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CASE REPORT

Huge parapharyngeal space tumor: Case report and short literature review

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ABSTRACT

The parapharyngeal space (PPS) has a special anatomical and clinical importance from the point of view of its content, communication and vicinity with major and vital structures. Tumors of the parapharyngeal space are uncommon, the most incriminated being the salivary gland tumors, neurogenic tumors, especially Schwannomas and paragangliomas, and lymphoreticular lesions. The correct diagnosis of the PPS tumors must include the radiologic evaluation represented by the computed tomography (CT) and the magnetic resonance imaging (MRI). The therapeutic plan depends on the imagistically identified aspect. We report a case of a 40-year-old male, who initially noted a massive tumor of the left side of the neck within 15 years. The cervical CT scan revealed, in the upper third of the left side of the neck, in the projection of the parotid and masticatory space and the submandibular region, under the lower jaw, medially at the level of cervical vertebrae C1 – C4, to the front of the sternocleidomastoid muscle, a volumetric formation irregular in shape, with clear and uneven contours, dimensions 40x65x85 mm, uniform density +30H. Transverse cervical approach was chosen for the surgical investigation.

KEYWORDS: parapharyngeal space, tumor, transverse cervical approach.

INTRODUCTION

The parapharyngeal space (PPS) has an inverted pyramid form, with the base oriented to the skull base and the upper part at the hyoid bone. It is a potential space where the medial wall is bounded by the superior constrictor muscle, the lateral wall by the ramus of the mandible and the posterior wall is lined by the prevertebral muscles[1]. Anteriorly, it is bound by the submandibular space, the pterygomandibular raphe and the medial pterygoid muscle fascia and posteriorly, by the retropharyngeal space, the deep fascia and the paravertebral muscle. In the superior part, the parapharyngeal space is bounded by a small portion of the temporal bone and inferiorly by the junction of the posterior belly of the digastric muscle and the hyoid bone. The medial wall is represented by the pharynx pharyngobasilar fascia, the pharyngeal wall and the buccopharyngeal fascia. The lateral wall is represented by the superficial layer of the deep cervical fascia, the deep lobe of the parotid gland, the posterior belly of the digastric muscle, the sphenomandibular and the stylomandibular ligaments.

The stylopharyngeal aponeurosis separates the parapharyngeal spaces into two compartments: the prestyloid space and the poststyloid space. Another anatomic important component is the cloison sagittale which separates the PPS from the retropharyngeal space. The parapharyngeal space communicates with several deep neck spaces: parotid, masticator, peritonsillar, submandibular, retropharyngeal.

PPS has a special anatomical importance also from the point of view of the content. The prestyloid compartment contains fat tissue, the retromandibular portion of the deep lobe of the parotid gland, minor or ectopic salivary gland, the trigeminal nerve branch to tensor veli palatini muscle, the ascending pharyngeal artery, the internal maxillary artery and the pterygoid venous plexus. The ca-

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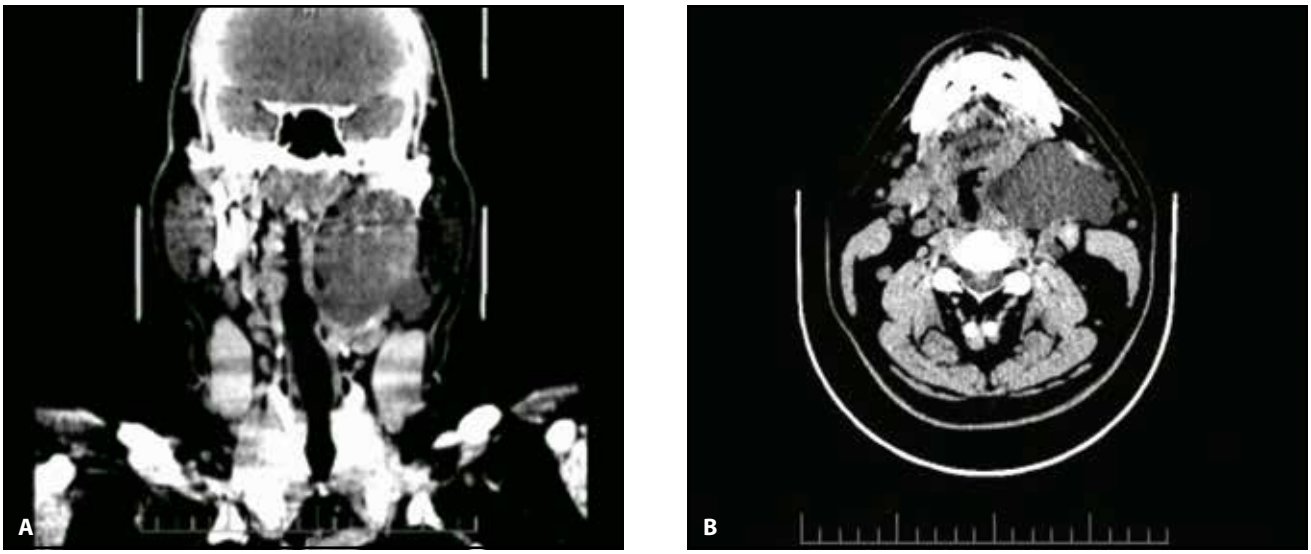


Figure 1. Contrast enhanced CT scan of the head and neck (coronal slide - A, axial slide - B). Tumoral mass, with irregular shape, clear, nonregular contours, 4.7 x 7.0 x 8, 1 cm, in the projection of the pterygopalatine fossa, above the submandibular salivary gland, anterior to the sternocleidomastoid muscle, medial from the parotid salivary gland, spreading vertebrally along the lateral wall of the oropharynx. The structure is homogeneous, with a density of +30 units, H. The surrounding tissue is not infiltrated. The surrounding bone structure shows no signs of destruction. The formation deforms the lumen of the naso-oropharynx. The left common carotid artery and the subclavian artery are moderately tortuous.

rotid artery, the internal jugular vein, the cervical sympathetic chain, glomus tissues, and the vagus, glossopharyngeal and hypoglossal nerves comprise the poststyloid compartment.

Tumours of the parapharyngeal space are uncommon, representing less than 1% of all head and neck tumors, 70-80% of them being benign[2]. There are different types of tumors which can develop in the PPS: salivary gland tumors, neurogenic tumors, especially Schwannomas and paragangliomas, and lymphoreticular lesions. 40-50% of the tumors arising in the PPS have salivary glands origin (the deep lobe of the parotid gland, the ectopic salivary gland nests, or the minor salivary glands of the lateral pharyngeal wall) and are located in the prestyloid space. In 80-90% of the cases, the salivary gland tumor identified in the PPS is the pleomorphic adenoma[1].

The most common symptoms of parapharyngeal space tumors are the presence of a neck mass, coughing, hoarseness and pharyngeal pain, abnormal sensation of the pharynx, tongue palsy, sleep apnoea and posterior neck pain.

The correct diagnosis of the PPS tumors must include the radiologic evaluation, which include the computed tomography (CT) and the magnetic resonance imaging (MRI). The therapeutic plan depends on the imagistically identified aspect. The MRI evaluation is useful in identifying the localization, origin, dimensions and relation with surrounding structures of the tumor. Depending on the prestyloid or poststyloid origin and/or localization, a potential diagnosis and tumor type can be established[3-5]. The presence of a tumor around the bifurcation of the carotid artery may require an angiography or a CT angiography in order to exclude a paraganglioma[3,6,7].

Surgery is the treatment of choice for PPS tumors, and the optimal approach is dictated by the clinical and imagistic findings. We have to remind that, due to its complex content consisting of major blood vessels, the cranial nerves (CNs), several multidirectional muscles, and vicinity of the jawbone and the skull base, the parapharyngeal space may be a difficult anatomic region to approach[3,8,9].

The major postoperative complications reported in the literature are the first bite syndrome, lower lip palsy, total facial nerve palsy, abnormal feeling of neck, or pharyngeal pain.

Given the information presented above, the diagnosis, surgical approach and complications associated with the surgical treatment of PPS tumors, we decided to submit a case report to contribute to improving the outcome of this pathology.

CASE REPORT

A 40-year-old male, initially noted with a tumor on the left side of the neck with a 15-year evolution, presented with a left intraoral mass noticed 1 month earlier and a discomfort and foreign body sensation when swallowing. Clinical examination revealed a left intraoral mass on the projection of the right palatine tonsil, displacing the soft palate

medially and extending to the oropharynx.

The ultrasonography showed a 40x40mm size tumor mass located in the left submandibular region. There was no significant lymph nodes enlargement in the neck area.

For a better characterization of the lesion, a native and contrast enhanced CT scan of the head and neck was performed. The CT scan revealed, in the upper third of the left side of the neck, in the projection of the parotid and masticatory space and the submandibular region, under the lower jaw, medially at the level of the cervical vertebrae C1-C4, anterior to the sternocleidomastoid muscle, a volumetric formation, irregular in shape, with clear and uneven contours, dimensions 40x65x85 mm, uniform density +30H (Figure 1). The tumor deformed the lumen of the oropharynx, displaced the left submandibular salivary gland inferiorly, closely adjoined the inner surface of the body of the lower jaw on the left side.

Clinical and paraclinical examination did not reveal any regional or distant metastasis lesions.

In this case, surgery was the treatment of choice, performed under general anaesthesia. After taking the informed consent from the patient, he was placed in supine position on the operating table, with the head extended and slight rotated to the right. Access to the parapharyngeal space was gained via an upper neck incision at the level of the hyoid bone. An oblique horizontal type skin incision was made, about 2 fingerbreadths below the angle of the mandible and continued parallel to the margin of the mandible and extended to the submental region. The carotid artery and the jugular vein were identified. The digastric and stylohyoid muscles were retracted. Due to the huge size of the tumor (40x65x85mm), in order to maximize the access in the operatory field, the submandibular gland was resected. The facial artery was identified posteroinferior to the gland where it emerges medially to the posterior belly of the digastric muscle. The facial artery was then ligated and divided superiorly to the posterior belly of the digastric muscle. After resecting the submandibular gland by retracting the posterior belly of the digastric posteriorly and the mandible superiorly, we passed a finger/instrument directly into the prestyloid PPS and the tumor was mobilized by blunt and gentle finger dissection. The tumoral mass was resected with surrounding tissue. The marginal mandibular nerve, terminal facial nerve branch, was identified and monitored visually without using nerve monitor and leaved because it was in no contact with the tumor.

The resected tumor had 90x80x70mm in size (Figure 2), with unregularly margin, homogeneous consistency and high density and was sent to the histopathological examination. The postoperative wound was inspected and stitched-up. We placed a drainage tube into the postoperative cavity, which was removed 5 days after.

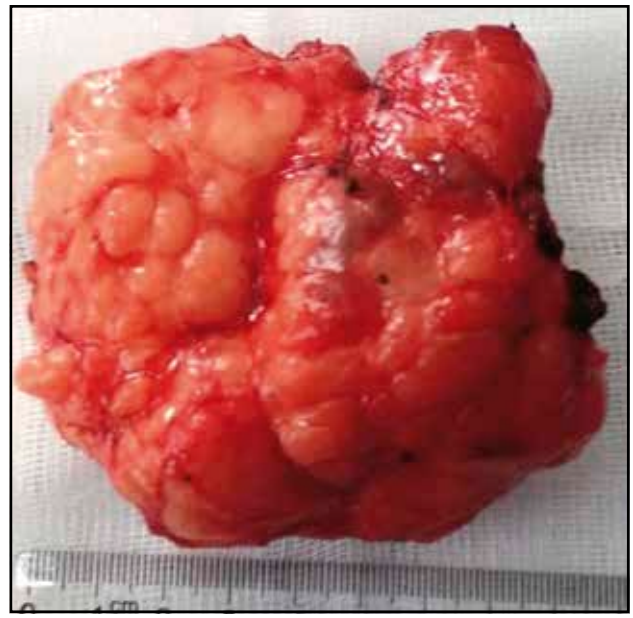


Figure 2. The 90x80x70mm size tumor.

The histopathologic examination of the tumor established the final diagnosis as pleomorphic adenoma of the parotid gland. Microscopically, the pleomorphic adenoma of the salivary glands has a highly variable appearance, being characterized by the presence of polygonal and fusiform myoepithelial cells with mixed proliferation, contained in a variable matrix stroma of myxoid, mucoid, cartilaginous or hyaline origin (Figure 3).

We administered antibiotics cephalosporins, a class of β-lactam antibiotics, and nonsteroidal anti-inflammatory drugs (NSAIDs) for one week postoperatively. Postoperatively, the patient accused pain in the mouth triggered by salivation or when taking the first few bites of food during a meal, the First bite syndrome. There were not any facial nerve injury symptoms. 6 months after surgery, the patient showed no local signs of recurrence.

DISCUSSIONS

Parapharyngeal space tumors may require several surgical approaches: transcervical-transmandibular, transparotid-transcervical, transoral. The proper surgical approach is established according to the size of the tumor, its location, extent, relationship with the nearby structures (critical neuro-

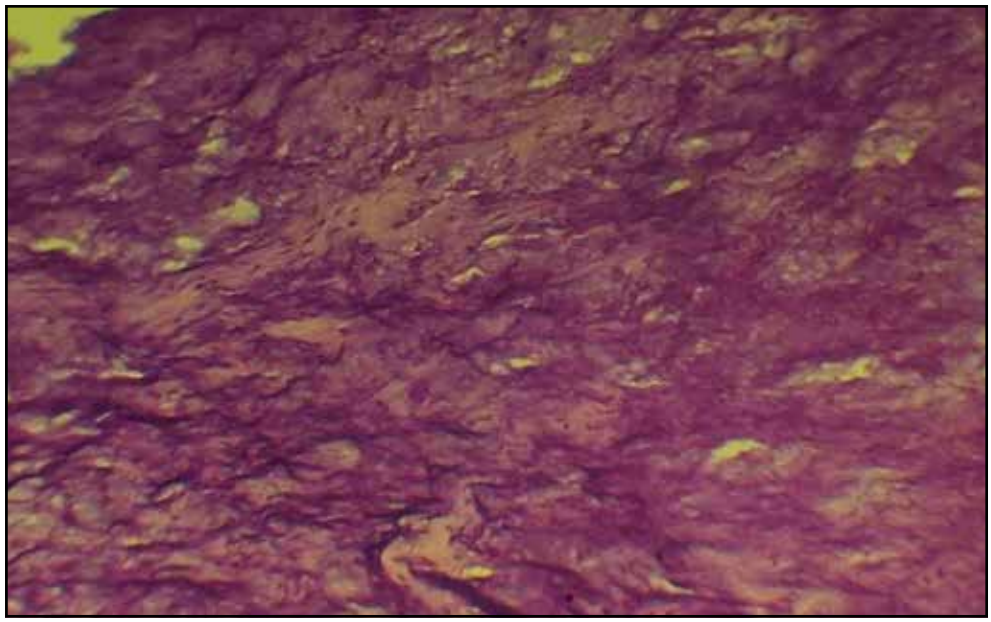


Figure 3. Microscopically, the pleomorphic adenoma of the salivary glands has a highly variable appearance, characterized by the presence of polygonal and fusiform myoepithelial cells with mixed proliferation, contained in a variable matrix stroma of myxoid, mucoid, cartilaginous or hyaline origin.

vascular structures, the skull base), vascularity, and histopathology result[10].

The transoral approach represents the tumor excision through the oral cavity via a mucosal incision over the bulging tumor. This approach provides limited surgical space and, in some cases, may require additional equipment, such as an endoscope or a microscope, in order to improve vision, because during this approach there is a higher risk of tumour spillage and neurovascular injury. This approach is preferred for benign tumours that occupy the prestyloid space, where the margin control is not paramount[11]. The transoral approach allows a better superior visual control of the dissection over the medial side and upper parts of the tumour located close to the skull base, other advantages being the short surgical and hospitalization time, the good functional outcome[12] and no external skin incision. The disadvantages comprise a poor visualization with a secondary risk of damage to major vascular structures, uncontrolled bleeding, cranial nerve injury, and tumour spillage[13], the need for nasogastric tube feeding, postoperative throat pain. Considering the limited surgical space, there are authors who suggest a combined transoral and transmandibular approach in case of massive tumours in order to achieve safe resection margin without tumour spillage[12].

Transmandibular approaches indications include high-grade malignant tumours, massive tumours, large vascular tumours, revision surgical cases, tumors extended into the superior portion of the parapharyngeal space, or with invasion of the skull base or the vertebral body. The classic transmandibular approach provides a wide exposure of the parapharyngeal space and involves a paramedian mandibulotomy, with incision across the floor of the oral cavity and pterygomandibular raphe to the upper gingivobuccal sulcus. In the literature, there are described some surgical variations in this approach, including osteotomies of the body of the mandible and / or at the condylar neck, both also necessitating a tracheostomy.

A transparotid approach can be essential in tumors arising within the deep lobe of the parotid gland, extraparotid salivary tumors, poststyloid tumors, including most neurogenic tumors and small paragangliomas. This surgical technique provides a good identification of the facial nerve and dissection of several or all its branches, being known that the parotid tumors may adhere to the facial nerve, increasing the risk of inadvertent permanent nerve injury during surgery[14].

The transcervical approach for removal of the parapharyngeal space tumors was first described in 1955[15]. The transverse cervical approach is a favourable technique in our practice, with the advantages of providing direct access to the tumor and an adequate control of the neurovascular structures from the neck. Nonetheless, it provides an optimal aesthetic outcome due to preservation of the mandibular continuity. This procedure is also characterized by minimal morbidity and hospitalization time.

The deep lobe of the parotid gland located in the prestyloid compartment is often affected by cancers, representing more than 50% of all parapharyngeal cancers[2]. Even though the incidence varies according to the literature, 67.7% to 84% of the neoplasms start in the parotid gland, 10% to 23% in the submandibular gland and the other cases in the sublingual and the minor salivary glands[6,15-18]. 95% of the parotid gland tumors occur in adults and are rare in paediatric patients[16]. Of all the benign tumors that can develop in the parotid gland, the pleomorphic adenoma is the most common entity.

To determine the extent of the disease, the local spread and the tumor type, the CT scan and the MRI are important diagnostic tools in the investigation. Based on the CT appearance and enhancement characteristics, it is difficult to accurately distinguish a minor salivary gland tumor from a neurogenic tumor. The MRI tissue signal characteristics alone cannot be reliably utilized in making this distinction. Internal carotid artery displacement remains the most reliable distinguishing feature. Minor salivary gland tumors will displace the internal carotid artery posteriorly and laterally, whereas neurogenic tumors will displace the internal carotid artery anteriorly and medially[19].

Along with the parotid gland tumors, there are different types of tumors which can develop in the parapharyngeal space, each of them with its own clinical and paraclinical characteristics. The neurogenic tumors emerge from the trigeminal nerve in the prestyloid compartment and the vagus nerve in the poststyloid space. From an imagistic point of view, the solitary neurofibroma is a well-defined oval or fusiform low-density lesion, with minimal contrast enhancement, while the plexiform neurofibromas are poorly circumscribed, trans-spatial, locally invasive, with low density lesions[20,21]. Solitary neurofibroma may be hardly differentiated from a schwannoma.

On the MRI, the glomus tumours appear as ovoid lesions, with well-defined margins and intense contrast enhancement due to their marked vascularity. An MRI 'salt and pepper' image is characteristic for this type of vascular tumor, the aspect being due to tiny foci of haemorrhage and flow voids within the lesion[20,21]. A differentiation has to be made between the glomus of the carotid body and the glomus vagale. Due to its location, the carotid body tumour widens the common carotid artery bifurcation, while in case of glomus vagale an anteromedial displacement of the internal carotid artery can be identified.

Another pathology which has to be excluded in case of parapharyngeal space lesions is represented by lymphomas. The CT image is a non-specific one as circumscribed homogeneous lesions isodense to muscle, with mild-to-moderate contrast enhancement, and posttreatment necrosis or calcification[20,21]. In lymphoma cases, involvement of the Waldeyer's ring can be a clue in the diagnosis.

Neoplastic infiltration of the parapharyngeal space by malignant tumours has also been reported. Nasopharyngeal carcinoma, oropharyngeal carcinoma, parotid gland and maxillary malignancies, skull base tumors such as meningiomas or chordomas can extend into the parapharyngeal space. The displacement and / or infiltration of the PPS, the internal carotid artery and internal jugular vein identified on the CT or MRI, can give valuable information about the localization of the lesion in one of the neck spaces.

The treatment of parapharyngeal space tumors in general and of pleomorphic adenoma in particular is essentially surgical. Depending on the localization, extension, infiltration, one of the above-described surgical approaches can be used. In our patient, we used the transcervical approach and during the surgical act one of our goals was not to damage and rupture the capsule of the pleomorphic adenoma in order to perform a radical resection. The preservation of an intact capsule is important from the recurrence perspective. The recurrence of the pleomorphic adenoma can be multilocal and may manifest up to 15 years after the initial resection. The risk of a reintervention is represented by the lesion of the facial nerve. In these cases, the intraoperatory facial nerve monitoring would help diminish such risk.

Considering that the cure rates are significantly lower in recurrence surgery (25% less reported in the literature), radiotherapy may be useful in selective cases[22]. In benign salivary glands tumors, the fractionate postoperative radiation dose is 60Gy to 66Gy, in case of close or positive margins, and of 70Gy in case of macroscopic tumors, even if the use of radiotherapy alone in this case does not give a chance of cure[23]. In patients diagnosed with high-grade carcinomas, the recommended doze is 60Gy even when there are negative excision margins[23].

Malignant transformation of pleomorphic adenomas is described in the literature, the degeneration being from both epithelial and mesenchymal components[24,25].

The particularity of our case is represented by the massive parotic gland tumor, with 15-year anamnesis, which was resected with saving the integrity of the mandible. The present case report has brought to light another issue: to do or not a prophylactic tracheostomy before surgery. Trans-

mandibular approaches usually indicate performing tracheostomy to cover alimentation and epithelization.

CONCLUSIONS

Parapharyngeal tumors are a rather rare pathology, initially being asymptomatic. Signs and symptoms are present in the advanced stages due to this tumor's peculiar anatomical location.

The diagnosis is based on radiological examination, CT or MRI imaging and, in some cases, conventional angiography may be used.

The treatment is eminently surgical, the chosen technique depending on the location and the size of the tumor.

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