The Surgical Strategy of Juvenile Nasopharyngeal Angiofibroma: A Case Report

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Case Report

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Abstract

Surgical resection to remove juvenile nasopharyngeal angiofibroma is challenging due to the high risk of massive hemorrhage. We report a comprehensive strategy which included preoperative devascularization by embolizing the feeding arteries and combined endoscopic and transoral approach was adopted.

Introduction

Juvenile nasopharyngeal angiofibroma (JNA) is an uncommon tumor which accounts for about 0.05% of all patients with head and neck neoplasms. Most reported cases are adolescent male patients aged between 9 and 19 years\(^1\). It originates mainly from the posterior part of the lateral wall of the nasal cavity, close to the superior border of the sphenopalatine foramen. JNA often progresses to fill the whole nasal cavity, eroding the maxillary sinus, and spread to the pterygoid region, invading the infratemporal fossa and the middle cranial fossa\(^2\). Surgical resection for JNA is the primary treatment while it is worth noting that the surgery for advanced JNA is challenging due to difficulties to control intraoperative bleeding\(^3\). Here we present an adolescent patient with advanced JNA and highlight the importance of comprehensive strategies in the treatment of JNA.

Case Presentation

Patient summary

A 14-year-old male weighing 45 kilograms presented to our otolaryngology clinic with recurrent epistaxis for the past 8 months. Epistaxis occurred frequently during sleep, flowing down the nasal cavity or into the oral cavity. He was forced to breathe through the mouth due to nasal obstruction. The result of incisional biopsy undertaken 2 months ago in the outpatient clinic showed juvenile angiofibromas. And then symptoms including bleeding and nasal congestion exacerbated. For the last 2 weeks he suffered from breathing difficulties while sleeping and felt pressure in the right ear. The 14-year-old’s general mental and physical health also deteriorated. He lost about 4kg in 2 months, and became depressed and socially withdrawn. Initially he refused treatment and was admitted to a local hospital for suicide attempt.

Examination and treatment

Anterior rhinoscopy revealed a reddish mass blocking nasal passage on the right side. Complete blood count was notable for hemoglobin (Hb) of 50g/L, hematocrit of 17.7%. Transfusion of 4 units of packed red blood cells (PRBC) was required due to severe anemia. Imaging examinations showed a tumor measuring 7.3*5.7*6.5 cm in the right nasal cavity and exhibiting heterogeneous enhancement, posterior invasion to the nasopharynx, as well as the pterygopalatine fossa (PPF) and the infratemporal fossa (ITF). (Fig. 1). There were multiple bony destructions, but no intracranial extension. The patient was diagnosed with JNA, and the angiofibroma was classified as Radkowski stage C, Önerci stage III. The
NSF-COR classification indicated that the tumor was stage NSF2R and recommended combined endoscopic and open surgical approaches. These approaches were adopted in this case.

Because of the advanced nature of the tumor, angiography employs superselective catheterization to adequately examine the tumor's blood supply and identify collateral vessels that may be dangerous for intracranial circulation. The vascular distribution to the tumor is provided by the internal maxillary branches of bilateral external carotid arteries (ECA), as well as a small contribution from the right internal carotid artery (ICA). Superselective bilateral embolization of the internal maxillary arteries (IMA) was performed to minimize collateral blood supply. Absorbable gel foam particles were used as embolic agent. However, both coils and gel foam particles were used for the right internal maxillary artery due to an arteriovenous fistula. After embolization, there were still a few arterial branches in the nasopharynx feeding the posterior part of the tumor. These vessels that originated from the ICA were not embolized. (Fig. 2)

The tumor was resected via sublabial Caldwell–Luc maxillectomy combine with an endoscopic resection under general anesthesia 48 hours after the embolization. Intraoral resection for tumor removal was performed, followed by an endonasal endoscopic resection and careful exploration of the Vidian canal and pterygoid process. No CT-guided navigation was utilized during the whole procedure.

Caldwell-Luc maxillectomy incision was conducted to open the anterior wall of the right maxillary sinus and expose the capsuled tumor. Oblation plasma technology was used to debulk the tumor and make a surgical space. The tumor capsule was separated from surrounding tissues with coblation enabling tumors to be removed in blocks from maxillary sinus, PPF and ITF. The right maxillary artery was identified and coagulated using bipolar forceps. Subsequently, only the mass in the nasal cavity was left. The tumor was debulked with Medtronic ENT power system, transected above the choanal and finally amputated from the nasopharynx mucosa. Attention was paid to protect the eustachian. Once the tumor was completely removed, it was pushed backward to the posterior nasal cavity and pulled out transorally. Afterwards the pterygoid process was drilled off, enabling probing of the Vidian canal. Intraoperative observation revealed the tumor related to the Vidian canal artery, suggesting that the tumor was also fed by this artery, and that most of the intraoperative bleeding came from the region at the posterior nasal cavity. The Vidian canal artery was coagulated using the coblator.

Total blood loss was 2000 mL. Eight units of PRBC were transfused in total during the 9-hour surgery.

Postoperative histopathology confirmed the diagnosis of JNA. After the procedure, the patient lost sensation in part of the right superior gum, which improved after removing the Bismuth iodoform paraffin paste (BIPP). The 14-year-old patient didn’t complain any discomfort during nasal endoscopy and nasal irrigation treatment. He was discharged from the hospital 10 days after operation. CT examination was carried out before discharge. Until now, the patient feels good and refuses to follow up.

The institutional review board approved publication of this case report and informed consent has been obtained from the patient's guardians, the case report and related images can be published.
Discussion

Although experienced teams with endoscopic technology and techniques can completely resect these tumors with minimal intraoperative bleeding and low rates of residual, some juvenile angiofibromas at later stages are challenging\(^{[3]}\). Considering the high risk of bleeding, in this case we assessed the patient's blood supply first through angiography.

Preoperative arteriography can be safely and effectively used as an adjuvant surgery to embolize the feeding artery before JNA surgical resection\(^{[4]}\). In the present study, preoperative digital subtraction angiography showed contributing vessels from bilateral ECA and involvement of right ICA. Based on the result, superselective embolization of both IMA. Minimal blush still remained after embolization, secondary to residual input from some small branches of the ICA, such as the Vidian canal artery.

In terms of surgical treatments, endoscopic excision is considered the first choice for early-stage tumors. As to more advanced tumors, endoscopic, endoscopic-assisted, or open surgical operations can be selected according to the invasion range of the tumor\(^{[3]}\). It is suggested that endoscopic plasma ablation is very suitable for stage I and stage II JNA tumors, and can reduce blood loss during surgery and shorten operation time\(^{[5]}\). In the present study, CT and MRI demonstrated a large, advanced tumor with significant expansion of the nasal cavity and extension into the ITF but no intracranial involvement. Combined endoscopic and sublabial Caldwell–Luc maxillectomy approach was selected. Application of plasma ablation at ITF and choanal was used for timely surgical debulking. At the same time, the auxiliary application of Medtronic ENT power system improved visualization and increased the space for surgical operation. With an experienced surgical team utilizing these techniques, the operation was performed uneventfully. Although the main feeding artery was embolized, a considerable amount of blood loss, most likely from the ICA, was recorded during the operation. This has raised some questions regarding the treatment of advanced JNA. For example, what percentage of intraoperative bleeding can be reduced by superselective embolization of ICA branches, and, how to make the right decision balancing between the volume of blood loss reduced by embolization and the multiple complications that embolization may cause.

Conclusions

Treatment in advanced JNA requires to formulate a comprehensive strategy. As shown in our case report, a personalized strategy was developed. Before all other measures, bilateral carotid angiography was necessarily undertaken to evaluate the vascular supply of JNA, and superselective embolization performed on both sides. Furthermore, plasma ablation is considered a better choice for early surgical debulking during operation. Finally, during advanced JNA treatment, it is important to pay attention on the patient's mental health and avoid biopsies if possible.

Abbreviations
Juvenile nasopharyngeal angiofibroma
pterygopalatine fossa
infra temporal fossa
hemoglobin
Bismuth iodoform paraaffin paste
internal maxillary arteries
internal carotid artery
external carotid arteries
Computed tomography
Magnetic resonance imaging

Declarations

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Authors’ contributions
YL and YP collected the clinical data. LSY refers to lots of literatures and drafted the manuscript. WYD and LSF provided imaging help. WYZ, SL, SSJ and CAP performed critical revision of the manuscript. The authors read and approved the final manuscript.

Availability of data and materials
All data generated or analyzed during this study are included in this published article.

Ethics approval and consent to participate
Not applicable.

**Consent for publication**

Informed consent has been obtained from the patient's mother for publication of the case report and associated images.

**Competing interests**

The authors declare that they have no competing interests.

**References**


**Figures**
Figure 1

Imaging examinations.

A tumor measuring 7.3*5.7*6.5 cm in the right nasal cavity and exhibiting heterogeneous enhancement, posterior invasion to the nasopharynx, as well as the PPF and ITF, with multiple bony destructions.
Figure 2

Preoperative angiography.

(A, B and C) Superselective catheterization followed by selective embolization with gel form particles of the left IMA as a supplying vessel.

(E, F and G) Identification of the dominant right IMA as well as an arteriovenous fistula, followed by embolization with gel foam particles and coils.

D and H. The carotid artery repeated angiography revealed a small amount of residual tumor staining in the right side.