Giant Parapharyngeal Space Pleomorphic Adenoma of the Deep Lobe of Parotid Presenting as Obstructive Sleep Apnoea: A Case Report & Review of the Diagnostic and Therapeutic Approaches

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Abstract

Introduction Salivary gland tumours constitute about less than 4 % of all head and neck tumours. Pleomorphic adenoma, also called benign mixed tumour, is the most common tumour of the salivary glands. About 80–90 % of these tumours occur in the major salivary gland mainly parotid gland and 10 % of them occur in the minor salivary glands.

Aims and Methods Aim of this case report is to discuss the unique case of giant parotid pleomorphic adenomas arising in the deep lobe involving the parapharyngeal space and difficulty in respiration at sleep during nights reported at this institute. The patient was undergoing treatment for obstructive sleep apnea syndrome when she reported at this institute for disturbed sleep. Diagnosis was based on computed tomography scan and magnetic resonance imaging and cytology by means of fine needle aspiration biopsy.

Conclusion An exhaustive pre-operative diagnostic algorithm is mandatory before approaching such lesions involving parapharyngeal space. Fine needle aspiration biopsy is, in our opinion, mandatory to avoid histological surprises. The surgical approach varies according to the location of the tumour and should provide excellent visibility with wide surgical exposure to secure local neurovascular structures.

Keywords Deep lobe pleomorphic adenoma · Obstructive sleep apnea · Tran cervical approach · Mandibular swing

Introduction

Pleomorphic adenoma is the most common salivary gland neoplasm, accounting for 63 % of all parotid gland tumours [1]. It derives its name from the architectural pleomorphism seen by light microscopy. It is also known as “mixed tumor, salivary gland type”, which describes its pleomorphic appearance as opposed to its dual origin from epithelial and myoepithelial elements [2].

Pleomorphic adenomas may occur at any age, but mainly they affect patients in the 4–6 decades. Male: female ratio is 2:3 [3]. Most tumours originate in the superficial lobe but, more rarely, these tumours may involve the deep lobe of the parotid gland, growing medially and occupying the parapharyngeal space. Deep lobe parotid pleomorphic adenomas are rare tumors that present a diagnostic and therapeutic challenge. Approximately 10–12 % of pleomorphic adenomas of the parotid are thought to arise from the deep lobe of the parotid [4].

Tumours located in the parapharyngeal space are relatively rare, and experience in early diagnosis and treatment is very limited because of absence of symptoms. Management of these tumours is more challenging due to the anatomical location of the parapharyngeal space [5] (Fig. 1).

Herewith a unique case of giant deep lobe parotid gland pleomorphic adenoma presenting with obstructive sleep apnoea, treated at our institute is presented. A review has been made of the literature, discussion of which focuses on the diagnostic and therapeutic approaches (Fig. 2).

Case Report

A 40-year-old female patient reported to the department of craniofacial surgery at Meditrina Institute of Medial

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Fig. 1 MRI Showing axial section

Fig. 2 MRI showing sagittal section

Sciences, Ramdaspath, Nagpur, India, complaining of the sensation of a foreign body in the pharynx which had been present for last 6 months. There was history of difficulty in

swallowing and respiration and frequent disturbed sleep during night since last 6 months. The frequency of disturbed sleep h increased since last 6 months and patient

Fig. 3 Showing transcervical approach with osteotomy prepared for mandibular swing

was not able to sleep during nights. She was previously diagnosed as case of Obstructive Sleep Apnoea Syndrome and was undergoing the treatment for the same.

Clinical examination showed no evidence of any pathology extraorally. Intraoral examination showed a swelling on the right side of the soft palate crossing the midline towards the left. It was firm in consistency with a normal overlying mucosa. No latero-cervical adenopathies were clinically appreciable. Computed tomography (CT) showed fairly defined, soft tissue lesion in the right parapharyngeal space with the lesion extending postero-laterally in between the right ramus of mandible and styloid process. The lesion was displacing the right lateral pterygoid muscle antero-medially. Medially it was bulging in the nasopharynx and the oropharynx, significantly compromising the airways. Magnetic Resonance Imaging showed large lobulated well defined homogeneously hypointense lesion on T1 WI and hyperintense lesion on T2 and STIR WI in the right parapharyngeal space. The lesion showed thin hypointense septae on T2WI with some hypointense soft tissue within. It measured $54 \times 65 \times 35$ mm (SI $\times$ RLO $\times$ AP) approximately. The lesion showed moderate to strong inhomogeneous enhancement on post contrast study. It was growing along the intero-medial axis. The deep lobe of the right side parotid gland was not separately identified from the lesion and it was abutting the neurovascular bundle of the parotid gland from the inner aspect. There was widening of the stylo-mandibular canal noted with truncation of the lesion at this point giving subtle dumbbell shape. Along the medial border it was indenting over the oropharynx and lateral pharyngeal wall smoothly with well defined fat planes between the lateral pharyngeal wall and

Fig. 4 Showing tumour mass after mandibular swing

lesion. There was significant narrowing of the oropharynx noted. There was complete effacement of the parapharyngeal fat seen on the right side. Anteriorly the lesion was smoothly indenting over the pterygoid muscles. Postero laterally the lesion was causing smooth scalloping over the ramus of mandible. Antero inferiorly it was abutting the mandibular gland without invasion. Postero-medially the lesion was indenting over the prevertebral muscles. Superiorly the lesion was not involving the base of skull or did not show any intracranial extension (Figs. 1, 2).

Fine needle aspiration cytology (FNAC), performed transorally, revealed a cytological picture of pleomorphic adenoma with some atypical neoplastic cells.

After obtaining the patient's informed consent, surgery was planned to approach the deeper lobe of parotid gland and parapharyngeal mass by transcervical approach, in order to achieve radical excision of the mass. After exposure of the parasymphysis, body, and ramus of the mandible mandibular swing access osteotomy was planned. The osteotomy cuts were planned by preserving the inferior alveolar neurovascular bundle and the medial surface of the mandible was exposed to visualize parapharyngeal structures (Figs. 3, 4). The tumor mass was visualized after further dissection and it was completely excised in toto. The swung mandible was repositioned after doing intermaxillary fixation and fixation was done by miniplates (Fig. 5). The measurements of the tumour were $5.5 \times 6.5 \times 3.5$ cm in size (Fig. 6) Histopathological examination revealed the features suggestive of pleomorphic adenoma. The patient's complaint of obstructive sleep apnoea was completely resolved from the second post operative day. The patient was discharged after 7 days with no facial nerve deficit.

Fig. 5 Showing fixation of osteotomy by miniplates

Discussion

Salivary gland tumours constitute about <4 % of all head and neck tumours. These tumours are commonly seen in adults [6]. It is the commonest benign salivary gland tumor; 84 % of the pleomorphic adenomas occur in the parotid, 8 % in the submandibular, and 4–6 % in the minor salivary glands. Among the various histological varieties of salivary gland tumours pleomorphic adenoma happens to be the commonest one constituting about 70 %. Most of pleomorphic adenomas involve the major salivary glands mainly parotid gland [7].

Pleomorphic adenoma is the most frequent parotid gland tumour, presenting a high rate of recurrence even if it resembles a benign neoplasm. Due to the few symptoms complained of by the patient and the possibility of extension into a hidden site, such as the parapharyngeal space, they can grow for a long time before being diagnosed, and the potential risk of malignant transformation increases over the years with an incidence of 1–7 % [8].

Deep lobe pleomorphic adenomas are generally discovered, during routine physical examination, as an asymptomatic mass. Indeed, they remain silent for a long time and the slow growth does not lead to symptoms even if the tumour is in contact, or displaces vital structures located in the parapharyngeal space such as vessels or nerves. In this reported case the uniqueness is that the patient was diagnosed as a case of obstructive sleep apnoea and was undergoing positive ventilator pressure therapy. The patient was examined intraorally when she was referred to our department where the tumour mass was clinically observed. Diagnostic imaging, such as CT or MRI, is mandatory in such cases in order not to miss any pathology present in the upper respiratory tract and to know the extent of the tumour. MRI was more informative in this case on account of its

Fig. 6 Showing excised specimen of 54 x 65 x 35 mm size

better definition of soft tissue, providing precise information concerning tumour margins as well as the relationship with the surrounding structures (Fig. 5).

Much controversy exists regarding the use of FNAC in the diagnostic procedures due to localization of these lesions and their relationship with the vascular and nervous structures that can be damaged by this kind of examination. In our opinion, FNAC is a reliable procedure that can guide the surgeon and be useful in choosing the right surgical approach [9, 10]. Even though it is not the first choice diagnostic tool, but it should be performed following diagnostic imaging in order to exclude a vascular lesion. Depending upon the location of the lesion, it can be performed intra-orally or percutaneously [11]. In our case, reliable results was obtained with FNAC intra-orally which was confirmed after excision of the lesion by histopathological examination. Open neck or trans-oral biopsies should be avoided, since opening the tumour capsule increases the risk of recurrence (Fig. 6).

Many different approaches have been described in the literature including intraoral and extraoral interventions for such space occupying pathologies. Approach for such tumours is always debatable and one must agree on the need to perform surgery requiring adequate exposure to identify and protect vital structures and ensure complete removal [12, 13].

Intraoral tumour resection can be achieved by trans-palatal approach but, in spite of the aesthetic benefit, it does not offer good control of the vascular and nervous structures on account of its small working area and it does not provide adequate exposure for removal of the tumour which is often extirpated fragmented, increasing the rate of recurrence. Therefore, this technique should be indicated only for small benign lesions (<3 cm) which occupy the anterior parapharyngeal space [15].

Deep lobe parotid tumors of all types should be removed by an external approach so that the facial nerve can be identified with precision and preserved undamaged. Access can be greatly improved by division of the stylomandibular ligament and styloid process, mandibulotomy, or by using an upper lateral cervical approach through the submandibular fossa [14]. Overall, transparotid, transcervical and transpharyngeal approaches are the extraoral approaches which are commonly used for such pathologies. Mandibulotomy can be performed to improve exposure.

Transparotid approach starts with a superficial parotidectomy with facial nerve preservation. The facial nerve is then separated from the deep lobe of the parotid gland and retracted. The dissection continued posteriorly and inferiorly around the mandible. Mandibulotomy can be performed if necessary to improve exposure [12].

Transcervical approach starts with a transverse incision at the level of the hyoid bone. The submandibular gland is often removed or retracted anteriorly. An incision through the fascia deep to the submandibular space allowed for entry into the parapharyngeal space and blunt dissection of the tumor. This approach frequently involves blind finger dissection in the parapharyngeal space and does not provide enough exposure for larger benign lesions extending cranially or those with a more aggressive growth pattern. This can be combined with mandibulotomy for better exposure. The key to the site of mandibulotomy is to avoid injury to the inferior alveolar nerve while providing access to parapharyngeal space.

Cervical transpharyngeal approach is used to excise large and highly vascular tumors. In this approach mandibulotomy is performed anteriorly and incision is made along the floor of the mouth up to the anterior pillar. The advantage of anterior mandibulotomy is the ease with which mandible can be swung laterally. It is thus known as mandibular swing approach [16]. In our case we used the combination of approach considering the extension of tumour and its relations with vital structures. Thus the damage to the facial nerve was avoided as we avoided the superficial lobe parotidectomy.

Some authors disagree with this approach as aesthetics are compromised by the labiotomy incision, and it also requires entering the oral cavity and involves salivary contamination of the wound [17]. In our opinion, a well-trained surgeon, paying attention to the suture, can achieve good aesthetical results without infection of the wound.

Conclusion

We present a rare case of obstructive sleep apnoea secondary to a deep lobe parotid pleomorphic adenoma and discuss its management, including the need for a thorough

examination and multidisciplinary approach. Only a few cases of pleomorphic adenoma of the deep lobe of the parotid gland causing OSA have been reported in the literature. Our case is not only extremely rare but also highlights that key diagnosis may be missed if a thorough assessment of the patient with proper intraoral examination is not performed. Deep lobe parotid pleomorphic adenomas are rare benign tumors that are often clinically indistinguishable from lesions of the superficial lobe of the parotid. Recurrence is rare following complete surgical excision via an external approach.

An exhaustive pre-operative diagnostic algorithm is required before approaching such a lesion. CT and MRI provide important information about the location and margins and can guide the surgeon in planning the right approach. FNAC in our opinion is mandatory to avoid any histological surprise. The surgical approach should provide excellent visibility with wide surgical exposure to secure local neurovascular structures in our experience. Combination of the approach without injuring the facial nerve is the key for managing such lesions.

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