-year-old woman. **B**, Right common carotid arteriography, lateral view, showed the hypervascular mass typically widening the carotid bifurcation. **C**, Superselective arteriography of the hypertrophied ascending pharyngeal artery with a 4F catheter. **D**, Right common carotid arteriography after embolization with PVA through the APA confirmed subtotal disappearance of the tumor stain; the small residual blush was from branches arising from the bifurcation.

was significantly less than that of the nonembolized group (600 mL, n = 11, $P = .022$). In the type III NP group, the average blood loss was 1100 mL. One of them was removed together with the involved ICA and ECA segments and an end-to-end anastomosis between ICA and CCA was performed. This patient had deficits of cranial nerve X and XII, but did not have episode of cranial ischemia. One NP was removed along with the bifurcation, whereas ICA reconstruction was not performed after the mean distal stump pressure of the ICA was verified to be higher than 60 mm Hg. This patient experienced central facial paralysis, but

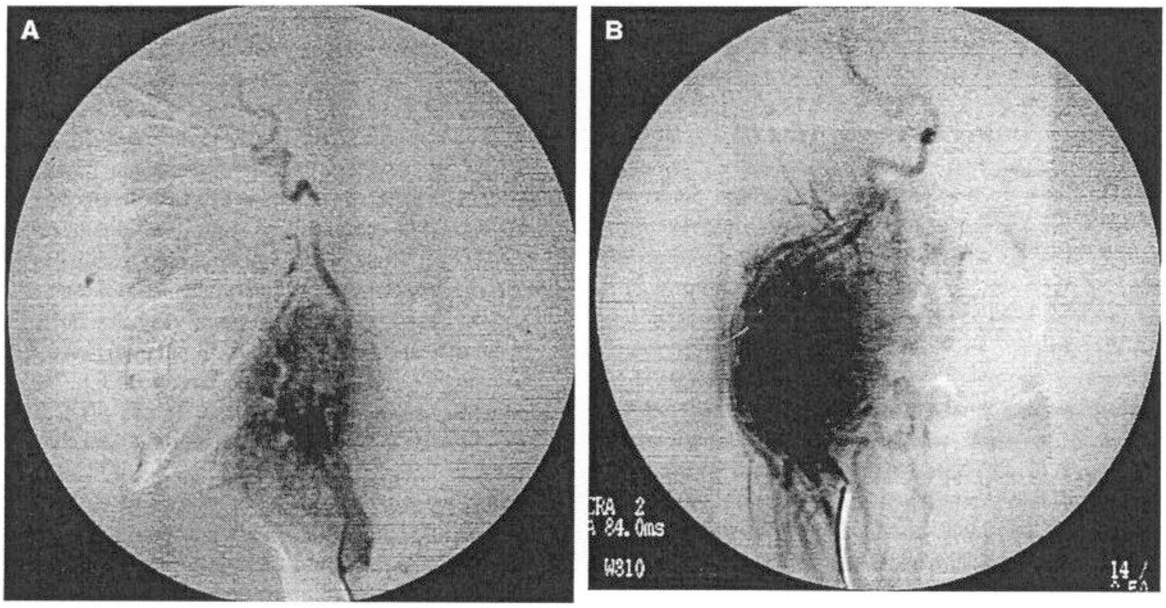

Fig. 5. **A**, Right common carotid arteriography in a 38-year-old woman, lateral view, showed a huge vascularized tumor enveloping the carotid bifurcation. **B**, Frontal view of the right common carotid arteriography, the main part of huge tumor located medially to the carotid artery.

recovered within 1 month and was well for 2 years despite a cranial nerve XII deficit. One large NP was solely dissected partially due to excessive bleeding, however this patient was lost to follow-up. The last one was removed along with the carotid bifurcation; this patient presented with multiple lung metastases at a subsequent follow-up, which resulted in the diagnosis of malignant paraganglioma.

The large hemangiopericytoma that involved the skull base was completely removed with 800 mL blood loss, and the carotid artery and cranial nerves were not involved. No recurrence was detected during 2 years of follow-up. The hemangioendothelioma was removed along with the involved facial nerve trunk. Also, major auditory nerve grafting was conducted. The intraoperative blood loss was 300 mL and no recurrence was detectable at 6 months.

DISCUSSION

Paragangliomas represent highly vascular tumors of neural crest origin that involve the walls of blood vessels or specific nerves within the head and neck. They may be defined as carotid body tumors, jugular paragangliomas, vagal paragangliomas, or tympanic paragangliomas, according to the site of origin. These tumors have multicentric diseases, which may include bilateral and unilateral multiple lesions. Malignancy occurs in 6% to 10% of the tumors, which is confirmed by the presence of metastatic lymph nodes or systematic metastases, not by the histologic appearance.[1,4,5] In our series, 2 cases (8.7%) had multicentric diseases, and 1 was considered malignant (4.3%). Two other vascularized tumors were described in this study, including one hemangiopericytoma and one hemangioendothelioma, which were only scarcely reported in the literature. Early detection and diagnosis of these tumor types is important, considering the surgical difficulties and risks. Doppler color-flow sonography is considered the first noninvasive imaging procedure in patients with suspected neck paragangliomas, due to its high sensitivity and specificity. CT scanning can delineate the relation of the tumor to adherent structures, in particular the skull base. MRI may better demonstrate the relation of the tumor to the adjacent internal jugular vein and carotid artery.[3,6] In our experience, the preferred diagnosis of a highly vascularized tumor in the neck sheath is paraganglioma. If the tumors are located in a deep extracranial space, they may be suspected as paragamglioma, angiofibroma, metastasis, or other vascularized tumors. DSA provides further verification of the diagnosis and provides a dynamic vascular map of the tumor, and should be used routinely for preoperative evaluation.

In this study, neck paragangliomas were divided into 3 types according to the relation of the tumor stain to the carotid artery on angiograms. Type I NPs were believed to be vagal paragangliomas, considering their relation to the carotid artery and cranial nerve X.[7] The arterial feeders were from ECA branches far beyond the bifurcation, and therefore facilitate superselective catheterization and embolization procedures. Type II NPs exhibited tumor blush just at the bifurcations, and displaced the ICA posterior-laterally. With the exception of 1 case, the bifurcations were enlarged and the ECA moved anteriorly, probably because the carotid body was located dorso-medially in the adventitial tissue of the bifurcation. Additionally, the ICA was preserved during operation in 15 of 17 cases. The ICA of type III NPs was sacrificed in 3 of 4 cases, and the other type III NP was only removed partially. Based on this, we believed that the ICA of type II NPs should be protected and preserved during operation, whereas for type III NPs, the ICA should be dissected together with the tumor if tolerance to carotid occlusion was verified.

A controversy exists concerning the size of tumors that should be embolized preoperatively, the embolic agents, and whether embolization is an alternative to surgery. Some are reluctant to embolize neck paragangliomas, because of the probability of complications.[1,3] Despite this, numerous teams believe that preoperative embolization may decrease intraoperative bleeding and facilitate surgery, especially for tumors larger than 3 cm in diameter, and the complication of this procedure is acceptably low. In this study, 6 cases with type II NPs were successfully embolized, and the intraoperative blood loss was significantly lower that the nonembolized cases. However, 2 patients with type III NPs experienced inadvertent embolization of the ophthalmic artery. One of these occurred after embolization of the ECA trunk and the other occurred after angiography. Considering the complications and risks, embolization should only be performed in the vessels that can be superselectively catheterized and determined not to allow free reflux of the contrast media into the internal carotid artery. Additionally, distal migration of emboli into the ophthalmic artery and central nervous system should be avoided.[8] We found that PVA, of varying particle diameters, and Gelfoam were safe and effective emboli agents. n-butyl-2-cyanoacrylate, more permanent polymerizing glue, must be delivered to the tumor through superselective catheterization with microcatheters. Casasco et al., Chaloupka et al., and Liang et al. reported successful devascularization of craniofacial tumors by percutaneous intratumoral injection of n-butyl-2-cyanoacrylate or ethanol.[9-11] We believe that neck paragangliomas are commonly amenable to conventional embolization and are thereafter removed safely. This works particularly well with the use of microcatheters. Intratumoral injection of glue may make the tumor adhere to adjacent tissues, precluding the intraoperative preparation of the tumor. Therefore, this technique should be reserved for huge paragangliomas or those involving the skull base that are unsatisfactorily embolized by an intraarterial approach.

We stress that the evaluation of the Circle of Willis is mandatory if the tumor encapsulates or invades the internal carotid artery. Many methods of determining tolerance to occluding carotid circulation have been described, including measurement of distal carotid stump pressure, Xenon CT scanning, EEG monitoring, and internal carotid balloon occlusion.[5,12] We rely on angiographic evidence of adequate contralateral cerebral blood flow, which is further confirmed by the preoperative Matas Test. Twenty-five patients in our study demonstrated evidence of tolerance to possible carotid clamping or sacrifice, via angiography. Four cases tolerated sacrifice of the involved ICA and bifurcations with (n = 3) or without (n = 1) reconstruction of the internal carotid circulation, and no signs of postoperative cerebral ischemia developed. One patient developed transient facial paralysis after ICA sacrifice. Internal carotid artery grafting was not performed at all, considering the reported 9.7% rate of strokes and a 2.4% incidence of mortality.[5]

In conclusion, employing a series of imaging techniques is optimum for safe and effective treatment of hypervascular head and neck neoplasms. Ultrasonography, enhanced CT scanning, and/or MRI confirm early screening of their high vascularization. Preoperative angiography further verifies the diagnosis of these tumors and provides important information concerning tumor blood flow and intracerebral circulation. Preoperative embolization decreases tumor blood flow and facilitates tumor removal. Additionally, meticulous vascular surgical or interventional radiological technique, careful perioperative nursing care, regular follow-up, and rehabilitation of any cranial nerve deficits are important factors in the treatment regimen.

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