

## CASE STUDY

### Cervical Schwannoma of the Vagus Nerve: Systematic Review and Two Illustrative Cases

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#### Abstract

**Background:** Cervical schwannoma is a rare and benign tumor exclusively developed from the cell of Schwann that rarely involve the vagus nerve.

**Methods:** We have conducted a retrospective review with strategy of the consulted references and database included articles published in Portuguese, English and French in PUBMED, MEDLINE, EMBASE, LILACS and SciELO, since 1936 until the present day, associated with a description of two illustrative cases. Both patients presented with a history of painless palpable mass in the cervical region.

**Results:** About 160 cases has been described in literature. Most cases of manifest between the third and sixth decades of the patient's life as a slow growing firm, painless mass in the lateral neck. Imaging is essential to management. Histopathological examination can reveals encapsulated, biphasic spindle cell neoplasia. The best choice of treatment for this type of tumor is complete surgical removal with preservation of the vagus nerve by means of intracapsular resection. In both illustrative cases the lesions were diagnosed by means of imaging studies and were totally resected. Surgical findings and histopathology confirmed the diagnosis of vagus nerve schwannoma. There was no recurrence at five years of follow-up. In one case there was dysphonia that solved within six months after the surgery.

**Conclusion:** Vagus nerve schwannomas are rare benign tumors. The enucleation technique is used to obtain the most functional preservation and complete excision without permanent deficit is possible even with the occurrence of large lesions.

**Key words:** vagus nerve; schwannoma; peripheral nerve tumor; head and neck; cervical vagus nerve

## **Introduction**

Schwannoma is a benign tumor exclusively developed from the cells of Schwann surrounding nerve fibers in the peripheral nervous system. These lesions may occur in the cervical region by up to 25% of cases and should be suspected in the presence of an isolated lateral cervical mass. Surgical treatment is the best choice, especially with a growing mass. The preservation of nerve function with complete resection is often possible since tumor develops extrinsically and displaces the remaining fascicles. The purpose of this report is present a systematic review associated with a description of two illustrative cases of this uncommon disease.

## **Methods**

The search strategy of the consulted references and database included articles published in Portuguese, English and French in PUBMED, MEDLINE, EMBASE, LILACS and SciELO. They were evaluated review articles, systematic reviews and meta-analyzes. The keywords used to find such results were: vagus nerve, schwannoma, peripheral nerve tumor, cervical vagus nerve. Papers excluded with cervical sympathetic chain schwannoma, jugular forame schwannoma, glossopharyngeal schwannoma, middle mediastinum, malignant cases, intrathoracic vagal nerve schwannoma, retroperitoneal schwannoma of the vagus nerve and papers containing vagus and other schwannomas without distinction.

This paper presents description of two illustrative cases of this disease.

## **Illustrative cases**

### **Case 1**

L.C.M.S., 34-year-old male, perceived the tumor in the right cervical region two years ago. There has been rapid growth of the lesion.

The physical exam revealed a painless nodule upon palpation in right cervical region, firm, insensitive and mobile. There was no associated dysphagia, dysphonia or neurologic deficit.

Biopsy by fine-needle aspiration cytology was performed and the histological findings were compatible with schwannoma.

Nuclear magnetic resonance imaging (MRI) of the cervical region was solicited and revealed expansive tumor measuring 7,7 x 3,4 x 3,0 solid with foci of heterogeneity in the inner compatible with cystic degeneration.

In June 2013, L.C.M.S. underwent microsurgery in which the lesion was totally resected by intracapsular enucleation and the nerve was preserved. Surgical exploration found a heterogeneous 8.0 x 4.0 cm mass causing anterior displacement of the internal jugular vein and vagus nerve and medial displacement of the carotid vessels, diagnosis pointed to benign schwannoma.

The immediate postoperative coursed with dysphonia.

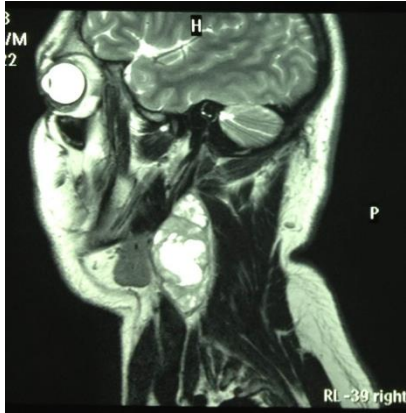


FIG 1 - MRI of the cervical region was solicited and revealed expansive solid tumor with foci of heterogeneity in the inner compatible with cystic degeneration.



FIG 2 -Surgical view.

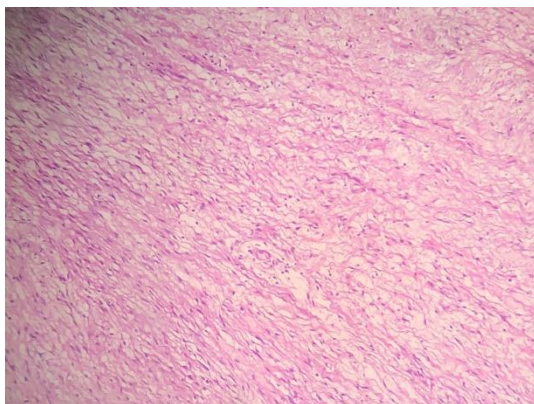


FIG 3 – Antoni B - pattern of growth, the tumor was less densely cellular stroma.

## Case 2

R.M.N.C., 35-year-old female, perceived the tumor in the right cervical region. The physical exam revealed a painless nodule upon palpation in right cervical region, firm, mobile in the horizontal but not in vertical direction. There was no neurologic deficit.

This case was not underwent to biopsy fine-needle aspiration cytology.

Computerized tomography showed a well-defined mass with anterior displacement of the common and internal carotid artery (4,0 x 2,0 cm) solid, located

in the carotid space, displacing previously vascular structures.

Using a skin crease incision, surgical exploration found displacement of the external jugular, a well encapsulated tumor arising from the vagus nerve was noted. Tumor was enucleated with sparing of the nerve, the lesion was totally resected and the nerve preserved.

In the immediate postoperative there was no neurological deficit.

Histopathology showed Antoni A palisading nucleus and verruca bodies suggestive of schwannoma.



FIG 1 - CT of the cervical region revealed expansive tumor solid located in the carotid space, displacing vascular structures.

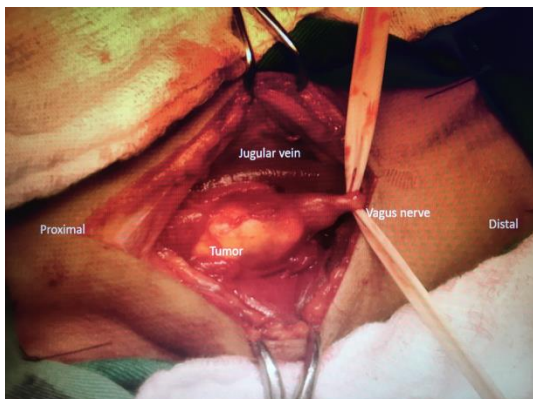


FIG 2 -Surgical view.

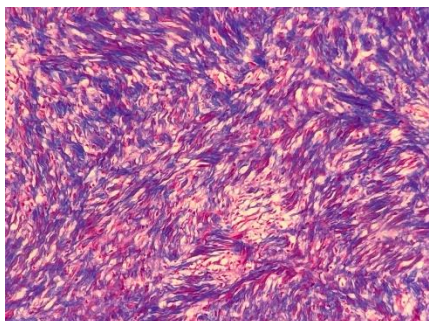


FIG 3 -Antoni A pattern of growth, elongated cells were arranged in areas of moderate to high cellularity.

**Table**

	n	AGE	GENDER	SIDE	RADIOLOGICAL FEATURES	TREATMENT
Sinakevic, 1934	1	-	M	R	-	Surgical resection
Cutler, 1936	2	1°: 38	M	L	10,5x7cm	Surgical resection
		2°: 69	M	L	6x4cm	Surgical resection
Heinlein, 1941	1	61	F	L		Surgical resection
Murley, 1948	1	19	F	R	3,5x2cm	Surgical resection
Slaughter, 1949	4	1°: 53	M	L	5x3 cm	Surgical resection
		2°:60	F	R		Surgical resection
		3°:65	F	L		Surgical resection
		4°: 65	F	L		Surgical resection
Paul, 1949	1	31	M	R	-	Surgical resection
Boisseau, 1955	3	1°: 35	F	L	10 x 4cm	Surgical resection
		2°:29	M	R	-	Surgical resection
		3°: 29	M	R	5	Surgical resection
Parnes, 1955	1	51	F	R	7 x 4 cm	Surgical resection

Conley, 1955	1	11	F	R	8 x 6 cm	Surgical resection
Altany, 1956	1	39	M	R	3 x 3 cm	Surgical resection
Gore, 1956	4	1°: 36	M	R	4,5 x 3 cm	Surgical resection
		2°: 36	F	L	4 x 3 cm	Surgical resection
		3°: 34	M	R	6,5 x 5 cm	Surgical resection
		4°: 55	F	L	6 x 3 cm	Surgical resection
Anderson, 1956	1	82	F	L	8 x 8 cm	Surgical resection
Penido, 1957	3	1°: 52	M	R	6 x 3 cm	Surgical resection
		2°: 58	M	R	-	Surgical resection
		3°: 59	M	L	-	Surgical resection
Haas, 1958	3	1°: 26	M	R	-	Surgical resection
		2°: 33	F	L	-	Surgical resection
		3°: 34	M	R	-	Surgical resection
Mitchell, 1958	1	51	F	R	-	Surgical resection
Zippel, 1960	1	36	F	L	-	Surgical resection
Horwich, 1962	2	1°: 34	F	R	10x5 cm	Surgical resection
		2°: 47	M	R	6 x 5 cm	Surgical resection
Bales, 1962	1	26	M	R	6,4 x 4 cm	Surgical resection
Pang, 1962	1	39	F	R	6,5 x 2,8 cm	Surgical resection

Leichtling, 1963	2	1°: 51	M	R	4,5 x 2 cm	Surgical resection
		2°: 48	M	R	6,5 x 4 cm	Surgical resection
Putney, 1964	1	71	M	L	4 x 4 cm	Surgical resection
Wayoff, 1964	1	59	F	R	-	Surgical resection
Vogl, 1966	1	59	M	L	0,5 x 0,5 cm	Surgical resection
Tanaka, 1967	3	1°: 36	F	R	-	Surgical resection
		2°: 50	F	L	-	Surgical resection
		3°: 56	M	R	-	Surgical resection
Rosenfeld, 1968	4	1°: 14	F	L	3 x 2 cm	Surgical resection
		2°: 32	F	R	3x2 cm	Surgical resection
		3°: 64	M	R	5x4 cm	Surgical resection
		4°: 45	F	R	4x3 cm	Surgical resection
Holland, 1968	1	53	F	R	5x2 cm	Surgical resection
Das gupta, 1969	2	-	-	-	-	Surgical resection
Katz, 1971	1	35	F	L	5,5x3 cm	Surgical resection
Reddick, 1973	2	1°: 24	F	L	2x1,5 cm	Surgical resection
		2°: 35	M	L	2x2.5 cm	Surgical resection
Andrè, 1975	3	1°: 64	M	L	4x3,5 cm	Surgical resection
		2°: 52	F	L	5 x 5 cm	Surgical resection

		3°: 38	M	L	6 x 5 cm	Surgical resection
Mair, 1976	1	71	M	R	3,5 x 3 cm	Surgical resection
Mukherjee, 1979	1	50	M	R	10 x 10 cm	Surgical resection
Pesavento, 1979	1	12	F	L	-	Surgical resection
Schulze, 1982	1	73	M	R	5 x 4 cm	Surgical resection
Chang, 1984	1	32	M	R	10 x 8 cm	Surgical resection
Gupta, 1984	1	40	F	R	6 x 4 cm	Surgical resection
St pierre, 1985	2	1°: 56	F	L	3 x 2 cm	Surgical resection
		2°: 61	F	-	4 x 3 cm	Surgical resection
Wood, 1986	1	62	F	R	5 x 3 cm swelling which was discovered during tracheal intubation for an eye operation	Surgical resection
Green, 1988	2	1°: 20	M	R	1 x 1 cm	Surgical resection
		2°: 36	F	R	4 x 3 cm	Surgical resection
Hussain, 1989	2	1°: 65	M	L	6 x 3 cm	Surgical resection
		2°: 63	F	L	4,6 x 3,5 cm	Surgical resection
Morrissey, 1990	1	54	F	L	CT: tumour was cystic and multilocular, extending from the base of the skull to the lower border of the piriform sinus in the parapharyngeal space.	Surgical resection
Peetermans, 1991	1	24	F	L	10 x 3 cm	Surgical resection
Park, 1991	4	1°: 49	F	L	10 x 5 cm	Surgical resection
		2°: 29	M	L	5 x 2 cm	Surgical resection



		3°: 25	F	L	8x4 cm	Surgical resection
		4°: 50	F	L	3 x 3 cm	Surgical resection
Walker, 1991	1	38	M	L	4 x 4 cm	Surgical resection
Galli, 1992	1	19	M	R	5,7 x 1,7 cm	Surgical resection
Nouls., 1993	1	54	M	L	5 x 4 cm	Surgical resection
Yumoto, 1996	1	33	M	L	5 x 3 cm	Surgical resection
Furukawa, 1996	5	1°: 11	F	-	-	Surgical resection
		2°: 36	M	-	-	Surgical resection
		3°: 42	F	-	-	Surgical resection
		4°: 50	M	-	-	Surgical resection
		5°: 53	F	-	-	Surgical resection
Gilmer-Hill, 2000	4	1°:55	F	L	CT scan: circumscribed, low density	Surgical resection
		2°:38	F	R	CT: circumscribed, cystic, enhancing	Surgical resection
		3°:50	F	R	CT: multilobulated, hyperintense, enhancing	Surgical resection
		4°:64	F	R	CT: circumscribed enhancing	Surgical resection
Fujino, 2000	2	1°: 26	M	L	5 x 5 cm	Surgical resection
		2°: 63	M	R	-	Surgical resection
Saydam, 2000	1	57	F	R	9 x 8 cm	Surgical resection
Kehagias, 2001	1	19	M	R	CT: mass within the right posterior triangle that displaced the internal jugular vein anteriorly	Surgical resection

Leu, 2002	2	1°: -	F	-	-	Surgical resection
		2°: -	M	-	-	Surgical resection
Heasley, 2003	1	63	M	R	3 x 3 cm	Surgical resection
Cunningham, 2003	1	45	M	R	4,5 x 4 cm	Surgical resection
Ford, 2003	1	38	M	L	5 x 4 cm	Surgical resection
Mevio, 2003	1	22	M	L	7 x 4 cm	Surgical resection
Rodríguez, 2003	1	55	F	R	CT: mass of 4 x 4 x 7.5 cm, encapsulated low density and with a cyst cavity occupying 25%	Surgical resection
Siddiqui, 2003	1	50	F	R	MR: heterogeneous mass in the neck, appearing completely encapsulated. The mass returned heterogenous high T2 signals from its substance. It also displaced the vascular and other soft tissue structures without any evidence of invasion	Surgical resection
Shetty, 2006	1	42	M	R	5 x 3 cm	Surgical resection
Szyfter, 2007	1	35	M	?	Ultrasound picture was unclear.	Surgical resection
Kang, 2007	6	1°:35	M	R	CT neck: vagal schwannoma	Surgical resection
		2°:39	F	L	CT neck: haemorrhagic thyroid cyst	Surgical resection
		3°:45	M	L	CT: enlarged lymph node near vagus nerve	Surgical resection
		4°:48	F	L	CT neck: vagal schwannoma	Surgical resection
		5°:48	F	R	MR: recurrent vagus known case of right schwannoma	Surgical resection
		6°:66	F	R	CT neck: schwannoma of the vagus nerve	Surgical resection

Singh, 2007	1	14	M	R	Ultrasound and fine needle aspiration cytology revealed a vascular lesion. He was meticulously evaluated with CT scan and digital subtraction angiography	Surgical resection
Nardello, 2007	1	63	M	R	3 x 2 cm	Surgical resection
Matejcek, 2008	1	33	F	R	MR: expansion location 10x4x1,4cm	Surgical resection
Nakano, 2008	1	59	F	L	CT: a nodule in the cervical-thoracic transition point, juxtaposed to the trachea and the left lower pole of the thyroid	Surgical resection
Peyvandi, 2008	1	32	M	R	CT: homogeneous round mass in the left side of the neck located in poststyloid region close to the carotid arteries and jugular vein	Surgical resection
Chiofalo, 2009	1	33	M	R	MR: well-circumscribed mass, with high and dishomogeneous signal intensity, on the right side of the neck, between the internal jugular vein and the carotid artery (3x3cm)	Surgical resection
Schupp, 2009	1	45	M	R	CT: Schwannoma of the vagus nerve with compression of the right hypoglossal nerve (6x3cm)	Surgical resection
Yong, 2010	1	65	F	R	CT: revealed a circumscribed non-enhancing heterogenous 4 x 4 x 7 cm mass splaying the right internal jugular vein and common carotid artery.	Surgical resection
Bilancia, 2011	1	44	M	R	MR: 2,5x1,7 cm, well-defined, oval-shaped lesion, medially displacing the common carotid artery and laterally displacing the internal jugular vein	Surgical resection
Lahoti, 2011	4	1°:10	M	R	CT: vagal schwannoma	Surgical resection
		2°:30	F	L	CT: thyroid cyst	Surgical resection
		3°:40	M	R	CT: vagal schwannoma	Surgical resection
		4°:48	F	L	CT: metastatic lymphnode	Surgical resection

Sreevatsa, 2011	3	1°:58	F	R	CT: a well-defined mass with anterior displacement of the common and internal carotid artery	Surgical resection
		2°:22	F	L	Carotid angiogram showed tumor blush and the carotids pushed anteriorly with splaying of bifurcation of carotid arteries	Surgical resection
		3°:13	F	R	CT: encapsulated parapharyngeal tumour	Surgical resection
Bergmark, 2011	1	50	F	R	3 x 3 cm	Surgical resection
Imperatori, 2011	1	67	F	L	3 x 3 cm	Surgical resection
Bhandary, 2011	1	50	F	L	6 x 4 cm	Surgical resection
Gibber, 2012	1	61	F	R	-	Surgical resection
Chiun, 2012	1	32	M	R	MR: right neck mass likely to be arising from the vagal nerve.	Surgical resection
Oyama, 2012	1	38	F	R	CT and MRI showed a schwannoma in the right parapharyngeal space	Surgical resection
Chai, 2012	1	32	M	R	7 x 6 cm	Surgical resection
Benmansour, 2013	1	32	M	R	CT: ovoid mass (6,3x5,0x4 cm) in the sterno-mastoid muscle is repressed and the internal jugular vein is compressed but still permeable  MR: 5,8x5,2x3,7 cm between the sterno-mastoid muscle and jugular-carotid vessels, roughly oval in shape, polycyclic contours, with a low signal on T1, a heterogeneous hyperintense T2 and enhanced so late and progressive after gadolinium injection	Surgical resection
Gaikwad, 2013	1	13	M	R	MR: well defined oval hypodense lesion in the right carotid space	Surgical resection
Mehta, 2013	1	25	M	R	CT: heterogenous enhancing mass lesion in right retro styloid parapharyngeal space extending up to carotid bifurcation deep to sternocleidomastoid causing antero	Surgical resection

					medial displacement of carotid vessels with obscuration of ipsilateral cervical part of internal jugular vein.	
Simsek G, 2013	1	55	M	R	Mass with high signal intensity on T1-weighted MRI and a heterogeneous, higher signal intensity on T2-weighted MRI	Surgical resection
Yilmazer, 2013	1	41	F	R	Non-vascular lesion, contrast non-enhancing, measuring 3 cm in diameter	Surgical resection
Bracale, 2013	1	34	M	R	3,5 x 2,3 x 2,6 cm mass, non-vascular lesion	Surgical resection
Sagar, 2013	1	17	F	L	Hypovascular mass, mild heterogeneous contrast enhancement	Surgical resection
Krishnamurthy, 2013	1	33	M	L	14 x 5.9 x 4.6cm mass, enhancing and vascular	Surgical resection
Sodhi, 2014	1	40	F	R	Large lobulated, heterogeneously enhancing lesion straddling across the jugular foramen, extending from cerebellopontine (CP) angle to the C3 vertebral body	Surgical resection
Hwang, 2014	1	44	F	R	CT: 4x3,5 cm well-defined mass with heterogeneous enhancement and a probable cystic component, and the common carotid artery was splayed apart from the internal jugular vein	Surgical resection
Agarwal, 2015	1	47	M	L	6x6 cm mass	Surgical resection
Tugrul, 2015	1	28	M	L	20 cm mass enhancing	Surgical resection
Vijendra, 2015	1	35	M	R	10x2x3 cm mass	Surgical resection
Kamath, 2016	2	57	F	L	4x4,5x3 cm mass,enhancing	Surgical resection
McDermott, 2016	1	37	M	R	11 cm mass, non vascular	Surgical resection
Abdulla, 2016	1	40	M	R	4,2x5,4x17,8 mass, 17.8 cm, poor contrast enhancement	Surgical resection
Giancarlo, 2016	1	31	M	R	4 x 3 cm	Surgical resection

Saini, 2017	1	34	F	L	Heterogeneous lesion non vascular	Surgical resection
Nagavalli, 2017	1	60	F	L	Diffusely enlarged and heterogeneous enhanced	Surgical resection
Carroll, 2017	1	31	M	L	6,2x5,5 cm mass	Surgical resection
Ansari, 2018	1	23	M	R	3,4x1,8cm mass, non vascular	Surgical resection
Kumar, 2018	1	18	F	R	Non vascular mass	Surgical resection
Mierzwiński, 2018	1	12	F	R	3,9x4,3x5,9 cm mass	Surgical resection
Cukic, 2018	1	60	F	L	8,6x7x5,6 cm non vascular	Surgical resection
Mat Lazim, 2018	1	22	F	R	5x4 cm mass highly vascularized	Surgical resection
Kumar, 2018	1	118	F	R	3,2x2,8 cm mass	Surgical resection
Dindigal, 2018	1	60	F	R	8x6 cm mass	Surgical resection
Bhattacharya, 2019	1	26	F	R	5cm x 4cm x 6.6cm mass, enhancing	Surgical resection

## Discussion

Cervical vagus schwannoma was first reported by Ritter in 1899 (apud Yumoto et al).<sup>110</sup> Since that, 160 cases has been described in the literature. This tumor most often presents as a slow growing asymptomatic solitary neck mass.<sup>61</sup>

Most cases of cervical schwannomas manifest between the third and sixth decades of the patient's life as a slow growing firm, painless mass in the lateral neck. Hoarseness, pain, or cough may be the presenting complaints. The tumor usually displace the carotid arteries anteriorly and medially, jugular vein laterally and posteriorly. Generally, these lesions are movable transversely but not vertically.<sup>61</sup> Those typical features were

found in our two cases.

Pre-operative diagnosis of schwannoma is difficult because many vagal schwannomas do not present with neurological deficits and several differential diagnoses for tumour of the neck may be considered, including paraganglioma, branchial cleft cyst, malignant lymphoma, metastatic cervical lymphadenopathy and neurofibromas.<sup>110</sup> Topographic features may be helpful in distinguishing origin nerve at initial presentation: the vagal schwannomas should be displaced between the internal jugular vein laterally and the carotid artery medially, where as schwannomas from the cervical sympathetic chain displace both the carotid artery and jugular vein without

separating them.<sup>43</sup> In our case 2, the tumor displaced the internal jugular vein laterally and anteriorly.

Imaging is essential to management. CT and nuclear MRI determine tumor size, extension and relation to the internal and external carotids.<sup>109</sup> The MRI is considered typical when the mass appears as a well-circumscribed tumor lying between the internal jugular vein and the carotid artery.<sup>48</sup> Radiological aspects in CT can show well defined mass compressed the internal jugular vein or internal carotid<sup>19</sup> or rare presentation with heterogenous enhancing mass lesion in right retro styloid parapharyngeal space extending up to carotid bifurcation deep to sternocleidomastoid causing antero medial displacement of carotid vessels with obscuration of ipsilateral cervical part of internal jugular vein.<sup>61</sup> MRI can show more details: dimensions, polycyclic contours, low signal on T1, a heterogeneous hyperintense T2 and enhanced so late and progressive after gadolinium injection among other things.<sup>19</sup>

The use of fine-needle aspiration and cytology is still controversial; the majority of authors does not recommend open or needle biopsy for these masses.<sup>68</sup> In this study, only one case was biopsied.

Histopathological examination can reveals encapsulated, biphasic spindle cell neoplasia. There are more cellular areas (Antoni A) where nuclei are sometimes palisaded around collagenized bands forming structures known as Verocay bodies, as well as collagen globules, often found in sheath-derived neoplasms peripheral nerve neural disease. In other

less cellular and loose stromal areas (Antoni B) cystic degeneration is often observed and hyalinized vessels. Fibrous capsule evidenced by Masson's trichrome staining<sup>98</sup>

In particular, cystic vagal schwannomas originating from the cervical vagus nerve are extremely rare<sup>68</sup> limited to 160 cases in the literature.

The best choice of treatment for this type of tumor is complete surgical removal with preservation of the vagus nerve by means of intracapsular resection.<sup>79</sup> The two patients here reported underwent surgery with intracapsular resection.

Nerve palsy is a common complication of the vagal schwannoma removal, but in many cases it recovers spontaneously when the neural integrity is preserved.<sup>79</sup> In our case 1, there was vocal cord palsy, which fully recovered in six months after the operation. In case 2 there was no vocal cord palsy or another complication. Horner's syndrome is also a usual postoperative complication<sup>111</sup> absent in our cases.

## Conclusion

Vagus nerve schwannoma is a rare benign tumor to be suspected in the presence of an isolated lateral cervical mass. Imaging studies are essential to the management.

Surgical treatment is the best choice, especially if the tumor is growing. The conservation of the original nerve is often possible.

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