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Technical Notes

Utility of sodium fluorescein in recurrent cervical vagus schwannoma surgery

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ABSTRACT

Background: Cervical schwannoma is a rare neoplasm that usually occurs like a nondolent lateral neck mass but when growing and symptomatic requires radical excision. Sodium fluorescein (SF) is a dye that is uptake by schwannomas, which makes it amenable for its use in the resection of difficult or recurrent cases.

Methods: We describe the case of a patient presenting with a recurrence of a vagus nerve schwannoma in the cervical region and the step-by-step technique for its complete microsurgical exeresis helped by the use of SF dye.

Results: We achieved a complete microsurgical exeresis, despite the presence of exuberant perilesional fibrosis, by exploiting the ability of SF to stain the schwannoma and nearby tissues. That happens due to altered vascular permeability, allowing us to better differentiate the lesion boundaries and reactive scar tissue under microscope visualization (YELLOW 560 nm filter).

Conclusion: Recurrent cervical schwannoma might represent a surgical challenge due to its relation to the nerve, main cervical vessels, and the scar tissue encompassing the lesion. Although SF can cross both blood–brain and blood–tumor barriers, the impregnation of neoplastic tissue is still greater than that of nonneoplastic peripheric tissues. Such behavior may facilitate a safer removal of this kind of lesion while respecting contiguous anatomical structures.

Keywords: Cervical carotid lodge, Cervical vagus schwannoma, Neck vascular-nervous fascicle, Sodium fluorescein, Transcervical approach

INTRODUCTION

Cervical schwannomas are rare tumors with a peak incidence between 30 and 50 years of age, regardless of gender. They originate from Schwann cells of peripheral nerve sheaths. Schwannomas of the carotid lodge can originate from either the vagus nerve or the sympathetic plexus.[23] The first case of vascular–nervous bundle schwannoma originating from fibers of the vagus nerve was described by Murley in 1948.[15] This is a benign, slow-growing tumor that usually arises solitary with the involvement of a single nerve. Multiple forms are characteristic in hereditary disorders such as Von Recklinghausen syndrome, although multiple vagus schwannomatosis might be present as well as in a patient without neurofibromatosis, as described by Abdulla and Sasi.[1] It is useful to distinguish between Schwannomas of the posterior compartment, which originate from cervical roots and grow in the vertebral canal and paravertebral spaces, and those of the anterior compartment that may arise from other neck nerves such as the vagus and its branches or, even more rarely, from the perivascular and perivisceral sympathetic plexus. Given the proximity to important neurovascular structures, radical exeresis may represent a surgical challenge. Sodium fluorescein (SF) is a soluble dye traditionally used in neurosurgery for the treatment of malignant tumors, but recently it has been effectively used also in benign tumor surgery. By exploiting alterations in the permeability of the blood–tissue barrier in tumors, it is uptake by the schwannoma and partly by pericapsular tissues where the vascular network may be impaired. Here, we report a case of a recurrent vagus schwannoma; surgically removed with the help of SF.

CASE PRESENTATION

A 57-year-old man, 80 kg body weight, with a previous history of cervical schwannoma (47 mm × 32 mm × 25 mm), previously treated in another institution with subtotal exeresis, 18 months before consulting. The tumor was located in the left carotid space posterior to the vascular-nervous bundle of the neck, attached to the anterolateral vertebral surface. In the previous 6 months, the patient had referred difficulty in swallowing and phonation. At follow-up 1 year after surgery, magnetic resonance (MR) documented the presence of scar tissue in the left carotid region with an oval-shaped lesion (37 mm × 18 mm × 22 mm) displacing anteriorly the vascular-nervous bundle; tightly adherent to the carotid artery and jugular vein sheaths, the latter appearing pervious although reduced in caliber due to compression [Figure 1]. The tumor appeared in MR imaging (MRI) as a T1W-hypointense and T2W-hyperintense solid-cystic mass, with irregular enhancement due to the presence of cystic areas within. On physical examination by palpation of the trigonum caroticum (trigonum anterius colli), the left carotid pulse was hypo-transmitted, and an oval mass was perceived displaying a tense-elastic consistency, mobile in planes, compressible, and nonpainful. The patient underwent microsurgical exeresis with the aid of sodium fluorescein (SF) through an anterior presternocleidomastoid approach [Figure 2]. The lesion was removed as also confirmed at postoperative MR control [Figure 3]. Upon awakening, the patient showed mild dysphonia and dysphagia, which

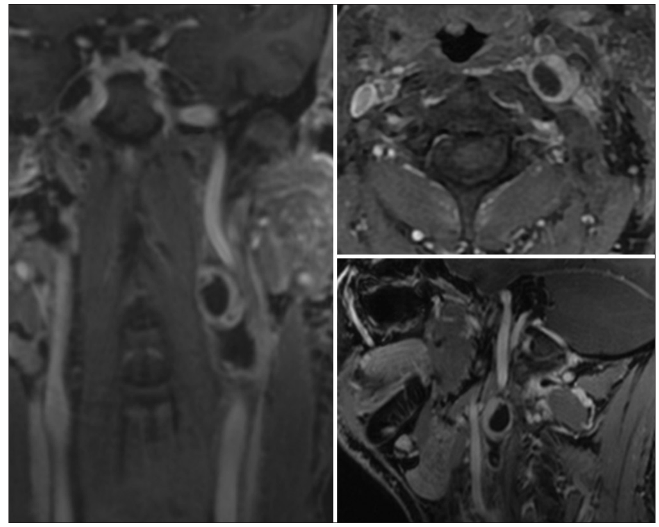

Figure 1: Preoperative magnetic resonance imaging. T1 weighted images + contrast. Dimensions of the oval-shaped lesion are 37 mm × 18 mm × 22 mm.

regressed in the following 48 h with the administration of corticosteroid therapy. The definitive histopathological study confirmed the diagnosis of neurofibroma (World Health Organization I).

Operative nuances

The patient was positioned in a supine position with the neck in extension and partially rotated to the right. Incision over the previous surgical presternocleidomastoid scar slightly extended upward toward the mandibular angle and downward beyond the plane passing through the cricoid (C6). An oblique incision was made on the platysma muscle, which appeared fibrotic and sclerotic. At this time, an intravenous bolus of 400 mg SF (5 mg/kg) was administered. Considering anatomical landmarks the sternocleidomastoid medial border and the cervical midline, blunt dissection of the carotid triangles (trigonus anterius colli) was performed. With ultrasound echography the internal jugular vein is immediately identified, appearing dilated over a stenotic tract and with a tortuous course. Proceeding with microsurgical dissection, the carotid artery was reached posteriorly; at this step, the lesion begins to be glimpsed in fluorescence (using the YELLOW-560 nm microscope filter) [Figure 2]. The dissection continued up to the C3–C4 plane, finding out the carotid bifurcation. This was done superiorly until the lesion's upper pole was reached; subsequently, the lower pole was identified and detached from the jugular vein. The space between the vessels and the vagus nerve was filled with scar tissue that was microscopically removed to free and mobilize the vessels from the nerve. The areolar tissue that normally fills visceral and vascular compartments was thickened and replaced by fibrotic tissue.

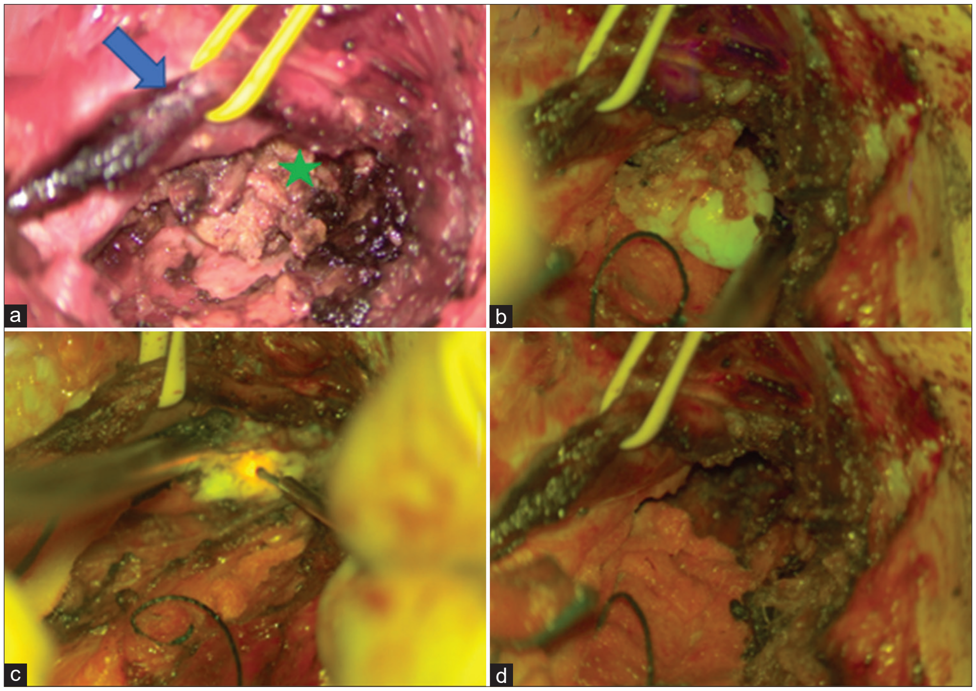

Figure 2: Intraoperative images of cervical vagus schwannoma: (a) Tumor exposure after dissection. The carotid artery is reflected with a vessel loop (blue arrow). A tumor is shown underneath the vascular complex (green star). (b) Tumor vision under yellow fluorescence. (c) The tumor under debulking maneuvers during resection and (d) surgical cavity after tumor total resection; we can observe there is no longer fluorescein uptake.

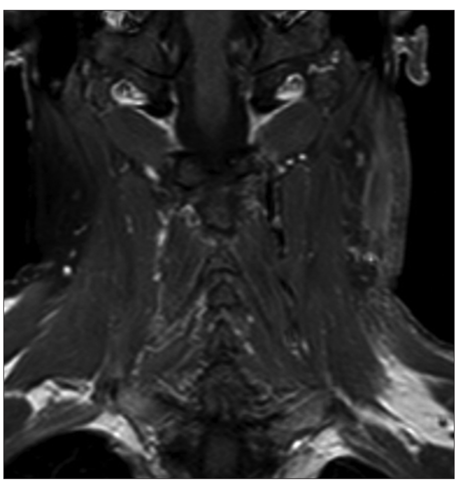

Figure 3: Postoperative magnetic resonance imaging; T1W + contrast. We appreciate the resection cavity on the left side, showing no macroscopic tumor remnant.

After checking the safety of the intended entry zone through electrophysiological monitoring the first capsule opening was performed, followed by central debulking employing an ultrasonic aspirator. With the help of fluorescence, it was possible to distinguish tumor from fibrosis and to identify the vital cleavage plane between the lesion and the vagus nerve. After generous debulking of the mass, the tumor capsule appears to be inwardly slackened, which facilitates the identification of the implantation base and its resection. Then the lesion was circumferentially dissected and piecemeal resected until gross total resection.

DISCUSSION

Schwannomas of the carotid lodge can originate from either the vagus nerve or the sympathetic plexus. Neurinomas of the cervical sympathetic chain are generally small and one of the first cases was described by Callum in 1949.[5] Recently, Adouly *et al*.[2] have treated at the same level as a giant schwannoma. These, if voluminous, can often occur with typical symptomatology of Horner's syndrome.[7,13] In our case, the patient presented dysphagia and phonation disorders, which are generally classical symptoms of vagus schwannomas. Gadolinium-enhanced MRI is to date the most sensitive diagnostic method because of the characteristic schwannomas homogeneous contrast-uptake. However, as in our case, there are aspects of inhomogeneous enhancement related mainly to necrotic-cystic areas that are found in older voluminous or recurrent schwannomas.[3] Some cervical schwannomas characterized by considerable vascularization may present the "intramass flow-void" sign due to dilated vessels with hyaline degeneration inside.[8] Dynamic contrast-enhanced computed tomography can also indirectly study these vascular engorgements in the arterial phase by recording parenchymal enhancement in the late phase,[19] but it remains a less sensitive and specific method than MR. The use of ultrasonography (US) cannot dispense with other imaging techniques because of the tumor sonographic features that are often very similar to enlarged ganglia. These generally appear as a homogenous, hypoechoic mass and may show posterior acoustic enhancement, but unusually they should show internal flow with a pseudocystic appearance.[11,12] However, US is useful not only in preoperative diagnosis but also intraoperatively to assess, as it was done also in our case, the distance of the vascular-nervous bundle from the platysma muscle and the tumor-arteries-veins relationships. Zhu *et al.*[24] used US-guided percutaneous microwave ablation for debulking and removal of cervical schwannoma. Surgical total resection is the first choice and most effective treatment, but also a very delicate procedure given the proximity of the tracheoesophageal axis and important neurovascular structures.[4]

Histologically, we distinguish between a true tumor capsule layer and an epineurium-perineurium capsule (tumor-complex capsule layer).[10] The inter-capsular resection, proposed by Hashimoto,[6] when feasible turns out to be an effective technique for preserving nerve function. The main surgical approaches for intracapsular enucleation are transcervical, postauricular, and transmaxillary. The transcervical approach appears to be associated with better preservation of the nerve of origin, less harm for adjacent vessels, and less overall morbidity.[9,23] The use of SF in schwannoma surgery has long been known, however, its application to remove a recurrence of the cervical vagus nerve has never been reported up-to-date, based on the best of our knowledge.[14,16,21] We performed an anterior approach by reopening the previous surgical scar and we reached the tumor below through fibrotic layers, proceeding by gradual steps: (1) Once the platysma was opened, identification of the vasculature bundle (the most superficial jugular vein and the deepest internal carotid artery); (2) debridement of scar adhesions between the vessels and the vagus nerve; (3) visualization of the tumor poles that were separated from the underlying plane; (4) intracapsular enucleation (debulking); (5) identification of the originating fiber of the vagus nerve; (6) debridement of further adhesions from the capsule; (7) dissection of the capsule from the nerve; and (8) segmental fragmentation of the capsule and last tumor portions removal. Proceeding from the most superficial planes down to depth, we created a surgical window encompassing tumor margins and directed it to the core of the lesion where debulking was started. This was facilitated by the fluorescence which was highlighted in the microscope thanks to the use of the YELLOW 560 nm filter [Figure 2]. SF is a soluble dye used in oncological neurosurgery, such as for gliomas,[20] since it allows a heterogeneous impregnation of neoplastic tissue to distinguish it from the surrounding healthy parenchyma, exploiting alterations in the permeability of the blood–brain barrier or the blood–tumor barrier (BTB). Finally, the same method seems to be having a good use in the removal of central and peripheral neurinomas.[16,21] In those cases, the SF is uptake by the schwannoma and partly by peritumor tissues. SF is a safe and well-tolerated dye, with almost no adverse effects even at higher dosages. It has even been used at concentrations as high as 20 mg/kg where it can be visible to the naked eye without the aid of microscope filters. A case of accidental overdosage was recently described where, despite the administration of 30 mg/kg (i.e., 10 times the recommended dosage), the patients did not experience adverse effects.[17] However, there are also a few cases reported of adverse events for a dosage higher than the standard one. Timing and dosage certainly play a key role in the uptake of lesion staining.[22] As also confirmed in retinal angiography studies, the quality of the fluorescence emitted is directly related to the concentration of SF that arrives at the lesion through the microcirculation.[18] Although the default dose used is 3 mg/kg, we preferred a slightly higher 5 mg/kg dose at the time of platysma incision benefiting in the following 40 min from excellent fluorescence. Such a choice was made in anticipation of possible capsule alterations, being a tumor recurrence and thick scarring along bordering tissues (pseudocapsule), which would have affected fluorescein uptake at that level. As the contrast is different between tumor and fibrosis, fluorescence allows to avoid excessive traction on the source nerve and carotid artery during surgical dissection. Moreover, since in recurrences, it is not always obvious to identify an accessible cleavage plan, this facilitated demarcation of the tumor borders, to perform debulking remaining within the mass and its removal.

CONCLUSION

Schwannomas of the cervical vagus nerve are rare pathologies that require surgical removal. Recurrences after a first surgical treatment could involve severe difficulties in finding a safe cleavage plane and in complete exeresis, due to distorted anatomy by profuse pericapsular scaring. Therefore, SF is an important tool aiding in identifying tumor borders and gradually proceeding to debridement from scar adhesions and intracapsular dissection.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The author(s) confirms that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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