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Surgical approaches for pleomorphic adenoma of the parapharyngeal space

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Abstract

Objective. This study aimed to analyse the outcomes of surgery for pleomorphic adenoma of the parapharyngeal space in relation to the surgical approach.

Method. This was a single-centre retrospective data analysis conducted from January 2008 to December 2020 on all patients who underwent operation for pleomorphic adenoma originating from the parapharyngeal space.

Results. Twenty-one patients with a mean age of 52.6 years were included. The transparotid-transcervical approach was the most common (52.4 per cent, $n = 11$) surgical approach, followed by transoral robotic surgery (28.6 per cent, $n = 6$) and conventional transoral surgery (19 per cent, $n = 4$). Post-operative complications included nine cases of transient partial facial nerve palsy and two cases of Frey's syndrome after the transparotid-transcervical approach and 2 cases of transient trismus and 1 pharyngeal wound dehiscence in the conventional transoral approach group. Complete macroscopic excision was always achieved, and no recurrence occurred during follow up.

Conclusion. These three approaches can provide adequate tumour visualisation, a high rate of clear excisional margins and an acceptable morbidity.

Introduction

Parapharyngeal space tumours are very challenging in terms of diagnosis and treatment. They represent only 0.5 per cent of all head and neck tumours.[1,2] There is an extensive diversity of histological tumour types arising from parapharyngeal space tumours, but reports show that overall 80 per cent of them are benign and 20 per cent are malignant.[3] Salivary gland neoplasms are considered to be the most frequent, followed by neurogenic tumours.[2] The pleomorphic adenoma is the most common tumour arising from the parapharyngeal space, accounting for 34 per cent of all cases.[3,4]

The parapharyngeal space is described as an inverted pyramid-like space extending from the skull base to the greater cornu of the hyoid bone.[5,6] The sphenoid bone and the petrous portion of the temporal bone define its superior limit (base). The junction of the greater cornu of hyoid bone with the posterior belly of digastric muscle constitutes its inferior limit (apex). The pterygomandibular raphe defines the anterior boundary, and the posterior aspect of the carotid sheath and the prevertebral fascia constitute the posterior limit. The medial boundary is formed by the buccopharyngeal fascia, which covers the superior pharyngeal constrictor muscle and the pharyngobasilar fascia plane. The medial pterygoid muscle and the condyle of the mandible define the lateral border.[6–8]

The parapharyngeal space is separated into pre-styloid and post-styloid compartments by the tensor veli palatine muscle and styloid process.[2,9] Parapharyngeal space tumours can be primary or metastatic and arise from any structure of the parapharyngeal space itself or adjacent structures. Pre-styloid space lesions mainly originate from the retromandibular portion of the parotid gland (deep lobe) and its surrounding adipose tissue or lymph nodes. As for post-styloid space, tumours may arise from the internal carotid artery, jugular vein, cranial nerves IX, X, XI or XII, sympathetic chain, and lymph nodes draining the oral cavity, oropharynx, paranasal sinuses and thyroid gland.[6] Tumours arising from the pre-styloid space are more frequent (59 per cent) compared with the post-styloid space (26 per cent). The rest (15 per cent) consist of lesions with indeterminate origin of both spaces.[3,7] The clinical presentation varies depending on the involved structures, but the most frequent presentations are neck mass and oropharyngeal bulge in the soft palate, which pushes the tonsil posteriorly.[1,2,6]

Pre-operative investigation includes computed tomography (CT) and magnetic resonance imaging (MRI); both are sufficient to identify tumour size, limits, extension, location and its relation to nearby structures.[2,6,9] The surgical approach usually depends on the involved structures and the tumour accessibility. The most common approaches are transmandibular-transcervical, transparotid-transcervical, conventional transoral and transoral robotic surgery.[2,6]

The literature regarding parapharyngeal space tumours is rich, but the heterogeneity of approaches and histology limits the level of evidence. The objective of the current research

Main Article

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Table 1. Symptoms at presentation and examination findings

Parameter	Value (*n* (%))
Symptoms at presentation	
– No symptoms/incidental findings	14 (66.7)
– Neck mass	4 (19.0)
– Pharyngeal pain	2 (9.5)
– Sleep apnoea	1 (4.8)
Examination findings	
– No clinical bulge	10 (47.6)
– Neck mass	4 (19.0)
– Oropharyngeal bulge	10 (47.6)
– Nasopharyngeal bulge	1 (4.8)

is to study the outcomes of the surgical treatment of pleomorphic adenoma of the parapharyngeal space in relation to the surgical approach.

Materials and methods

This single centre retrospective study was conducted from January 2008 to December 2020 at Toulouse University Hospital. We included all consecutive patients treated surgically for a pleomorphic adenoma occupying the parapharyngeal space. We only studied pleomorphic adenoma because it is the most common histopathology and to have a better consistency in terms of clinical outcome comparison.

Patient charts were reviewed retrospectively regarding patient characteristics, clinical presentation, diagnostic investigation, surgical approach, intra-operative data, post-operative complications, histopathology and follow up. Computed tomography and/or MRI scans were reviewed for eligibility. The surgical approach was chosen according to the tumour size, location and extension.

The surgical approaches were categorised into several groups (i.e. transparotid-transcervical, conventional transoral (without robotic or endoscopic assistance) and transoral robotic surgery). Transparotid-transcervical approach included a parotidectomy with facial nerve dissection, along with a cervical extension for a better exposure of the parapharyngeal space.

Results

A total of 21 patients (11 males and 10 females) were included over a period of 13 years. The mean age at diagnosis was 52.6 years (range, 24–75). In the majority of patients (66.7 per cent, $n = 14$), the tumour was incidentally found following a radiological investigation in the area of the head and neck, with either a normal clinical examination (47.6 per cent) or an oropharyngeal bulge (47.6 per cent) in the majority of patients (Table 1).

In terms of clinical investigation, MRI was conducted in all cases for diagnosis and pre-operative evaluation for the surgical approach. A CT scan was performed in 12 patients (57.1 per cent). The radiological findings of all tumours were consistent with a pre-styloid space lesion. The lesions were in contact with the skull base in 12 patients (57.1 per cent). The oropharyngeal bulge was noted radiologically in 14 patients (66.7 per cent).

Fine-needle aspiration cytology (FNAC) was performed transorally ($n = 9$) or transcervically ($n = 5$) without radiological guidance in 14 patients (66.7 per cent) who had a clinically accessible mass. The result suggested pleomorphic adenoma in all cases. Overall, the pre-operative investigation was suggestive of pleomorphic adenoma in all patients.

The surgical approaches used were the transparotid-transcervical approach (52.4 per cent, $n = 11$), transoral robotic surgery (28.6 per cent, $n = 6$) or the conventional transoral approach (19 per cent, $n = 4$) (Figure 1 and 2). No mandibular swing or combined approaches were required. The en bloc tumour excision was achieved with no intra-operative macroscopic capsular rupture in all but 3 patients (14.3 per cent). The first patient operated on with the transparotid-transcervical approach had a mass in contact with the skull base, for which the access was limited, leading to capsular rupture. The other two cases underwent transoral robotic surgery and had focal capsular spread. The fact that both tumours had prior biopsy breaching the capsule may be a predisposing factor for intra-operative capsular rupture. However, the excision was deemed macroscopically complete in all three patients, and no residual tumour was seen on the MRI conducted at one month and one year post-operatively.

The mean operating time of each approach was as follows: transparotid-transcervical, 246.6 minutes; transoral robotic surgery, 190.8 minutes; and transoral surgery, 108.8 minutes. The transparotid-transcervical approach was significantly longer than the conventional transoral approach and transoral robotic surgery ($p < 0.01$). In addition, transoral robotic surgery was longer than the conventional transoral approach ($p = 0.0004$). There was no documentation of significant blood loss intra-operatively in our series. The length of hospitalisation showed no significant difference between the three groups, with a median of five days in all three surgical approaches.

The final histopathology was confirmed as pleomorphic adenoma in all patients. Most tumour subtypes (87.5 per cent) were stroma rich or myxoid. Adverse pathological features were seen in only two patients who presented with an incomplete capsule alongside either pseudopodia ($n = 1$) or satellite nodules ($n = 1$). Clear margins could not be confirmed in only the 3 patients with a capsular rupture (14.3 per cent).

The most common complication was facial nerve palsy (42.9 per cent, $n = 9$) in the transparotid-transcervical group, which was limited to the marginal mandibular nerve (33.3 per cent, $n = 7$) or was diffused for two patients (9.5 per cent) with House–Brackmann grade IV facial paralysis. The weakness was transient and resolved during follow up of up to nine months in all cases in the transparotid-transcervical group. It was attributed to prolonged retraction of the nerve during the dissection in the parapharyngeal space. Transient limitation of mouth opening was seen in 2 patients (9.5 per cent) of the conventional transoral group. It was likely related to prolonged use of the mouth gag. It was noted that no instances of first-bite syndrome occurred. Two patients (9.5 per cent) presented with Frey's syndrome. Both patients underwent a transparotid-transcervical approach for a tumour involving the superficial lobe, without possibility of preserving the superficial musculoaponeurotic system. In the transoral robotic surgery group, one patient presented with wound dehiscence at day seven, which required a transoral revision surgery for wound closure.

After a mean follow up of 21 months (range, 1–60), no patient experienced recurrence. Our protocol of follow up included a clinical examination one month and one year after surgery and an MRI at one year post-operatively. In

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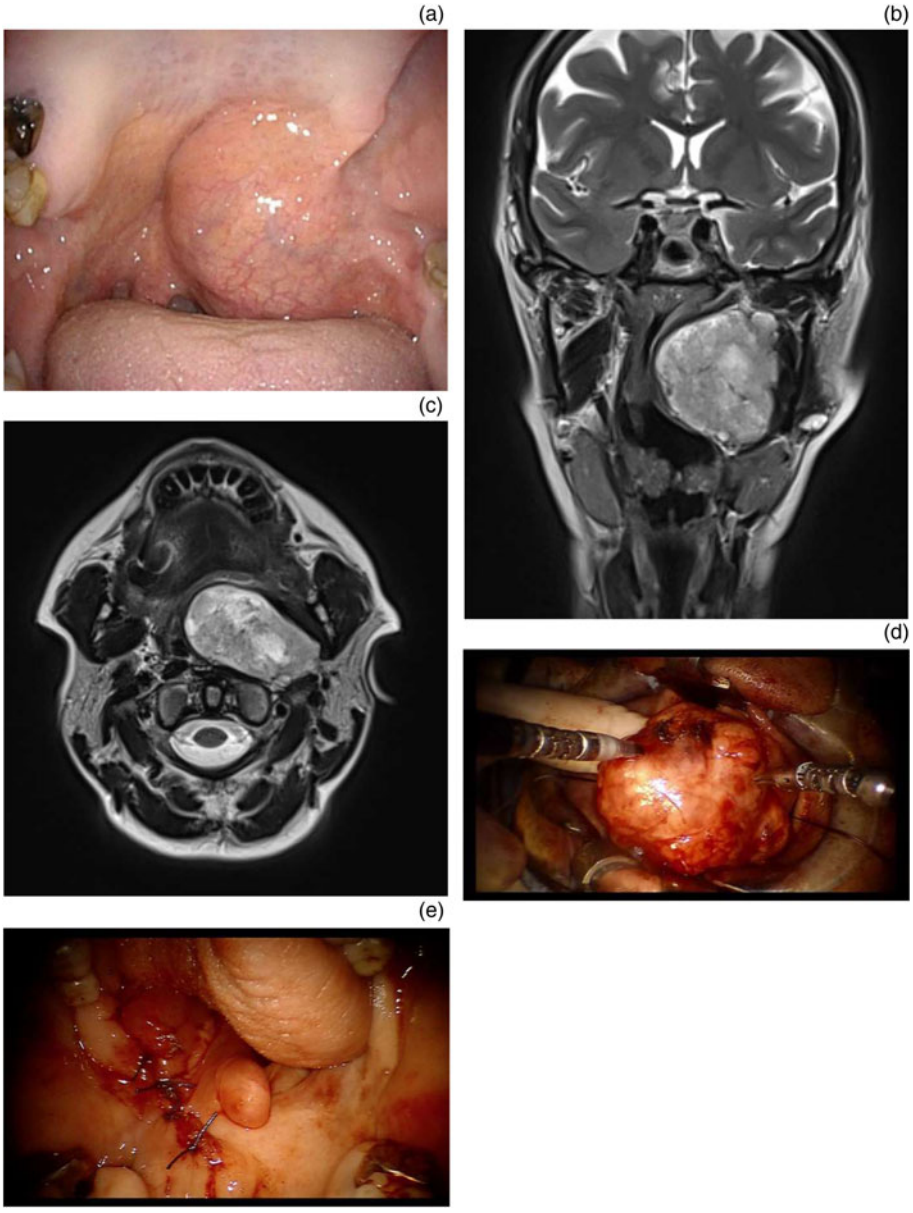

Fig. 1. Pleomorphic adenoma of the left parapharyngeal space in a 52-year-old woman. (a) Clinical examination showing an oropharyngeal bulge, (b) pre-operative coronal plane magnetic resonance imaging (MRI) scan, (c) pre-operative axial plane MRI scan showing a 5 cm mass occupying the parapharyngeal space, (d) intra-operative view of final dissection and delivery of the tumour with the finger and (e) intra-operative view of primary closure of the oropharynx.

case of intra-operative capsular rupture, an MRI was performed at one month to confirm the completeness of excision. Most patients were then referred to their primary care physician for further follow up.

Discussion

The clinical presentation in our series was consistent with what is found in the literature.[1,3,10] The majority of our patients were asymptomatic at the time of diagnosis, which is a strong indicator that parapharyngeal space lesions are underdiagnosed. In terms of clinical investigation, MRI and FNAC were sufficient for an adequate clinical diagnosis and to determine the preferential surgical approach. Magnetic resonance imaging is the preferred imaging as it provides sufficient information concerning the tumour origin, nature, size and its relation to nearby vital structures. Moreover, it is well documented that MRI is superior to CT in diagnosis and assessment of pleomorphic adenoma of the parapharyngeal space.[11,12] Therefore, we can suppose that MRI is sufficient to suggest the diagnosis of pleomorphic adenoma pre-operatively. However, we believe that FNAC can still provide a significant input, despite limited access to the parapharyngeal space. It largely relies on the experience of the cytologist and the surgeon or radiologist taking the sample.[13–15]

Understanding the anatomy of parapharyngeal space is crucial for the correct diagnosis and, most importantly, to decide how to approach the parapharyngeal space for the tumour excision. The majority of parapharyngeal space tumours are of salivary origin and are located in the pre-styloid compartment.[3,4,10] There are several factors determining the surgical approach to a parapharyngeal space tumour (e.g. its size, location and histopathology). Nevertheless, the decision is always subjective and depends on the surgeon's preference and comfort level.[16] An attempt to standardise surgical approach decision-making was made by Kanzaki and Nameki before the advent of transoral robotic surgery and relying on the pre-operative imaging and the division of the parapharyngeal space into six compartments.[17]

The transoral approach was first described by Harry Ehrlich in 1950 and is the most debated surgical approach in the literature.[18,19] It is associated with minimal access and poor vision of the operating field,[20] and it was only used during the first

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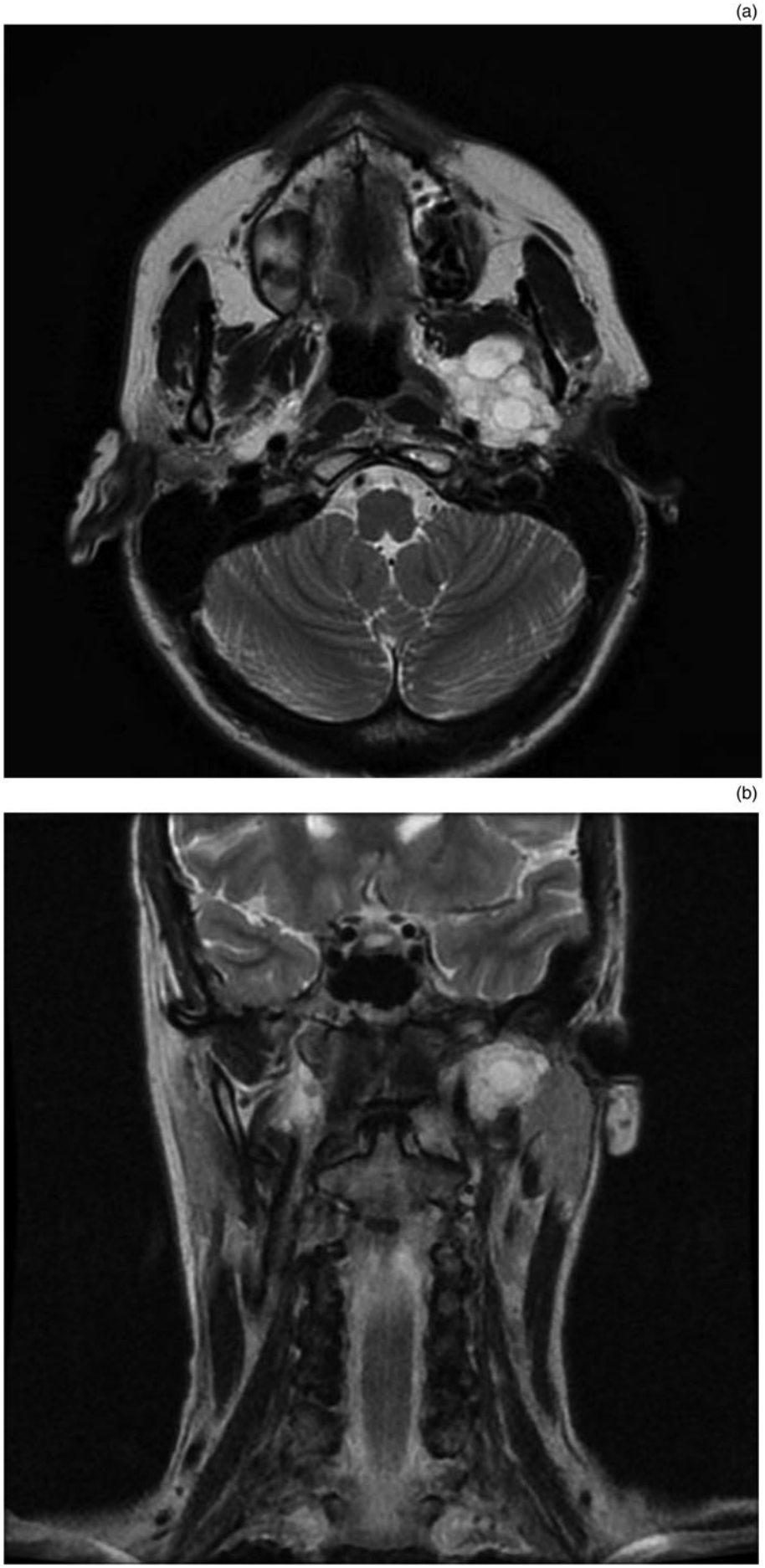

Fig. 2. Pleomorphic adenoma of the upper portion of the left parapharyngeal space in a 52-year-old woman, discovered incidentally on magnetic resonance imaging (MRI). (a) Pre-operative axial plane MRI slice, (b) pre-operative coronal MRI slice showing a mass limited to the upper portion of the parapharyngeal space, extending to the skull base and (c) intra-operative view after facial nerve dissection and tumour excision.

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Fig. 2. Continued.

years of our study period, prior to the use of transoral robotic surgery in our centre. Transoral robotic surgery is a relatively new approach, with the first documented use in 2007.[21] The Da Vinci® robotic system has the advantages of a magnified three-dimensional visualisation, tremor-reduction technology and wristed instruments with seven degrees of freedom.[16] Some studies suggest these advances have reduced the risk for tumour rupture, incomplete excision and uncontrollable bleeding intra-operatively.[20] For centres where transoral robotic surgery is not available, an endoscope-assisted transoral approach seems to provide satisfactory access.[22] The pre-operative investigation is crucial before making the decision of a transoral approach. The evaluation of the mouth opening is the most important aspect to anticipate the quality of both exposure and working space. Pre-operative imaging is paramount to rule out carotid artery encasement and bony erosion of the skull base. The main contraindications for transoral approaches are trismus, macroglossia and maxilla-mandibular defects.[23,24] Other studies advised against transoral robotic surgery for tumours bigger than 6 cm.[23] Another advantage of transoral robotic surgery is to limit the need for a transpalatal approach. The entrance door to the parapharyngeal space is made by a linear mucosal incision around the tonsillar fossa, preserving the palatoglossus, palatopharyngeus and superior constrictor muscles. Dividing the pterygoid muscles can improve the mouth opening and transoral exposure. There is usually no need for a mucosal reconstruction as a primary closure can be achieved.

The main risks for all transoral approaches are neurovascular injury, tumour spillage or implantation, incomplete excision, and surgical site infection.[18,25] In our series, two patients out of six from the transoral robotic surgery group presented with intra-operative capsular rupture. Both patients had undergone transoral incisional biopsies prior to the surgery, which was likely to induce a capsular fragility. A history of biopsy should be considered as an argument in favour of a transoral approach, in order to avoid seeding along the transcervical incision and to obtain a safe mucosal margin intra-orally. Moreover, follow-up imaging did not show any evidence of residual or recurrent disease. Some authors claim that tumour spillage during a transoral approach allows for a thorough washing and prevents tumour dissemination in the neck.[26]

The transparotid-transcervical approach was the most common in our series and is preferred by many for the good access and visualisation it provides.[27–29] The keystone for this surgery is to preserve the facial nerve and limit the retraction injury. In order to improve the exposure, the authors recommend a nasotracheal intubation for a complete jaw closure during the surgery and an extension of the classical Blair's incision in the submandibular area to provide a better exposure of the major blood vessels and cranial nerves. The posterior belly of digastric muscle, stylohyoid muscle and stylomandibular ligament are commonly divided. The submandibular gland is retracted anteriorly or excised, and the mandible is protracted to identify the major neurovascular structures of the post-styloid space.[8,30] A parotidectomy is performed, as completely as possible posteriorly and in the deep lobe. Some studies advocate for preserving the superficial lobe of the parotid gland whenever feasible, with the aim of reducing the risk of Frey's syndrome and for a better cosmetic outcome.[31] The main indication for this approach is a mass reaching the skull base and/or merging laterally within the deep lobe of parotid gland with ill-defined contour on radiological investigations.[4,5]

The transcervical approach is the most frequently used for excision of parapharyngeal space mass of all origins, most notably those arising from the post-styloid compartment.[18] The parapharyngeal space is accessed through a submandibular incision at the level of the hyoid bone, which can be further

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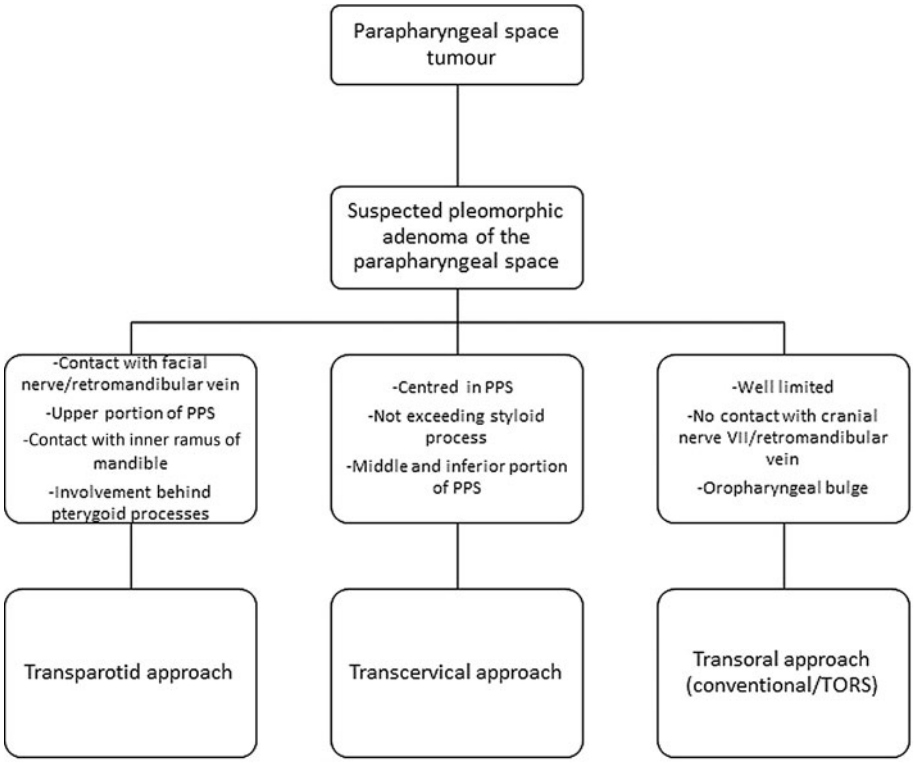

Fig. 3. Proposal of decision-tree for management of pleomorphic adenoma of the parapharyngeal space (PPS). TORS = transoral robotic surgery.

extended to the submental area for mandibulotomy with lip splitting. Many surgical steps are similar to those of the transparotid-transcervical approach (i.e. retraction of the submandibular gland, division of the posterior belly of the digastric muscle, the stylohyoid muscle and stylomandibular ligament and protraction of the mandible). The facial nerve is not identified. The mylohyoid muscle can be transected for further exposure of parapharyngeal space.[12] However, none of our cases underwent an exclusively transcervical approach because all tumours originated from the deep lobe of the parotid gland, which justified a parotidectomy in our opinion. Some studies suggest the transcervical approach should be avoided for masses bigger than 4 cm because the mandible is a barrier for tumour manipulation and extraction.[3,6,18,27,32] Furthermore, it is not recommended for tumours with a long vertical dimension and with radiological suspicion of cranial foramen invasion as they are associated with a high risk of internal carotid artery laceration during blunt dissection.[12] Overall, many authors support the transcervical approach because it can provide good control of the lesion while avoiding dissection of the facial nerve, but it is mainly reserved for tumours with limited attachment to the deep lobe of the parotid.[5,33]

Mandibulotomy is essentially indicated for malignant tumours, recurrent neoplasms, large benign tumours and highly vascular neoplasms with a need for vascular control.[12] There are three ways of conducting the osteotomy: through the body (lateral mandibulotomy), and midline and paramedian approaches. Following the osteotomy, the mandible is swung and the floor of the mouth is stretched. An incision in the floor of the mouth is made 1 cm medial to the gingiva across the mucosa, soft tissues and muscles of mouth floor and extended up to the anterior tonsillar pillar. It carries a possible risk of inferior alveolar nerve anaesthesia, malocclusion, teeth loss, malunion or non-union of the mandible, and in some cases it might require a tracheostomy.[30,34] No mandibulotomy was performed in our series, although it is reported in 2.0 to 20.5 per cent of cases in the literature.[8,27,33,35,36] From our standpoint, mandibulotomy is now unnecessary for the vast majority of pleomorphic adenomas of the parapharyngeal space. Additionally, no combined approach was necessary in our series for a complete tumour manipulation and excision. As a result, the morbidity and complications of the mandibular split were avoided, and the length of hospitalisation was limited.

- Parapharyngeal space tumours can be excised with various surgical approaches
- Accessibility and involved structures, shown on magnetic resonance imaging and computed tomography, are the main determining factors for surgical approach
- The transparotid-transcervical approach is chosen when there is no oropharyngeal bulge or there is intra-parotid location or lateral extension behind the pterygoid process or inner ramus of mandible
- The chosen approach must provide adequate tumour visualisation in order to achieve clear excision margins

The histopathological subtypes of pleomorphic adenoma are divided into three groups based on their epithelial element: mucoid, myxoid or chondroid.[37] The most common is the myxoid subtype, which is documented to have a higher rate of recurrence and is associated with incomplete and thin capsules.[38] However, there are a number of pathological differences between a pleomorphic adenoma originating from the superficial or deep lobe of the parotid gland. Deep lobe pleomorphic adenomas tend to be bigger, have a thicker capsule and there is less invasion of the capsule by the tumour.[39] The rate of recurrence documented in the literature ranges between 0 and 10.5 per cent.[5,12,27,31,40,41] Our rate of complete surgical excision and the absence of recurrence during the follow up seem to validate our management. Indeed, a concession can be made on the surgical exposure in order to avoid the morbidity of a systematic transcervical approach or a mandibulotomy.

The complications were dominated by facial nerve palsy in our series because of the prolonged retraction of the nerve

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during tumour dissection in the parapharyngeal space. This occurs from 5.2 to 30.8 per cent of cases in the literature. Permanent facial nerve palsy has been reported after up to 7.7 per cent of procedures.[2,4–6,27,31]

The limits of our study are its retrospective design and the small number of patients, which prevented statistical comparison between groups and a short follow-up period. Indeed, these tumours evolve slowly and late recurrences can still occur.

Our decision tree for management of pleomorphic adenoma of the parapharyngeal space was based on clinical examination and radiological findings (Figure 3). The transparotid-transcervical approach was preferred when close contact to the facial nerve was suspected. It should be preferred to the transcervical approach for salivary tumours. Transoral robotic surgery is a good option for selected, well-defined masses in the parapharyngeal space with oropharyngeal bulge clinically and radiologically. The classic transoral approach should be avoided if transoral robotic surgery is available because it is associated with significant risk of neurovascular injury with limited capability for intra-operative management. Indications for mandibulotomy must remain exceptional.

Conclusion

Pleomorphic adenomas of the parapharyngeal space are rare tumours, often asymptomatic and largely underdiagnosed. Pre-operative assessment is paramount in order to choose the most suitable surgical approach. In selected patients, transparotid-transcervical, transoral robotic surgery and conventional transoral approaches can provide adequate tumour visualisation, a high rate of clear excisional margins and an acceptable morbidity. Indications for mandibulotomy for pleomorphic adenoma of the parapharyngeal space should be exceptional.

Competing interests. None declared

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