Cervical Sympathetic Schwannoma: Report of Two Cases and Review of the Literature

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Abstract- This study describes two cases of cervical sympathetic schwannoma operated at department of vascular surgery and review the literature to clarify the demographic, diagnostic, and therapeutic features of these rare lesions for the first time. Two 32- and 59-year-old ladies both presenting with a painless cervical mass were referred to our service with the initial diagnosis of carotid body tumor. At the operation, mobile masses which could be easily dissected from the surrounding arteries and veins, except for the cervical sympathetic trunk were observed. Micro-surgical techniques helped us with removing the lesions with saving the sympathetic trunk in both cases. No permanent deficits were observed post-operatively in patients. Angiographic studies can provide the only pre-operative clues to diagnose a sympathetic schwannoma. Total removal of the lesion at the expense of sacrificing the sympathetic nerve is associated with minimal neurologic deficits which are well tolerated by the patient.

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Introduction

Para-pharyngeal masses have a wide range of differential diagnosis such as carotid body tumor, thyroid lesions, paraganglioma, branchial cleft cyst, and cervical schwannoma. Cervical sympathetic schwannoma (CSS) is a very unusual diagnosis for a cervical mass. These tumors are usually diagnosed intra-operatively and before the surgery more common lesions such as thyroid or carotid body tumors rank higher in the diagnostic list.

Non-specific clinical presentation, examination and appearance on imaging which is usually dismissed as other cervical masses, make their pre-operative diagnosis difficult. Here, the authors report two new patients with CSS operated at department of vascular surgery and then review the available literature to discuss demographic and clinical features, diagnostic modalities, management, and prognosis of these tumors.

Materials and Methods

PubMed database was searched for the keywords "cervical sympathetic schwannoma" and all the articles were evaluated up to March 2011. The earliest report was published in 1952. Full-texts of the articles were purchased from the data banks available at the Tehran University of Medical Sciences electronic resources (www.tums.ac.ir). The abstracts of non-English articles and the English articles whose full-text were not accessible were evaluated for the required information. Data including demographic information, symptoms and examinations at presentation, tumor size, diagnostic imaging, pre-operative diagnosis, surgical findings, post-operative morbidity, and follow up were all registered in SPSS (version 13.0, Inc., Chicago, IL). Non-parametric Mann-Whitney U test was used for comparing the medians. Data are expressed as mean ± standard error.

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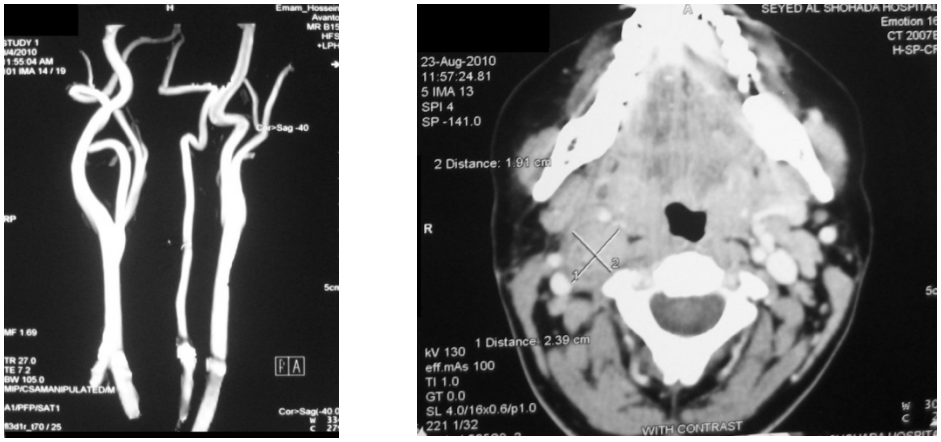
Case 1

A 32-year-old lady was referred with a painless, slowly progressive mass at her right mandibular angle since one year before. She had been previously operated on the same side where some reactive lymph nodes was removed. On her physical examination, 2.5 x 3cm non-tender, mobile mass was palpated which was pulsatile but no bruit could be heard. No neurologic symptoms including hoarseness or ptosis were observed. Her cervical CT-scan and angiography are showed in figure 1.



1.a 1.b

Figure 1. CT-scan and CT angiography of the patient presented in case one

Axial contrast-enhanced CT scan shows the round lesion in the right retro-mandibular area. Note the carotid vessels splayed by the lesion (fig 1.a). 3-D reconstruction of the cervical CT-angiography depicts the splayed internal and external carotid arteries by the tumor. No signs of hypervascularity are at the tumor site which is in favor of a schwannoma rather than carotid body tumor (fig 1.b)

She was diagnosed with carotid body tumor (CBT) and planned for surgery. At the operation, there was splaying of internal and external carotid arteries (ICA and ECA) but these vessels, internal jugular vein (IJV), and vagal nerve could easily be freed from the tumor. It became clear that the tumor had originated posterior to the carotid bifurcation and by splaying the branches resembled a CBT. The tumor originated from the sympathetic trunk. The nerve sheath was opened, and the tumor was totally removed without injuring the nerve trunk. However, the patient woke up with Horner's syndrome which fully recovered after two months. Pathological studies showed a benign schwannoma.

Case 2

A 59-year-old woman presented with a painless cervical mass since four years ago. She had undergone thyroidectomy for a multi-nodular goiter. Her physical examinations were unremarkable except for a non-tender, mobile, and soft mass at her left mandibular angle. CT-angiography showed splaying of the ECA and ICA. She underwent surgery with a diagnosis of CBT. The tumor could easily be dissected free from ICA, ECA, IJV, and hypoglossal and vagal nerves. The lesion was adherent to the sympathetic trunk from which it was dissected gently. No morbidity was observed post-operatively, and the tumor pathology was reported to be schwannoma.

Results

Data of 48 patients from the literature besides our two cases were collected (table 1). Of all the 50 cases, 59.5% were female, and the female/male ratio was 1.47. The average age was 44.35 ± 2.45 with the age range of 13 – 77 years old. No difference was detected between males and females regarding age at presentation using MWU ($p<0.05$).

Of all cases, 91.8% presented with cervical mass, 6.1% with ptosis because of Horner's syndrome, and 2.0% with shoulder or cervical pain and paresthesia. The volume of the tumor was 32.93±5.99 cm3 and 70.50±63.76 cm3 in patients presented with cervical mass and in the others, respectively ($p>0.05$, MWU). The average of maximum diameter was 4.67±1.76 cm. Duration of symptoms was 43.10±14.44 months. Tumors were distributed equally on both sides (right to left ratio = 1.05). No immobile masses were reported. Only 5.9 % of the masses were tender. 58.8% of the lesions were pulsatile on examination, but none had bruit on auscultation. Horner's syndrome signs or

symptoms were observed in 28.0% of patients. Tumors were reportedly enlarging in 22.0 % of the subjects. Only 6% of patients had dysphagia at presentation.

Table 1. Characteristics of the patients reviewed from the literature along with our two patients

No.	Author	Publication year	Age (years)	Gender	Side	Presentation	Size (cm)	Fine needle aspiration	Pre-op diagnosis
1	Al-Mashat[1]	2009	70	Male	Left	neck mass	-	-	-
2	Aydin[2]	2007	74	Female	Right	neck mass	7.5 × 5.5	inconclusive	Tuberculosis
3	Aygenc[3]	2002	13	Female	Right	neck mass	-	-	-
4	Bocciolini[4]	2005	42	Male	Left	neck mass	4.5 × 3	spindle shaped	VS/PG/LAP
5	Burduk[5]	2006	28	Female	Left	neck mass	-	-	-
6	Cashman[6]	2008	35	Female	Right	thyroid swelling	2.6 × 2.7 × 6.0	inconclusive	Thyroid Mass
7	Casserly[7]	2009	45	Female	Left	neck mass	2 × 3	Inconclusive	CBT
8	Cheshire[8]	2007	34	Female	Left	shoulder pain	1.6	-	-
9	Fornaro[9]	2006	49	Male	-	neck mass	-	-	-
10	Ganesan[10]	1997	77	Female	-	neck mass	-	-	-
11	Giustolisi[11]	2006	-	-	-	Horner	-	-	-
12	Hood[12]	2000	60	Male	Right	neck mass	4 × 5	inconclusive	VS/PG/LAP
13	Jain[13]	2008	46	Female	Left	Horner	3.5 × 2.5	-	CSS
14	Kahraman[14]	2009	60	Female	Left	neck mass	9 × 7 × 6	-	-
15	Kamal[15]	2007	25	Male	Right	neck mass	5 × 4	-	-
16	Kang[16]	2007	42	Female	Left	neck mass	-	-	CBT
17	Kang[16]	2007	43	Male	Left	neck mass	-	-	CBT/LAP
18	Kang[16]	2007	55	Female	Left	neck mass	-	inconclusive	Branchial cyst
19	Kang[16]	2007	64	Female	Right	neck mass	-	-	CBT
20	Kang[16]	2007	67	Female	Right	neck mass	-	schwannoma	LAP
21	Kara[17]	2002	20	Female	Left	neck mass	2 × 2	-	-
22	Karpati[18]	1998	53	Female	Right	neck mass	3	branchial cleft cyst	Branchial cyst/LAP
23	Lin[19]	2007	-	-	-	neck mass	-	-	-
24	Lin[19]	2007	-	-	-	neck mass	-	-	-
25	Lin[19]	2007	-	-	-	neck mass	-	-	-
26	Mandel[20]	2008	48	Female	Left	neck mass	-	-	CSS/CBT
27	Nazari	2012	32	Female	Right	neck mass	2.5 × 3	-	CBT
28	Nazari	2012	59	Female	Left	neck mass	-	-	
29	Ozlugedik[21]	2007	59	Male	Left	neck mass	6.5 × 4.3	-	CSS/VS
30	Ozlugedik[21]	2007	32	Female	Right	neck mass	4	-	LAP
31	Panneton[22]	2000	33	Male	-	neck mass	-	-	-
32	Panneton[22]	2000	49	Male	-	neck mass	-	-	-
33	Politi[23]	2005	46	Female	Left	neck mass	5 × 5 × 7	-	-
34	Rohaizak[24]	2002	24	Female	Right	neck mass	3 × 3	inconclusive	-
35	Rosner[25]	2001	47	Male	Right	neck mass	4 × 2	-	CBT/VS
36	Souza[26]	2000	35	Male	-	neck mass	-	-	-
37	Tomita[27]	2009	21	Male	Right	neck mass	6 × 3 × 2	Inconclusive	-
38	Tomita[27]	2009	56	Male	Right	neck mass	5×4×4	inconclusive	-
39	Tomita[27]	2009	60	Male	Right	neck mass	6×4×3.5	schwannoma	-
40	Tomita[27]	2009	52	Female	Right	neck mass	6.5×4.5×4.5	inconclusive	-
41	Tomita[27]	2009	54	Female	Right	neck mass	4.5 × 3 × 3	inconclusive	-
42	Tomita[27]	2009	27	Female	Right	neck mass	3 × 2.5 × 2.5	inconclusive	-
43	Tomita[27]	2009	25	Male	Left	neck mass	4 × 3 × 3	-	-
44	Tomita[27]	2009	44	Male	Left	neck mass	3.5 × 2 × 4.5	-	-
45	Tomita[27]	2009	33	Male	Left	neck mass	4 × 3.5 × 4.5	inconclusive	-
46	Uzun[28]	2005	25	Female	-	-	-	-	-
47	Wax[29]	2004	-	-	-	neck mass	-	-	-
48	Wax[29]	2004	-	-	-	neck mass	-	-	-
49	Wax[29]	2004	-	-	-	neck mass	-	-	CBT
50	Wax[29]	2004	-	-	-	Horner	-	-	CBT

Angiographic studies including conventional DSA, CT-Angiography, and MR-Angiography were performed in 10% of cases which showed no vascularity for any of the tumors. In some instances, enhanced MRI or CT-scan images were used to evaluate the relation of the tumor and the surrounding vessels. Overall, the splaying of the common carotid artery (CCA) bifurcation or anterior dislocation of CCA and internal jugular vein were reported in 34% of cases. Imaging studies showed well-circumscribed tumors with variable degrees of enhancement where their bright appearance on T2 sequence and vivid enhancement led some authors to the diagnosis of Schwannoma, however, specific differentiation of the origin as the vagus or sympathetic trunk was impossible.

FNA was performed in 34% of cases, of which 76.5

% were inconclusive; 17.6% were reported schwannoma or spindle cells with fusiform nuclei, and 5.9% were diagnosed with branchial cleft cyst. Differential diagnoses in different articles in decreasing order were carotid body tumor, vagal schwannoma, and reactive or metastatic lymph-adenopathy. Other less common diagnosis included branchial cleft cyst, paraganglioma, and thyroid swelling. Of the articles whose authors reported their pre-op diagnosis (40% of all reports), 90% had not included CCS in their diagnosis list. Pathologic features of schwannoma were observed in all cases.

Sympathetic trunk was preserved in only 4 % of all cases, and horner's syndrome happened in 88.9% of cases. Also, first bite syndrome occurred in 20 % of cases. The horner's syndrome was still present at follow-up examinations except for our first patient which fully recovered at two months follow up visit. The average follows up time was 16.87±14.80, and no recurrence was reported during this time period.

Discussion

Extra-cranial schwannomas are very rare lesions and even rarer are the CSSs. Of the few case series of head and neck schwannomas published, 8 – 25% were from the sympathetic chain and the nerves of origin in other cases were cervical or brachial plexus, hypoglossal, vagal, or accessory (16,30-32). Considering the fact that vagal schwannoma is more common than CSS, it can be one of the main differential diagnoses for CSS pre-operatively.

CSS is reported in the wide range of age from childhood up to 70s (10,33). Our patients were 32 and 59 years old who were not too far from the average age in our analysis (44.35±2.45). Also, in accordance to the female predominance in the literature, both of our patients were females. One case was in the right and the other in the left side as these lesions occur equally on both sides. These lesions most commonly present as a cervical mass and according to their origin from the sympathetic trunk; Horner's syndrome is observed in some cases. The presentation of Horner's syndrome is usually with ptosis as it was reported in 6.1% of the cases (11,13,29).

This ptosis may help with clinically differentiating these tumors from the vagal schwannoma. CCS is usually a painless, mobile, and soft mass which can be pulsatile on examination. Though, these tumors are not vascular; anterior displacement of CCS may induce a pulsation but auscultation of the mass reveals no bruit. Unfortunately, none of these features differentiates these tumors from other cervical masses.

On CT-scan and MRI studies, the findings are those of a schwannoma - a well delineating mass with variable enhancement and hyper-signal T2 images – but not characteristics for the nerve of origin. The location is usually in the parapharyngeal space where cross-sectional studies evaluating the relation of the tumors to the great vessels are useful for detecting the nerve of origin (34). One third of the reported cases in our analysis showed splaying of the CCA or anterior displacement of CCA and IJV without splaying them. However, this number may have underestimated the true prevalence of the displacement of these vessels as imaging findings were not fully described in some cases. On the contrary, vagal nerve schwannoma displaces these vessels posteriorly or results in splaying of CCA and IJV.

Also, vascularity of the tumor may help in the diagnosis: a lesion splaying CCA, which is hyper, vascular is most likely a CBT where if hypo vascular, schwannoma is a more favourable diagnosis (35). Although, the relation of the vessels to the tumor is of paramount importance for pre-op diagnosis, angiographic studies are not necessary for such evaluations. However, in cases where carotid body tumors are at differential diagnosis, angiographic imaging is inevitable. Also, the relation between IJV and the carotid artery can be evaluated by using ultrasound which also aids with direct visualization of the vagus nerve and its relation to the tumor (36).

Although FNA plays a significant role in approaching a non-vascular cervical mass, it seems to have a limited value in diagnosis of schwannoma as in more than two-third of cases the results were inconclusive or even misleading with the wrong diagnosis (2,18). Thus, although FNA does not help with diagnosis of schwannoma and its management, it may still be performed when other lesions rank high in the differential diagnosis.

Cases included in our analysis were all pathologically proved to be schwannoma with conventional Antony A and B regions and in some cases confirmed with immunohistochemistery. Some other rare lesions of the cervical sympathetic trunk such as malignant triton tumor or melanocytic schwannoma have been reported whose pre-op diagnosis may not change our surgical plan, but affects our decision to implement adjuvant therapy (37,38).

CCS is usually an intra-operative diagnosis because due to the rarity of the tumor and unspecific imaging

and clinical features, other pathologies are ranked at the top of the differential diagnosis list. In both of our patients, the initial diagnoses wer e carotid body because of the location and pulsation of the mass and the angiographic features. In our review, pre-op diagnosis was mentioned in only 40% of the reports, of which 90% were diagnoses other than CCS. We believe that even this figure is far beyond reality due to the missed values, and pre-operative diagnosis is made in a smaller percentage of the patients. The most common initial diagnosis in the literature was carotid body tumor followed by the vagal schwannoma, lymph-adenopathy, branchial cleft cyst, paraganglioma, and thyroid mass.

At surgery, these tumors are almost always well circumscribed and can be easily dissected apart from carotid artery, jugular vein, and vagal nerve. However, they are tightly adherent to the sympathetic trunk which confirms the diagnosis but, on the other hand, makes their dissection extremely difficult and in some cases, impossible (39).

Although a classification to consider the relation between the nerve of origin and the tumor is devised to predict the resectability and post-operative functional deficit, it gained no widespread usage (31). In our review, all the surgical attempts were associated with sacrificing the sympathetic trunk and consequent Horner's syndrome post-operatively. Considering the Horner's syndrome makes little, if at all, inconvenience to the patient, most authors do not recommend nerve grafts for reconstruction of the sympathetic trunk. Sural and greater auricular nerves are recommended by some as possible sources for graft (10). However, we highly recommend meticulous microsurgical techniques to preserve the nerve and, therefore, to prevent the Horner's syndrome. Although we removed the tumor with saving the nerve in both of our cases, one developed horner's syndrome post-operatively which resolved completely in two months. As almost all these tumors are benign, they have a favourable outcome if total removal is achieved and no adjuvant therapy is recommended. Accordingly, our analysis showed no recurrences during the 16.87 ± 14.80 month follow-up. Though, damaging the cervical sympathetic trunk results in persistent Horner's syndrome, it is usually well tolerated by the patients (10).

CCS is a very rare pathology, and its diagnosis requires careful examination of the patient and diagnostic images. We need to think of it, to reach the correct diagnosis. With proper surgical techniques, excellent results can be achieved with causing no harm to the patient.

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