Endoscopic approach for excision of juvenile nasopharyngeal angiofibroma: complications and outcomes

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Abstract

The purposes of this study are to report the efficacy of the endoscopic approach for juvenile nasopharyngeal angiofibroma (JNA) and to compare its related intra- and postoperative complications with findings from traditional approaches in the literatures. This study is a retrospective report of 47 cases of JNA that were treated with nasal endoscopic surgery between 1998 and 2005. According to the staging system by Radkowski et al (Arch Otolaryngol Head Neck Surg. 1996;122:122–129), the staging of the included patients were the following: 21 in stages IA to IIB, 22 in IIC, 3 in IIIA, and 1 in IIIB. Five patients were embolized before surgery. The mean blood hemorrhage in embolized patients was 770 mL, whereas in nonembolized patients, it was 1403.6 mL. In the follow-up period (mean, 2.5 years), the recurrence was found in 9 patients (19.1%), and mean time of recurrence was 17 months after surgery. The rupture of cavernous sinus occurred in 2 cases with no mortality. The mean hospital stay was 3.1 days in all cases and 1.8 days in embolized patients. The findings of this study demonstrate that endoscopic resection of JNA is a safe and effective technique because of decrease in blood loss, hospitalization, and recurrence rate, especially in tumors that are not extended through intracranial space. It is therefore strongly recommended that this modality is implemented as the first surgical step for tumors with stages I to IIIA of the Radkowski’s staging system.

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1. Introduction

Juvenile nasopharyngeal angiofibroma (JNA) is a histologically benign tumor or tumor-like lesion [1] that has local invasions. It is a vascularized lesion without any capsule. Juvenile nasopharyngeal angiofibroma is almost always seen in males, and its peak age is around the adolescence age groups. Although this tumor is the most common benign neoplasm of the nasopharynx, some existing reports indicate the presence of angiofibroma in nasal septum, middle turbinate, hard palate, and alveolar ridge [2-5]. It is responsible for less than 0.05 percent of all head and neck tumors [6]. The tumor usually originates from the superior border of the sphenopalatine foramen, which is formed by the junction of the trifurcation of palatine bone, horizontal wing of vomer, and the roof of pterygoid plates [7]. The origin of the tumor is of the greatest importance because it elucidates its pattern of distribution. In addition, it affects the surgical approach by which it will be managed. Initially, the tumor grows in the submucosal plane of the nasopharyngeal roof, reaching the septum and posterior aspect of nasal space, creating a mass that may cause nasal airway obstruction. As the tumor grows, the anterior face of sphenoid sinus is affected and becomes invaded by the tumor. Angiofibroma may laterally extend to the pterygomaxillary fissure and cause bowing of the posterior wall of the maxillary sinus. Further, it may involve the infratemporal fossa and middle cranial fossa. The simultaneous involvement of bilateral
sphenopalatine foramen by JNA is also reported [8]. The diagnosis of JNA is essentially based on clinical and radiologic examination [7]. During initial evaluation, computed tomography (CT) scan with and without contrast reliably assesses tumor extensions. Magnetic resonance imaging studies, however, may be more accurate than CT in assessing the intracranial extensions. The preoperative angiography is also helpful for the evaluation of feeding vessels and also allows embolization of these vessels. Although radiotherapy, hormone therapy, cryotherapy, electrocoagulation have all been recommended in the literature, surgery remains the treatment of choice for JNA [6]. The conventional surgical approaches including transpalatal, lateral rhinotomy, midfacial degloving, transmandibular, transhyoid, with or without extensions such as the upper lip split or concomitant craniotomy and infratemporal fossa approaches, have been examined. The management of nasopharyngeal angiofibroma has always been regarded as a formidable challenge to endoscopic surgeons as well as head and neck surgeons [9]. Kamel et al [10], in 1996, described transnasal endoscopic approach for the limited angiofibroma lesions. In that study, the eligible tumors for endoscopic surgery were limited to tumors of nasopharynx, nasal cavity, ethmoid, and sphenoid sinuses. Based upon the recent findings reported in the literature, not only is endonasal surgery combined with a preoperative embolization of the arterial supply suggested for small- and middle-size JNAs, but it is also used for large tumors extending to the pterygopalatine fossa and medial aspect of the infratemporal fossa. Note that minimal intracranial extension is not an absolute contraindication [11]. The endoscopic resection provides several advantages over more traditional surgical techniques, which include the avoidance of facial incisions and plating of the maxilla and the minimization of bone removal. These maneuvers may result in facial growth asymmetry when performed on adolescents. The endoscopes permit a multiangle and magnified view of the tumor and surrounding structures [12]. The purposes of this study are to report the efficacy of the endoscopic approach for JNA at all stages of Radkowski’s classification (see Table 1) [13] and to compare the related intra- and postoperative complications with findings from similar studies and other approaches in the literatures. This study is a retrospective report of 47 cases of JNA that were treated with nasal endoscopic surgery for a given period, as discussed in the following sections.

2. Materials and methods

Between August 1998 and May 2005, 47 patients with JNA who had undergone exclusive endoscopic surgery or endoscopic-assisted surgery at the Tertiary Department of Otolaryngology, Head and Neck Surgery of Amiralam hospital were studied.

At first, patients with JNA in various stages were selected; however, once a patient with a significant bleeding in stage IIIB was encountered, similar patients with advanced intracranial involvement were referred for radiotherapy.

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Table 1

<table>
<thead>
<tr>
<th>Stage</th>
<th>Description</th>
</tr>
</thead>
<tbody>
<tr>
<td>IA</td>
<td>Involvement limited to the nose and/or nasopharynx</td>
</tr>
<tr>
<td>IB</td>
<td>Extension into one or more sinuses</td>
</tr>
<tr>
<td>IIA</td>
<td>Minimal extension into the pterygopalatine fossa</td>
</tr>
<tr>
<td>IIB</td>
<td>Occupation of the entire pterygopalatine fossa with or without erosion of the orbital apex</td>
</tr>
<tr>
<td>IIC</td>
<td>Involvement of the infratemporal fossa with or without extension to the cheek or posterior to the pterygoid plates</td>
</tr>
<tr>
<td>IIIA</td>
<td>Erosion of the skull base (the middle cranial fossa/base of the pterygoids); minimal intracranial extension</td>
</tr>
<tr>
<td>IIIB</td>
<td>Erosion of the skull base; extensive intracranial extension with or without cavernous sinus invasion</td>
</tr>
</tbody>
</table>

Fig. 1. (A) Preoperative contrast-enhanced coronal paranasal sinus CT scan of the patient with stage IIIB JNA. (B) Computed tomography scan was taken about 2 years post endoscopic operation of the same patient.
Neurosurgeons were always present when patients of stage IIIA or IIIB tumors were being operated on.

All patients who underwent JNA endoscopic surgery were evaluated endoscopically in 2 weeks after surgery, then monthly, for the first 6 months, and annually afterward. An annual CT scan evaluation had begun since the third month postoperation. The mean follow-up period for each patient was about 2.5 years, and all subjects were examined with both endoscopic techniques and CT scan, as noted earlier. If an unusual situation was encountered during endoscopy while these patients were observed for recurrence, the findings were confirmed with a contrast-enhanced CT-scan imaging. The diagnosis of JNA was based on the clinical manifestations and the data obtained from CT scan imaging (Fig. 1), and sometimes with or without magnetic resonance imaging.

Among 47 patients who were included in our study, 31 (66%) cases were primarily treated (see Table 2), but the remaining 16 (34%) cases were treated secondarily in our center (see Table 3), which were previously operated on, using conventional or endoscopic methods.

The surgical technique used in these 47 cases was mainly based on exclusive endoscopic (in 43 cases) or endoscopic-assisted methods (in 4 cases), as described below:

After the patient became generally anesthetized in a hypotensive method, the tumor bulk was pushed to the nasopharyngeal space using a meticulous mechanical dissection. This was done with a medium-size freer and long meshes soaked with adrenalin solution. When progressing toward the nasopharyngeal area, every accessory division of the tumor was handled exclusively. For example, the tumor extension toward pterygomaxillary fissure was approached via removing the posterior wall of the maxillary sinus and force insertion toward pulling it out of that region with a large size endoscopic forceps. If there were more extensions lateral to the pterygomaxillary fissure and/or infratemporal fossa, it was necessary to assist intranasal forces with a lateral to medial directed force through an incision made in the posterior part of the ipsilateral gingivobuccal sulcus. Force was applied by fingertip through the incision line. The intrasphenoidal segment of the tumor was also another challenge. Surgicel

### Table 2

**Patients who were primarily treated with endoscopic surgery**

<table>
<thead>
<tr>
<th>Patient</th>
<th>Age</th>
<th>Symptoms and signs</th>
<th>Tumor location</th>
<th>Tumor stage*</th>
<th>Cranial nerve injury</th>
<th>Hospitalization days</th>
<th>Follow-up (mo)</th>
<th>Embolization</th>
<th>Intraoperative bleeding (mL)</th>
<th>Transfused blood (U)</th>
<th>Tumor recurrence</th>
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<td>–</td>
<td>5</td>
<td>70</td>
<td>–</td>
<td>750</td>
<td>1</td>
<td>–</td>
<td>–</td>
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<tr>
<td>2</td>
<td>14</td>
<td>NO + E</td>
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<td>2</td>
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<td>3</td>
<td>+</td>
<td>–</td>
</tr>
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<td>1</td>
<td>70</td>
<td>+</td>
<td>300</td>
<td>–</td>
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<tr>
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<td>52</td>
<td>–</td>
<td>1500</td>
<td>4</td>
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<tr>
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<td>Left IIIA, II, III, VI</td>
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<td>16</td>
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<td>16</td>
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<td>–</td>
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<td>–</td>
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<td>21</td>
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<td>16</td>
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<td>40</td>
<td>–</td>
<td>1000</td>
<td>2</td>
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<td>22</td>
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<td>300</td>
<td>–</td>
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<td>300</td>
<td>–</td>
<td>+</td>
<td>–</td>
</tr>
<tr>
<td>24</td>
<td>10</td>
<td>NO + E</td>
<td>Left IIC</td>
<td>–</td>
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<td>750</td>
<td>1</td>
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<tr>
<td>25</td>
<td>12</td>
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<td>Left IIC</td>
<td>VNP</td>
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<td>7</td>
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<td>–</td>
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<td>26</td>
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<td>NO + E</td>
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<td>–</td>
<td>1250</td>
<td>3</td>
<td>+</td>
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<tr>
<td>27</td>
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<td>Left IB</td>
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<td>28</td>
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<tr>
<td>30</td>
<td>21</td>
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<td>15</td>
<td>–</td>
<td>4500</td>
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<tr>
<td>31</td>
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<td>Left IB</td>
<td>–</td>
<td>3</td>
<td>26</td>
<td>–</td>
<td>300</td>
<td>–</td>
<td>–</td>
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</tr>
</tbody>
</table>

NO indicates nasal obstruction; E, epistaxis; H, headache; PND, posterior nasal discharge; VNP, vidian nerve problem (ie, xerophthalmia); AION, anesthesia in the territory of infraorbital nerve.

* Radkowski’s staging.
meshes were used when encountering massive cavernous bleeding. The anesthetic team was also ready to manage extensive bleeding. After the main parts of the tumor were moved into the oropharynx and nasopharyngeal area, it was removed completely by using a mouth gag instrument and a large-size forceps intraorally. Insulated suction electrocautery and forceps were used for establishment of an acceptable hemostasis. These tumors were never resected in a piecemeal manner. At the end of the operation, a search was conducted for any residual tissues and abnormal bleeding sites that could denote remnant disease. Prophylactic drilling of the clivus or pterygoid root and sphenoid diploe was also performed on patients who had obvious bone involvements in these locations.

Among 47 patients that underwent endoscopic surgery, 2 (4.2%) cases were operated on by the combined approach of “endoscopy and transpalatal open surgery”; 2 (4.2%) other cases were operated on by the combined surgical approach of “endoscopy and ipsilateral gingivobuccal incision surgery”; and 43 cases (91.6%) were operated on exclusively by the endoscopic approach. The histopathological examination after surgery revealed angiofibroma in all patients. The mean follow-up was 33.1 months, with a range of 27 to 98 months.

3. Results

3.1. Symptoms

The most common complaint was nasal obstruction that we observed in 43 patients (91.5%). The second most common symptom was epistaxis, which occurred in 26 cases (55.3%). Visual acuity disturbances and proptosis were found in combination with nasal obstruction and epistaxis in 1 patient (2.1%). One patient had tinnitus, another case had purulent postnasal discharge, and finally, facial swelling occurred in 4 cases (8.5%).

3.2. Age and sex

The age criterion was the age at which the patient was first recognized to have JNA. Mean age was 17.1 years. All patients were male.

3.3. Staging

The patients were categorized according to Radkowski’s classification (see Table 1) [13]. The staging of the included patients were the following: 21 in stages IA to IIB, 22 in IIC, 3 in IIIB, and 1 in IIIB.

3.4. Hospitalization

Mean hospital stay was 3.1 days for all cases and 1.8 days for the patients with embolization. The time at hospital was 1 to 2 days for 24 cases (51.1%), 3 to 5 days for 20 cases (42.6%), and more than 5 days for 3 cases (6.4%). The last 3 cases, among them the oldest patient, had to be treated more than 5 days because of complications (postoperative pancreatitis, massive hemorrhage, and trauma to the optic nerve).

3.5. Blood loss and embolization

The criterion for postoperative hemorrhage was the occurrence of bleeding during the first 48 hours after endoscopic surgery. According to this definition, there were 5 cases (10.6%) of bleeding that were controlled with supportive measures such as ice packs, elevation of the head of the bed, and parenteral rehydration and topical decongestants. The mean blood loss in all patients was 1336.2 mL; in stages IA to IIC, 890 mL; in stage III, 3450 mL. Thirty-two
patients (68.1%) required blood transfusions during the operation. A total of 169.2 blood units (200 mL blood in each unit), of which 74.2 U (43.9%) was of autologous sources and 95 U (56.1%) was in the form of packed red cells, transfused intraoperatively.

Five patients (10.6%) were embolized, whose mean blood loss was 770 mL. In 2 cases (4.2%), temporary postembolization complications were observed. One patient had a decrease in visual acuity in the left eye, and another one had left hemifacial and parotid swelling in combination with left eye chemosis and decrease in left eye visual acuity. Both cases recovered completely. Forty-two patients (89.4%) were not embolized and had a mean hemorrhage of 1403.6 mL compared with 770 mL blood loss in the embolized patients. There was no postoperative hemorrhage observed in the 5 embolized patients. Among those 5 cases, 1 was at stage IIA, 2 at stage IIB, 1 at stage IIC, and 1 at stage IIIA.

3.6. Other complications

In 2 patients (IB and IIA), unilateral cheek paresthesia was observed ipsilateral to the side of surgery postoperatively. In 2 (4.2%) cases (IIA, IIC), a reduction in lacrimation was found on the side of the endoscopic surgery. The transient damage to cranial nerves III and VI and a decrease in visual acuity on the side of surgery were seen in 1 patient (IIIA), who recovered completely later on. Overall, there were 5 (10.6%) cases with either cranial or peripheral nerve injury, among which only 1 patient (IIIB) had a serious damage to the cranial nerves with an acceptable recovery after 1 year. There were 2 cases with cavernous sinus injury, 1 with stage IIIA and other with stage IIIB. No dural or meningeal injury or brain abscess happened in these series of patients. In 13 cases (27%), nasal synchia was found postoperatively, which was treated endoscopically in an ambulatory setting.

3.7. Tumor recurrence

During the mean duration of 33.1 months follow-up, 9 patients (19.1%) had recurrence.

4. Discussion

The endoscopic surgery in the nasal cavity and paranasal sinuses has been primarily used to cure nonneoplastic diseases. Along with evolution of surgeon’s skill in endoscopic operations, these approaches take into the consideration for surgical approach to the nasal neoplasms including JNA.

Seldom does this tumor have any connections to the surrounding structures. The availability of the nutrient vessels of JNA and the ability to search for the places where the remnants of this tumor can potentially lead to recurrence are increased by endoscopic techniques [14]. This approach, if meticulously carried out, is therefore considered a very effective way to treat JNA, and as such, it causes less disability in patients, decreases the duration of hospital stay, and lowers the intraoperative bleeding rate. Other advantages of endoscopic approach is least morbidity, including avoiding any surgical scar on the face, resecting the least amount of normal soft tissue, and avoiding the destruction of facial bones and late facial deformities [12,15,16].

In this report, a study of 47 patients with a mean follow-up period of 33.1 months had been carried out. The maximum number of patients in case series reported thus far has been only about 20 cases, with a mean follow-up period of 22 months [17].

In this section, general data and the comparison of primarily and secondarily treated patients will be discussed.

Mean age of the patients in this study is 17 years. In the literature, the most common age at which JNA occurs is between 10 and 25 years. Even in ages more than 25 and less than 7 years, atypical JNA is considered [18].

Endoscopic surgery plays a great role in reducing the intraoperative bleeding because the intraoperative hemorrhage that occurs in open surgeries is to a large extent due to the incisions that are made to approach the tumor rather than the extraction of the tumor itself [16].

This study demonstrates that the intraoperative hemorrhage level will not be high in patients especially in whom the tumor has little invasion inside the cranium. The mean blood transfusion in stages IA to IIC was acceptable (890 mL), compared to 1000 to 1500 mL in previous studies [19].

4.1. Role of embolization

In this study, in embolized cases, a significant amount of declining in the hemorrhage level (45.5%) was observed, especially when compared to the mean hemorrhage level of the remaining 42 cases (770 vs 1403.6 mL, respectively). Regarding literatures, the average intraoperative hemorrhage level in endoscopic surgeries performed on embolized patients is reported less than 770 mL, that is, 350 mL and 372 mL [16,17]. In addition, embolized patients had both less hospitalization days (1.8 vs 2.2 days) and less postoperative hemorrhage rate (0% vs 11.9%) significantly. Regarding the studies by Lloyd et al [20] and Borghesi et al [21], the risk of incomplete excision of JNA in the setting of preoperative embolization is higher. In addition, the postembolization complications were encountered in 2 of 5 embolized patients; so, we did not persist in performing arterial embolization in all of our cases.

4.2. Cranial nerve injury

In this study, there was 1 case (stage IIIA) of serious damage to the cranial nerves II, III, and VI; 2 cases (4.2%) of hemifacial numbness; and 2 cases (4.2%) of decreased lacrimation. However, in a long-term study on 44 JNA patients with open surgery for 11 years, most of the other complications such as infraorbital nerve dysesthesia were reported [22].
4.3. Dural injury and brain abscess

In this study, no cases of dural or meningeal injury and no case of mucocele or brain abscess were detected.

4.4. Cavernous sinus injury

In this study, 2 cases (4.2%) (stages IIIA and IIIB) experienced cavernous sinus damage. The operation was completed successfully, and both patients have survived. Because of the significant bleeding in the patient with stage IIIB, it is suggested to refer such patients with advanced intracranial involvement, especially in the region of cavernous sinus or encaement of internal carotid artery, for radiotherapy.

Juvenile nasopharyngeal angiofibroma is a tumor with predilection for pushing but rarely invading dura. It is believed that open surgical approach using an anterior cranial approach with microsurgical techniques is the method of choice for surgical management of such tumors with intracranial extensions [23]. It is believed by the authors that the endoscopic approach to these stages of JNA is still the preferred method because under the endoscopic magnification of the surgical field, meticulous dissection of the tumor from adjacent dura and cavernous sinus can be performed. It is under such conditions that if the surgeon decides to resect the tumor completely from the cavernous sinus, he or she can manage massive bleeding by endoscopic sealing the puncture site with Surgicel or any other autologous grafting materials. However, with or without endoscope, the surgeon must be ready to overcome a massive hemorrhagic burst; so, sufficient blood for transfusion and several large intravascular lines must be available before proceeding with surgery in this dangerous location.

4.5. Other complications

In the literature, there are reports of other complications such as lacrimal duct stenosis, secretory otitis media, diplopia [22], and sphenoid mucocel [24], but there was none encountered by the team in this study. One of the minor complications of all endoscopic surgeries is synchia formation, which occurs due to the irritations and abrasions in the confronting nasal mucosa and fusion of these injured walls together. In this study, there were 13 cases (27%) that experienced intranasal synechia formation and recovered with ambulatory endoscopic managements.

4.6. Recurrence of angiofibroma after endoscopic surgery

During the follow-up period, 9 patients (19.1%) experienced recurrence. The mean interval between the surgery and the relapse was 17 months. This percentage was ranging from 7% to 39.5% in other studies [23]. Pryor et al [25] reported overall recurrence rate of 24% in patients treated using conventional methods.

4.7. Comparison between the primarily and secondarily endoscopic surgical approaches to the JNA

As mentioned earlier, in this study, 31 (66%) patients without a previous history of surgery and 16 (34%) patients with previous surgery, which had been led to relapse, underwent an endoscopic surgery. There was no significant difference between the recurrence rates of JNA in 2 groups: 19.3% in the primarily treated patients vs 18.7% in the secondarily treated patients. There was a significant statistical difference of the postoperative hemorrhage level between the 2 groups (P = .027) that is in favor of less blood loss in the primarily treated group (see Table 4).

Another significant difference was in the hospitalization period between these 2 groups (see Table 5) (P = .049), which was in favor of a shorter hospital stay in the primarily treated group.

5. Conclusions

The main advancement in the treatment of JNA is the introduction of the endoscopic approach. This allows complete resection of the lesion with low morbidity.

Considering the results of this study, which contained the largest number of JNA cases that were managed with endoscopic surgical approaches, it becomes evident that intraoperative hemorrhage level, hospitalization duration, surgery complications, and even recurrence rate can be significantly reduced. The preoperative embolization of the patients, if done by an expert interventional radiologist, can dramatically decrease the intraoperative hemorrhage rate.

The authors believe that the endoscopic approach for the resection of JNA is the main surgical modality for tumors with stages I to IIIA of the Radkowski’s staging system.
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