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ORIGINAL ARTICLE

Juvenile Nasopharyngeal Angiofibroma Surgical Treatment in Paediatric Patients☆

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KEYWORDS
Juvenile angiofibroma;
Surgery;
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Abstract
Introduction: The juvenile nasopharyngeal angiofibroma is a highly vascularised benign neoplasm of complex treatment in its surgical preparation, surgery to be performed, risks and recurrences.
Objective: The aim of the study was to determine the management and surgical treatment for the pathology of juvenile nasoangiofibroma.
Materials and methods: We reviewed the clinical histories and images of the patients who underwent surgery with a pathology result of juvenile nasoangiofibroma in the period from January 2008 to December 2016.
Results: Sixty-one cases were treated; all of them treated using the same surgical access by means of a Le Fort I osteotomy. All of the patients were male, with an average age of 13.3 years. The Andrew–Fish classification was used for staging the cases, most were staged as grade II and I.
Conclusions: The described approach provided extensive surgical access, which was adequate for the different stages of the tumour. It requires experience to be able to resect the tumour with the least possible bleeding.

PALABRAS CLAVE
Nasoangiofibroma;
Cirugía;
Le Fort

Tratamiento quirúrgico del angiofibroma nasofaríngeo juvenil en pacientes pediátricos

Resumen
Introducción: El nasoangiofibroma juvenil es una neoplasia benigna sumamente vascularizada de tratamiento complejo tanto en su preparación quirúrgica, cirugía a realizar, riesgos y recurrencias.

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Objetivo: Determinar el manejo y tratamiento quirúrgico para la patología de nasoangiofibroma juvenil.
Materiales y métodos: Se revisaron las historias clínicas e imágenes de los pacientes intervenidos por la especialidad de cirugía de cabeza, cuello y maxilofacial con resultado de patología compatible con nasoangiofibroma juvenil, en el periodo de enero 2008 a diciembre del 2016.
Resultados: Se intervinieron 61 casos, todos ellos tratados con el mismo acceso quirúrgico por medio de una osteotomía Le Fort I. La totalidad de los pacientes fue del sexo masculino con un promedio de edad de 13,3 años. Se utilizó la clasificación de Andrew-Fish para la estadificación de los casos. Obteniendo el grado II y I la mayor cantidad de casos.
Conclusiones: El abordaje descrito provee un acceso quirúrgico extenso, el cual es adecuado para los diferentes estadíos de la tumoración. Requiere de experiencia para poder llevar a cabo la resección de la tumoración con el menor sangrado posible.

Introduction

Juvenile nasoangiofibroma or juvenile nasopharyngeal angioifibroma is a benign neoplasm which begins in the sphenopalatine hole, found in the nasopharynx where the sphenopalatine artery is located, which is the terminal branch of the internal maxillary artery. The literature describes its presentation in males during puberty, although cases have been reported of younger ages than puberty and exceptionally in females.[1–14]

This type of tumour is highly vascularised due to its origin in the sphenopalatine artery. Its growth is predominantly asymptomatic and varies depending on the size of the tumour. Clinical symptoms of patients are advanced stage tumours, initially with nasal obstruction, generally unilateral and with episodes of profuse nasal or oral cavity bleeding. It grows through the ostium and invades the paranasal cavities, the oropharynx, the infratemporal fossa and then the pterygomaxillary fossa, the orbit through orbitary fissues and the middle skull base, compromising the cavernous sinus, the optic chiasm and the pituitary fossa, even extending to the anterior cranial fossa which in these cases would be intracranial but extra-meningeal. As a result, facial deformity, exophthlamos, reduction of visual acuity and headaches occurs.[3–8]

This is the most common of nasopharyngeal tumours and represents from .05% to .5% of head and neck tumours, with a presentation of 1/150,000 males aged between 10 and 24 years, with an average of 15 years. This tumour is benign, although due to its growth in the paranasal cavities it is usually bone destructive.

Nasal obstruction is the most common symptom (91%), followed by epistaxis in 63% of cases and intracranial compromise, facial deformity and ocular proptosis in more advanced stages. Intracraneal extension presents in between 20% and 36% of cases and is usually extradural.[8–20]

There are several classifications: that of Fisch and its modification with Andrews are the most commonly used, together with the Chandler classification (Table 1).

Treatment is mainly surgical. Preoperative embolisation, in view of arterial irrigation, is currently used as standard to reduce bleeding during the operation due to the vascular nature of the tumour, and this is highly favourable for reducing intraoperative bleeding. Reports on its usage vary between 24 and 48 hours prior to surgery, and up to 3–7 days prior to surgery.[5–13]

Several surgical approaches have been described over the years, with the most reported being the transpalatine route, the midface degloving route, Le Fort I osteotomy and the endoscopic route. Endoscopic surgery has been used for stages I and II due to the smaller size of the tumour, although it is now combined with open surgery and for treatment of residual tumour and/or relapses of advanced stages, depending on the spread and size of the tumour.[8–24]

No technique is exempt from complications and sequelae. All carry risks because of the nature and location of the tumour.[16–33]

Although intracranial extension is common, intradural involvement is infrequent, with a plane existing between tumour and dura mater, which is dissectible. If there is still compromise at dura mater level its reconstruction is imperative in the same surgical intervention.[15–25]

Radiotherapy is only used for residual post-surgical conditions or tumours which are not surgically resectable, and which due to their location may compromise vital structures. The following complications must also be borne in mind: panhypopituitarism, delayed growth, temporal lobule necrosis, cataracts, blindness, malignancy of the tumour or skin cáncer.[22]

Recurrence is reported in 6%–50% of cases, with variation depending on the surgical technique. Generally, between 6% and 7% for stages I–II and between 40% and 50% for stages III–IV. It is in stages III and IV where recurrence is greater due to the difficulty in handling tumour size.[26–30]

Materials and Methods

Study design was observational, descriptive and retrospective. The medical histories of all the patients operated on

Table 1 Chandler and Andrews–Fisch Classifications for Nasopharyngeal Angioifibroma.

Chandler Classification	
Stage I	Tumour limited to nasopharynx.
Stage II	Tumour extension to nasal cavity and/or sphenoids.
Stage III	Tumour extension to one or more of: antre, etmoids, pterygomaxillary fossa infratemporal, orbit or cheek.
Stage IV	Extension to cranial cavity.
Andrews–Fisch Classification	
Stage I	Tumour limited to the nasal cavity and/or nasopharynx.
Stage II	Tumour extension to the pterygopalatine fossa or maxillary, ethmoid or sphenoid sinuses, igopalatine or maxillary, ethmoid or sphenoid sinuses.
Stage IIIa	Extension into infratemporal fossa or orbit without intracranial compromise.
Stage IIIb	Extension into infratemporal fossa or orbit with extradural (parasellar) intracranial involvement.
Stage IVa	Intracranial tumour without infiltration of the cavernous sinus, optic chiasm or pituitary fossa.
Stage IVb	Intracranial tumour with infiltration of the cavernous sinus, optic chiasm or pituitary fossa.

Sources: Refs.[34,35]

for juvenile nasoangiofibroma were reviewed from January 2008 to December 2016.

Inclusion criteria was to have undergone surgery for juvenile nasoangiofibroma, as confirmed by the result of pathological anatomy and exclusion was all patients who had not been operated on, with an illness which did not correspond and medical files which did not contain sufficient data for an appropriate or complete record of the cases.

Results

The clinical records of 61 surgical interventions on 55 patients were reviewed, where 5 were relapses of the condition and one case was operated on for a different diagnosis, resulting in surgery for nasoangiofibroma.

100% of cases corresponded to males, with an average of 13.3 years and a mean of 12 years. With 33 cases (54%) between 12 to 14 years, 18 (30%) from 15 to 17 years, 8 cases (13%) between 9 and 11 years and only 2 cases (3%) under 9 years of age.

Nasal obstruction was the most common symptom, followed by epistaxis and palatine ballooning in 92%, 56% and 43% of cases respectively, with facial deformity and reduction of visual acuity in 13% and 8% each. Exophthalmos presented in 5% of the total and one case was asymptomatic (2%).

With regard to location and extension of the nasoangiofibroma the nasopharynx was compromised totally in all cases (Table 2).

According to the Chandler classification stage III was the highest presentation followed by stage II, whilst in the Andrews–Fisch classification stage II and I were the stages with the most cases (Table 3).

The total average of blood loss was 818 cc per intervention, with 2.8 units of globular transfused units to the patient and with preoperative haemoglobin of 12.8 g% and postoperative on the seventh day of 11.5 g%.

Save in one case, where the patient was admitted to hospital with an orbital tumour and exophthalmos, who was operated on with coronal access with orbitonasal osteotomy and osteosynthesis, and later operated on for a residual tumour, all the patients were intubated submentally which consisted in inserting a ringed endotracheal tube with a balloon orally, and later a submental incision was made in the midline and at 1–1.5 cm behind the posterior edge of the chin, and with blunt dissection a haemostats clamp was removed through the floor of the mouth at lingual frenulum level, the tube balloon was deflated, externalized towards the neck to be inflated again, and the same procedure was

Table 2 Involvement of the Craniofacial Cavities.

Extension	N.°	%
Nasopharynx	61	100
Nasal cavity	58	95
Sphenoid sinus	32	52
Maxillary sinuses	30	49
Pterygomaxillary fossa	26	43
Oropharynxy	21	34
Zygomatic fossa	12	20
Orbital cavity	12	20
Genian region	9	15
Middle skull base	9	15
Ethmoid cell	2	3
Anterior skull base	2	3
Total	61	100

Table 3 Cases according to Chandler and Andrews–Fisch Classifications.

Chandler	N.°	%	Andrews-Fisch	N.°	%
Stage I	1	2	Stage I	18	30
Stage II	17	28	Stage II	20	33
Stage III	34	56	Stage IIIa	14	23
Stage IV	9	15	Stage IIIb	7	11
Total	61	100	Stage IVa	2	3
			Total	61	100

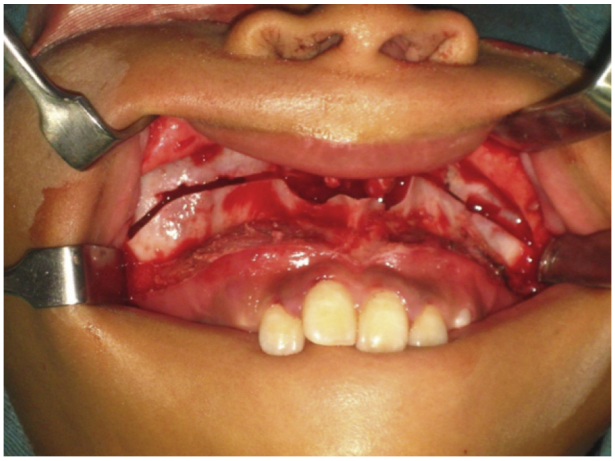

Figure 1 Le Fort I osteotomy.

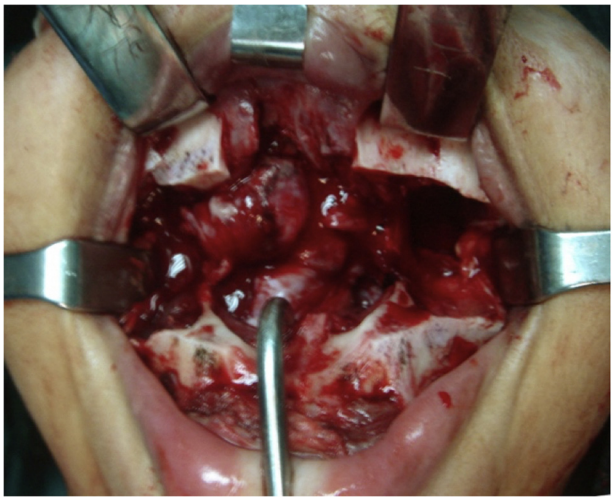

Figure 2 Le Fort block descent and tumour exposure.

repeated for the proximal end of the endotracheal tube; it was then sutured and the airway passage re-established.

Following this, with a superior vestibular approach, a Le Fort I osteotomy was performed (Fig. 1). This descends and impacts the mucosa of the nasal floor of the compromised side, allowing access to the nasal cavity and the paranasal sinuses (Fig. 2), resection of the tumour and review of the operating site (Fig. 3). After this the respective osteosynthesis is performed and anterior and posterior nasal packing. For the osteosynthesis several inputs are used, such as titanium plates and screws, resorbable material and surgical wire, with detailed reduction and bony attachment.

Osteosynthesis was performed with titanium plates and screws in 48 cases (79%), resorbable in 10 cases (16%) and surgical wire in 3 cases (5%) regardless of the staging of each case, so as to restore the anatomy of the third part of the midface region and dental occlusion. There were no major differences from using the different methods of bony attachment regarding deviation of the mid maxillary and mandibular line, poor consolidation, infection or growth of the facial skeleton and dental refill, with the upper jaw remaining stable with the methods used.

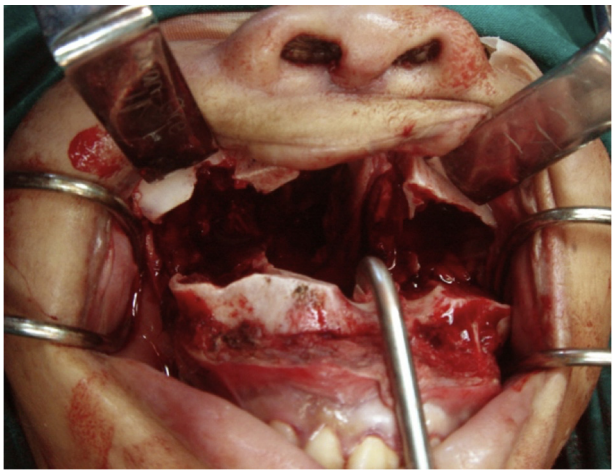

Figure 3 Nasal cavity and paranasal draining.

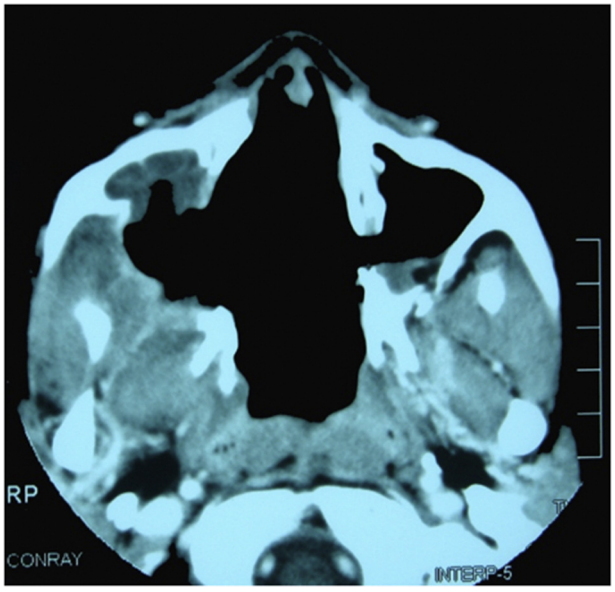

Figure 4 Tomography after surgery.

We used the Andrews–Fisch classification as a comparative parameter of the surgical procedure and for presentation of results.

Eighteen cases corresponded to grade I which presented with a blood loss volume of between 251 and 500 cc in 44% of cases, and required an average of 1.7 packed red blood cells per operation. Two cases required further intervention with surgery again being performed with a Le Fort I approach 7 and 13 months after primary surgery.

Twenty cases corresponded to grade II, with mean a blood loss of between 501 and 750 cc, and an average transfusion of 2.95 packed red blood cells per patient. One case relapsed after 4 months, and endoscopic resection was performed due to the yet small size of the tumour. There were no surgical complications in this group.

Fourteen grade IIIa patients were operated with a higher blood loss of between 751 and 1000 cc, and which required 3.1 packed red blood cells per patient. Again there was one relapse one year after surgery and a Le Fort I resection was then performed.

Table 4 Volume of Blood Loss, Number of Transfusions and Relapses According to Stages.

Andrews–Fisch	Grade I		Grade II		Grade IIIa		Grade IIIb		Grade IVa	
Surgical Bleeding	N.° Cases	%	N.° Cases	%	N.° Cases	%	N.° Cases	%	N.° Cases	%
0–250 cc	3	17	1	5	0	0	0	0	0	0
251–500 cc	8	44	5	25	1	7	1	14	0	0
501–750 cc	3	17	7	35	3	21	1	14	0	0
751–1000 cc	3	17	1	5	7	50	3	43	0	0
1001–1250 cc	1	6	6	30	3	21	2	29	1	50
1251 or above	0	0	0	0	0	0	0	0	1	50
Total	18	100	20	100	14	100	7	100	2	100
Transfusion	Cases	%	Cases	%	Cases	%	Cases	%	Cases	%
1 Packed red blood cells	9	50	2	10	1	7	0	0	0	0
2 Packed red blood cells	4	22	7	35	4	29	1	14	0	0
3 Packed red blood cells	5	28	4	20	3	21	3	43	0	0
4 Packed red blood cells	0	0	4	20	5	36	2	29	2	100
5 Packed red blood cells	0	0	3	15	1	7	1	14	0	0
Total	18	100	20	100	14	100	7	100	2	100
Cases With Relapses	N.° Cases	%	N.° Cases	%	N.° Cases	%	N.° Case	%	N.° Cases	%
Le Fort	2	11	0	0	1	7	0	0	1	50
Endoscopic	0	0	1	5	0	0	0	0	0	0
Total	18	100	20	100	14	100	7	100	2	100

For grade IIIb the highest blood losses obtained were between 751 to 1000 cc and from 1000 to 1250 cc in 3 and 2 cases respectively, increasing the average of packed red blood cells transfused to 3.4 per case and with no relapses in this category (Fig. 4).

Finally, in group IVa bleeding was higher in both cases, with 4 packed red blood cells being required in each case. Furthermore, in one case there was a relapse and the patient was operated on one year after surgery with the La Fort I approach. Radiotherapy was only used on this case, where the residual tumour was located at the anterior and mid skull base, with respective follow-up of the tumour through magnetic resonance, maintaining its size, and with no further symptoms or posterior growth.

Finally, total relapse for the treated disease was 10% for all cases, i.e. the 6 patients.

Regarding embolisation, 33 patients (54% of cases) were embolised for 7 days prior to surgery in 10 cases and 4 days in 5 cases, obtaining an average of 5.4 days prior to surgery. Twenty eight cases were not embolised (46%), 11 grade I and 10 grade II according to Andrews–Fisch classification.

There was one permanent facial paralysis postembolisation in a IIIa case and one transitory postembolisation blindness in a IIIb case which was resolved in 3 weeks.

The average haemoglobin level prior to surgery and 7 days after surgery for each grade was as follows: for grade I the average prior haemoglobin level was 12.6 g% and posterior level was 11.4 g%; for grade II it was 12.4 g% and 11.1 g% respectively. In the grade IIIa we had the presurgical average of 12.2 g% and posterior of 11.8 g%, with similar values for grade IIIb of 12.1 g% and 11 g% before and after intervention. Grade IV showed a greater difference with 12.5% prior haemoglobin level and 10 g% after surgery (Table 4).

Discussion

Juvenile nasoangiofibroma is a benign tumour with high vascularisation and as described in all publications with a predominance exclusively in males, except very sporadically in reported exceptions.

Similarly to the publications with this type of approaches, in the presented cases the Le Fort I osteotomy enabled us to access the nasal cavity, the paranasal sinuses, the nasopharynx, the oropharynx and also the genian region, the pterygomaxillary fossa, the zygomatic fossa and extradurally, the skull base, anatomical spaces which are involved because they are invaded by advanced stage tumours. Although access may therefore be gained to the medial and lateral regions of the face, this has the limitation that in the cavernous sinus, chiasma and pituitary region, which it is necessary to access for tumour resection and haemostasia, better access is gained with a craniotomy.

Due to the location of the tumour and the regions involved there is standard usage of intubation through the region underneath the chin so as to separate the airway for anaesthesia and ventilation of the patient from the surgical site. Few reports with this type of procedures have been found.

In initial stages prior embolisation may be omitted, but in more advanced stages there is greater use of embolisation, because the higher the stage, the larger the tumour and the heavier the bleeding during surgery. Protocol

dictates between 3 and 7 days as the ideal postembolisation time for performing surgery, and these times coincide with other similar studies. We should highlight that in 3 cases surgery had to be put forward due to spontaneous bleeding and in 3 cases delayed due to unexpected administrative complexities.

No significant differences were found regarding resorbable or non resorbable osteosynthesis material (titanium plates and screws or surgical wire) for postoperative stability of the Le Fort I segment and restoration of the facial skeleton anatomy, nor did any other complications present from the material used, the use of which had been reported in different studies for the treatment of fractures and/or bone reconstructions.

The administration of replacement red blood cells is necessary during surgery in all stages and particularly in advanced stage tumours.

With regards to complications, the patient with temporary postembolisation blindness had already presented with a reduction of visual acuity and compromise of the orbit with exophthalmos prior to surgery. The patient recovered on the seventh day after surgery. Radiotherapy used on one patient was because of a residual macroscopic disease at the anterior and middle skull base.

As described in the literature, relapse is always potentially present, and especially in stages above II, because no technique is exempt from relapse. Several reports for the same disease describe embolisation as standard in all patients who will undergo endoscopic surgery and in almost all of them for open surgery.

We consider that endoscopic surgery is highly indicated for stage I and II tumours, where due to the size and medial location of the tumour endoscopy is a less gruelling surgical technique with lower blood loss, but previous embolisation is required and is of great use for the removal of early detection tumour recurrences.

The technique we have described does not necessarily require an embolisation. It is also ideal for tumours in stages with extension towards lateral regions to the nasal cavity and nasopharynx.

Conclusions

Nasoangiofibroma mostly occurs in adolescence, as described in the literature.

Treatment of this condition is surgical, with prior embolisation being of great use, especially in stages III and IV. The management and prognosis of the disease is appropriate and similar to international reports, with open surgery being proposed for all stages, and no limitations derived from the need for embolisation.

It is important to consider the amount of blood products required to replace blood loss during surgery, particularly due to the vascular nature of the tumour.

Le Fort I osteotomy offers broad surgical access with viewing and direct palpation of the structures occupying the paranasal sinus, skull base, pterygomaxillary fossa, genial fossa and infratemporal fossa and above all with no extraoral scars. The resorbable materials present with the same characteristics for osteosynthesis as the non resorbable ones.

Recurrences were in keeping with the statistics reported for the different techniques used.

Radiotherapy is an option which should be assessed as additional treatment for residual disease with compromise of vital structures.

Conflict of Interests

The authors have no conflict of interests to declare.

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