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Preoperative embolisation of head and neck paragangliomas — a single-centre experience

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ABSTRACT

Introduction. Paragangliomas are neuroendocrine tumours commonly located in the abdomen, thorax, head and neck. The definitive treatment for these tumours is surgical resection, which in some cases can be very challenging due to the involvement of critical neurovascular structures and their high vascularity. Therefore, pre-operative embolisation may be performed to reduce the risk of complications. This study aimed to present our experience with endovascular embolisation of head and neck paragangliomas (HNP).

Material and methods. In this single-centre study, we reviewed data from consecutive patients with HNP who underwent pre-operative embolisation from 2017 to 2023. The efficacy of embolisation, the method of embolisation, as well as the rate of complications, were noted.

Results. A total of 27 patients (15 females) with an average age of 47 years underwent selective embolisation of HNP. Satisfactory embolisation, defined as occlusion of > 75% of the blood supply, was achieved in 22/27 cases (81.5%). The most commonly used embolic agents included coils and microspheres. With the exception of minor vessel dissections in two patients and embolic agent migration in two patients causing reversible occlusion of the intracranial vessels, there were no other complications associated with embolisation. No neurological deficits occurred in relation to the endovascular procedure.

Conclusions. The results of our study indicate that endovascular embolisation of HNP prior to surgical resection is a safe and efficacious procedure, with a relatively low complication rate and associated morbidity.

Keywords: head and neck tumours, paraganglioma, endovascular, embolisation

Introduction

Paragangliomas are rare, mostly benign, and highly vascular neuroendocrine tumours of the autonomic nervous system [1]. The most common locations of paragangliomas include the abdomen, thorax, head and neck [2]. Functionally, they can be divided into parasympathetic and sympathetic tumours [3]. Whereas the former are usually asymptomatic, hormonally inactive, and located mostly at the skull base (distribution of the IXth and Xth cranial nerves), the latter are highly active, symptomatic, and are found more often in the abdominal and pelvic region [4]. Both CT and MRI play important roles

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in the diagnosis of paragangliomas, the assessment of the extent, and the relationship to other structures, as well as for pre- and post-treatment evaluation and therapy responses [5]. Regardless of the location and hormonal activity, over 90% of cases are benign lesions, and total resection is considered the primary and definitive treatment [6].

Head and neck paragangliomas (HNP) include carotid body (the most frequent), jugulo-tympanic, and vagal tumours [7, 8]. They are generally benign and asymptomatic, but larger tumours can cause symptoms such as syncope, pulsatile tinnitus, hearing loss, dysphagia, or hoarseness of voice that result from progressive cranial nerve dysfunction [9]. Surgical treatment is challenging due to the high vascularity of the lesion and the involvement of critical neurovascular structures, which carry the risk of significant intraoperative blood loss, cranial nerve paralysis, and ischaemic events [10]. Therefore, some centres use pre-operative embolisation of the tumour, with the aim of reducing blood loss during surgery [11, 12]. It has been demonstrated that besides blood loss reduction, operations are shorter, and embolisation does not increase the risk of serious complications. On the other hand, other authors have failed to show any advantages of endovascular treatment prior to surgical resection [13].

This study aimed to present our experience with endovascular pre-operative embolisation of HNP with the goal of adding to the ongoing debate regarding their impact on surgical resection of these tumours.

Clinical rationale for study

Pre-operative embolisation of highly vascular head and neck tumours is a well-established method aimed at reducing intraoperative blood loss that is routinely performed in many centres worldwide. However, as far as the endovascular treatment of HNP is concerned, reports on its efficacy, safety, and impact on surgical resection are not unanimous. In our study, we present a single-centre experience with particular attention paid to the technical details and clinical outcomes. We also discuss the available literature.

Material and methods

Study participants

This retrospective and single-centre study was designed to evaluate the procedural and clinical outcomes of pre-operative endovascular embolisation of HNP in patients who underwent surgery from 2017 to 2023. The study was approved by local institutional review boards, and was conducted in compliance with the Declaration of Helsinki. Written informed consent for the procedure was acquired the day before from each patient. Inclusion criteria were: (a) age ≥ 18 years; (b) HNP diagnosis based on clinical history and computed tomography and/or magnetic resonance imaging, which was further confirmed by digital subtraction angiography (DSA); (c) surgery performed 24–48 hours after the endovascular embolisation; and (d) postoperative histopathological confirmation of the paraganglioma diagnosis. Exclusion criteria were: (a) previous surgical treatments of the lesion, and (b) patients with temporal bone paragangliomas, Fisch type A or B. All patients were screened for endocrine activity before embolisation and surgery. Patients with active tumours received alpha-blockers at least two weeks before treatment. Medical records, including demographics (age and gender), preprocedural details (complaints, and site of the lesion), and imaging records (CT and/or MRI) were collected.

Paraganglioma classification

The Shamblin classification was used for carotid body paragangliomas, and the Fisch classification was used for temporal bone paragangliomas. All lesions were evaluated based on surgical and pre-operative contrast-enhanced imaging (CT and/or MRI).

Endovascular procedures

All interventions were performed by interventional neuroradiologists with more than five years of experience in endovascular embolisation. Procedures were conducted as inpatients in the angio suite, via monolateral femoral access, and under local anaesthesia. Selective DSA of the common, internal, and external carotid arteries, as well as the vertebral arteries, were acquired to visualise arterial feeders.

A balloon test occlusion (BTO) was performed using an occlusive balloon, which provided temporary (i.e. up to 15 minutes under neurological surveillance) occlusion of the internal carotid artery (ICA) after the administration of heparin (5,000 units administered intravenously once the vascular sheath was placed in the common carotid artery, immediately before the BTO).

Afterwards, under general anaesthesia, superselective injections were carried out depending on the feeder's origin, with an endovascular strategy customised on a case-by-case basis depending on the angioarchitecture. The presence of a dangerous anastomosis between the extra- and intracranial circulation was carefully assessed. Endovascular embolisations were achieved; the type of microcatheter and embolic used depended on local availability and operator preference (Fig. 1).

In feasible cases, the direct puncture technique was applied, as shown in Figure 2. In these cases, occlusive balloons were used again. The percutaneous approach was applied in cases of carotid body tumours located relatively superficially with a favourable relation to the carotid vessels and classified as Shamblin II or IIIA.

Technical success was defined as complete or near complete (i.e. occlusion of > 75% blood supply to the lesion) occlusion at the last DSA run, with noticeably slower blood inflow. In all cases, repeated ICA injections were performed

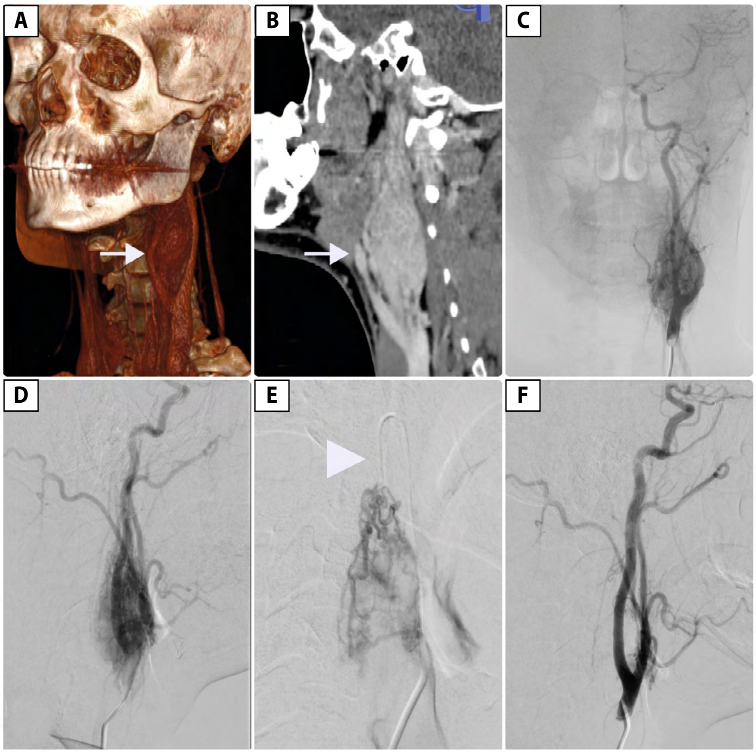

Figure 1. 19-year-old male patient diagnosed with left-sided carotid body paraganglioma (white arrow): **A** — CT 3D reconstruction, **B** — sagittal view of contrast-enhanced CT. Diagnosis was confirmed in DSA (**C**, **D**). Selective catheterisation and embolisation of arterial feeders was performed with particles (**E** — white arrow). Control DSA confirmed complete occlusion of lesion (**F**)

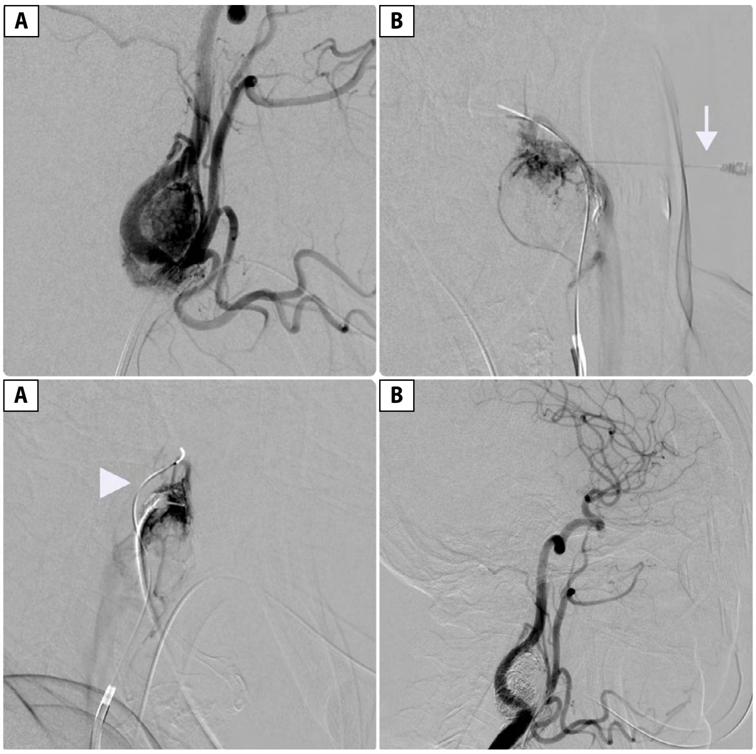

Figure 2. 61-year-old female patient diagnosed with left-sided carotid body paraganglioma (**A**) underwent pre-operative percutaneous embolisation with liquid embolic agent. Under US and DSA guidance, lesion was punctured (**B**) and embolisation was carried out with an inflated balloon preventing non-target migration (**C**). Control DSA confirmed complete occlusion of vascular blood supply to lesion (**D**)

to exclude potential complications (vessel occlusion and/or narrowing).

Intraprocedural complications were evaluated. Mechanical closure devices or manual compression of the site of the puncture were applied, based on the patient's vascular condition. AngioSeal (Terumo, Somerset, NJ, USA) or Perclose ProGlide (Abbott Vascular, Temecula, CA, USA) devices were used, with the latter being preferred in cases of narrower vessels and/or significant atherosclerotic changes. If deemed necessary, subsequent procedures were scheduled. Surgery was scheduled within 24-48 hours after the embolisation.

Surgical procedures

All surgeries were performed by experienced head and neck surgeons under general anaesthesia. In one case of carotid rupture during surgery, a vascular surgeon participated in the surgery. In cases with intracranial extension, surgery was performed in cooperation with a neurosurgeon.

Patients with temporal bone paragangliomas, Fisch type A or B, were operated upon without pre-operative embolisation, and were therefore not included in this study. Patients with Fisch type C and/or D tumours were operated upon using a subtemporal Fisch type A approach with anterior facial nerve transposition. The sigmoid sinus was routinely closed, and the jugular bulb was excised with the tumour. Intraoperative bleeding was controlled using Surgicel pads (Ethicon, Neuchatel, Switzerland) and/or Floseal hemostatic matrix (Baxter, US).

In patients with carotid paragangliomas, the tumours were excised after meticulous preparation of the tumour from the adventitia of the external, internal, and common carotid arteries after separation from the hypoglossal and vagal nerves.

Postoperative care included control of systolic blood pressure and pain and a postoperative clinical neurological evaluation. Patients after surgery usually stayed overnight in an observation room at the ENT Department, and none of them required treatment in the Intensive Care Unit. The diagnosis was confirmed by histopathological examination.

Results

Patient demographics and clinical characteristics

A total of 27 patients underwent pre-operative embolisation and surgical resection of HNP tumours between 2017 and 2023 in our centre. Fifteen of the patients were female (55.6%) and 12 were male (44.4%). The youngest patient was 19, while the oldest was 67, with a mean age of 46.8 ± 15.7. In terms of tumour location, 51.9% of the patients (14/27) had left-sided tumours, with 40.1% (11/27) of tumours being right-sided. Two patients (8.0%) had bilateral lesions.

Pre-procedural imaging findings and endovascular treatment

All patients underwent imaging examinations that included a CT and/or MRI. All paragangliomas were then confirmed using DSA. The large majority of lesions were supplied by ipsilateral branches of the external carotid artery (22/27, 81.5%). From this group, the main arterial feeders identified during diagnostic angiography were the ascending pharyngeal artery (23/27, 85.2%), the occipital artery (10/27, 37%), the superior thyroid artery (5/27, 18.5%), the maxillary artery (4/27, 14.8%), and the posterior auricular artery (2/27, 7.4%). Other blood supply originated from the ipsilateral internal carotid artery and the vertebral artery (two patients in each group, 14.5%). One lesion was supplied by branches of all three vessels (4%). The most commonly used embolic materials included microspheres, glue (N-butyl cyanoacrylate mixed with lipiodol), and coils. In three cases (11.1%), the direct puncture technique was applied. In these patients, embolisation with Squid (Balt, Montmorency, France) was performed.

In terms of periprocedural complications, two patients (7.4%) had minor vessel dissections. Both occurred during superselective catheterisation of the targeted vessel. Neither of them precluded the success of catheterisation and embolisation. Apart from this, two cases (7.4%) had an occlusion of the distal middle cerebral artery branches during the control DSA run. Both patients received 5 mg of glycoprotein IIb/IIIa inhibitor (Abciximab ReoPro; Eli Lilly and Co., Indianapolis, IN, USA). Complete recanalisation was noted in both cases. Every procedure was completed using a control head DynaCT performed on an angiographic table to exclude periprocedural intracranial haemorrhage.

The final DSA run disclosed complete or near complete embolisation (i.e. occlusion of > 75% of blood supply to the lesion) in 22 cases (81.5%). In five patients (18.5%), an occlusion of 50–75% of the tumour's vasculature was observed. Three patients (11.1%) underwent staged treatment due to the complexity of the vasculature (treatment involved 2-3 procedures). Demographic details, pre-procedural imaging findings, and endovascular treatment results are set out in Table 1.

Discussion

Pre-operative endovascular embolisation is widely used in a multidisciplinary approach to hypervascular head and neck tumours [14]. Its primary aim is to reduce perioperative blood loss and decrease morbidity and mortality. Although HNP are lesions that can benefit greatly from endovascular embolisation, the role of pre-operative embolisation and its impact on surgery remains a matter of ongoing debate [15].

The primary aim of our single-centre study was to evaluate the efficacy and feasibility of minimally invasive pre-operative embolisation of HNP in a consecutive group of patients. In

Table 1. Demographic details, pre-procedural imaging findings and endovascular treatment results

Patient	Age	Sex	Location*	Classification	Side**	Blood supply ***	Technique ****	Embolic material	% of occlusion	Complication
1.	67	Female	TBP	FISCH C1	R	ECA	Endo	NBCA*****	95	0
2.	41	Female	CBT	SHAMBLIN IIIA	R	ECA	Endo	Microspheres	50	0
3.	32	Male	TBP	FISCH C2	R	ECA	Endo	Microspheres	75	0
4.	24	Female	CBT	SHAMBLIN I	L	ECA	Endo	Microspheres	90	0
5.	65	Female	TBP	FISCH C2	L	ECA	Endo	Microspheres + NBCA	90	0
6.	56	Female	CBT	SHAMBLIN I	R	ECA	Endo	Microspheres	100	0
7.	19	Male	CBT	SHAMBLIN III	L	ECA	Endo	Microspheres + NBCA	100	0
8.	59	Female	CBT	SHAMBLIN II	R	ECA	Endo	Microspheres	75	0
9.	42	Female	TBP	FISCH C2Di1	R	ECA	Endo	Microspheres + coils	85	Non-target ICA embolisation
10.	22	Male	TBP	FISCH C2	L	ECA	Endo	Microspheres	80	0
11.	64	Female	CBT	SHAMBLIN IIIA	R	ECA + ICA	Direct	Squid	90	0
12.	61	Female	CBT	SHAMBLIN II	L	ECA + ICA	Direct	Squid	90	0
13.	35	Male	CBT	SHAMBLIN III	R	ECA	Endo	Microspheres + coils	100	Non-target ICA embolisation
14.	37	Female	TBP	FISCH C2	R	ECA	Endo	Microspheres + gel sponge	100	0
15.	39	Male	CBT	SHAMBLIN III and IIIA	Bilateral	ECA	Endo	Microspheres + coils	90	Feeding artery dissection
16.	41	Female	CBT	SHAMBLIN II	L	ECA	Endo	Microspheres	60	0
17.	69	Female	TBP	FISH C4Di2	L	ECA + VA	Endo	Microspheres + NBCA	50	0
18.	43	Female	TBP	FISH C1	L	ECA	Endo	Microspheres	85	0
19.	25	Male	TBP	FISH C1	L	ECA + ICA + VA	Endo	Microspheres	70	0
20.	64	Male	TBP	FISH C1	L	ECA	Endo	Microspheres	75	0
21.	58	Male	CBT	SHAMBLIN II	R	ECA	Endo	Microspheres + coils	50	0
22.	63	Male	TBP	FISH C2	L	ECA	Endo	Microspheres + gel sponge	75	ICA dissection
23.	52	Female	TBP	FISH C1	L	ECA	Endo	Microspheres + coils	75	0
24.	45	Male	CBT	SHAMBLIN II	Bilateral	ECA	Endo	Microspheres + coils	90	0
25.	68	Male	CBT	SHAMBLIN II	L	ECA	Direct	Squid	90	0
26.	39	Male	TBP	FISH C2	L	ECA	Endo	Microspheres	100	0
27.	30	Female	TBP	FISH C4Di2	R	ECA + VA	Endo	Microspheres + NBCA	80	0

*TBP — temporal bone paraganglioma; CBT — carotid body paraganglioma; **R — right; L — left; ***ECA — external carotid artery; ICA — internal carotid artery; VA — vertebral artery; ****Endo — endovascular embolisation; Direct — direct percutaneous puncture technique; *****NBCA — N-butyl cyanoacrylate our series, we observed a relatively high rate of satisfactory (defined as the exclusion of 75% of arterial supply) occlusion (22/27, 81.5%). Our results are in line with findings from a recent systematic review. In 2021, De Marini et al. evaluated 22 studies with over 300 HNP and reported a successful devascularisation rate of 79% [12]. In 2023, Schartz et al. published a meta-analysis that compared the two most common approaches to HNP embolisations (direct puncture and arterial embolisation), and observed a higher rate of technical success in the direct puncture group (91.5%) [16].

This observation led to another aspect of our analysis, namely the method of embolisation. Several authors have attempted to compare the two most commonly used techniques i.e. transarterial embolisation and the direct puncture technique. The main advantage of the former method over the latter is the wide variety of embolic materials (particles of various sizes, coils, N-butyl cyanoacrylate, and gelfoam) that enable the customisation of the therapy [17]. Although the navigation of tiny tumour vasculature via the transarterial approach is not easy, successful experiences with transarterial embolisation have been reported [18, 19]. Perhaps a combined transarterial and transvenous approach might be the solution in cases of particularly challenging vasculature, as described by Brahimaj et al. [20]. On the other hand, according to some authors, the direct puncture technique enables complete embolisation regardless of the lumen of the arterial feeders, and many centres have implemented this technique with a high rate of success [21–23].

In our centre, we mainly perform embolisation using an endovascular approach, as this method is preferred by head and neck surgeons. In their experience, the value of transarterial embolisation extends beyond the reduction of bleeding during surgery, especially in cases of jugular bulb paragangliomas, as the embolisation particles that are visible during the drilling of the infiltrated bone help in identifying the extent of the tumour within the bone and differentiate it from granulation or bone marrow tissue. On the other hand, stiffened casts of liquid embolic materials used during the direct puncture technique can impede the preparation of the tumour, according to some opinions. However, we did not find any relevant papers that could confirm these findings in the literature.

The result of the balloon occlusion test provides additional information for the surgeon. This test aims to evaluate the efficacy of the intracranial collaterals in maintaining perfusion of the affected vascular territory during temporary occlusion, and can influence the decision regarding the extent of tumour removal, internal carotid preparation, and the eventual ligation of both common carotid artery branches [24, 25]. Although there is no consensus on its standardised use in the pre-operative embolisation of HNP and its potential to increase the risk of vessel injury during balloon inflation [26], it is routinely performed prior to the embolisation procedure in our centre.

As far as the procedural complications are concerned, in our retrospective experience, we have noted two types of complications: non-target embolisation and vessel injury. Overall, the complication rate was 14.8%, which is higher than previously reported [12]. Although every procedure began with an initial DSA aiming to disclose potential anastomoses between the internal and external carotid arteries and its branches, all measures were undertaken to prevent the reflux of embolic particles during the embolisation. Occlusion of intracranial branches probably occurred through a small perforating artery unidentified on the preliminary diagnostic angiography. These events were noted during transarterial embolisation.

In our centre, we do not routinely use temporary balloon occlusion for transarterial embolisation, which has been reported to impede the accidental migration of the embolic material on the one hand, but has on the other hand been associated with an increased risk of injury to already weakened vessel walls [27]. Apart from this, migration of the embolic material has been reported despite the use of the balloon occlusion technique, which is why it is not currently recommended on a large scale [28, 29]. Fortunately, in both of the described cases, complete recanalisation was achieved after glycoprotein IIb/IIIa inhibitor therapy. Both patients recovered with no neurological deficits.

Similarly, both of the dissections were noted in patients treated using a transarterial approach. Nonetheless, in both cases, the injury was focal, non-flow-limiting with no further sequelae on consecutive DSA runs, and therefore the surgical resection proceeded as planned without complications.

Our study has certain limitations. First and foremost, it had a retrospective and single-centre design. Due to this fact, the precise data on surgical intra-operative blood loss is lacking and cannot be compared between the groups of complete and incomplete embolisation. Secondly, our sample was relatively small which limits the validation of the data. Finally, we were unaware of the potential complications occurring after 48 hours from the embolisation as the patients underwent surgical treatment.

Conclusions

Pre-operative embolisation continues to be a valuable tool in the management of patients with HNP. The results of our study indicate that it might be a safe and efficacious procedure with a relatively low complication rate and associated morbidity.

However, the final decision on the use of embolisation should be made having balanced the risk of complications against the advantages of reduced bleeding during the preparation of the tumour.

From our experience, we suggest that the decision on the treatment modality should be based on the location, stage, and type of the paraganglioma, as well as the age of the patient and the presence of any neural deficits or hearing impairments.

Clinical implications and future directions

Our single centre experience demonstrates that pre-operative minimally-invasive treatment of HNP enables complete or near-complete occlusion of the lesions in a majority of cases, is associated with an acceptable rate of complications, and can be performed in the staged treatment of these tumours. Future prospective studies are necessary to compare the two most commonly used approaches, i.e. percutaneous or combined, to improve the outcome and lower the risk of adverse events.

Article information

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