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RESEARCH ARTICLE

PRIMARY AND LARGE PLEOMORPHIC ADENOMA OF THE PARAPHARYNGEAL SPACE: A CASE REPORT

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Abstract

Primary pleomorphic adenoma (PA) of the parapharyngealspace (PPS) is relatively rare. It shows a real diagnostic and therapeutic challenges. We report a case of a 25-year-old male, who was consulting for dysphagia and sleep apnea syndrome evolving over eight months. Physical examination revealed a submucosal mass arinsing from the lateral wall of the left oropharynx, narrowing its lumen. Imaging revealed a heterogeneously enhancing tumor in the left PPS, separated from the deep lobe of the parotid, measuring 60/47 mm. The patient underwent combined transcervical and transoral approach, complete tumor removal was insured. Histopathologic study confirmed the diagnosis of PA. No postoperative complications and no recurrence were noticed during 24 months of follow up. Primary PA arising in the PPS is of rare occurrence. The diagnosis seems difficult and remains based on histopathological examination. A suitable surgical approach must be required to insure complete and safe removal of this tumor.

Introduction: -

The pleomorphic adenoma (PA), formerly known as the "mixed tumor", affects the main salivary glands. Its location in the accessory salivary glands known as atypical location, does not exceed 9% of cases, pleomorphic adenoma arising de novo in the para-pharyngealspace (PPS) is very rare. It shows difficulties in term of its origin (primary or originating from the deep lobe of the parotid), presents challenges of diagnosis and surgical managment due to the non specific symptoms of this entity and the complex anatomy of this space.We share our experience with an exemplary case of primary and large PA of the PPS and we discuss its diagnosis and surgical approaches.

Case report:

A 25 year -old male, with no clinical history, presented to our head and neck department, with an eight months history of dysphagia, odynophagia, sleep apnea syndrome and pharyngeal speech, with no alteration of general state. Physical examination revealed a submucosal mass arinsing from the lateral wall of theleft oropharynx, displacing the soft palate and the right anterior tonsillar pillar anteromedially, and narrowing the oropharynx (Fig. 1), on palpation, this mass was firm, with poorly defined limits, fixed, painless, and no-pulsatile, with nocervical expression.

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There were no lymphadenopathy or cranial nerve deficits. Cervical and facial CT- scan showed an homogeneous hypodense well defined oval lesion occupying the left parapharyngeal space and causing luminal narrowing, measuring 60 mm / 47 mm, not taking the contrast, without bone lysis, without invasion of the soft tissues, the parotid gland had normal appearance and there was no cervical lymphadenopathy (Fig.2). Magnetic resonance imaging (MRI) with angiography demonstrated a well demarcated heteregenously mass occupying left prestyloid space in contact with carotid arteries without spacing of their bifurcation, this lesion compromised pharyngeal space and was extended to the skull base without intracranial invasion. It measured 60 mm/47 mm, with moderate contrast enhancement, it was separated from the deep lobe of the parotid (Fig.3). An endobucal biopsy was performed under general anesthesia, the histological study showed dual epithelial and conjunctive tumoral proliferation in favor of pleomorphic adenoma. The patient underwent large surgical excision of the tumor by combined transoral and trancervical approach, avoiding mandibulotomy. The excision of the tumor was complete used an extracapsular blunt dissection through left paramedian pharyngotomy, the large vessels were controlled through a lateral left neck incision (Fig.4). The anatomopathological study of the piece confirmed the diagnosis of pleomorphic adenoma. Immediate postoperative outcomes were unventhfull, there were no swallowing, no cranial nerve paralysis, no hematoma, no infection, the feeding was taken orally after 7 days, then, the patient was discharged home. The trismus was disappeared within 6 months, there was no sign of clinical or radiological recurrence for a follow-up of 24 months (fig. 5, fig. 6).

Discussion:-

Generally, PA of PPS is an extension of a PA developed from the deep lobe of the parotid [1], when is primary located in PPS, is exceptional, only a few cases have been reported in the literature according to our knowledges. [2,3]. Two theories could explain the ectopic situation of the PPS PAs: the first is based on the pharyngeal extension of the deep lobe of parotid in the prestylian space [1]. Our case does not correspond to this description. The second theory uses a study by C. Micheau, reporting that salivary tissue can be found all the way up the neck. The presence of ectopic salivary tissue maybe due to scattering of salivary lobules from the main salivary glands, but separated by other structures [4], which would be the case of our patient.

Primary PA of PPS remains asymptomatic, if the tumor does not exceed 2.5 cm in diameter, the tumor syndrome is progressive and dominates the clinical presentation, as was the case withour patient. The presence of cranial palsy of the cranial nerves should be alarming to look for malignant tumor.

MRI and CT scan provides the most useful information about the nature of the tumor, its margins, as well as appreciating the relationship of the tumor to the surrounding structures. The MRI angiography shows the position of the carotid artery relative to the tumor and thus helps in determining the surgical approach [5]. In our case, we had eliminated the vascular character by comparing both MRI angiography and CT scan outcomes, then, we had performed a biopsy to retain the diagnosis of PA.
Some authors, such as İstemihan et al, perform fine-needle aspiration cytodiagnosis to support the diagnosis of PA of PPS [6], but, it should be practiced after eliminating a vascular tumor with imaging .We have conducted a biopsy, because of the benign and non-vascular aspect of the mass, this attitude had been avoided by some authors considering the great risk of haemorrhage, nerve injury, capsule rupture and recurrence of PA [7].

Several surgical approaches have been described for the management of PPS tumors: the more common among them for the excision of PPS PAs are the transcervical, transoral, transparotid transcervical, the transmandibular and endoscopic robotic assisted approaches [8].The best surgical approach is that one which allows good exposure of the tumor, to ensure its complete resection. The surgical management was difficult in our case, given the deep location of the tumor, the large vessels and nerves passing through the region, on the one hand, the size exceeding 50 mm in diameter and the extension to the skull base on the other hand.

The transcervical transparotid approach is indicated according to Horowitz et al. in PPS PA swith large attachment to the deep lobe, or when the tumor originated from the deep lobe of the parotid, but the risk here is the facial nerve damage. For the same authors, the transcervical approach alone, allows avoiding this risk, is indicated when this attachment with the deep lobe of the parotid is narrow [9].
The transmandibular approach is rarely used but is rather reserved for tumors necessitating wide exposure of the PPS, such as very large lesions, malignant transformations, revisions and masses that were exposed to radiotherapy [10]. The transoral approach is reserved for very small tumors, but it has some disadvantages, it can lead to major bleeding since control of the external carotid artery traversing the PPS is extremely difficult. also, it does not allow a

good exposure, so excision of the tumor may be incomplete. However, recently, the endoscope-assited transoral approach (Da Vinci system) provides less operative trauma and offers the advantage of wide exposure and superior precision that may overcome those risks [11]. With the intention of having a good exposure, to guarantee a complete extracapsular dissection of the mass and avoiding the risk of vascular injuries, our therapeutic strategy was based on combined approach associating left paramedian pharyngotomy and high cervical incision to control large vessels of the neck, this approach reduces comorbidity and allows good outcomes, has been advocated by most authors who have reported this rare entity of primary PA of PPS [3].

Conclusion:-

Primary PA of PPS remains very rare, this case highlights the importance of a detailed radioclinical study of parapharyngeal space tumors to rule out the vascular nature of the tumor, good anatomical knowledge of the region is for sure the onlyway for complete treatment without iatrogenesis.

Conflicts of interest: -

The authors declare that there is no conflict of interest in this work.

Figure 1: The soft palateis displaced anteromedially by the parapharyngeal mass, narrowing the oropharyngeal lumen.

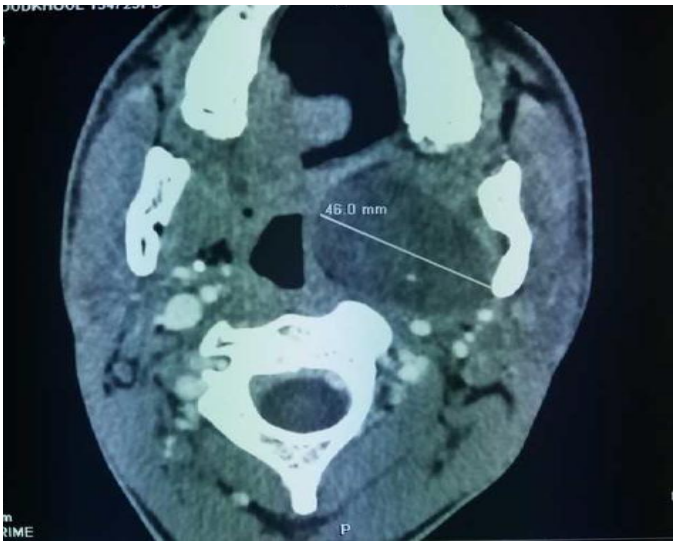

Figure 2:- axial CT-scan showing an oval, well-definedhomogeneous and hypodense lesionoccupying the left PPS, withoutbonelysis or invasion of adjacent structures.

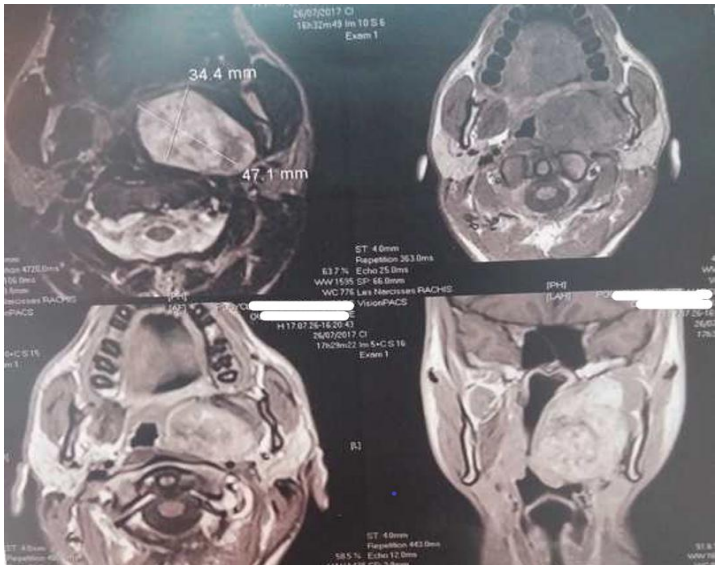

Figure 3: - Magnetic resonance imaging showed moderately and heterogeneously enhanced tumor of left PPS and the ralationship of the mass with great vessels of the neck.

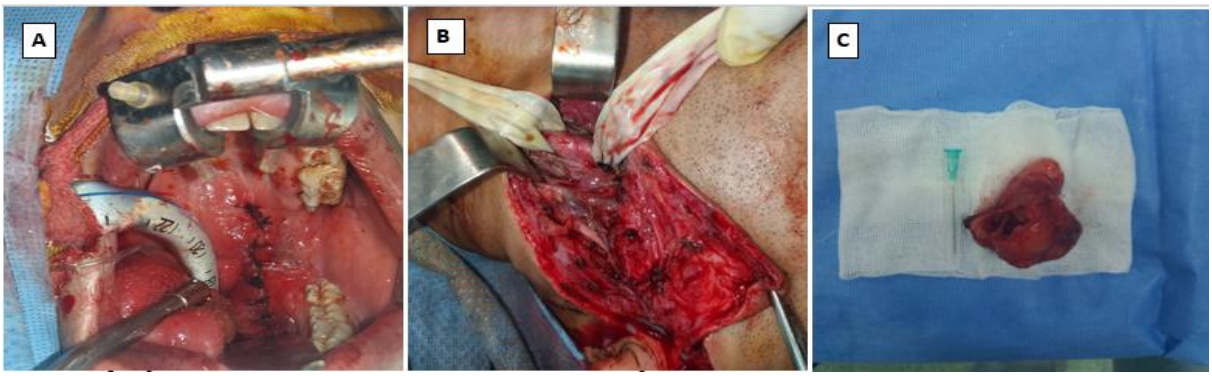

Figure 4: - peroperative images: transoral approach after tumor resection (A),transcervical approach and control of large vessels of the neck (B), the excised specimen (C).

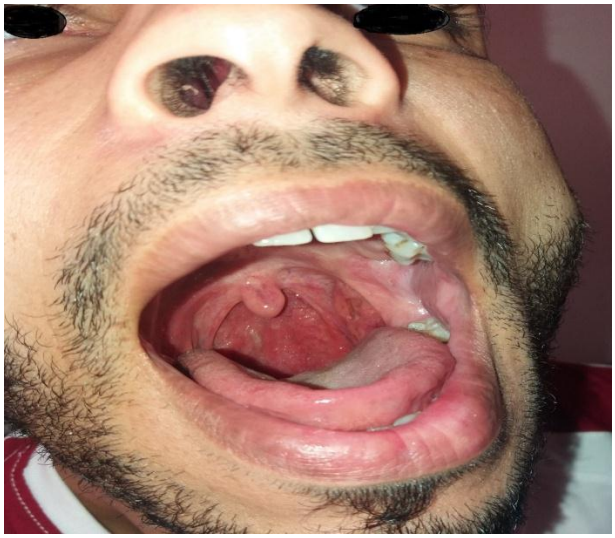

Figure 5: - Clinical post-operative outcomes.

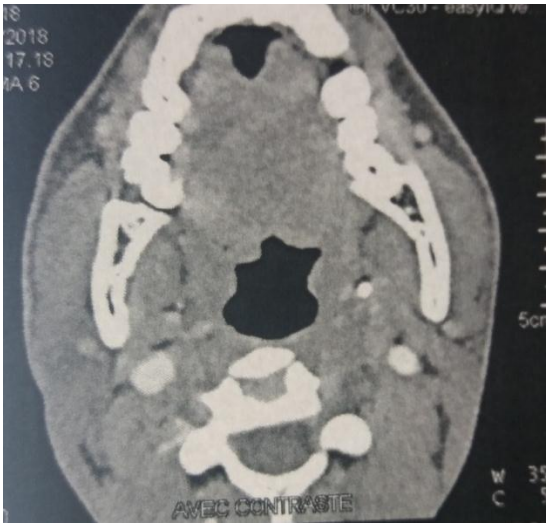

Figure 6: - Radiological post-operative outcomes.

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