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Case Report

A Rare Case of Isolated Schwannoma in Infraorbital Nerve with Aesthetic and Extracorporeal Zygomatic Bone Approach

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Abstract

Schwannomas of the head and neck are uncommon tumors that arise from cranial, peripheral, or autonomic nerves. In these cases, only seventeen cases of infra-orbital nerve schwannoma have been previously reported. We report a rare case of a large schwannoma arising from infra-orbital nerve in a female patient at 54 years old, eroding the anterior wall of maxillary sinus, involving the maxillary sinus, the floor of the orbit, the pterygoid fossa and extending into infratemporal fossa. Treated by surgery, with transconjuntival-transcanthal approach, and orbitectomy with extracorporeal zygomatic bone.

Keywords: *Schwannoma, infraorbitary nerve, periorbital surgery, extracorporeal zygomatic bone, rare cases.*

INTRODUCTION:

Schwannoma or neurilemoma is a well-differentiated and generally solitary benign tumor, characterized by proliferation of cells of Schwann from a peripheral nerve initially described in 1910 by José Verocay (1876-1927) [1,2,3].

This coined the term neurinoma [4] later in 1935, Arthur Stout coined the term neurilemmoma (5]. Friedmann finally, suggests that the term schwannoma should be preferred [6]. They originate from any nerve covered with schwann cell sheath. Schwannomas constitute 25–45% of tumors of the head and neck.

The trigeminal nerve is rarely associated with schwannomas; especially in the infraorbital nerve area [7]. About 4% of head and neck schwannomas present as a sinonasal schwannoma [8]. The symptoms primary is: sensation of retrocular pressure, proptosis and pain [9,10,11].

Malignant peripheral nerve tumors are rare, but they have a very high rate of recurrence and a low 5-year survival rate because of extension through the superior orbital fissure to the midbrain region.

Although countless cases have been reported, only 10 are of the infraorbital nerve are reported by Kumar [12]. Finally, we identified 6 articles published between 2015 and January 2025 [7,13,14,15,16,17] summarizing 18 cases with this one we present.

In the medical literature, only seventeen cases of schwannomas originating from the infraorbital nerve (table 1), two of them corresponded to the malignant variety. The authors experienced a rare case of schwannoma arising from the left infraorbital nerve of the trigeminal a 54-year-old female patient, so we report this case with a review of the literature. This article describes the clinical and radiologic features of peripheral nerve sheath tumors and the surgical treatment.

Table 1. Published cases to date of infraorbital nerve schwannoma.

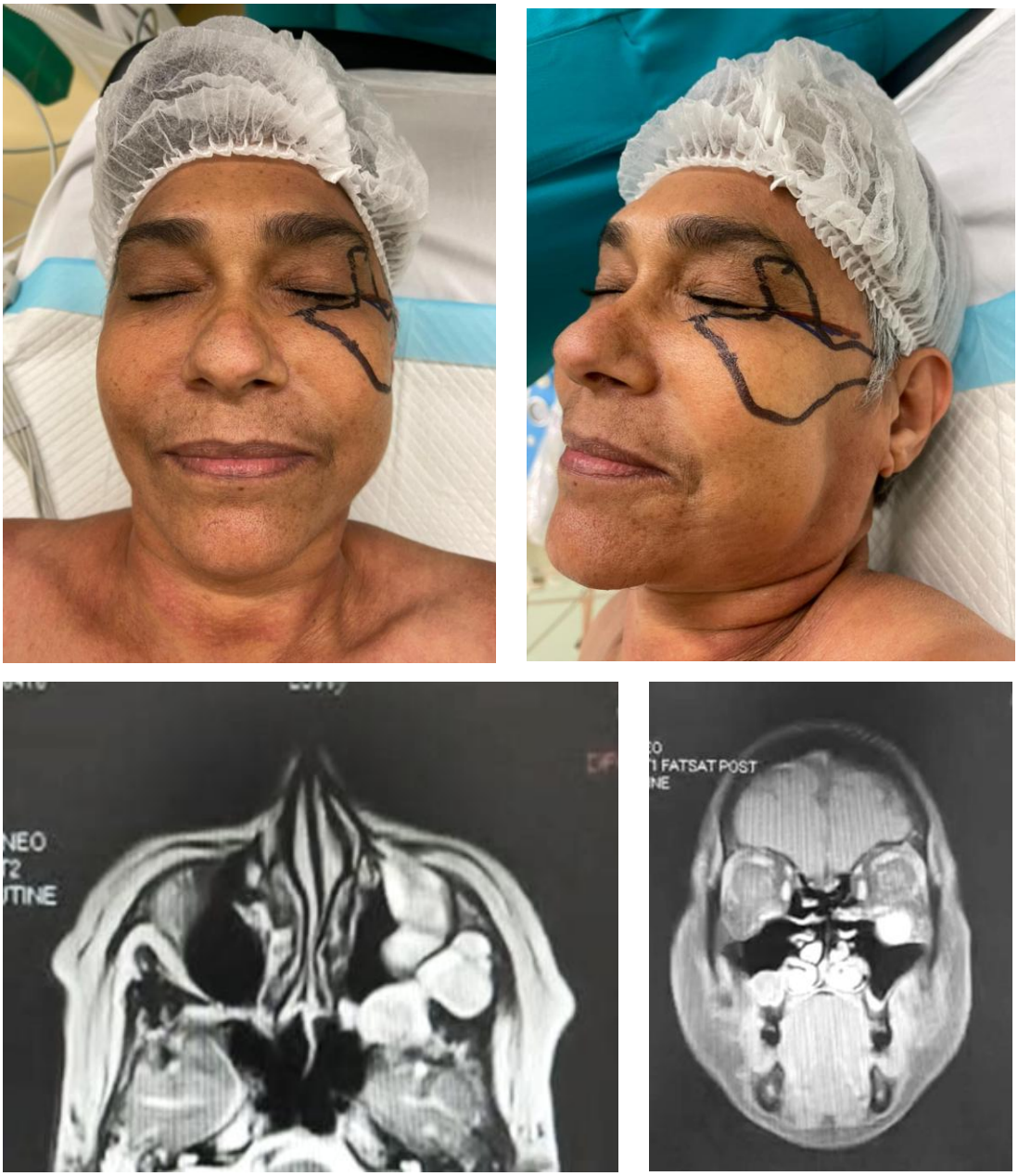
	Cases	Localization	Age /Sex	Surgical Approach
1	Tezer *et al*. (2006)	Lower orbit	16/F	Subciliar
2	Garg *et al.* (2008)	Lower orbit	35/F	Dieffenbach/orbitotomy
3	Karkas *et al*. (2008)	Maxillary Sinus, infratemporal fossa, part of nasal cavity	14/M	Weber-Fergusson/osteoplastic maxillectomy
4	Choi *et al.* (2009)	Maxillary Sinus	54/F	Cadwell-Luc/subciliary
5	Clarençon *et al.* (2009)	Cheek	45/F	Extra-oral skin incision
6	Raviraj (2011)	Infraorbital subcutaneous	8/M	Weber-Fergusson, Dieffenbach extension
7	Ha *et al.* (2013)	Infraorbital subcutaneous	20/F	Intra-oral
8	Kinger *et al.* (2013)	Cheek	30/F	Intra-oral
9	Kok *et al*. (2013)	Nasolabial subcutaneous	12/M	Intra-oral
10	Kumar, (2015).	Cheek	40/M	Extra-oral skin incision
11	Kurniawan *et al.* (2018).	Cheek	41/M	Intra-oral
12	Choi, *et al.* (2018).	Cheek	45/M	Subciliar
13	Pirimoglu & Kantarci (2018).	Cheek	18/F	ND
14	Reyna-Rodríguez & Chavez-Alvarez (2021)	Cheek	40/M	Extra-oral skin incision
15	Shafie, *et al*. (2022).	Floor Orbit	58/M	Transconjunctival
16	Mallick, *et al. (*2023)			
17	Ayushree *et al.* (2023)	Cheek	13/M	Rethi-Meyer
18	Our Case (2025)	Maxillary Sinus, floor orbit, infratemporal fossa	54/F	Transconjuntival/Canthotomy/ crow's foot fold incision/extracorporal zygomatic bone

CASE PRESENTATION

In 2022 we will receive a consultation a 59-year-old female patient with a history of ductal cancer in situ of the right breast -managed with modified radical mastectomy and anastrozole- and a family history of breast and liver cancer, who presents a painful lesion in the left infraorbital region of three months of evolution, is received for consultation.

At the clinical examination does not reveal any disease or alteration, but on the palpation at the infraorbital region it shows a solid mass, the patient did not complain of other symptoms such as tenderness, and did not have any trauma history.

Computed axial tomography of the skull reports an isodense, osteolytic lesion located in the left infratemporal fossa with extension to the floor of the orbit and sinus maxillary, the entire course of the infraorbital nerve.



Clinical appearance of the patient, marking the zygomatic bone and the incision for the approach. The magnetic resonance image shows a homogeneous lesion occupying the right maxillary sinus, as well as the infratemporal fossa and causes erosion of the bone of the floor of the orbit.

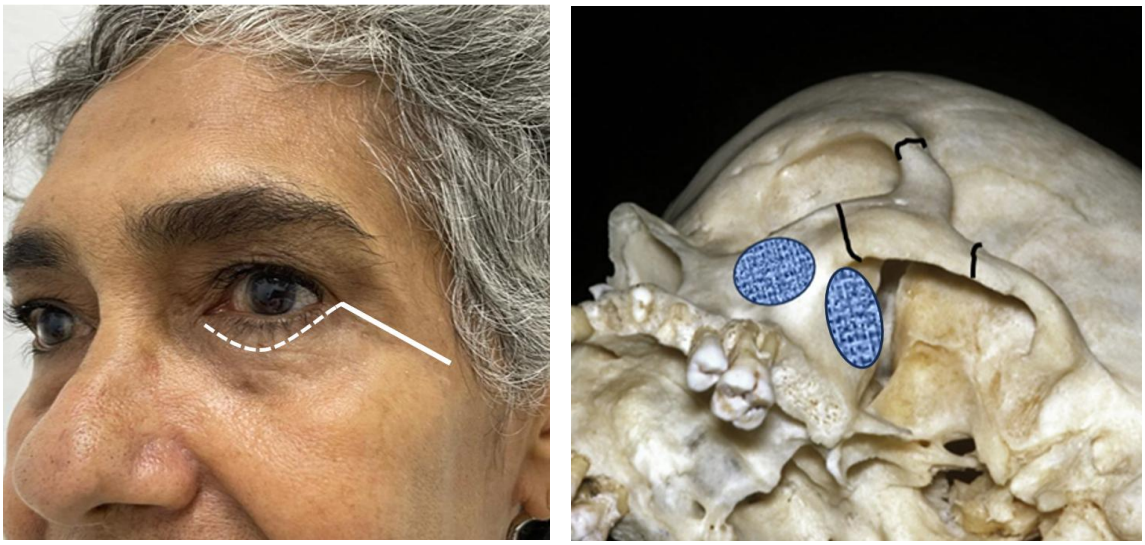
The magnetic resonance image shows a homogeneous lesion occupying the right maxillary sinus, as well as the infratemporal fossa. Multi-lobulated lesion causing erosion of the lateral and posterior wall of the maxillary sinus, contacting the medial pterygoid muscle, enhanced intensely and heterogeneously both in normal sequences and in fat saturation sequences where it is more evident.

Description of the Procedure

The removal of tumors should aim to provide a sufficiently wide access route to be able to remove the tumor in its entirety, and above all, prevent any recurrence.

Due to its location in the maxillary sinus and its extension to the infratemporal fossa we perform a combination of incisions for a broad approach with minimal aesthetic consequence. The patient is placed supine position. Depending on the location of the lesion, the patient's head can be rotated 30 to 90˚ to the side contralateral to the surgical incision. The neck is extended toward the floor until the malar eminence is the highest point in the operative field.

We used a transconjunctival-transcanthal approach -*Berke, 1953; Velpeau-Reese,1971*-, and an extracorporeal mobilization of the zygomatic bone -*Gangolphe, 1900*-.



Transconjunctival-transcanthal approach with extension to the temporal region. Skull showing in black lines, the sites where the zygomatic osteotomy was performed and in blue circle, the area of bone erosion.

Under general anesthesia, the approach is performed, incising the conjunctiva of the left eye from the lower eyelid to the external canthus and then inferiorly towards the orbital rim. Another incision of skin on the lateral edge of the eye was made up to the periosteum. Then, a straight incision was made in the temporal region. After the dissection of planes, the periosteum of the zygomatic bone is incised and the floor of the orbit is desperiotized. the masseter muscle was desinserted.

An ostectomy of the zygomatic bone is performed, the experimentation has shown to Van Merris (1901) that, in order to penetrate the sphenomaxillary suture, it is necessary to begin the section not at the frontal suture, but a little higher up. We performed it below the frontomalar suture because it was considered unnecessary to make it higher.

To maintain a fixed point on the malar contour, it was decided to perform the anterior ostectomy behind the zygomatic foramen. The cut on the external side of the orbital rim is made taking care to keep it perpendicular to the bone plane to avoid any slippage towards the interior of the orbit. The next cut is made from back to front, starting at the inferior sphenoid fissure and exiting through the body of the zygomatic bone.

Once the entire bone tripod has been removed, the surgeon detaches it from the various muscular or aponeurotic adhesions that still hold it in place, the temporalis muscle retracted.

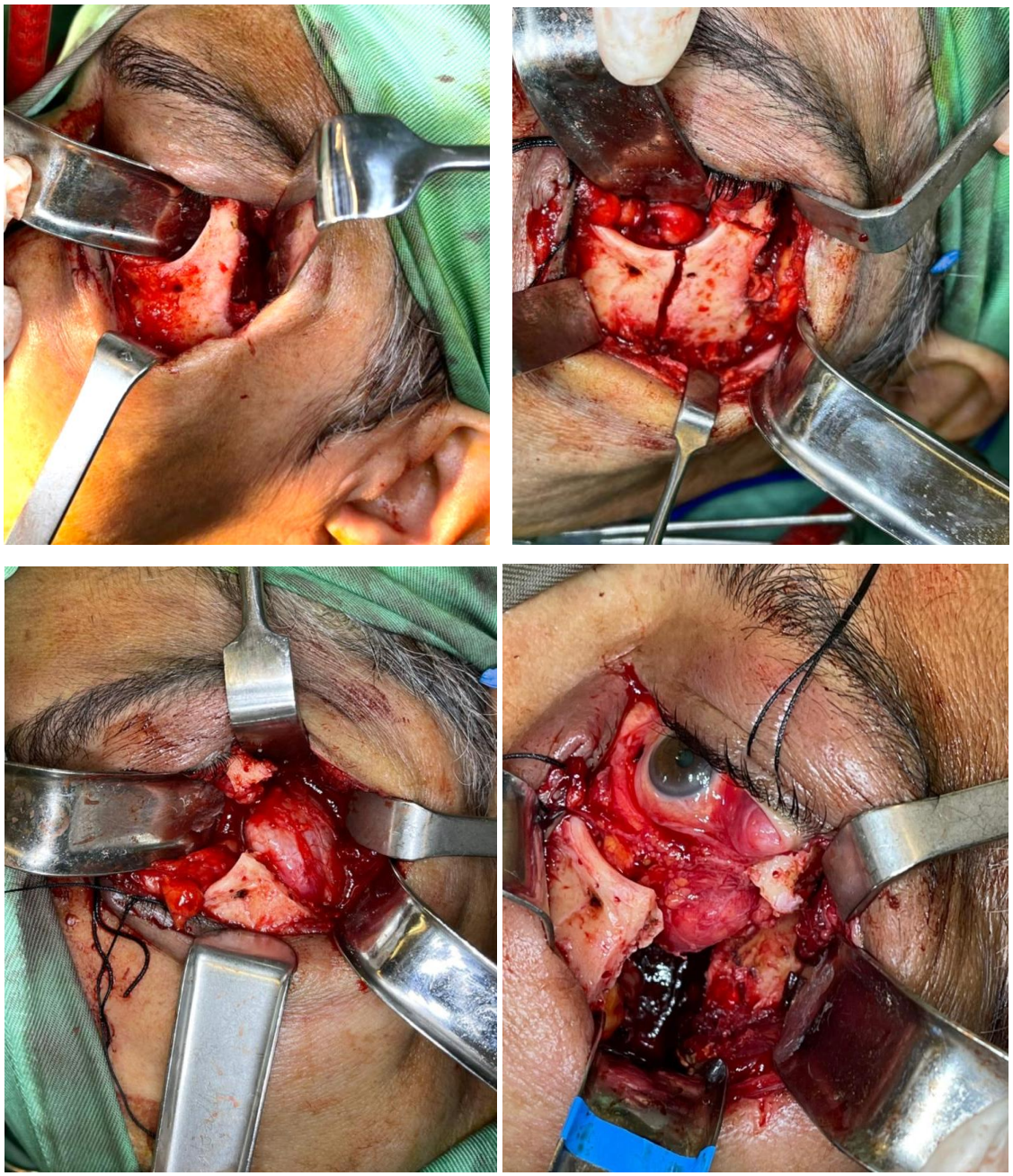
The tumor, which was well encapsulated, was carefully dissected and the mass was completely excised, paying attention to the margins along the border with the surrounding tissues.

No evidence of the invasion of the tumor tissue into neighboring tissues was observed. We were careful to spare nerve from excision and used small blunt scissors, bipolar electrocautery and magnification, then resect the tumor lesion, which invaded the floor of the orbit, maxillary sinus and infratemporal fossa.

At the time of exploration, the lesion was found to emanate from the nerve trunk branch of infra-orbital nerve at the pterygoid fossa, which was dissected and can´t preserved. An orbital floor mesh is placed and the zygomatic bone is repositioned, fixed with microplates and screws, 1.5mm. system.

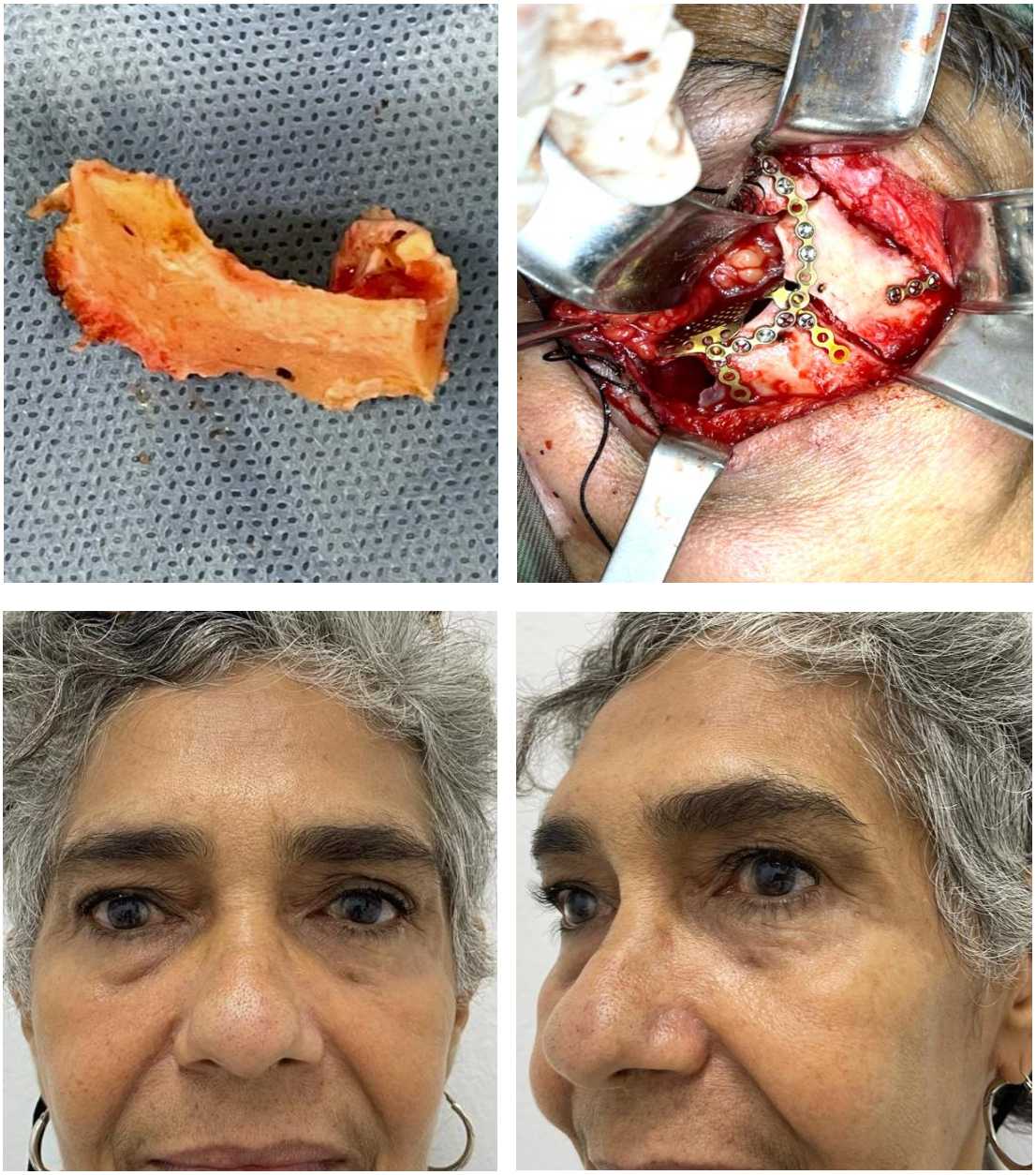
Finally, the external canthal ligament was sutured to the zygomatic bone suture. the conjunctiva and deep planes are closed with 6-0 vicryl threads, and the skin is closed with 5-0 nylon thread.

The patient was discharged on the second day after surgery, asymptomatic with eyelid swelling and without ocular neurological deficit. managed with steroids, NSAIDs and antibiotic therapy.



Top images: Exposure after deperiostization of the zygomatic bone, it can be observed zygomaticofacial foramen. The ostectomy was performed below the frontomalar suture and leaving part of the malar body (zygomatic buttress) for better reconstruction and maintaining good exposure of the tumor. Photographs below: circumscribed tumor. On the right, surgical bed where the maxillary sinus and pterygoid process can be seen.

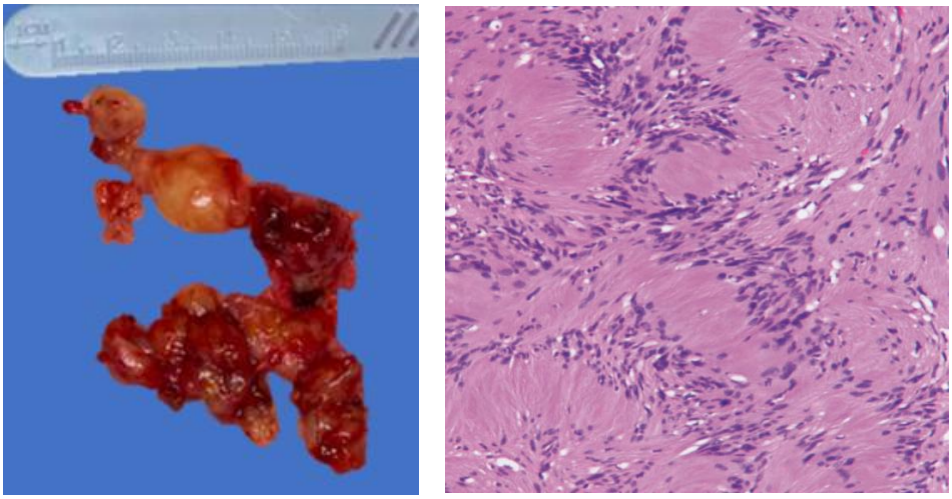
The patient's condition improved well postoperatively with no specific complications. After 4 years of follow-up, he just had a complaint of mild anesthesia in the left maxillary and the left lateral nasal area with no clinical evidence of recurrence.



Above: Zygomatic bone and arch extracorporelly. Reposition and fixation of the zygomatic bone, note the erosion on the anterior wall of the maxillary sinus. Bellow: Clinical appearance of the patient 6 months later.

Biopsy Results

Biopsy reports proliferation of spindle cells with little cytoplasm and spindle-shaped nuclei, with some visible nucleoli on a collagenous background, without evidence of atypia, necrosis or mitotic figures. Immunohistochemistry reports positive staining for vimentin and S100, corroborating the diagnosis of Schwannoma.



Macroscopic lesion aprox. 7 x 5 cm. The histological sections are predominantly composed of Antoni A tissue and sometimes form Verocay bodies.

DISCUSSION

Schwannomas are well-differentiated tumors that derive from Schwann cells in the sheaths of peripheral nerves. Orbital schwannomas are rare; their presentation is more common in the supraorbital or trochlear nerves. These tumors are slow-growing, which makes their invasion of bone structures unlikely. Schwannoma derived from infraorbital nerves arises within the orbit or maxillary sinus.

Neoplasms originated from cells composed of the nerve sheath are schwannoma and neurofibromatosis. Schwannoma typically arises solely and has a well-wrapped shape. Its transformation to malignant cell is very rare and its reoccurrence was low if it is completely removed. Neurofibromatosis is commonly multicentric and only 4% of it is wrapped by capsule. Compared to schwannoma, it is more difficult to separate nerves and the mass and has high rate of transformation to malignant cell. It is often related to Von Recklinghausen's disease, and in this case, it shows about 8% of malignant transformation [7].

Schwannoma's classical histopathological arrangement is called Antoni A and Antoni B and was first described by the Swedish neurologist Nils Antoni (1920). The palisading arrangement of Schwann cells in a stack-like arrangement with an acellular eosinophilic center is eponymically called Verocay bodies because they were first described in 1910 by the Uruguayan neuropathologist Jose Verocay [22].

Diagnosis is generally made by the patient's medical history, symptoms, and imaging tests, and can be confirmed by histopathologic examination through special immunohistochemical staining.

Clinical symptoms vary depending on the distribution of the mass and the origin of the dominant nerve, but patients mostly complain of compression, pain or sensory disturbance due to the mass. In patients, there were no specific symptoms other than the palpable mass in the right cheek area and discomfort when wearing glasses [7].

Ultrasonography, computed tomography (CT), and MRI are diagnostic techniques for schwannoma. Ultrasonography and CT are not sensitive and specific for the diagnosis. Schwannomas are usually isodense or slightly hyperdense and are uniformly intense enhancing on CT images. MRI commonly reveals low signal intensity on T1-weighted images and high signal intensity on T2-weighted images. Post-contrast MR images often show homogenous intense enhancement pattern [23,24,25,26,27,28].

In principle, a schwannoma should be removed by surgical excision because of its resistance to radiation therapy, and in other literature, the transfacial approach has been introduced as an approach to trigeminal schwannomas [29].

It is the mass arose from sensory nerves so it does not mostly affect eye movement or visual acuity if it arises within the orbital. The main symptoms are exophthalmos and pain. The mass slowly grows and does not invade surrounding bone structure; however, it sometimes pushes or corrodes the bone wall by pressure.

Other cases, such as the one reported by Shafie *et al.* present orbital displacement and facial asymmetry, without changes in visual acuity [30].

Despite invasion of the orbital floor and zygomatic bone, the patient described did not present changes in vision or displacement of the eyeball, but did present discomfort in the left malar region. Changes in eye movements may be due to compression of the extraocular muscles [31].

The treatment of schwannomas is exclusively surgical and the approach depends on the extent and location of the tumor. Various surgical approaches have been described for tumours of the infratemporal fossa, depending upon their extension, and for space-occupying lesions of the orbit and adjacent extracranial skull base.

About the Approach

Roux (1861) and Kronlein (1888) [20,32] performed the surgery with the zygomatic pedicled to the soft tissues. Many other approaches have been performed since that time, depending on the size, location and histopathological variety.

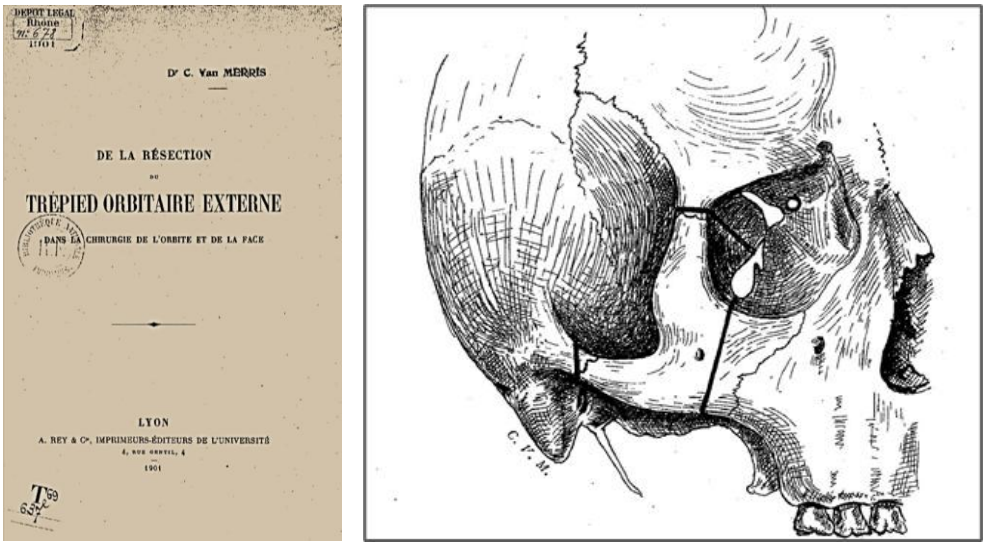
Surgery for primary inferolateral orbital tumor extending to the infratemporal fossa usually involves a combination of transorbital and preauricular approaches [33].

Other approaches used include external approach with skin incision, Caldwell-Luc approach for tumor within the maxillary sinus, subciliary or eyelid crease incision, Weber-Fergusson incision combined with osteotomy for larger lesions [12] zygomatic-transamadibular .

In view of the cosmesis, making no compromise in the adequacy of exposure and clearance of tumor. Our case, due to its location in the maxillary sinus and its extension to the infratemporal fossa we perform a combination of incisions for a broad approach with minimal aesthetic consequence.

The approach used in the case to be presented is wrongly attributed to Berke (1953) was Velpeau, had already used a transcanthal (transcomissurale) approach to access the orbit [18]. Willard Parker in 1845 [19] and then Herman Knapp's original 1874 description of a transconjunctival approach to an optic nerve tumor [18] but is to Reese in 1971 to whom it is attributed.

Apparently Czermak had published something before 1905 performed extracorporeal removal of the zygomatic, but his operation does not appear to have been performed on a living person at 1901 according to van Merris [19], although it was described in a patient by Czermak in 1905 [20, 21] five years after Michel Gangolphe (1858-1919) did it in 1900 and introduced the technique in the XIII Congress of Surgery in Paris in 1901 and that he called: *Résection du Trépied Orbitaire Externe* (Resection of the External Orbital Tripode); Later that same year his disciple, van Merris, wrote a thesis on this technique [18].



van Merris´s thesis where Ganglophe´s technique was described. Bone sections from the Gangolphe operation (Resection of the external orbital tripod).

In this case, the tumor was somewhat distant from the location where it was palpable, and there were concerns about the invasion into surrounding tissues or difficulty of dissection.

However, considering several factors such as safety problems like facial nerve damage, cosmetic problems such as scar formation, and familiar plastic surgical procedures, we approached it through subciliary incision, and since the tumor did not invade the neighboring tissue and was well dissected, it was possible to excise it sufficiently.

We used in our case a transconjunctival-transcanthal approach extending with bony removal of the total zygomatic bone extracorporeally.

CONCLUSION

Schwannoma is a benign tumor arising from neuroectodermal schwann cell of cranial, intraspinal, peripheral and autonomic nerve sheaths. This schwannoma, also known as a neurilemmoma, is rarely transformed into malignant tumor.

It can arise from all parts of the body; however, 25-45% of schwannoma arises in the head and neck area such as oral cavity, mastoid, middle ear, larynx and paranasal sinuses [34].

Its location may cause compressive symptoms or displacement of adjacent structures [11, 35]. They may present as asymptomatic, slow-growing masses, but may occasionally cause paresthesia or compressive symptoms at the site where they appear. In the head and neck, schwannomas usually originate from sensory nerves and rarely arise from the sheaths of the infraorbital branches of cranial nerve V.

Since this tumor is not very susceptible to radiation, treatment consists of surgical resection and the approach is dictated by the size and the site of the lesion. Various approaches can be implemented, including intraoral incisions, Caldwell-Luc, sub- ciliary, Weber Ferguson, sub labial transoral or endoscopic approaches, Le fort I maxillary osteotomy approach, osteoplastic maxillectomy approach, midfacial degloving, and limited transzygomatic or limited rhytidectomy approach [16].

The wide surgical field obtained by removing the zygomatic bone exposes the deep plane of the region formed anteriorly by the orbital cavity and posteriorly by the three temporal, zygomatic and pterygomaxillary fossae. This provides a considerable means of approach for operating in these cavities.

Few cases of schwannomas of the infraorbital nerve have been reported. This rare case of isolated schwannoma in infraorbital nerve (only seventeen case) and our case done with extracorporeal zygomatic bone surgical resection is presented.

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