

Outcomes of endoscopic surgery, endoscopic-assisted surgery and open surgery in the treatment of juvenile nasopharyngeal angiofibroma

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Abstract

Introduction: The therapeutic procedures for juvenile nasopharyngeal angiofibroma (JNA) still remain a challenge.

Aim: To present the outcomes of JNA treatment at the Department of Otolaryngology in Poznan, Poland, between 2000 and 2008. This study contrasts endoscopic surgery and endoscopic-assisted surgery with open surgery in cases of small and large JNA.

Material and methods: We reviewed the histories of patients who were managed surgically for JNA between 2000 and 2008 at the Department of Otolaryngology in Poznan, Poland. The size of the tumour was measured prior to surgery, using contrast-enhanced computed tomography or magnetic resonance imaging. The extent of the disease was then assessed according to the staging scale proposed by Radkowski et al.

Results: Fifteen patients underwent surgery for JNA. Of 8 patients with small tumours, 4 patients underwent open surgery, and 4 patients underwent endoscopic surgery. Of the remaining 7 patients with large tumours, five underwent open surgery and two of them endoscopic-assisted open surgery. In the one-year follow-up of patients who underwent endoscopic surgery exclusively or endoscopic-assisted open surgery no recurrence was found. Of the 9 patients who underwent open surgery, tumour recurrence was observed in 4 cases.

Conclusions: Selection of the surgical technique in JNA depends on the extent of the tumour and the equipment available. The endoscopic technique is useful for selected small tumours and open surgery and/or endoscopic-assisted open surgery is useful for large tumours. Repeat JNA surgeries may be performed using the open technique, the endoscopic technique, or the endoscopic-assisted open technique.

Key words: juvenile nasopharyngeal angiofibroma, exclusively endoscopic surgery, navigation.

Introduction

Juvenile nasopharyngeal angiofibroma (JNA), a rare tumour developing in adolescent and young adult males, accounts for 0.05-0.5% of all head and neck tumours [1, 2]. Juvenile nasopharyngeal angiofibroma is composed of histologically benign connective and vascular tissues, and is characterised by local infiltration [3]. Although androgens and the co-existence of familial adenomatous polyposis (FAP)

are thought to play an important role, the pathogenesis of JNA still remains unclear [4, 5]. Epistaxis and nasal obstruction are the most common manifestations. Juvenile nasopharyngeal angiofibroma can be a life-threatening condition, as the epistaxis tends to be profuse and difficult to control, and the tumour tends to spread towards the base of the skull and intracranially.

Juvenile nasopharyngeal angiofibroma develops in the posterolateral wall of the nasal cavity near the

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superior border of the sphenopalatine foramen [6]. The tumour can extend medially to the nasal cavities or the nasopharynx and laterally to the pterygopalatine fossa and the infratemporal fossa. From the pterygopalatine fossa, the tumour may further extend into the orbit through the inferior orbital fissure. From the infratemporal fossa, the tumour may grow upwards through the base of the pterygoid process into the area between the foramen ovale, rotundum and lacerum, reaching the middle cranial fossa. Juvenile nasopharyngeal angiofibroma can erode the lateral and posterior wall of the sphenoid sinus and extend as far as the pituitary gland [7]. Juvenile nasopharyngeal angiofibroma is always extradural, even if it extends into the cranium [8].

As a part of the diagnostic evaluation of JNA, contrast-enhanced computed tomography (CT) and magnetic resonance imaging (MRI) are used to assess the extent of the disease. Angiographic embolization is performed as a preparatory procedure prior to surgery. The maxillary artery, the ascending pharyngeal artery, and branches of the internal carotid artery are the primary feeders of the tumour [2].

Surgical resection is the treatment of choice for JNA, although radiotherapy, hormonal therapy, cryotherapy, electrocoagulation and chemotherapy have also been used. Many surgical approaches have been reported, including the transpalatine, transzygomatic, transmandibular, infratemporal and transfacial approaches (lateral rhinotomy, Denker's method, midfacial degloving) [9]. The surgical techniques and equipment currently being used for JNA resection include navigation (computer-assisted surgery – CAS, image-guided surgery – IGS), coupled with endoscopic resection, especially in the case of

recurrent tumours, the frequency of which can be up to 50% [10].

Aim

The purpose of this paper was to present the outcomes of JNA treatment at the Department of Otolaryngology and Laryngological Oncology in Poznan, Poland, between 2000 and 2008. Our case studies are summarised here based on size of the JNA and surgical technique. This study contrasts endoscopic surgery and endoscopic-assisted surgery with open surgery in cases of small and large JNA.

Material and methods

We reviewed the histories of patients who had been managed surgically for JNA between 2000 and 2008 at the Department of Otolaryngology and Laryngological Oncology in Poznan, Poland. Patients with a diagnosis of histologically confirmed juvenile angiofibroma were analyzed retrospectively.

The size of the tumour was measured prior to surgery using contrast-enhanced computed tomography (CT) or magnetic resonance imaging (MRI). The extent of the disease was then assessed according to the staging scale proposed by Radkowski *et al.* (Table I) [11].

Starting in 2005, the patients underwent pre-op angiographic embolization to reduce intraoperative blood loss [12].

In our Department of Otolaryngology and Laryngological Oncology in Poznan, Poland, 15 patients underwent surgery for JNA between 2000 and 2008 (Table II). The age ranged from 9 to 56 years with the mean age of 18. Eight patients had small tumours (lower than Radkowski stage IIB) (Figure 1)

Table I. Staging of juvenile angiofibroma according to Radkowski *et al.*

IA	Confined to the nose and/or nasopharyngeal vault
IB	Extension into one or more paranasal sinuses
IIA	Minimal extension into the pterygopalatine fossa
IIB	Full occupation of the pterygopalatine fossa, with or without erosion of orbital bones
IIC	Extension through the pterygomaxillary fossa into the cheek and temporal fossa or posterior to pterygoid plates
IIIA	Erosion of skull base – minimal intracranial extension
IIIB	Erosion of skull base – extensive intracranial extension with or without cavernous sinus invasion

Table II. Patients undergoing surgery for juvenile nasopharyngeal angiofibroma at the Department of Otolaryngology and Laryngological Oncology, Poznan, Poland, 2000-2008

Patient number	Age	Radkowski stage	First surgery	Recurrences	Surgical technique	Other treatment	Embolization
1	13	IIB	2000	2003	OS	Rx 2003	No
2	15	IB	2000	–	OS		No
3	20	IB	2002	–	OS		No
4	11	IIIA	2003	2007, 2008	OS/ES/EAOS		No/Yes
5	14	IIB	2002	2003	OS		No
6	16	IB	2003	–	OS		No
7	20	IIB	2004	–	OS		No
8	17	IIA	2004	–	OS		No
9	9	IIB	2005	2005, 2007, 2008	2xOS/EAOS/OS	Fisch t. C	Yes
10	14	IB	2007	–	ES		Yes
11	10	IIB	2007	–	EAOS		Yes
12	19	IA	2007	–	ES		Yes
13	19	IB	2007	–	ES		Yes
14	22	IIC	2008	–	EAOS		Yes
15	56	IA	2008	–	ES		Yes

Rx – radiotherapy, OS – open surgery, EAOS – endoscopic-assisted open surgery, ES – endoscopic surgery

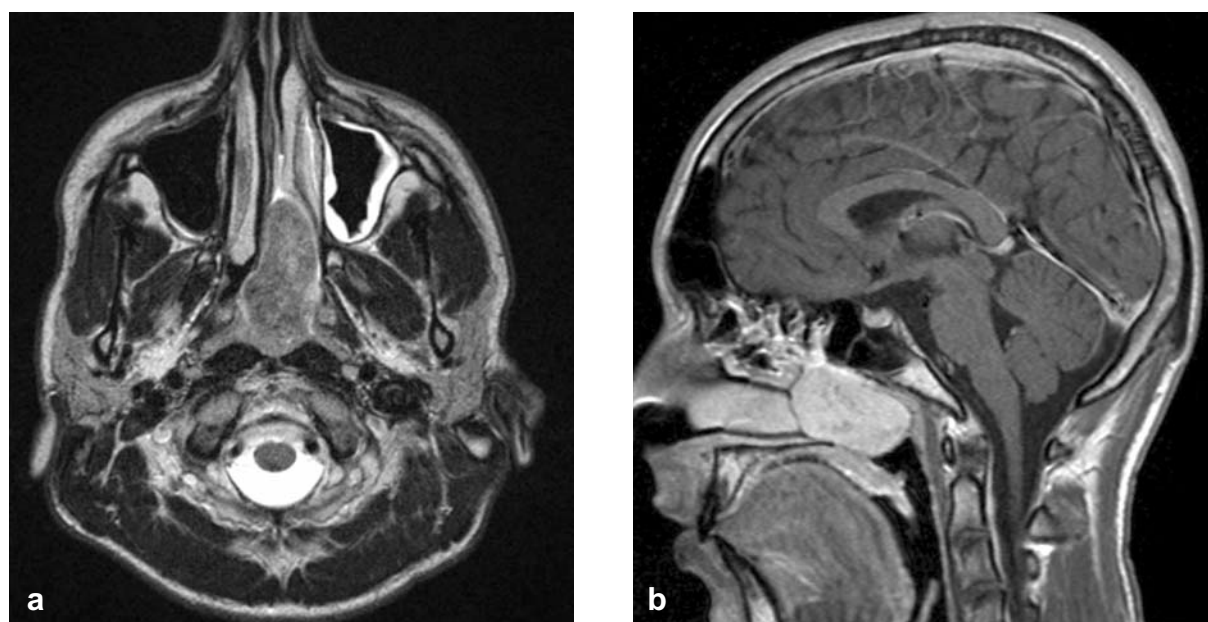


Figure 1. Computed tomography scan. Axial (a) and sagittal (b) views of a juvenile angiofibroma with stage IA according to Radkowski *et al.* requiring an exclusively endoscopic approach

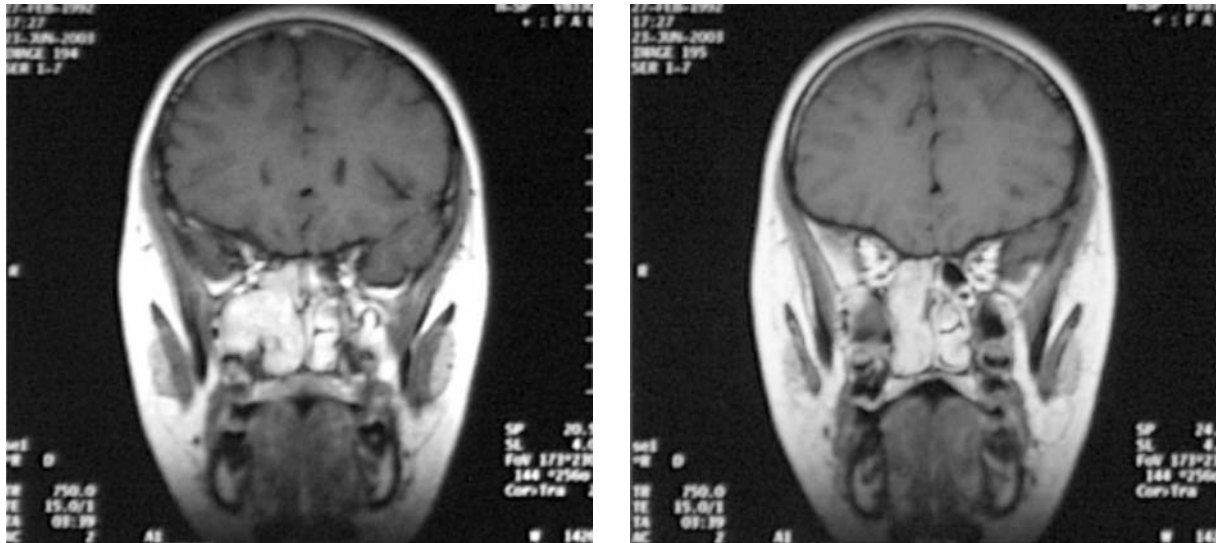


Figure 2. Magnetic resonance imaging scan. Coronal view of a juvenile angiofibroma with stage IIC according to Radkowski *et al.* requiring endoscopic-assisted open surgery

and seven had large tumours (Radkowski stage IIB or higher) (Figure 2).

Selection of the surgical technique depended on the extent of the tumour and the equipment available. Endoscopic surgery was performed on small JNAs (lower than Radkowski stage IIB) starting in December 2006. Manipulation difficulties during endoscopic resection were overcome by enlisting a second operator and using the four-hand technique [13-15]. Large JNAs (stage IIB or higher), which presented problems for endoscopic surgery, were resected using endoscopic-assisted open surgery, which facilitated both surgical techniques. In cases where the tumour had invaded vital structures such as the skull base or the parasellar region, computer-assisted surgery (CAS) made it considerably easier to perform radical surgery.

Endoscopic resection of juvenile nasopharyngeal angiofibroma

The surgery is performed by two operators using “the four-hand technique” under general anaesthesia and controlled low systolic blood pressure [15, 16] (Figure 3).

Ethmoidectomy is initially done in order to better visualize the tumour, followed by a wide meatal antrostomy to reveal the posterior wall of the maxillary sinus. The middle turbinate and, if necessary, the inferior turbinate are then resected. The tumour is then bluntly dissected off the nasal

septum and the nasopharyngeal mucosa until the stalk is visualised in the vicinity of the sphenopalatine foramen. If the tumour extends into the pterygopalatine fossa, the posterior wall of the maxillary sinus is removed in order to extricate the tumour from this region. The released tumour is then removed through the nasal or oral cavity. Bleeding is controlled by coagulation and nasal packing. Nasal packing is left in place for 24 or 48 h depending on the intensity of bleeding.

Endoscopic-assisted open approach

This surgery is based on the external approach combined with endoscopic technique. According to Carrau *et al.* [17], in endoscopic-assisted procedures

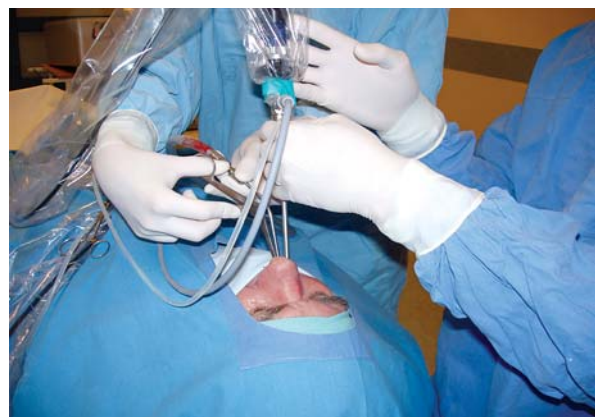


Figure 3. Intraoperative view of the “four-hand technique”

Table III. External approaches used for surgery of juvenile nasopharyngeal angiofibroma at the Department of Otolaryngology and Laryngological Oncology, Poznan, Poland, 2000-2008

Patient number	Radkowski stage	Open surgery	Recurrences		
			1 st	2 nd	3 rd
1	IIB	Denker's method	Rx	–	–
2	IB	Denker's method	–	–	–
3	IB	Denker's method	–	–	–
4	IIIA	Denker's method	ES	EAOS = lateral rhinotomy + ES	–
5	IIB	Denker's method	Denker's method	–	–
6	IB	Denker's method	–	–	–
7	IIB	Denker's method	–	–	–
8	IIA	Denker's method	–	–	–
9	IIB	Denker's method	Denker's method	EAOS = midfacial degloving + ES	Fisch t. C
11	IIB	EAOS = midfacial degloving + ES	–	–	–
14	IIC	EAOS = midfacial degloving + ES	–	–	–

Rx – radiotherapy, OS – open surgery, EAOS – endoscopic-assisted open surgery, ES – endoscopic surgery

Table IV. Number of JNA patients by surgical technique and extent of the tumour in the case series at the Department of Otolaryngology and Laryngological Oncology, Poznan, Poland, 2000–2008

JNA	Small tumours (< IIB*)		Large tumours (≥ IIB*)	
Total (n = 15)	8	54%	7	46%
Open surgery	4	50%	5	71%
Endoscopic surgery	4	50%	0	0%
Endoscopic-assisted open surgery	0	0%	2	29%

*Radkowski et al. staging system for juvenile angiofibroma

at least some of the resection is performed under endoscopic control.

Results

Fifteen patients underwent surgery for juvenile nasopharyngeal angiofibroma at the Department of

Otolaryngology and Laryngological Oncology in Poznan, Poland, between 2000 and 2008. Of 8 (54%) patients with small tumours (lower than Radkowski stage IIB), 4 patients (50%) underwent open surgery (Denker's method) and 4 patients (50%) underwent endoscopic surgery. Of the remaining 7 patients (46%) with large tumours (Radkowski stage IIB or higher), five (71%) underwent open surgery (lateral rhinotomy, Denker's method, or midfacial degloving – Table III) and two of them (29%) endoscopic-assisted open surgery (Table IV).

Post-op follow-up visits took place every 3 months. The follow-up period ranged from 1 year (endoscopic surgery and endoscopic-assisted surgery only) to 9 years, for a mean of 4 years and 6 months. Endoscopic assessment of the postoperative cavity was performed at each visit, and an imaging study (CT or MRI) was done once a year.

No recurrence was observed during the postoperative period in the 8 patients (53%) with small JNAs, but there were 4 patients with recurrences (26%) among the 7 patients (47%) with large JNAs (Table V). A single recurrence was observed in 2 patients (patient #1: radiotherapy 3 years after primary surgery, patient #5: repeat surgery 1 year

later); two subsequent recurrences were seen in one patient (patient #4: two repeat surgeries after a 4-year interval); and there were three subsequent recurrences in one patient (patient #9: three repeat surgeries: 2, 4 and 5 years after primary surgery). The third repeat surgery in patient #9, performed outside the Otolaryngology Department in Poznan, used the Fisch type C infra-temporal fossa approach with petrosectomy and reconstruction of the facial nerve with the sural nerve (skull base surgery) (Table II).

In the small angiofibroma group (lower than Radkowski stage IIB), no recurrences were observed for any of the surgical techniques used. In the group with larger angiofibromas, i.e. those occupying the infratemporal fossa or extending into the skull base, there were 4 recurrences, and these patients underwent repeat surgery (endoscopic surgery and endoscopic-assisted open surgery, with three successful outcomes). One patient was referred for surgery using the Fisch type C approach.

The JNA recurrences seen in four patients were confined to the nasopharynx and the maxillary sinus, as in patient #1, or else penetrated intracranially, as with patients #4 and #9. Table VI provides a more detailed description of JNA recurrences. In the one-

Table V. Number of JNA recurrences by extent of the tumour at the Department of Otolaryngology and Laryngological Oncology, Poznan, Poland, 2000-2008

JNA	Small tumours (< IIB*)		Large tumours (≥ IIB*)	
Total (n = 13)	8	53%	7	47%
Recurrences	0	0%	4	26%

*Radkowski et al. staging system for juvenile angiofibroma

year follow-up of patients who underwent endoscopic surgery exclusively (patient #10, stage IB; patient #12, stage IA; patient #13, stage IB) or endoscopic-assisted open surgery (patient #11, stage IIB), no recurrence was found. Of the 9 (60%) patients who underwent open surgery, tumour recurrence was observed in 4 (26%) cases (Table VII).

The following complications were observed in the open surgery group: bleeding necessitating blood transfusion, nasolacrimal canal obstruction, hypoesthesia of the cheek. Patients who underwent endoscopic surgery reported no postoperative symptoms.

Table VI. Location of JNA recurrence in the four patients treated at the Department of Otolaryngology and Laryngological Oncology, Poznan, Poland, 2000-2008

Patient number	1 st recurrence	2 nd recurrence	3 rd recurrence
1	Nasopharynx, maxillary sinus		
4	Pterygopalatine fossa, buccal fossa, temporal fossa	Minimal intracranial invasion	
5	Nasopharynx, sphenoid sinus, pterygopalatine fossa		
9	Nasopharynx, sphenoid sinus, pterygopalatine fossa	Pterygopalatine fossa, buccal fossa, temporal fossa	Massive erosion of skull base

Table VII. Number of JNA recurrences by surgical technique at the Department of Otolaryngology and Laryngological Oncology, Poznan, Poland, 2000-2008

	Open surgery		Endoscopic surgery		Endoscopic-assisted open surgery	
Total (n = 15)	9	60%	4	27%	2	13%
Recurrences	4	26%	0	0%	0	0%

The duration of the hospitalization period ranged from 4 to 7 days and 2 to 3 days in open surgery and endoscopic surgery groups, respectively.

Eight patients (#4, #9, #10, #11, #12, #13, #14, #15) underwent preoperative angiography with embolization of the maxillary artery branch, the ascending pharyngeal artery, or the internal carotid artery. The only post-procedural complication was post-embolization headache. Intraoperative blood loss in patients undergoing preoperative embolization decreased 10-fold.

Discussion

Surgical management of juvenile nasopharyngeal angiofibroma is associated with the following difficulties: risk of intraoperative bleeding, large tumour size, difficult location, and risk of recurrence. In light of these challenges, the surgical technique should only be selected after a thorough assessment of the extent of the tumour. The extent of JNA, graded according to one of the scales proposed in the literature, should be determined by a thorough analysis of the imaging studies available (contrast-enhanced CT and MRI), with particular focus on the spatial relationships between the tumour and important anatomical structures.

In this paper we have reported on the experience of the Department of Otolaryngology and Laryngological Oncology in Poznan, Poland, over the past eight years with the surgical management of JNA using traditional open surgery, endoscopic surgery, and endoscopic-assisted open surgery. During the period under analysis, we introduced embolization, and endoscopic procedures and equipment navigated on the basis of three-dimensional images (CAS, IGS).

Of the traditional external approaches, we used lateral rhinotomy, Denker's method and midfacial degloving (Table III). Classic surgery was performed in 4 (50%) of the 8 (54%) small JNAs and 5 (71%) of the 7 (46%) large JNAs (Table IV). According to the literature data, open surgery ensures the best exposure of JNA located in the nasopharynx or the medial infratemporal fossa, and it is therefore reserved for tumours that have not extended intracranially [18]. However, our experience suggests that the traditional technique may also be used for tumours with minimal intracranial extension (patient #4, stage IIIA). Like Danesi *et al.* [15], we observed

that midfacial degloving was more beneficial than lateral rhinotomy because it avoids postoperative scars.

The endoscopic technique has been used in our Department for two years and we employed it on 4 patients (50%) with small JNAs. Recent reports indicate that there is an increasing trend to use endoscopic management for large JNAs [19-25]. Only a few authors have experience with extensive JNAs. Mann *et al.* [16] indicate that they are in favour of using endoscopic surgery for Fisch stage III JNA, but fail to provide details. Onerci *et al.* [19] analyse 12 of their own cases, 4 of which were Radkowski stage IIIA tumours. In half of these cases, the tumour was not removed because it had invaded the cavernous sinus. Wormald and Van Hasselt [18] also report their experience with endoscopic surgery, but none of the 7 cases of JNA were classified as Radkowski stage IIIA or higher. Nicolai *et al.* [20] report on 15 cases of JNA (including 4 cases of Fisch stage III) treated with endoscopic surgery alone, and Roger *et al.* [22] present 20 cases of JNA, including 9 cases of Radkowski stage IIIA, treated using the above method. Both these reports state that in order to prevent potential recurrence, it is essential to debride thoroughly the base of the sphenoid bone using a drilling technique. It seems logical that the endoscopic technique is not appropriate for large JNAs that extend intracranially, laterally from the cavernous sinus or posteriorly from the alae of the sphenoid bone [20, 21, 23].

Like Nicolai *et al.* [20], we believe that the major advantages of endoscopic surgery include: extension of the picture, visual access at various angles, avoidance of scars that would impair growth of the cranial bones, and shortening the duration of surgery. In the event of manipulation difficulties, enlisting a second operator considerably improves the surgical conditions (the "four-hand technique" has been published, by us, in the literature) [23-25]. The option of converting from the endoscopic to the open technique should be considered and explained preoperatively to the patient.

The combination of open and endoscopic surgery (endoscopic-assisted open surgery) provides a very detailed view of the surgical field and makes it possible to remove advanced JNA tumours. Our experience with the endoscopic-assisted technique is limited to two patients (patient #11, stage IIB; patient #14, stage IIC). However, like Herman *et al.* [10], we

believe that endoscopic-assisted surgery reduces the risk of tumour recurrence.

We observed four recurrences of JNA during the mean post-op follow-up period of 4 years and 6 months. All the recurrences were observed in patients with large tumours and accounted for 44% of the JNA cases that involved open surgery (Table V). Our results parallel the international literature, where tumour recurrences account for 13 to 50% of all cases of JNA patients undergoing open surgery [3, 10, 26]. Liu *et al.* [27] point to incomplete resection as the source of recurrence, which in their opinion proves it is necessary to perform complete resection. On the other hand, Danesi *et al.* [15] and Herman *et al.* [10] accept the residual tumour concept and advocate a “wait and watch” approach during the postoperative period.

No tumour recurrences were observed among the patients with small JNAs in our Department who were managed with endoscopic surgery and endoscopic-assisted surgery (Tables V and VII), which is consistent with the findings of other authors. Based on our data, we can conclude that the endoscopic technique is definitely superior to the open surgery technique in the case of small tumours. We hypothesise that the advantage results from achieving an extended, polygonal endoscopic view of the tumour and the surrounding structures [27]. There are reports in the literature of the endoscopic technique being used for large JNAs with intracranial extension [19, 21, 22, 28]. However, as already mentioned, these are isolated reports on a small number of patients with high recurrence rates. The question, then, is: What are the upper limits on endoscopic management of JNA? Perhaps further experience with the state-of-the-art image-guided endoscopic technique (CAS) and thorough control of local bleeding will allow us to raise the bar.

In the repeat surgeries performed at the Department of Otolaryngology and Laryngological Oncology in Poznan, Poland, the techniques were selected based on the extent of the recurrence (Table VI) and the equipment available. These techniques included the traditional approach, endoscopic approach, endoscopic-assisted approach and the Fisch type C infratemporal fossa approach (the latter surgery being performed outside our Department). It should be stressed that primary surgery (using the open technique) was not a contraindication for repeat surgery using the endoscopic technique (Table II).

There are numerous reports on preoperative embolization of afferent tumour blood vessels aimed at reducing intraoperative blood loss [29-31]. However, Lloyd *et al.* [26] point out that embolization makes it impossible to precisely delineate the tumour, which makes complete resection impossible and hence increases the risk of recurrence. Nevertheless, most authors, with whom we agree, perform embolization 24-48 h before surgery.

Conclusions

1. Selection of the surgical technique in juvenile nasopharyngeal angiofibroma depends on the extent of the tumour and the equipment available. Our experience shows that the endoscopic technique is very useful for selected small tumours (Radkowski stage IA to IIA) and open surgery and/or endoscopic-assisted open surgery is useful for large tumours (Radkowski stage IIB or higher). Repeat JNA surgeries, depending on the extent of recurrence, may be performed using the open technique, the endoscopic technique, or the endoscopic-assisted open technique.
2. The endoscopic technique, as compared with the open technique, results in a shorter hospitalization period and lower number of complications. Endoscopic surgery is an alternative to open surgery in the case of small JNAs.
3. Preoperative embolization significantly reduces intraoperative blood loss.

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