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Preoperative embolisation of carotid body paraganglioma

Przedoperacyjna embolizacja przyzwojaków tętnicy szyjnej

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S u m m a r y

Introduction. Paragangliomas are rare, slow growing neuroendocrine tumours that arise from the extra-adrenal autonomic paraganglia. In the head and neck region the dominating location is carotid bifurcation – carotid paraganglioma. Surgical excision is the most common treatment. Excessive vascularity resulting from marked hypertrophy and hyperplasia of blood vessels in the tumour besides common involvement of lower cranial nerves makes surgery demanding and difficult. Despite recent advances in surgical techniques and in the imaging diagnostics the intraoperative haemorrhage and risk of neurovascular complications remains considerable. Therefore preoperative endovascular embolisation is proposed to reduce intraoperative bleeding and aid excision. Preoperative embolisation benefits are still under dispute and due to the rarity of carotid paraganglioma experience in its surgical treatment is limited. Analysis of individual series carries an added value in promoting progress in this field.

Aim. The aim of this study is to review an experience of a single centre in the use of preoperative embolisation in resecting carotid paraganglioma with particular attention to the embolisation technique and its effectiveness.

Material and methods. This is a retrospective analysis of surgical treatment of carotid paragangliomas supported with preoperative tumour embolisation. It is based on four patients with pathologically confirmed carotid paraganglioma.

Family history, clinical presentation, imaging diagnostics, Shamblin classification, course and method of preoperative embolisation, surgical treatment, complications and the early outcome were reviewed. All lesions were represented by a painless cervical mass, without neurological symptoms. Imaging included computed tomography, magnetic resonance and magnetic resonance angiography. Tumour size and Shamblin group were assessed from the MR images. The tumours were located in the posteromedial wall of the common carotid artery at the carotid bifurcation. Three patients were Shamblin group II and one in group III.

Tumour vascularity and vessels feeding the lesion were identified on preoperative digital subtraction angiography of the common carotid artery in all patients. In patients consenting to surgery it was followed by tumour embolisation using superselective catheterisation and particle occlusion of he tumour-feeding vessels. Embolisation procedure is described in detail.

Results. Angiogram revealed a highly vascularised tumour mass displacing internal and external carotid arteries in all cases. The lingual and ascending pharyngeal arteries were the dominant feeding vessels to a carotid body tumours. No anastomoses between the branches of the external and internal carotid arteries were detected. The follow-up arteriogram showed complete tumour devascularisation with normal flow in the carotid arteries. There were no major complications of the procedure. During selective and super-selective catheterisation and embolisation no major technical difficulties were encountered. Complete subadventitial resection of the tumour was achieved in all patients. There was no operative mortality nor morbidity. Preoperative embolisation of the tumour reduced intraoperative bleeding facilitating dissection, reduced blood loss and time of surgery. Median intraoperative blood loss was 270 mL. There were no recurrences or delayed complications at the follow up of 24-76 months.

Conclusions. The results of the study confirm that the surgical treatment of carotid paragangliomas may benefit from embolisation aimed at decreasing tumour blood flow. Intraoperative tedious ligation of feeding arteries can be replaced by preoperative selective embolisation.

The adopted method of embolisation proved it to be safe and efficient. Review of the literature indicates that standard definitions and uniformity across reports would aid in establishing best practice measures.

S t r e s z c z e n i e

Wstęp. Kłębczak jest rzadko występującym, wolno rosnącym nowotworem neuroendokrynnym pozanadnerczowym wywodzącym się z autonomicznych zwojów nerwowych. W obrębie głowy i szyi najczęściej związany jest z tętnicą szyjną wewnętrzną i jej rozwidleniem – przyzwojakiem szyjnym. Bogate unaczynienie guza, jego związek ze ścianami tętnic szyjnych i obrastanie dolnych nerwów czaszkowych sprawiają istotne trudności przy ich operacyjnym usuwaniu. Postęp techniki operacyjnej i metod obrazowania nie wpłynął istotnie na zmniejszenie tych trudności, z których ryzyko krwawienia należy do najważniejszych. Zmniejszenie tego ryzyka upatruje się w przedoperacyjnej embolizacji naczyń nowotworu. Korzyści tego sposobu postępowania nadal stanowią przedmiot dyskusji. Z powodu rzadkości występowania liczba publikacji i doświadczenie poszczególnych ośrodków są ograniczone, dlatego analiza własnych doświadczeń poszerza postęp w tej dziedzinie.

Cel pracy. Praca ma na celu przegląd doświadczenia własnego z chirurgicznego usuwania przyzwojaków szyjnych po uprzedniej embolizacji przeznaczyniowej, a także szczegółową analizę metody embolizacji przeznaczyniowej.

Materiał i metody. Retrospektywna analiza przebiegu leczenia operacyjnego 4 chorych z histopatologicznie potwierdzonym przyzwojakiem szyjnym, u których wykonano przedoperacyjną embolizację guza. Analizowane dane dotyczą: historii rodzinnej, objawów klinicznych, wyników diagnostyki obrazowej, ryzyka operacyjnego według klasyfikacji Shamblina, przebiegu embolizacji i operacji oraz występowania powikłań we wczesnym okresie pooperacyjnym. Wszystkie guzy objawiały się jako bezbolesne masy na bocznej powierzchni szyi. Chorzy nie mieli zaburzeń neurologicznych. U wszystkich wykonano obrazowanie tomografią komputerową, rezonansem magnetycznym (MR) i angiografię rezonansową. Wielkości guza i kategorię w skali Shamblina określano na podstawie MR. Troje chorych miało guz w II stopniu Shamblina, a jedna chora w III stopniu. Unaczynienie guza i ustalenie naczyń go zasilających ustalono na podstawie cyfrowej subtrakcyjnej angiografii. Następnie chorzy mieli wykonywane superselektywne cewnikowanie naczyń zasilających i embolizację naczyń guza cząstkami Embozene®.

Wyniki. Angiografia wykazała bogato unaczynione guzy przemieszczające gałęzie tętnicy szyjnej wspólnej. Tętnice językowa i gardłowa wstępująca były głównymi tętnicami zasilającymi nowotwór. Nie ujawniono połączeń pomiędzy tętnicami szyjnymi wewnętrzną i zewnętrzną. Angiografia po embolizacji wykazywała zubożenie unaczynienia guza i pełną drożność tętnicy szyjnej. Nie zanotowano trudności technicznych ani powikłań w czasie i bezpośrednio po embolizacji. Guzy u wszystkich chorych zostały całkowicie usunięte podwyściółkowo. Nie obserwowano żadnych powikłań operacyjnych. Krwawienie śródoperacyjne było umiarkowane, średnio wynosiło 270 mL. U żadnego chorego nie wystąpiły nawrót ani odległe powikłania w okresie obserwacji pooperacyjnej od 24 do 76 miesięcy.

Wnioski. Wyniki badania potwierdzają przydatność przedoperacyjnej embolizacji w leczeniu przyzwojaków szyi. Zmniejszenie napływu krwi do guza ułatwia jego wydzielanie i zmniejsza utratę krwi. Embolizacja superselektywna naczyń przyzwojaka okazała się metodą skuteczną i bezpieczną. Przegląd piśmiennictwa wskazuje na konieczność standaryzacji pojęć używanych w opracowaniach z tej dziedziny, co pomoże w wypracowaniu najlepszych zasad postępowania z chorymi z kłębczakami szyi.

INTRODUCTION

Paragangliomas (glomus tumours) are rare, slow growing neuroendocrine tumours that arise from the extra-adrenal autonomic paraganglia. Malignant transformation does not exceed 10% of cases. Paragangliomas of the head and neck constitute a distinct group of those tumours, creating specific diagnostic and therapeutic challenges. They represent less then 0.5% of all head and neck tumours. They occur 3 times more often in females then in man 2/3 of them between 40 and 60 years of age. Around 25% are multifocal. Location of paragangliomas in the order of frequency is as follows: carotid body (chemodectoma) (60-67%), tympanicum in the middle ear, jugulotympanicum, jugulare, vagale (1, 2). Thus the dominating location is carotid bifurcation.

Paragangliomas are basically spontaneous, but may be associated with hereditary mutations in the genes encoding the succinate dehydrogenase (SDH) enzyme complex and a number of other susceptibility genes: *NF1*, *RET*, *VHL*, *SDHA*, *SDHB*, *SDHC*, *SDHD*, *SDHAF2* (*SDH5*), and *TMEM127* (3).

Surgical excision is the most common treatment. Excessive vascularity resulting from marked hypertrophy and hyperplasia of blood vessels besides common involvement of lower cranial nerves makes surgery demanding and difficult. Therefore preoperative endovascular embolisation may be used to reduce in-

traoperative bleeding and aid excision. Some tumours invade the walls of the carotid artery considerably reducing their resectability (4, 5). Radiotherapy is used for palliation of unresectable tumours (6).

Despite recent advances in surgical techniques and in the imaging diagnostics the intraoperative hemorrhage and risk of neurovascular complications remains considerable (7, 8).

In an attempt to reduce the vascularity of the tumour to decrease intraoperative bleeding and thus the technical difficulty preoperative embolisation is utilised, but its benefits are still under dispute (9, 10).

Due to the rarity of carotid paraganglioma experience in its surgical treatment is limited and series in the individual centres are small. Analysis of individual series carries an added value in promoting progress in this field.

AIM

The aim of the study is to review an experience of a single centre in the use of preoperative embolisation and its influence on the course of operation of carotid paraganglioma. Assessment of the embolisation technique and its effectiveness is another goal of the study.

MATERIAL AND METHODS

This retrospective analysis reports experience in surgical removal of carotid paragangliomas supported with preoperative tumour embolisation. Four patients were enrolled into the study: 3 women and 1 man with confirmed diagnosis of carotid paraganglioma. Their age ranged from 20 to 47 years (median 31).

The clinical records, radiological reports and images and pathological examinations of all patients provided data on the family history, clinical presentation, imaging diagnostics, Shamblin classification, course and method of preoperative embolisation, surgical treatment, complications and the early outcome were reviewed.

Family history was negative for cancer. All lesions were represented by a painless cervical mass, without neurological symptoms.

Imaging in all patients included computed tomography, magnetic resonance (MR) and magnetic resonance angiography (MRA). Tumour size and Shamblin group were assessed from the MR images. The tumours were located in the posteromedial wall of the internal carotid artery at the carotid bifurcation.

The size of the tumours was measured on MR images in the cranio-caudal and frontal and sagittal planes at the level of the maximum tumour extent. The cranio-caudal diameter did not exceed 67 mm, frontal 33 mm and sagittal 35 mm.

Surgical risk was assigned according to the Shamblin classification (11). Group I tumours are localised and do not involve the surrounding major vessels, in group II are adherent or partially surround the vessels and in group III at least one of the major vessels is completely surrounded or encased in the tumour. Three patients were classified to Shamblin group II and 1 to Shamblin group III.

Tumour vascularity and vessels feeding the lesion were identified on preoperative digital subtraction angiography of the internal carotid artery in all patients. In patients consenting to surgery it was followed by tumour embolisation accomplished in the Department of Interventional Radiology and Neuroradiology.

Carotid paraganglioma diagnosis was confirmed on histopathology and immunohistochemistry.

Method of embolisation

In all patients embolisation procedure followed angiography. Under local anaesthesia common femoral artery was punctured in the groin with the use of Seldinger technique. The common carotid artery was accessed with guiding catheter 6F MPC Envoy® ensuring stable position. Through the guiding catheter a selective catheter was introduced to the distal aspect of the common carotid artery and 10 ml of contrast media (Visipaque 320® GE HealthCare) was injected. Selective digital subtraction arteriography of the internal and external carotid artery with emphasis on defining the dominant feeding arteries to the tumour was performed. Multiple views with magnification were helpful. Access to the feeding arteries was achieved with using microcathethers. Selective injection of the contrast media showed rich pathological vascularity of the tumours. Embolisation of the feeding vessels was performed with the use of microparticles 250-500 μm (Embozene®). Contrast was added to particles to help image the pathway of flow. Embolisation was completed by a control common carotid artery angiography.

Surgical resection was performed within 48 hours of embolisation.

Under general anaesthesia tumours were exposed through a transverse cervical incision.

All neurovascular structures were identified and periadventitial dissections of the carotid artery were performed. Proximal and distal control of the internal carotid artery (ICA) and external carotid artery (ECA), and common carotid artery (CCA) were performed before tumour resection. Any feeding vessels supplying the tumour were ligated early.

RESULTS

Angiogram of a carotid body tumour revealed a highly vascularised mass displacing internal and external carotid arteries, splaying of the carotid bifurcation (fig. 1). The lingual and ascending pharyngeal arteries were the dominant feeding vessels to a carotid body tumours (fig. 2, 3). No anastomoses between the branches of the external and internal carotid arteries were detected. The follow-up arteriogram showed complete tumour devascularisation with normal flow in carotid arteries (fig. 4). In all four cases complete occlusion of the arteries feeding the tumour has been achieved. There were no major complications of the procedure. In one patient a local subcutaneous haematoma in the site of percutaneous catheter introduction occurred. It resolved spontaneously. During selective and super-selective catheterisation and embolisation no major technical difficulties were encountered. This has been attributed to the use of microcatheters and experience of the operator.

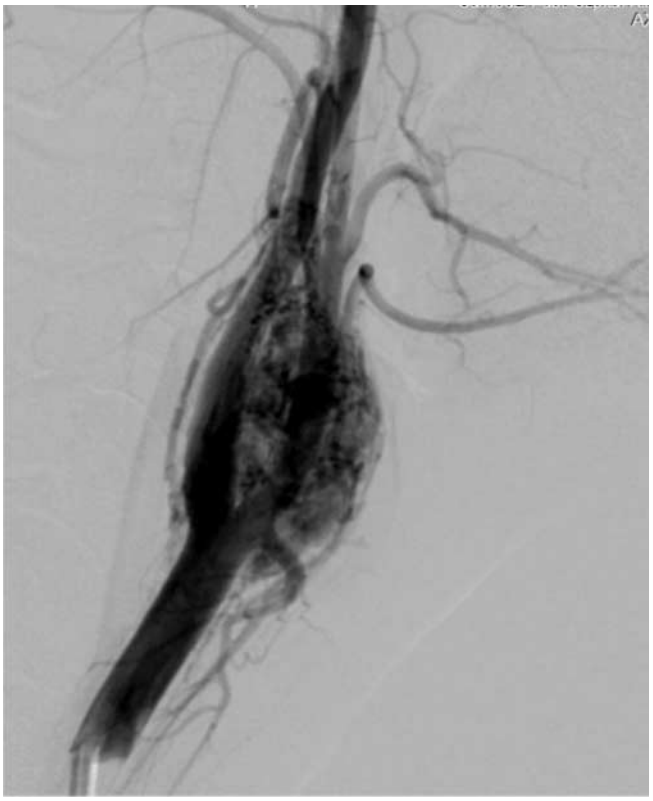

Fig. 1. Arteriography of the common carotid artery. Multiple pathological arteries at the level of carotid artery division. Highly vascularised mass is splaying internal and external carotid arteries

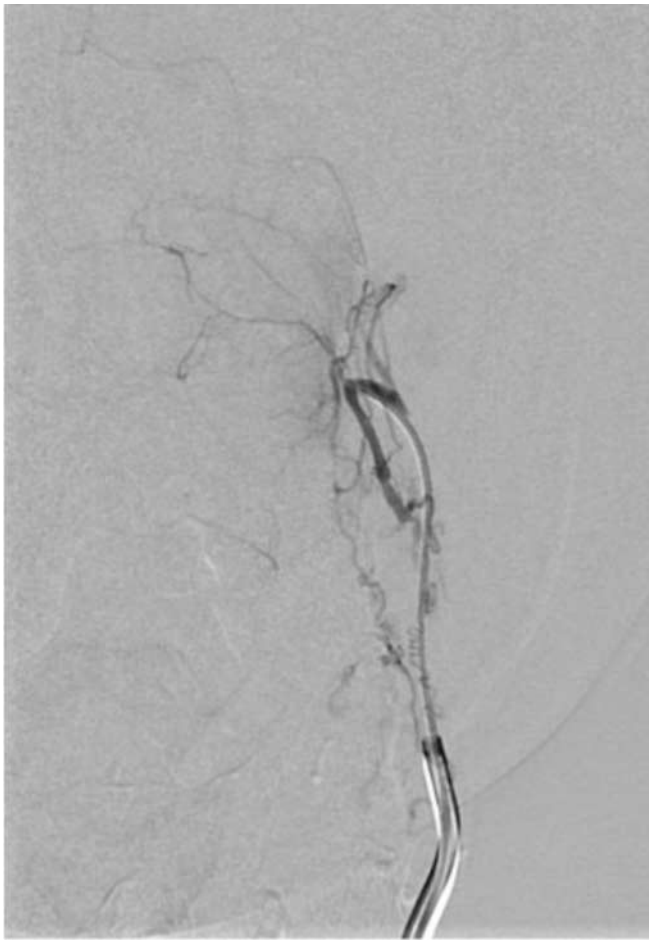

Fig. 2. Selective catheterisation of the ascending pharyngeal artery

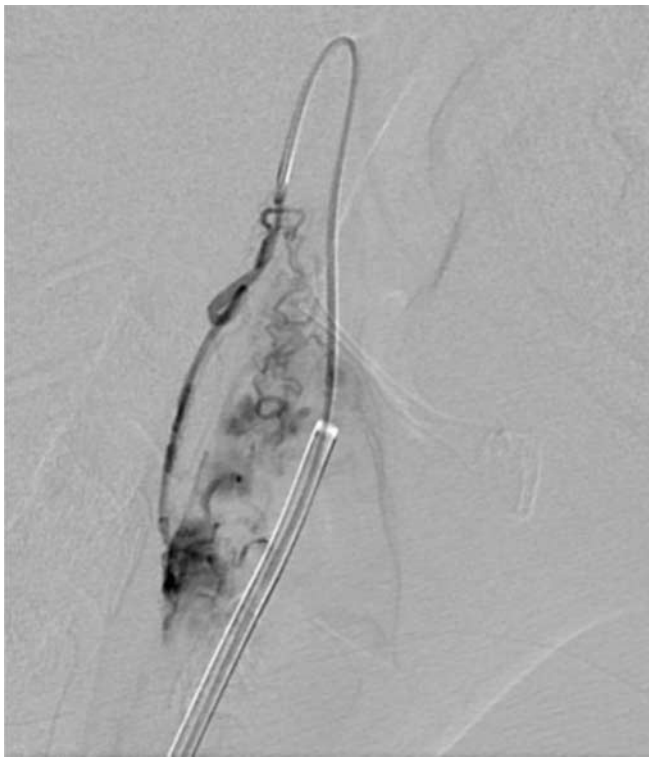

Fig. 3. Superselective catheterisation of the branches of the lingual artery

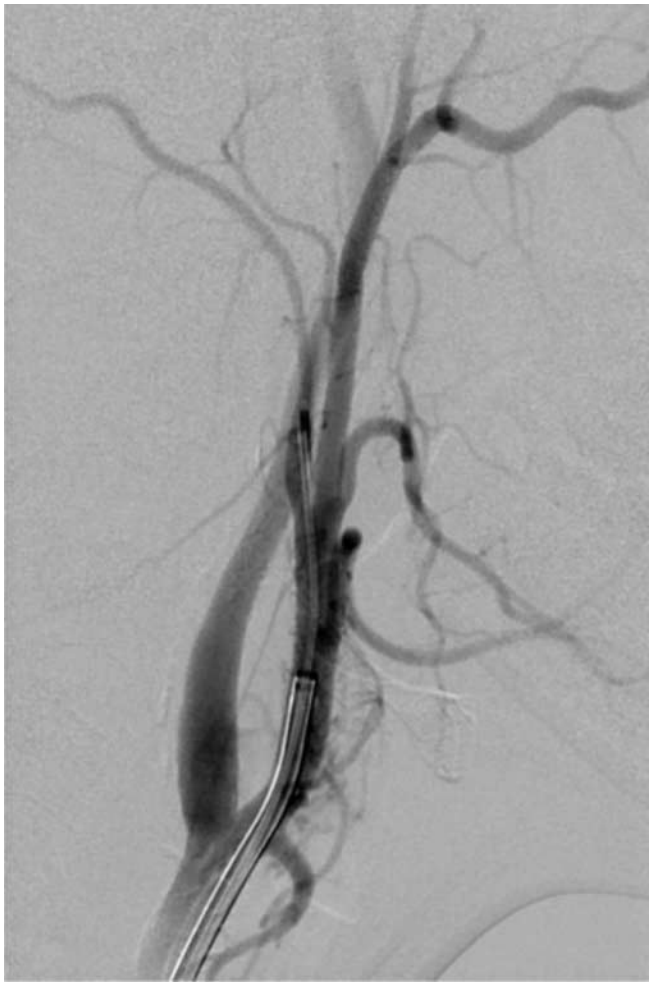

Fig. 4. Control arteriography after successful embolisation of all pathological arteries is showing normal flow in both carotid arteries

Complete subadventitial resection of the tumour was achieved in all patients. There was no operative mortality nor morbidity. No perioperative stroke, clinically significant bradycardia or hypotension were observed. Preoperative embolisation of the tumour reduced intraoperative bleeding facilitating dissection, reduced blood loss and time of surgery. Median intraoperative blood loss was 270 mL No revision was needed for bleeding.

There were no recurrences or delayed complications at the follow up of 24-76 months.

DISCUSSION

Carotid paragangliomas are rare tumours and the experience of individual centres in the diagnosis and surgical treatment is limited (7, 9, 10-13). The patients in the presented series fit into the reported demographic and clinical characteristics of those tumours (1, 2).

The series describes only Shamblin I and II tumours, thus not requiring carotid reconstruction or permanent or temporary occlusion. Preoperative embolisation of this highly vascular tumour was reported to facilitate operation and reduce blood loss (4, 9, 10, 14-16). In the reported series of preoperatively embolised paragangliomas the bleeding has not been extensive and blood loss did not exceed 550 mL. There were no postoperative bleeding recurrences which may be attributed to total tumour excision, meticulous haemostasis but also to the preoperative embolisation.

Several techniques of carotid body tumour embolisation via the endovascular route or through direct percutaneous puncture have been described in the literature. Embolisation was shown to facilitate tumour removal and diminish the need for transfusions (14-17).

Percutaneous embolisation with liquid embolic material such as glue or Onyx was shown to be safe and effective. It may better obliterate the tumour bed in contrast to the endovascular embolisation (18-20). However in both methods there is a potential risks of glue displacement into brain through supplying arteries during injection and even delayed migration has been reported (21, 22). Extreme caution must be payed to collaterals and anastomoses between external carotid artery and internal carotid artery and vertebral artery. Selective catheterisation of the external and internal carotid branches is thus required to adequately delineate the blood supply.

Choosing particles as an embolic material, it is important to adjust their size to efficacy and complication rates. Small particles penetrate more distally into the tumour capillaries but can cause injury to the vasa nervorum leading to cranial nerve palsies or enter the intracranial circulation through anastomoses (23, 24).

Effects of embolisation may be transient or permanent depending on the embolic material used. Therefore the timing of surgery after embolisation is very important. Histologic examinations proved thrombus formation and giant cell reaction within first 7 days after embolisation. Henceforth recanalisation of immobilised vessels starts. Consequently, surgery should be carried out 1 to 7 days after embolisation to maximise the benefits of the embolisation procedure (25). It also reduces the time for collateral circulation development.

Minor complications attributable to the embolisation are usually limited to puncture site in the groin like haematoma or localised pain and swelling which resolve spontaneously.

Major complications of embolisation of extracranial tumours are rare (26).

Stroke and intracranial haemorrhage, cranial nerve palsy, tissue damage have been reported in 3 to 6% during embolisation (23, 26). Major complications require additional therapy and usually prolonged hospitalisations.

In paraganglioma embolisation care must be exercised to avoid embolic particle migration to internal carotid artery through the common tumour – internal carotid anastomoses, particularly in tumours close to the ICA artery (17). This has not happened in the presented series when an appropriate embolisation technique with superselective microcatheter was employed.

CONCLUSIONS

The results of the presented series evaluation confirm that the surgical treatment of carotid paragangliomas may benefit from embolisation aimed at decreasing tumour blood flow. Due to the rarity of these hypervascular tumours there are no randomised controlled trials evaluating safety and efficacy of preoperative embolisation. Standard definitions and uniformity across reports would aid in establishing best practice measures.

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