The Journal of Laryngology & Otology
December 2001, Vol. **115,** pp. 981–987

Long-term oncological results in 47 cases of jugular paraganglioma surgery with special emphasis on the facial nerve issue

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Abstract

Oncological and functional results were assessed in 47 type C and/or D jugular paraganglioma operated on between 1984 and 1998 using the classical infratemporal fossa type A approach (mean follow-up = 66 months). In 24 instances, however, the facial nerve was not re-routed. Total resection was achieved in 33 cases (70 per cent). In 25 patients available for follow-up this resulted in a 92 per cent cure rate while two patients (eight per cent) developed recurrences that are being followed-up clinically and radiologically. Sub-total resection, leaving infracentimetric tumour remnants after being coagulated, was achieved in 14 cases (30 per cent). In 11 patients available for follow-up, only three cases developed tumour regrowth (27 per cent) that was controlled by salvage irradiation or surgery while in the other cases tumour remnants remained stable (73 per cent). Symptomatic post-operative lower cranial nerve impairment was observed in 23 per cent. When results were analysed depending on whether the facial nerve had been re-routed (n = 18) or not (n = 24), the incidence of facial paralysis HB grade III or more at one year was 33 per cent and eight per cent, respectively. Total resection was achieved in 56 per cent when the facial nerve was re-routed versus 75 per cent when it was not, the difference being due to a higher incidence of large tumours in the first group.

The present study suggests that: 1) surgical resection of jugular paraganglioma provides overall satisfactory results, i.e. a 86 per cent rate of either cure or tumour remnant stabilization, but carries a significant risk of iatrogeny; 2) complete tumour removal should not be attempted, especially in patients over 60 years of age with no pre-operative neurological deficits, since leaving infracentimetric tumour remnants has no major detrimental effect on the final outcome; 3) facial nerve transposition carries a significant risk of cosmetic sequelae while it does not provide significant advantages in terms of tumour resection and long-term oncological control.

Key words: Paraganglioma; Jugular Veins; Facial Nerve; Surgical Procedures, Operative; Treatment Outcome

Introduction

Since the first description by Rosenwasser[1] in 1945, paraganglioma arising from the jugular foramen have been approached by various surgical techniques. While an inferior mastoidectomy-anterior hypotympanic approach, with preservation of the external ear canal may be proposed in cases of limited extension to the infralabyrinthine compartment,[2,3] more extensive surgical procedures are needed when the tumour extends into the petrous apex or intracranially. For managing such tumours, the infratemporal fossa (ITF) type A described by Fisch[4] in 1978 is now considered as the standard procedure. This technique includes anterior transposition of the facial nerve that is supposed to improve access to the jugular foramen, the infralabyrinthine compartment and the ascending carotid artery as well as to the posterior fossa.

Undoubtedly, this sophisticated procedure facilitates tumour removal and provides excellent oncological results.[5] However, anterior mobilization of the facial nerve may cause facial paralysis leading to long-term cosmetic sequelae which is hardly acceptable in the management of a benign tumour. In an extensive analysis of the literature, Selesnick *et al.*[6] found that short re-routing, i.e from the stylomastoid foramen up to the second genu, yielded long-term grade I-II function in 91 per cent, while long re-routing, i.e. from the stylomastoid foramen up to the geniculate ganglion, resulted in grade I-II function in 73 per cent. Recently, Pensak and Jackler[7] have questioned the necessity of facial

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Accepted for publication: 12 June 2001.

nerve re-routing and suggested that satisfactory tumour resection could be achieved leaving the facial nerve *in situ*.

The aim of the present study was two-fold: 1) to analyse the oncological and functional results in 47 cases of jugular paraganglioma, type C and/or D according to Fisch's classification operated between 1984 and 1998 using most of the main steps of the ITF type A approach; 2) to study the influence of facial nerve mobilization, short or long, on the incidence of facial paralysis and oncological results by comparing the cases operated with, or without, facial nerve mobilization.

Results strongly suggests that facial nerve transposition is not necessary to achieve satisfactory control of these tumours.

Materials and methods

Clinical features

The patient population of this study comprises 47 cases of jugular paraganglioma, types C and/or D operated between January 1984 and October 1998. All patients underwent pre-operative biological and neuroradiological assessment, i.e. catecholamine levels, computed tomography (CT), arteriography, and (since 1990) magnetic resonance imaging (MRI).

The ages of the 47 patients ranged from 18 to 73 years (mean age = 46 years). Twenty-eight patients were female and 19 male. There were 26 left-sided tumours. Ten patients had multiple vagal or carotid body paragangliomas. In three cases, tumours were bilateral. Five patients had incomplete or complete facial paralysis pre-operatively. Four cases had increased catecholamine levels with clinical signs.

Table I and Figures 1–3 summarize the main clinical features of these patients: classification according to Fisch, pre-operative cochleo-vestibular symptoms and function, pre-operative lower and XIIth cranial nerve function.

Follow-up consisted of annual clinical examination, CT with injection and (since 1990) MRIs were performed annually in the first two years following surgery and, when possible, at three, five and more years post-surgery.

Surgical treatment

All patients underwent endovascular embolization two to four days prior to surgery. Four patients had direct *in situ* embolization by the technique developed in our institution.[8] The technique used most of the main steps of the ITF approach, type A, as described by Fisch.[4] These main steps were: combined transcervical-transmastoid approach with

TABLE I
CLASSIFICATION OF THE 47 PARAGANGLIOMAS

	C1	C2	C3
	4	15	6
De 1		3	2
De 2	3		3
Di 1		5	2
Di 2		2	2

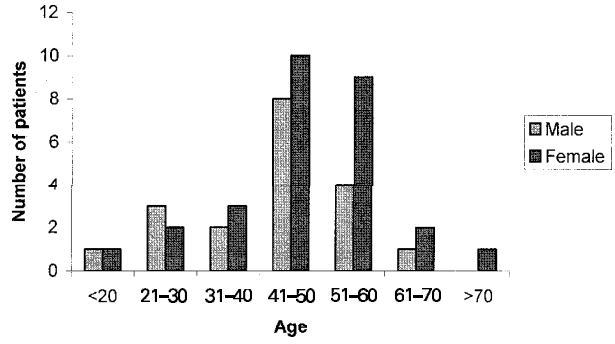

FIG. 1
Age and sex distribution of the 47 paragangliomas.

transection and exclusion of the external auditory canal; identification of the main vessels and nerves in the digastric area; identification of the trunk of the facial nerve in the parotid region; extensive radical mastoidectomy with sacrifice of the external auditory canal; canal wall-down procedure and exenteration of the invaded petrous air cell system; sacrifice of the tympanic membrane, malleus, and incus; skeletonization of the ascending, genu or horizontal internal carotid artery (ICA) when needed; fat obliteration of the middle-ear spaces. Nine cases had transposition of the second and third portions of the facial nerve (long transposition) as in the classical ITF approach, and nine cases had transposition of the third mastoid portion (short transposition).

In 19 cases, the cochlea was sacrificed either because of tumour invasion (n = 13) or to gain access to the intercochleo-carotid cell tract (n = 6). In 14 patients, a juxtacondylar approach including resection of the transverse process of C1 was used to control feeding pedicles coming from the vertebral artery and to improve the postero-inferior exposure of the jugular foramen as previously described.[9] A complementary neurosurgical procedure with resection of tumour in the posterior fossa was performed in 14 cases. This was done at the same time in nine cases and as a secondary stage in the five remaining cases.

In five cases, the facial nerve was sacrificed because of massive tumour invasion. While these patients do not add further data concerning the issue of facial nerve mobilization and paralysis, they were included to provide a broader oncological database.

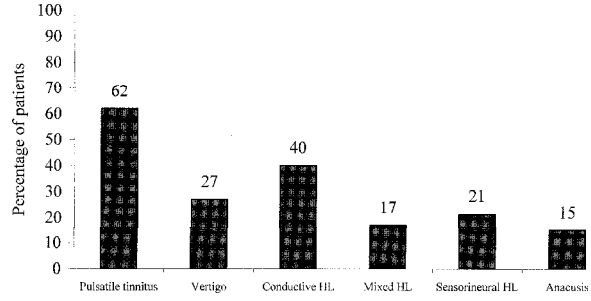

FIG. 2
Pre-operative cochleovestibular symptoms and function.

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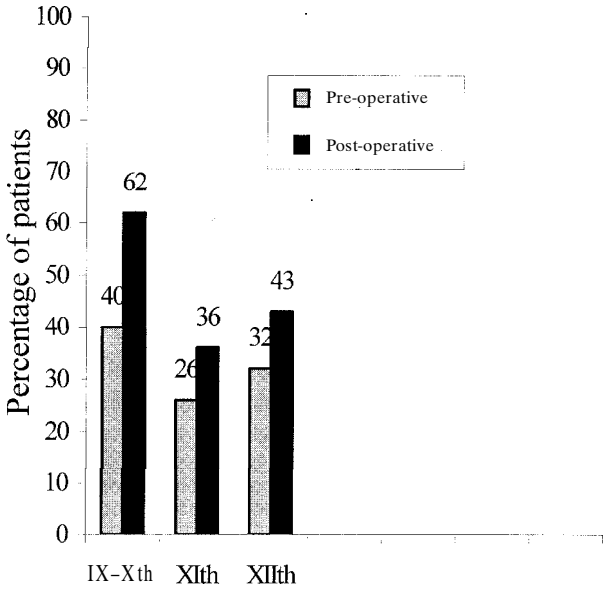

FIG. 3
Cranial nerve assessment.

Since 1994, the surgical procedure was modified in that patients underwent basically the same surgical steps except that the facial nerve was not transposed. This concerned the 24 last cases. The mastoid portion of the Fallopian canal was skeletonized and the infralabyrinthine and retrofacial cells exenterated (a procedure similar to the Fallopian bridge technique described by Pensak and Jackler[7]).

Results

Oncologic results

Total resection, i.e. no tumour remnants were seen at the end of surgery, was achieved in 33 cases (70 per cent). Eight patients were lost for follow-up within two years. Twenty-three patients were tumour-free clinically and radiologically in follow-ups at two to 15 years. Two patients are being followed clinically and radiologically without additional treatment because of asymptomatic and very slow growing recurrent growth.

Subtotal resection, i.e. infracentimetric remnants were left in place after having been coagulated, was achieved in 14 cases (30 per cent). Two of these patients died, one from acquired deficiency syndrome (AIDS) and one from a post-therapeutic neurovascular accident. Eight patients had a stable or regressing tumour on radiological follow-up at 17 months to 14 years. Three patients had recurrences that were treated by radiotherapy in two cases and surgery in the other. Tumour remnants and recurrences were located in the petrous apex adjacent to the cavernous sinus, around the genu and ascending ICA, the nervous compartment of the jugular foramen and in the posterior fossa. One patient was lost for follow-up. Table II summarizes these data.

TABLE II
ONCOLOGICAL RESULTS

	Total resection (n = 33)	Sub-total resection (n = 14)
Stable remnants	–	8
Recurrence	2	3
Lost for follow-up	8	1
Death	–	2

Three patients developed a contralateral tumour that was irradiated and satisfactorily controlled for the 11 to 14 years that they were observed.

Analysis was then performed to determine whether leaving the facial nerve in place influenced the quality of tumour removal. To address this issue, we compared oncological results in 18 patients who underwent subtotal (II° and III° portions) or partial (III° portion) facial nerve rerouting and in 24 patients where the facial nerve was left in place.

Results show that: 1) when the facial nerve was rerouted (n = 18), total resection was achieved in 10 cases: six patients were free of disease, two had recurrences, two were lost for follow-up. Subtotal resection was achieved in eight cases: four had stable remnants, two developed recurrences; 2) when the facial nerve was left in place (n = 24), total resection was achieved in 18 cases: 15 patients were free of disease, three were lost for follow-up. Subtotal resection was achieved in six cases: four were free of disease, one had a recurrence and one was lost for follow-up.

Facial outcome

Transposition of the facial nerve resulted in immediate facial paralysis (House and Brackmann (HB) grade V–VI) in 10 out of eighteen (56 per cent) patients at one month post-operation. More precisely, mobilization of the second and third portions yielded a complete paralysis in five out of nine cases of long transposition. Complete paralysis also occurred in five out of nine cases of short transposition. At one year post-operation, the incidence of grade IIII–IV paralysis in these patients was 33 per cent. i.e. three patients after long transposition and three after short transposition.

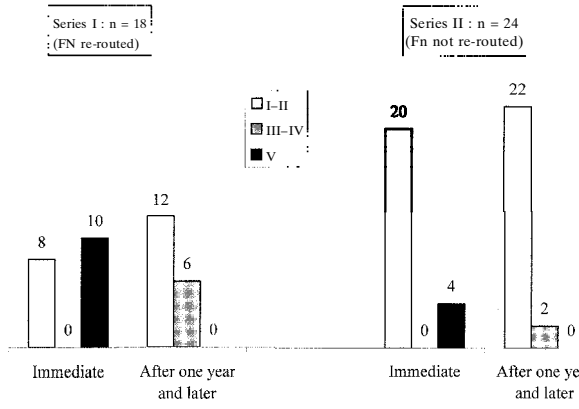

FIG. 4
Post-operative facial nerve assessment (House and Brackmann grades).

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When the facial nerve was left in place (n = 24), 20 patients had no facial paralysis (HB grade I and II) in the one month post-operation. Four patients (16 per cent) sustained HB grade V–VI facial paralysis. One of these patients had intracranial extension (Di2) requiring prolonged dissection of the tumour in the cerebellopontine angle. At one year, two patients were graded HB II, one was graded HB III and the other was graded HB IV.

Thus, the incidence of grade III–IV paralysis in 24 patients was eight per cent at one year post-operation. Figure 4 summarizes these data.

Other sequelae and complications

One patient suffered hemiplegia in the immediate post-operative period. This event was due to migration during surgery of a balloon placed by the neuroradiologist in the ICA to suppress the blood supply. It is most likely that the mobilization occurred during dissection of the vertical and genu segments of the artery which were surrounded by the tumour. The patient died 17 months later. No radiological follow-up was available. One patient became human immunodeficiency virus (HIV) positive as a result of a blood transfusion. She was lost to oncological follow-up after six months, and died in her home country a few years later.

Figure 3 summarizes the post-operative status of the lower and XIIth cranial nerves in these patients. Four patients developed aspiration and pneumonia and required tracheostomy. These four patients had undergone a combined neurosurgical approach. They all recovered after two to four months. A laryngoplasty medializing the paralysed vocal folds was subsequently performed in two of these four patients and in seven other cases to improve a marked dysphonia. Functional vocal results were satisfactory in four cases, moderate in three cases, but poor in the two remaining cases. Swallowing problems were encountered in some of these patients, that subsided in a few weeks without any complementary surgical procedure.

Seven patients had CSF leak through the skin incision. In two instances, CSF leaked through the tracheostomy. Five patients developed meningitis. They all recovered after two to four months.

Discussion

The present study concerns 47 patients presenting jugular paraganglioma type C and/or D who underwent the main steps of the ITF type A approach described by Fisch.[4] These 47 patients were part of a series of 89 patients with the same type of paraganglioma. The remaining 42 patients were not included in this series either because some of them were referred for recurrences (n = 11) and/or because they were managed either by radiotherapy (n = 17) or by various surgical procedures that could not be compared with that used in the present series (n = 25), namely transmastoid-transcervical or transmastoid-transtympanic approaches combined, when needed, with a transtemporal or suboccipital exposure.

Tumour removal and oncologic result

Total resection was achieved in 33 cases (70 per cent). Out of 25 patients available for follow-up for more than two years, 23 (92 per cent) were tumour free with an average follow-up of 66 months (two to 15 years). Such a cure rate is in agreement with those of Gjuric *et al.*[10] who, in 51 per cent of complete tumour removal, obtained a 90 per cent cure rate or of Patel *et al.*[11] who reported an 83 per cent figure. In a recent retrospective review of 34 patients presenting types C or CD tumours, Fisch and coworkers[5] achieved a complete tumour removal in 83 per cent and observed a recurrence in three per cent of the cases.

A salient finding of our analysis, probably the most important one, is that leaving infracentimetric tumour remnants has no major effect on the final outcome. Out of 14 patients with subtotal resection (30 per cent), 11 were available for follow-up. Only three (27 per cent) developed recurrences that were treated either by radiotherapy (n = 2) or by surgery (n = 1) while in eight patients, the tumour remnant showed stability or very slow recurrent growth which is still under radiological follow-up (17 months to 15 years). These figures are slightly higher than those of Fisch and coworkers[5] who reported subtotal resection in only six out of 36 patients (17 per cent). Two patients had to be referred for additional radiotherapy and four showed no further growth of the tumour remnant.

It may be argued that recurrences are usually seen after five years and that follow-up of some patients is too short to properly assess the long-term incidence of a subtotal removal. In our experience recurrences can be detected much earlier. With the new imaging techniques, the first post-operative control may detect remnants which will sometimes become clinically patent a few years later. We now perform a post-operative CT scan with injection as early as the day following surgery in order to avoid the inflammatory reaction that develops a few days later, and are able to identify tumour remnants of 1 cm or even less.

In light of these results, it is now our policy to deliberately leave infracentimetric remnants in patients over 60 years of age, with no pre-operative cranial deficit, when complete tumour removal would carry a risk of major injury to either the ICA or lower cranial nerves, or increase the seriousness of the required surgical procedure and the risk of CSF leakage through the dura opening.

Neurovascular, cranial nerve and other complications

The present series confirms that surgical resection carries a significant iatrogeny.[12,13]

The massive hemiplegia due to migration during surgery of the balloon placed in the ICA is a very rare complication that has not been reported in skull base literature. This dramatic complication which eventually proved fatal led us, when the ICA has to be sacrificed because of major tumoral encasement, to replace the very slippery and unstable balloons by coils.

CSF leaks have been frequently reported after intracranial tumour resection or previous radiotherapy.[11,14] Various technical procedures have been proposed to avoid them. However, the most efficient still remains to stage the neurosurgical step in case of a large intracranial extension.

Eleven patients (23 per cent) developed severe aspiration, pneumonia and/or marked dysphonia or swallowing problems. This is in agreement with our previous study on 19 surgically treated patients, of whom five (26 per cent) presented permanent symptomatic post-operative lower cranial nerve dysfunction.[15] This confirms that patients who are previously neurologically intact do not cope easily with sudden post-operative loss of lower cranial nerves.[16] Conversely, pre-operative palsies might encourage aggressive operative removal as these patients have usually accommodated the resulting deficits.[14]

Thanks to spontaneous evolution, or to various rehabilitative procedures, the debilitating consequences of these paralyses are usually overcome within a few weeks with minimal long-term sequelae. On this particular point, we do not agree with Briner *et al.* (1999)[5] who state that dysphagia remains the major problem related to surgery of jugular paraganglioma. Rather from our standpoint, we consider facial paralysis to be the major residual complication.

Facial nerve re-routing and facial outcome

The position of the facial nerve hampers direct surgical exposure to the jugular foramen. Re-routing the facial nerve anteriorly has been advocated by many authors. Capps[17] in 1952, Shapiro and Neues[18] in 1964, Gejrot[19] in 1965, and Glasscock *et al.*[20] in 1974, among others, occasionally used anterior displacement of the facial nerve to facilitate resection of the tumour. It was not until 1978 that Fisch,[4] reporting the ITF approach, described the technique and indications for systematic management of the facial nerve according to tumour extension. Basically, the facial nerve is gently mobilized from the stylomastoid foramen to the geniculate crest and transposed in a new Fallopian canal drilled along the anterior epitympanum and in a groove made in the parotid gland.

According to this author,[21] mobilization of the facial nerve may cause 'some degree of paresis but complete paralysis is rare'. In their recent survey of 20 cases of anterior transposition, Fisch and co-workers[5] were able to preserve normal facial function in 80 per cent (thus giving a 20 per cent long-term impairment). While such results are undoubtedly achieved by some highly experienced surgeons, complete facial paralysis leading to long-term cosmetic sequelae may occur in other hands.

This is indirectly attested by the numerous articles reporting surgical and technological refinements aimed at limiting the occurrence of facial paralysis. Thus, over the years various authors have proposed mobilization of the mastoid portion only, *en bloc* displacement of the mastoid portion of the facial nerve with its surrounding connective tissue in the region of the stylomastoid foramen[22] and with the posterior belly of the digastric muscle,[23] replacement of the mobilized portion at the end of surgery in its original position,[23] or permanent electromyography (EMG) recording.[24] Despite these technical advances, it remains an unfortunate reality that 'a facial dysfunction still regularly occurs after facial nerve rerouting ... proportional to the length of nerve mobilized.'[6]

The mechanism of facial paralysis is most likely vascular. Disruption of the normal arterial supply and interruption of the venous drainage passing through the periosteum in the region of the stylomastoid foramen are responsible for acute devascularization of the nerve. From an experimental standpoint, Mostafa and Samir[25] have shown in guinea pigs that facial nerve re-routing induced constant histopathological changes, namely swelling of axons, Schwann cell proliferation, myelin sheath vacuolation, and cellular infiltration, similar to those described in other forms of facial nerve injury. These changes persisted for an extended period of time.

Thus, the question arose as to whether surgical removal of jugular paraganglioma could be achieved without facial nerve rerouting. This was suggested by Maniglia *et al.*,[3] Martin and Prades,[26] and recently by Pensak and Jackler.[7] These last authors described the 'Fallopian bridge technique' and stated that in the majority of cases satisfactory exposure can be provided without facial nerve re-routing.

A salient finding of our study is that re-routing the facial nerve, be it short or long, may have deleterious and long-term effects on nerve function. At one year post-operation, we found a 33 per cent incidence of HB stages III and IV. This figure calls into question our own surgical expertise, since faulty technique could be responsible in a procedure which in other hands yields a very low rate (10 per cent) of grade III and IV dysfunction occurring only in patients already operated on or irradiated.[27] However, this is not likely the case since: 1) the incidence reported here is close to the 27 per cent of patients with HB grades III and IV reported by Selesnick *et al.*,[6] and 2) severe paralysis occurred equally throughout the period under consideration, i.e. independently of our increasing experience. We also did not find any difference in the severity of the facial paralysis whether a short (mastoid portion) or long (mastoid and tympanic portions) transposition was performed. This is consistent with the notion that the main factor of paralysis is devascularization of the facial nerve in the stylomastoid foramen region. In contrast, leaving the facial nerve in place led to only an eight per cent incidence of long-term grade III–IV facial paralysis. This result suggests that, even without mobilization, surgical dissection of the facial nerve in the stylomastoid region and in the posterior fossa carries an inescapable risk of facial paralysis. It remains to be determined whether pre-operative embolization increases this risk as recently suggested by Marangos and Shumacher.[28]

Facial nerve re-routing and tumour removal

This study raises two principal issues concerning the effect of facial nerve mobilization or non-mobilization on the surgical procedure. First, does non-mobilization of the facial nerve endanger the patient's life during surgery? Analysis of our series suggest that re-routing the facial nerve anteriorly is not a prerequisite for management of the ICA. In both instances, facial nerve re-routed or not, we faced 11 cases of serious haemorrhage during dissection of the ascending portion of the ICA, mainly due to a rupture of the carotid-tympanic branch. Bleeding was controlled by adequate packing of the vessel, without serious consequence.

Second, does re-routing the facial nerve facilitate the removal of the tumour? Undoubtedly, mobilization of the facial nerve away from the surgical field does facilitate the dissection of the infralabyrinthine compartment, the jugular foramen, and the initial portion of the ascending carotid artery. But it does not affect the quality of tumour removal. A figure of 56 per cent of total or near total resection was achieved when the facial nerve was re-routed versus 75 per cent when it was not. This difference may be accounted for by the fact that the former series included larger tumours than the latter one. In any case, it suggests that leaving the facial nerve in place does not restrict tumour removal. In both instances, the tumour remnants were located in regions that would not have been better exposed by facial nerve re-routing.

Thus, the issue raised by our study is not whether mobilization of the facial nerve induces a higher incidence of facial paralysis – this would depend on the skill of the surgeon and the difficulty of the case. The issue here is whether surgery without mobilization compromises the quality of the oncological outcome. Our data indicate that this is not the case.

Surgical implications

From a practical point of view, a satisfactory exposure of the jugular foramen may be obtained without facial nerve re-routing provided that five technical requirements are maintained: clear identification of the facial nerve trunk in the posterior parotid area, complete removal of the tympanic ring, resection of the styloid process, retraction of the temporomandibular joint, and extensive skeletization of the mastoid portion of the Fallopian canal (the Fallopian bridge technique described by Pensak and Jackler).[7]

For some authors,[29,30] transposition of the facial nerve is reserved for cases in which the paraganglioma invades the horizontal portion of the carotid artery and petrous apex. In those instances, we found that the main limiting obstacle is the cochlea and not the facial nerve. This is especially true in the case of poorly pneumatized petrous bone. In this anatomical circumstance, sacrificing the anterior labyrinth seems more efficient than re-routing the facial nerve anteriorly, for adequate exposure to the medial face of the artery and to the petrous apex. This provides a satisfactory access to the intercochleocarotid cell tract which is the main pathway for anterior tumoral extension.

Regarding type D paraganglioma, we agree with Pensak and Jackler[7] that the presence or absence of intracranial extension has little relevance to the decision whether or not to re-route the facial nerve. In that case of posterior fossa invasion, the juxtacondylar approach removing the transverse process of C1 and the surrounding tissues is important.[9] Along with the exenteration of the inferior and retrolabyrinthine cells and retraction of the sigmoid sinus, it improves markedly the postero-inferior exposure to the jugular foramen and posterior fossa, medial to and below the facial nerve.

Finally, re-routing the facial nerve is advocated to allow a one-stage removal of a posterior fossa extension. In our experience, we prefer to stage the neurosurgical step mainly because of the risk of CSF leakage through the dura opening.

Conclusion

Facing a benign tumour, treatment should improve or cure presenting symptoms and prevent further neurological deficits. Because of its deep location in the skull base, paraganglioma of the jugular foramen raises a surgical challenge aimed at complete tumour removal with minimal morbidity. The results of our study show that surgical resection provides overall satisfactory oncological results, i.e. an 86 per cent rate of either cure or tumour remnant stabilization in 36 patients available for follow-up. However, it carries a risk of post-operative neurological deficits.

To improve exposure and, thus, tumour control, anterior transposition of the facial nerve has been proposed. In assessing the 'cost-effectiveness' ratio of facial nerve re-routing, the present study suggests that facial nerve mobilization carries a significant risk of cosmetic sequelae while it does not provide significant advantages in terms of tumour resection and long-term oncological control. Questioning this main step of the infratemporal approach described by Fisch should be considered by surgeons not as a challenge of their skills but rather as an attempt to avoid patients suffering from disfiguring paralyses. In agreement with previous authors, we propose that management of paraganglioma should be tailored to the symptoms, the pre-operative neurological status, the extent and, above all, the nature of this tumour.

Acknowledgements

We thank Dr Guy Rowley and Sidney Wiener for editorial assistance.

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Pr P. Tran Ba Huy takes responsibility for the integrity of the content of the paper.
Competing interests: None declared