Parapharyngeal Space Tumor: Submandibular Approach Without Mandibulotomy

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Abstract

Introduction Among all head and neck tumors, only 0.5% occur in the parapharyngeal space and 80% are benign tumors. Surgery is the cornerstone of treatment. The deep location of this space and surrounding structures has given rise to a large number of surgical approaches for resection of parapharyngeal neoplasms. Our experience using the submandibular transcervical approach on 14 patients who had surgery upfront for treatment of these lesions is described.

Material and methods This is a retrospective, descriptive case-series analysis of patients with parapharyngeal space tumors treated from January 2009 to July 2016, in whom carotid body tumors were excluded. Fourteen patients were included, 11 females (76.9%) and 3 males (23.1%), with a mean age of 46.9 years (SD 14.85).

Results The most common symptom reported at patient's presentation to our clinic was a foreign body sensation in the oropharynx and pain. Navigation-guided surgery was used in two cases. Mean tumor size was 4.7 cm (SD 1.785). Complete resection of lesions was performed in all cases, and the major complication was massive bleeding in one case (7.1%).

Conclusion According to the present analysis, the submandibular transcervical approach is an effective and safe technique that allows resection of large parapharyngeal tumors, even those close to the skull base. It has minimal complication rate, prevents morbidity associated

mandibulotomy and allows extension to a transparotid, transmandibular and even to an infratemporal fossa approach. Navigation is indicated in tumors < 2 cm. The submandibular transcervical approach should be considered upfront for neoplasms ≤ 7 cm, preferentially ≥ 0.5 cm distant from skull base.

Keywords Parapharyngeal space tumor · Submandibular approach · Salivary gland tumor · Sarcoma

Introduction

The parapharyngeal space is a deep space in the neck with the shape of an inverted pyramid that extends from the skull base to the hyoid bone. It runs laterally to the 4 pharynx and is bound posteriorly by the prevertebral fascia, medially by the pharyngobasilar fascia and the superior constrictor muscle of the pharynx and laterally by the medial pterygoid muscle, the ascending branch of the mandible, the superficial cervical fascia and the submaxillary gland. It can be divided into two compartments: pre-styloid or anterolateral and post-styloid (posteromedial) separated by the tensor veli palatini muscle and the styloid process. This space is limited anteriorly and superiorly by the posterolateral wall of maxillary antrum and continues above and laterally with the infratemporal fossa, separated from this by the medial pterygoid fascia [1–3]. Tumors of the parapharyngeal space (PPST) are an uncommon problem, comprising only 0.5% of all head and neck tumors, with 70–80% of these tumors corresponding to benign lesions [2]. A wide histologic variation has been described in this location; however, the majority of tumors are salivary gland neoplasms (40–50%) in the anterolateral division and neurogenic (20%), in the posteromedial. The most

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common salivary gland tumor is pleomorphic adenoma, while schwannomas comprise the most common neurogenic lesion in this space, specifically in the posteromedial portion [1].

Due to the histology of the vast majority of lesions localized in this space, surgical resection is the initial treatment. The main objective of surgery for PPST is to obtain adequate visualization of the lesion to ensure complete resection with preservation of adjacent structures. The purpose of this review was to describe our experience, results and limitations of the management of PPST via the submandibular transcervical approach.

Methods

We performed a retrospective descriptive study of a case series in patients with parapharyngeal space tumors treated from January 2009 to July 2016. Data were obtained from the electronic records of our Institution. Included in the study were patients who underwent PPST resection with the submandibular transcervical approach, excluding patients who had undergone other surgical approaches and patients with diagnosis of carotid body and/or glomus jugulare tumors, or patients who had received previous treatment.

Fourteen patients were included, and the following demographic characteristics were analyzed: gender; age; histology; size; location; initial symptomatology; physical examination; postoperative progress; surgical morbidity; and hospitalization days. All patients underwent preoperative examinations as well as imaging studies [computed tomography scan (CT) or magnetic resonance imaging (MRI)] to define the exact location of the tumor and decide on management; two patients underwent a positron emission tomography scan (PET scan) because the primary tumor was a medullary carcinoma and an esthesioneuroblastoma, respectively.

Surgery was performed with the transcervical approach, which is described as follows: a transverse curvilinear incision is performed on the Langer lines 3 cm below the mandibular border of the affected side, and a subplatysmal dissection of upper and lower flaps is performed. The upper flap dissection starts with identification and elevation of the submandibular capsule to identify and preserve the marginal branch of the facial nerve. The anterior border of the sternocleidomastoid muscle is incised and displaced posteriorly to dissect the X and XI nerves, the internal jugular vein and carotid system bifurcation, referring these with vascular loops. Subsequently, the XII nerve is dissected and identified as well. Displacement or resection of the submandibular gland depends on the size of the parapharyngeal tumor to be resected. Vascular control, as well as

dissection of neural structures of the neck, is accomplished with use of loops and bipolar cautery. Careful palpation of the neoplasm, ascertaining its mobility and size, follows, and in very large lesions, digastric and stylohyoid muscles are transected. Sharp, blunt and bipolar dissection of the tumor is performed, with the aid of digital dissection, and gentle traction is applied, ascertaining complete encircling, vision, and control of the upper pole of the neoplasm. Once this is performed, resection of the tumor is carried out. Copious irrigation follows, ensuring complete resection of the lesion. In all cases, a closed drain is left in the surgical bed (Fig. 1a–d).

Results are presented as percentage of the total of cases analyzed, and mean, median and standard deviation values were calculated.

Results

Fourteen patients with PPST treated surgically by the submandibular transcervical approach were included in our series. Gender, clinical findings, pathology report and location of tumors of this cohort are shown in Table 1. Patients mean age is 46.9 years [range 20–75 years (SD 14.85)]. Mean size of the parapharyngeal lesions resected was 4.75 cm [range 1–8 cm (SD 1.785)]. One patient had dysarthria due to changes in tongue motility, and two additional patients reported pain in the parotid region. In one case, the parapharyngeal tumor was found in a CT requested for a left maxillary lesion (ossifying osteoma), and there was no symptomatology in the oropharynx (case 6). The most frequent finding on physical examination was bulging of the posterior or lateral wall of the oropharynx with displacement of the uvula toward the contralateral side.

Mean hospital stay was 5.1 days, and median follow-up of our cohort was 15.5 months (range 13–82 months).

Presumptive diagnosis was based on clinical characteristics, imaging and lesion location in the pre- or post-styloid space. The imaging study utilized in 14 patients was CT, and in four an MRI was performed as well. No patient had a carotid angiogram, and two patients underwent a PET-CT (Figs. 2, 3, 4, 5). No patient in our series had a preoperative biopsy.

Two patients had additional tumors. Patient 4 had a 1-cm lesion of the hard palate which was resected at the time of the primary procedure, and this latter lesion was subsequently reported to be a basal cell adenoma (BCA). Patient 6 had another 5 × 6 cm lesion in the left maxilla, compatible with an ossifying fibroma on imaging; an inferior maxillectomy was performed afterward, and the final pathology report confirmed the clinical radiologic

Fig. 1 **a** Tumor view after submandibular gland removed, **b** total tumor view after digital dissection, **c** parapharyngeal space after tumor resection, **d** hemostatic agent inside the parapharyngeal space

diagnosis. The surgical approach for parapharyngeal lesions was performed as described in the Methods section.

Blood loss was < 50 mL in the majority of cases, but in one case (Patient 13), in whom minor infiltration of skull base was observed in the preoperative image (Fig. 5), massive bleeding ensued (2000 mL), which was successfully controlled by external carotid artery ligation and the use of hemostatic agents; final pathology report was alveolar sarcoma, and this was the only primary malignant tumor in this series. The patient received stereotactic adjuvant radiotherapy. In nine patients (64.28%), a submandibular gland resection was performed, and in five (35.7%) the gland was preserved. Patient 10 had a history of medullary carcinoma and a solitary metastasis in the parapharyngeal space, which was detected by PET-CT scan. After a multidisciplinary discussion in our department, a decision was made to perform surgical resection of the lesion, and the patient received adjuvant radiation therapy. Patient 12 had previous history of esthesioneuroblastoma, and after surgery, adjuvant chemotherapy was administered.

During follow-up, one patient with a diagnosis of pleomorphic adenoma had a second primary lesion (with the same histology), at 50 months of follow-up, in the superficial lobe of the ipsilateral parotid gland, which was treated with superficial parotidectomy without complications.

Discussion

The parapharyngeal space is a complex anatomic region because of its deep location, and where vital structures location renders surgery a challenge for the head and neck surgeon. Anatomically, the parapharyngeal space is divided into two compartments: pre- and post-styloid (anterolateral and posteromedial) [3]. This classification permits, on the basis of preoperative imaging and analysis of the anatomic structures involved, a presumptive diagnosis and better planning of the surgical approach [3]. Clinical examination of the parapharyngeal space is difficult; consequently, imaging studies are critical for evaluation of PPST. A contrast CT, as well as an MRI, and angiography

Table 1 Patients characteristics, treatment and follow-up

N/G	Age	Symptoms	Physical exam	Image findings	Histology	Size	Complications	Adjuvant treatment	Follow-up (months)
1/F	38	Otic pain Odynophagia	OT	Pre-S	Pleomorphic adenoma	7 × 6 cm	None	None	67
2/F	63	FBS Odynophagia	OT	Pre-S	Pleomorphic adenoma	5 × 4 cm	Frey Sx	None	56
3/M	41	Parotid pain	OT	Pre-S	Pleomorphic adenoma	4.5 × 3 cm	None	None	62
4/F	66	FBS	OT Palatal tumor	Post-S	Schwannoma/BCA	4 × 3 cm	None	None	14
5/F	53	FBS Dysarthria	OT Cervical tumor	Post-S	Schwannoma	3 × 2 cm	Odynophagia Dysarthria	None	16
6/F	48	FBS Dysphonia	OT Maxillary tumor	Post-S	Schwannoma	6 × 4 cm	Odynophagia	None	14
7/F	25	FBS	OT Cervical tumor	Post-S	Schwannoma	5 × 3 cm	None	None	15
8/M	20	FBS	OT	Pre-S	Pleomorphic adenoma	6 × 5 cm	Odynophagia	None	82
9/F	47	FBS Odynophagia	OT Cervical tumor	Post-S	Schwannoma	6.5 × 4 cm	Odynophagia	None	18
10/F	41	A	None	Pre-S	Medullary carcinoma	2 × 2 cm	None	RT	13
11/F	75	Otic pain FBS	OT	Pre-S	Pleomorphic adenoma	7 × 6 × 2.8 cm	None	None	13
12/M	53	None	None	Post-S	Esthesioneuroblastoma	1 × 1.5 cm	None	CH	13
13/F	42	Pain	OT and parotid tumor	Pre-S	Alveolar sarcoma	6.6 × 5.2 cm	Massive bleeding	RT	13
14/F	45	None	None	Post-S	Schwannoma	2 × 2.5 cm	None	None	26

G gender, F female, M male, FBS foreign body sensation, OT oropharyngeal tumor, Pre-S pre-styloid tumor, Post-S post-styloid tumor, BCA basal cell adenoma, Frey Sx Frey syndrome, RT radiotherapy, CH chemotherapy

in selected cases (as in carotid body tumors not included in the present series), are vital for diagnosis and for the design of the therapeutic approach [4]. All cases in our series had a preoperative MRI or CT scan; PET-CT scan could be indicated in special situations where metastases are suspected as in cases 10 and 12. PPST usually presents as an asymptomatic mass with dimensions of > 2.5 cm. Symptoms depend on the space affected. PPST found in the pre-styloid compartment produces otitis media, hoarseness, nasal obstruction, dysphagia or dyspnea; those from the post-styloid compartment may manifest compression of cranial nerves IX, X, XI and XII causing speech difficulty, dysphagia, dysarthria or Horner's syndrome [4]. Cranial

nerve palsy, pain or trismus is suggestive of malignancy [4]. In our series, different from others, the most frequent symptom of pre-styloid tumors was oropharyngeal FBS (foreign body sensation). Critical points to assess before surgery are: intraoperative exposure for an optimal dissection and preservation of neurovascular structures as well as complete resection of the neoplasm, with minimal postoperative morbidity or adverse results, both functional and esthetic [5]. The approaches described at present offer direct access to the parapharyngeal space sometimes with excellent exposure of the adjacent structures and facilitate optimal resection of the tumor; however, not all have minor morbidity and recovery is frequently prolonged and

Fig. 2 Case 2: CT scan axial view of a pre-styloid well-delineated lesion with isodense uptake. Pathology diagnosis was pleomorphic adenoma. Case 3: axial view CT scan of left-sided, mostly hypodense tumor mass with origin in deep lobe of the parotid gland. Note pre-styloid location of lesion. Final diagnosis was pleomorphic adenoma. Case 4: homogeneous contrast-enhanced neoplasm in right

parapharyngeal space. Note distance from floor of middle fossa of > 0.5 cm. Final pathology report: schwannoma of sympathetic chain. Case 5: CT scan axial view of right-sided isodense attenuated tumor mass, with well-delimited margins, with anterior displacement of the styloid process. Final diagnosis was schwannoma

requires lengthy rehabilitation periods [6]. The surgical approach to this space is a matter of controversy [1, 5], with the transcervical–transparotid approach being the most widely used; however, other surgical procedures have been described such as transoral, transcervical submandibular, modified transcervical (transcervical–transmastoid, transcervical with mandibulotomy), transmandibular and lateral approaches to the skull base [6–8]. Prasad et al. [8] suggest an algorithm depending on the location of the tumor depending on the tumor location at the vertical axis of the parapharyngeal space, so that neoplasms near the skull base require a transmastoid

infratemporal approach. Those PPST of the middle-third could be safely resected by the transparotid approach and those of the lower third by the transcervical approach. A drawback of our series is the relatively small number of cases, but we refer only to non-vascular lesions resected via a submandibular transcervical approach. There are series like the one from Iglesias-Moreno [9] where carotid body and glomus jugulare tumors are included, and other extensive surgical approaches are used for lesions with a mean size of 5 cm, where a transcervical submandibular technique with lower morbidity rate could be applied. We think that main situations where a different technique from

Fig. 3 Case 6: CT scan of right post-styloid well-circumscribed tumor mass, with hypoattenuated areas alternating with contrast-enhanced punctiform areas (heterogeneous pattern). Hypodense lesion is observed as well on the anterior maxilla, well circumscribed with immersed bone spiculae. Final diagnosis was schwannoma and ossifying fibroma, respectively. Case 7: shows a CT scan of a poorly attenuated post-styloid right-sided tumor mass. Carotid artery (ca) and

internal jugular vein (IJV) pushed forward by lesion. Final histology: schwannoma. Case 8: isodense pre-styloid neoplasm with well-delimited borders, producing minimal protrusion of parotid gland. Final pathology report was pleomorphic adenoma. Case 9: shows a coronal CT scan of a heterogenous contrast-enhanced right parapharyngeal mass, well circumscribed, with a > 0.5 cm separation from the middle fossa floor. Final histology diagnosis was schwannoma

the submandibular procedure could be indicated are in tumors ≥ 8 cm or ≤ 2 cm, or in which a more extensive exposure is needed because of a high vertical position of the lesion which does not permit an adequate resection. This fact has been reported as well by Basaran et al. [10], who reports on 44 PPST where even carotid body tumors are included, and who essentially performs a

submandibular transcervical approach except for 5 cases where a transparotid dissection is added.

A recently revisited technique in the management of these lesions is the transoral approach [7]; this procedure is suitable for small tumors because exposure tends to be limited and vascular control in case of hemorrhage is not easily obtained. Additionally, transoral excision facilitates rupture of the tumor capsule, dissemination in case of

Fig. 4 Case 10: on PET-CT shows nodular, well-circumscribed lesion located in the left post-styloid compartment, 3 cm in diameter with radionuclide uptake SUV max ≥ 6 . Final histologic diagnosis was metastatic medullary thyroid carcinoma. Case 11: a CT scan shows a pre-styloid isodense, nitid contoured tumor mass on right

parapharyngeal space, with a final pathology report of pleomorphic adenoma. Case 12: a PET-CT that shows a left 3 cm, pre-styloid parapharyngeal space node, with high radionuclide uptake, SUV max ≥ 6 . Final diagnosis was metastatic esthesioneuroblastoma

Fig. 5 Case 13: an MRI of a large left-sided parapharyngeal neoplasm with invasion to left middle fossa floor, 8 cm in diameter, well circumscribed and encapsulated in lower quadrants. Final

diagnosis was reticular sarcoma. Case 14: an MRI shows coronal gadolinium-enhanced view of left-sided tumor mass 3 cm in diameter, with final pathology report of schwannoma

malignant tumors, as well as infection due to contamination with oral cavity fluids. In a series of 26 patients treated by the transoral approach, minimal morbidity was reported if—and when—the patient was properly selected, with small tumors, and without close location or invasion of large vessels [7]. Although there are recent reports of endoscopic assisted and/or robotic-transoral approaches, there are no firm conclusions regarding these new technologies [11, 12], which could be indicated for small (≤ 2 cm), benign and medially located lesions. At our

institution, it is done only in highly selected cases and under transoral laser microsurgery [13].

The transcervical submandibular approach without mandibulotomy offers major advantages regarding functional and esthetic results, with rapid recovery periods [14]. This is particularly important in malignancy cases where adjuvant therapy with radiotherapy will not be delayed because of the bone sections healing process, as illustrated in case 13 where the final pathology report was a reticular sarcoma. The submandibular transcervical approach has

been recommended for small lesions (≤ 2 cm), but in our experience location and palpation of these tumors are difficult, with frequent confusion with normal tissue or bony landmarks. Moreover, in this situation, as exemplified in cases 10 and 12 we used navigation for complete resection of the lesions. Consequently, we propose that best cases for the transcervical approach are tumors 3–6 cm in diameter, specifically regarding the vertical dimension, with clear separation from the skull base (≥ 0.5 cm). In large tumors, close to the skull base, adequate vascular control is required, in addition to median mandibulotomy, in order to achieve minimal bleeding and no morbidity when possible.

Other approaches described for management of large tumors have the disadvantage of a larger dissection, with osteotomies that delay recovery. Lazaridis and Antoniadis [6] described mandibulotomy with coronoidectomy for better exposure and tumor and vascular control; however, morbidity as a result of a double fracture, parotidectomy and postoperative rehabilitation tend to be considerable. Judson et al. [15] proposed a double mandibulotomy at the level of the body and ascending branch avoiding damage of the alveolar nerve; however, this maneuver requires the use of a plate to reconstruct or splint the double osteotomy. There are few studies comparing approaches with or without mandibulotomy, and the majority favor non-performance of a mandibular section. The morbidity described in these studies is considerably greater in patients who undergo mandibulotomies, consisting mostly of facial branch damage, malocclusion and defects in union of the mandibular segments sectioned [2]. On the other hand, the transcervical approach (transparotid or submandibular) makes complete resection possible, with the potential of extending the procedure for lesions close to the skull base (with resection of the parotid and submandibular gland), with good functional and cosmetic results. In our series of 14 submandibular transcervical approaches, eleven patients had submandibular gland resection done, and in three patients, this was preserved. No patient had a transparotid approach, independent of tumor location or histology. Zhi et al. [16], in a series of 51 patients operated on for benign PPST with a transcervical approach, obtained excellent exposure, even in post-styloid tumors and with minimal operative morbidity. Kuet [17] in a retrospective series that included 1293 cases reported that the most common approach used was the transparotid (54%), mainly in cases of post-styloid or located near the skull base tumors. The second most used approach was the transcervical (27%), for small- to medium-sized tumors, and located in the inferior third of the parapharyngeal space. Our series included tumors with a diameter of up to 8 cm, some located in the post-styloid space, excluding jugular foramen tumors, which were completely resected without the need to perform a parotidectomy.

Nerve damage is a critical point in PPST surgical procedures. Depending on tumor size, location and histology of the, as well as the approach performed, the cranial nerves most susceptible to damage are IX, X, XI and XII. As described in our surgical technique, these are dissected and referred prior to tumor resection. Occasionally, nerve resection is performed, due to the nature of the neoplasm, mostly in neurofibromas, schwannomas or paragangliomas. Presutti et al. [14] reported a series of 18 patients, seven operated on via the transcervical approach and 11 by the transparotid approach. Four patients had paresis of the hypoglossal nerve, and three patients had transient cord palsy due to neuropraxia of the vagus nerve. Two additional patients developed dysphagia due to IX nerve damage, and one had paresis of the marginal facial branch. In our series, the most common adverse event was odynophagia in four patients; additionally, in two cases the glossopharyngeal nerve was resected, in one the hypoglossal nerve and in one additional patient the vagus nerve, because of schwannomas originating in these cranial nerves. All patients received physical therapy, with an average recovery time of 6 weeks, with quality of life similar to that of the preoperative period, since these patients already had paresis of the nerves involved.

80% of PPST are benign. Most series report a 2% recurrence frequency. All malignancies received chemo- or radiation therapy in an adjuvant setting [18].

Conclusion

In our series of patients operated via the submandibular transcervical approach, complete resection of large tumors was performed, either pre- or post-styloid located, with short hospital stay, minimal morbidity and postoperative recovery time, with no complications associated with a mandibular osteotomy. In addition, the transcervical approach is unique in that it permits extending the operating field based on the surgical findings; for example, it can be easily converted to a transparotid or transmandibular approach to facilitate visualization of the parapharyngeal space when needed. Navigation-oriented surgery could be indicated in lesions < 2 cm. In our experience, the best indication for the submandibular approach in PPST is in 2–7-cm vertical diameter non-vascular neoplasms, with ≥ 0.5 cm separation from the skull base.

Compliance with Ethical Standards

Conflict of interest The authors declare that they have no conflict of interest.

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