

Parapharyngeal space tumors: a serial case study

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Abstract

Objective: Primary parapharyngeal space (PPS) tumors are one of the most challenging head and neck tumors to diagnose and treat. We analyzed our experience in patients with PPS tumors who were treated in our hospital over 13 years.

Methods: We retrospectively reviewed 16 patients with PPS tumors between 2006 and 2018. The study included clinical symptoms, histological types, surgical approaches, adjuvant therapies, postoperative complications, and prognosis.

Results: The mean age of the patients was 49.63 ± 17.03 years. A palpable neck mass (56.3%) was the most common symptom. In our series, 78.6% of the tumors were benign, and of these, schwannomas were the most common (6/14, 42.9%). Three surgical approaches were used in our patients, including transmandibular (57.1%), transcervical (21.4%) and transparotid (21.4%) approaches. Few complications were reported, including hoarseness and numbness. Adjuvant therapy was administered depending on pathological parameters of the tumors. No recurrence was observed during a mean follow-up of 60.6 months in patients with malignant tumors.

Conclusions: Radiological studies of PPS tumors are essential for diagnosis and surgical planning. Excision of PPS tumors using appropriate surgical approaches provides good outcomes.

Keywords

Parapharyngeal tumor, transmandibular approach, transparotid approach, transcervical resection, neck mass, mandibulotomy, facial palsy

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Introduction

Primary parapharyngeal space (PPS) tumors account for approximately 0.5% of all head and neck tumors.¹ Approximately 80% of these tumors are benign.² The differential diagnosis of PPS tumors includes benign or malignant salivary gland neoplasms, neurogenic tumors, and other miscellaneous tumors. The most common benign neoplasms are pleomorphic adenomas of the salivary gland, followed by paragangliomas and neurogenic tumors.^{1,3–5}

The symptoms and signs of PPS tumors are subtle. PPS tumors are usually diagnosed only when they become large enough to be detected.⁶ Because of the location, these tumors are difficult to approach directly. Therefore, imaging studies play a central role in diagnosis and preoperative planning. Head and neck magnetic resonance imaging (MRI) and computed tomography (CT) with contrast are the most common tools used to diagnose PPS tumors.^{4,7}

Surgical excision is the primary treatment for PPS tumors.^{7,8} The surgical approaches for PPS tumors include transcervical, transparotid, transoral, and transmandibular approaches. Any one, or a combination of these approaches, has been used to manage PPS tumors depending on the location of the tumor and the surgeon's preference.^{9–11} The PPS is an anatomically complex region, which contains several vital structures, including the carotid artery, jugular vein, and cranial nerves. PPS tumors originate from a variety of histologies, including neurogenic, vascular, and salivary gland tumors. Therefore, there is no single best treatment approach for PPS tumors.

In this study, we examined the surgical approach (transcervical, transparotid, and transmandibular approaches), tumor location, treatment outcome, and complications in patients with PPS tumors who were treated in our hospital over 13 years.

Materials and methods

Patients

We retrospectively studied patients who were treated for PPS tumors in the Head and Neck Surgery Department of Chang Gung Memorial Hospital in Linkou, Taiwan, between 2006 and 2018. All patients who were suspected to have PPS tumors were recruited and their medical records were reviewed. The data obtained included the patients' age and sex. symptoms/signs, tumor size, preoperative investigations (MRI and CT), surgical histopathological approaches, findings, complications, follow-up duration, adjuvant treatments, and outcomes.

All research was carried out in compliance with the Helsinki Declaration. This study was approved by the institutional review board of Chang Gung Memorial Hospital (IRB No.: 201801371B0). This was a retrospective study and the data in this study do not disclose individual participant's identities. Therefore, no consent was required from the patients.

Surgical approaches

The transparotid, transmandibular, and transcervical approaches are the main surgical approaches used in our hospital. For the transparotid approach, we made a preauricular incision and separated the parotid gland from the sternocleidomastoid muscle. Sharp dissection was used to isolate the facial nerve safely. A facial nerve monitor was used throughout the procedure. First, the superficial lobe of the parotid gland was removed to increase tumor exposure. The tumor was then dissected to its deepest part and excised as a whole (Figure 1). For the transcervical approach, an incision was made in the neck and the sternocleidomastoid muscle was retracted backward to expose the tumor, great



Figure 1. Photographs of the transparotid approach. (a) The facial nerve was isolated (arrow) and the superficial lobe was removed to allow entry into the parapharynx space (asterisk). (b) The tumor (acinic cell carcinoma) was removed with the capsule intact.

vessels, and nerves. The tumor was excised and the vessels or nerves were preserved if possible. In the transmandibular approach, the neck skin was elevated first, the lip was split, and then mandibulotomy (midline osteotomy) was performed. Blunt dissection of the constrictor and pterygoid muscles to expose the tumor was performed, and the tumor was excised while preserving the great vessels (usually the internal jugular vein and carotid artery) and nerves. After the tumor was removed, the mandibulotomy was fixed using miniplates.^{10,12}

The surgical plans and approaches were tailored to individual patients according to the tumor location, size, and relationship with nearby anatomical structures.

Chemoradiation therapy

Three patients with malignancies underwent postoperative radiotherapy (60–65 cGy) or concurrent cisplatin-based chemotherapy and radiation therapy (CCRT) because of aggressive pathological parameters, including advanced tumor stage, perineural invasion, lymph node metastasis, and positive resection margins.

Results

Study population

The study included 16 patients (7 men and 9 women) with a mean (\pm standard deviation) age of 49.63 \pm 17.03 years. The patients' demographic data are shown in Table 1. The most common symptom was a neck mass (56.3%) and three (18.8%) patients presented with hoarseness due to preoperative vocal cord paresis on the lesion side. The mean tumor size was 4.42 \pm 1.14 cm. In total, 14 (87.5%) patients underwent surgical excision, one refused further treatment, and one chose regular follow-ups alone.

Radiological examination

All of the patients underwent preoperative CT (n=13) or MRI (n=3) head and neck scans (Figures 2 and 3). All of the patients underwent CT imaging (Figure 2) or MRI (Figure 3) postoperatively. Most of the tumors had a well-demarcated border in the parapharynx and appeared as slightly enhanced on CT scans. Neurogenic tumors generally appear homogeneous, whereas the border of parapharynx malignancies may be smooth while the mass is heterogeneous.

No.	Age (years)	Sex	Symptoms	Pathology	Tumor size (cm)	lmaging study	Approach
Ι	44	F	Neck mass	Schwannoma	5	СТ	Transmandibular
2	60	F	Lump in throat	Paraganglioma	4	CT	Transmandibular
3	25	F	Neck mass	Schwannoma	3.7	СТ	Transcervical
4	25	Μ	Neck mass	Lost to follow-up	_	CT/MRI	
5	63	Μ	Neck mass	Parotid gland carcinoma ex-pleomorphic adenoma	3.5	СТ	Transmandibular + ND level II
6	79	F	Lump in throat	Schwannoma	5.5	MRI	Transmandibular
7	53	F	Incidentally detected by MRI	Cavernous hemangioma	4	СТ	Transmandibular
8	27	F	Neck mass	Malignant carotid body paraganglioma	4	MRI	Transcervical
9	49	Μ	Oral ulcer and pain	Observation	3	СТ	
10	55	F	Sore throat	Basal cell adenoma	5	СТ	Transmandibular
11	35	F	Husky voice	Schwannoma	3.8	CT	Transmandibular
12	38	Μ	Neck mass	Schwannoma	5.I	CT	Transparotid
13	45	Μ	Neck mass	Left neck spindle cell hemangioma	3.4	СТ	Transcervical
14	88	Μ	Neck mass	Schwannoma	7.5	СТ	Transparotid
15	49	F	Incidentally detected by MRI	Cavernous hemangioma	3.8	MRI	Transmandibular
16	59	Μ	Neck mass	Acinic cell carcinoma	5.2	СТ	Transparotid

F, female; M, male; ND, neck dissection; CT, computed tomography; MRI, magnetic resonance imaging.

Vascular tumors are generally more conspicuous on T2-weighted images. Moreover, the great vessels are generally displaced by tumor growth. Preoperative angiography was performed in cases of clinically suspected great vessel invasion or when the tumor was hypervascular. Moreover, embolization was considered for hyper-vascularized tumors to prevent massive intraoperative bleeding.

Pathology of patients who underwent mandibulotomy

The pathologies of the eight patients who underwent mandibulotomy (8/14, 57.1%) included cavernous hemangioma (n = 2), basal cell adenoma (n = 1), paraganglioma of the parapharynx (n = 1), and schwannoma (n = 3) (Table 1). The remaining patient among these eight patients (case #5), had a carcinoma ex-pleomorphic adenoma of the parotid gland and underwent neck level II lymph node dissection, as well as mandibulotomy (Figure 4). The neurosurgeon used the zygomatic approach to dissect the coronoid process and then retract the temporal muscle with its coronoid insertion to protect the internal jugular vein and carotid artery.¹³ Using this method, the potential for great vessel injury was minimized.¹⁴



Figure 2. Computed tomographic coronal sections of a carcinoma ex-pleomorphic adenoma of the left parotid gland. (a) Preoperative scan (arrows). (b) No recurrence was observed 60 months after surgery and concurrent chemoradiotherapy.



Figure 3. Magnetic resonance imaging axial sections of a right parapharynx schwannoma. (a) Preoperative scan showing the schwannoma (arrow). (b) Postoperative scan showing no recurrence at 1 year after surgery.

Surgical approaches

The three patients with malignant tumors and surgical excisions received postoperative adjuvant therapy (radiation or chemoradiation therapy, Table 2). For neurogenic tumors in our patients, we excised the tumor by enucleation and intended to preserve nerve function as much as possible.

The transmandibular surgical approach was used more often than the transparotid

and transcervical approaches in our patients (Table 3). The transparotid approach was used in three (3/14, 21.4%) patients. Two of these patients had parapharyngeal schwannomas and one had acinic cell carcinoma, which originated from the deep lobe of the parotid gland and extended into the PPS. The transcervical approach was used in three (3/14, 21.4%) patients. One of these patients had a spindle cell hemangioma of the neck, one had a malignant carotid body paraganglioma, and one had an ancient schwannoma of the vagus nerve.

Complications

Three patients presented with preoperative vocal cord paresis, which progressed into



Figure 4. The transmandibular approach in a left parapharyngeal space tumor. The mandible was split by midline osteotomy (arrowheads). The pterygoid muscles (arrow) were dissected and the parapharyngeal space (asterisk) was dissected and identified.

paralysis after surgery. In one patient, lip splitting and mandibulotomy resulted in a hypertrophic scar. Postoperative facial palsy was observed in two patients. Of these two patients, one had grade VI palsy (House–Brackmann grading), which improved 4 months after surgery, and one had grade V facial palsy, which recovered 6 months after surgery.

Pathology

The tumor was excised in 14 patients with proven pathology, and the remaining two patients did not receive further treatment and the nature of their tumors was not known. Malignant tumors were detected in the final pathology of three (3/14, 21.4%) patients, including one case each of carcinoma ex-pleomorphic adenoma of the parotid gland, malignant carotid body paraganglioma, and acinic cell carcinoma. The pathological findings were benign tumors in 11 patients, including schwannoma (6/14, 42.9%), cavernous hemangioma

 Table 2. Disease status of malignant parapharyngeal lesions.

Pathology	Treatment	Follow-up duration (months)	Current status
Malignant carotid body paraganglioma	Surgical wide excision + RT	126	No recurrence
Carcinoma ex-pleomorphic adenoma of the parotid gland	Surgical wide excision $+$ CCRT	66	No recurrence
Acinic cell carcinoma	Surgery + CCRT	38	No recurrence

CCRT, concurrent chemoradiotherapy; RT, radiotherapy.

Table 3. Comparison of surgical approaches and tumor size.

	Transmandibular approach	Transcervical approach	Transparotid approach
Number (%)	8 (57.1)	3 (21.4)	3 (21.4)
Mean size, SD (cm)	4.06 (±0.81)	3.7 (±0.30)	5.97(±1.36)
Largest size (cm)	5.5	4.0	7.5

SD, standard deviation.

(2/14, 14.3%), paraganglioma (1/14, 7.1%), basal cell adenoma (1/14, 7.1%), and spindle cell hemangioma of the neck (1/14, 7.1%).

Outcomes

The mean follow-up period for patients with benign tumors was 27.3 months (range, 1–72 months) after surgery. No tumors recurred in these patients. The mean follow-up period for this group was 60.6 months, and no tumor recurrence was observed.

Discussion

Our sample of 16 patients included seven (43.75%) men and nine (56.3%) women, which is similar to the sex prevalence reported in previous studies.⁹ Of the 14 patients who underwent surgery, 21.4% had malignant tumors and the tumors were benign in 78.6%, which is consistent with previous studies.²

PPS tumors are one of the most challenging head and neck tumors to diagnose because their symptoms are subtle. In our study, a neck mass was found in approximately half of our patients and was the most common symptom, which is consistent with previous findings.^{8,15,16} The second most common symptom was a lump in the throat. In our series, the mean PPS tumor size at the time of diagnosis was 4.42 cm and the largest tumor was 7.5 cm. Most patients were initially asymptomatic and the tumor was not discovered until it became large enough to be palpated or otherwise detected. All of the tumors in our sample exceeded 3.0 cm. Our findings support the suggestion that PPS tumors are difficult to diagnose.

Because of the difficulty in initially detecting PPS tumors, imaging studies are essential for diagnosis. CT and MRI are useful tools for investigating the tumorhost interface, involvement of major neurovascular structures, glandular tissue, and the relationship to the craniofacial skeleton, including the skull base.² CT scans were performed in 81.3% of our patients. In our experience, MRI provides better resolution for identifying neurovascular structures than does CT.

In our study, 78.6% (n = 11) of the PPS tumors were benign. Of these, neurogenic tumors (all schwannomas) were the most common and comprised approximately half of these benign tumors. This finding is consistent with several previous studies.^{4,13,15} However, this finding differs from that of the largest systemic review performed to date, which showed that salivary gland tumors were the predominant benign PPS tumor.² In our series, paraganglioma accounted for 18.18% (n=2/11) of the benign PPS tumors, which is similar to the percentage reported in previous studies 10% - 40%). 10,15,17Our (range, series included three cases of hemangioma, which accounted for 27.3% of the benign tumors. This percentage is considerably higher than that reported in a systematic $(8\%)^2$ review and previous case series (4.8%).¹⁵

We found that three patients had malignant PPS tumors, including carcinoma ex-pleomorphic adenoma, malignant carotid body paraganglioma, and acinic cell carcinoma. The patient with malignant carotid body paraganglioma received radiotherapy after surgery, and the remaining two patients underwent CCRT after surgery (Table 2). All patients underwent regular follow-up, and no recurrence was noted after adjuvant therapies were completed.

The transmandibular, transcervical, and transparotid surgical approaches were used to manage PPS tumors in our patients (Table 1). Overall, the transmandibular approach was performed the most often in our study. The surgical approach was selected according to the location and size of the tumor (Figure 5 and Table 3). The transcervical approach provides adequate exposure in the bifurcation of the carotid



Figure 5. Schematic of the surgical approaches used in our study.

artery and has the least cosmetic sequela. In tumors in which the superior border is high and under the inner surface of the mandible, surgical dissection in the highest and deepest part is difficult. The transparotid approach dissects all of the branches of the facial nerve and provides the best protection of nerves. A scar is usually not apparent in this approach. However, this approach is also limited by its surgical exposure in the superior region near the skull base. The transmandibular approach was used for tumors in a high position that compressed the carotid artery or internal jugular vein, the transparotid approach was used for tumors that were located behind the mandible ramus, and the transcervical approach was used in all other cases. Two important considerations are vascular control and exposure of the cranial nerves. The transmandibular approach provides good access to the base of the PPS (Figure 4). Therefore, if unexpected bleeding occurs during dissection, this approach offers an adequate surgical field to control the bleeding. Although some surgeons may have concerns about possible facial, lingual, or hypoglossal nerve injuries during osteotomy and dissection of the soft tissue surrounding the mandible, experienced surgeons can avoid these complications.

Of the three patients who had postoperative vocal cord paralysis, the only complaint was hoarseness, and other symptoms, such as swallowing difficulties or dyspnea, were not reported. Previous studies have shown that vagal nerve injury is the most common complication of surgery, affecting 14% of patients.² In fact, nerve injury may be considered an expected sequel of poststyloid PPS tumors because most are neurogenic.² The pathologies of the three patients (Nos. 1–3) with vocal cord paralysis included an ancient schwannoma of the vagus nerve, a schwannoma of a vagus nerve branch, and a paraganglioma in which the tumor was close to the carotid artery and vagus nerve. One patient (No. 11) underwent cosmetic surgery for scar revision. Additionally, three other patients had schwannomas, which originated from the sympathetic nerve. One of these three patients developed Horner's syndrome.

Facial palsy is a common complication in management of PPS tumors.⁸ One patient (No. 14, Table 1) had a 7.5-cm schwannoma located in the deep lobe of the parotid gland that was tightly adhered to the facial nerve. We were able to dissect the facial nerve branches during surgery and preserve the nerve. The postoperative facial palsy was temporary and recovered 4 months after surgery. A patient with acinic cell carcinoma (No. 16, Figure 1) developed facial palsy after the facial nerve was mobilized during surgery. The palsy persisted for 3 months and gradually recovered.

Our primary goal was complete surgical excision of malignant PPS tumors in patients with no contraindications. Adjuvant therapy (radiotherapy and CCRT) was administered to patients with malignant PPS tumors that had aggressive pathological parameters, including advanced tumor stage, perineural invasion, lymph node metastasis, and positive resection margins. No recurrence occurred during follow-up in patients who underwent surgery for benign or malignant tumors.

There are some limitations to our study. This was a retrospective study with a likelihood of insufficient data collection and a high attrition rate. Moreover, the number of cases was relatively small. Comparison of results by statistics between different surgical techniques with such a limited number of patients is difficult.

Conclusion

Surgical management of PPS tumors is challenging because of their deep location and relationship with nearby neurovascular structures. In our study, approximately 80% of PPS tumors were benign. Radiological studies of PPS tumors are essential for diagnosis and surgical planning. Excision of PPS tumors using appropriate surgical approaches provides good outcomes.

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Authors' contributions

KHL and SFH conceived the idea for the manuscript, conducted a literature search, and drafted the manuscript. CKY and SFH organized the manuscript and critically revised the manuscript. KHL, CKY, SCC, and CTL collected the data. KHL, CKY, SCC, CTL, and SFH read and approved the final manuscript.

Availability of data and materials

The datasets used and analyzed during the current study are available from the corresponding author on reasonable request.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

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