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Giant follicular cysts extended in pterygo-maxillary fossa, antro-naso-ethmoidal and orbital space associated to exophthalmos and diplopia in young patients

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ABSTRACT

Follicular cysts develop from the enamel epithelium of an un-erupted tooth. Two cases of extremely large and extended follicular cysts related to the upper impacted third molars in young patients and treatments are described. **Case 1** Female, aged 16 with swelling of the right cheek and oral vestibule, right exophthalmos and diplopia. Spiral Computed Tomography (CT) showed a massive lesion occupying the maxillary sinus with extension into the pterygo-maxillary space, due to the destruction of the posterior antral bone wall. Above the lesion was compressing the orbital floor. Transantral surgical excision was performed approaching to the pterygo-maxillary space and orbital structure also using the operating microscope. 5-years follow-up shows good restoring of the involved structures without relapse. **Case 2** - Female, aged 22, with swelling of the left cheek and oral vestibule, left nasal obstruction, orbital pain and diplopia. Spiral CT-MR integrated study allowed a very accurate analysis: the lesion occupied the entire maxillary sinus, the pterygo-maxillary space, the left nasal cavity, ethmoidal structures with posterior orbital compression. Transantral surgical excision was performed approaching to the pterygo-maxillary space; nasal-ethmoidal and orbital structures were approached by endoscopic technique. Follow-up shows good clinical, anatomical and functional conditions. Histological examination confirmed in both cases the diagnosis of follicular cyst. Giant follicular cysts require an accurate preoperative study due to the delicate structures that may be involved. In the reported cases, the operating microscope and endoscopic surgical procedures were needed in the delicate surgical steps to perform the detachment in deep areas.

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Fig. 1. a. Clinical and radiological aspect of the patient at the admission. b. Spiral CT showing the involvement of the hard palate, the pterygo-maxillary region and the ethmoidal-orbital right structures, both on axial and coronal scan. c. MRI integrated study shows extension of the lesion (T2 sequences).

1. Introduction

Follicular cysts of the jaws are osteolytic lesions developing from the follicular dental epithelium. Compared to the other dentigerous cysts, they own a growth, differentiation and degeneration potential greater than radicular cysts. Due to their tendency to expand rapidly and to displace the teeth germs, when they are associated with impacted superior third molars, their extension can include the antral cavity and the surrounding anatomical structures. This event occurs as a result of compression of the cyst on the osseous thin maxillary boundary walls, causing the thinning of the cortical bone [1,2].

Thus cystic expansion into the pterygo-maxillary area, the nasal cavity, the ethmoidal and the orbital regions can cause morphological, functional, nervous and inflammatory complications. In particular, ocular issues as diplopia can arise; eyesight reduction, including blindness represent dangerous complications due to severe orbital infection [2]. We reported two cases of follicular cysts of the maxilla, unusual for dimensions involving meso- and sovrastructure describing the clinical diagnostic strategies and the surgical procedures adopted.

2. Cases presentation

2.1. Case 1

A caucasian female 16 years old patients was admitted to Maxillofacial Surgery Operative Unit of University of L'Aquila - Italy presenting a swelling of the right cheek and oral vestibule, referred to be appeared few weeks earlier, right exophthalmos and diplopia (Fig. 1a). Orthopantomogram (OPG) showed very deep impacted right upper third molar and an opacity of the maxillary sinus area. Spiral Computed Tomography (CT) showed a massive homogeneous lesion, involving the whole maxillary sinus with extension into the pterygo-maxillary space. The posterior antral bone wall appeared completely resorbed. Above the lesion was compressing the orbital floor (Fig. 1b). Due to the delicate nature of the involved structures,

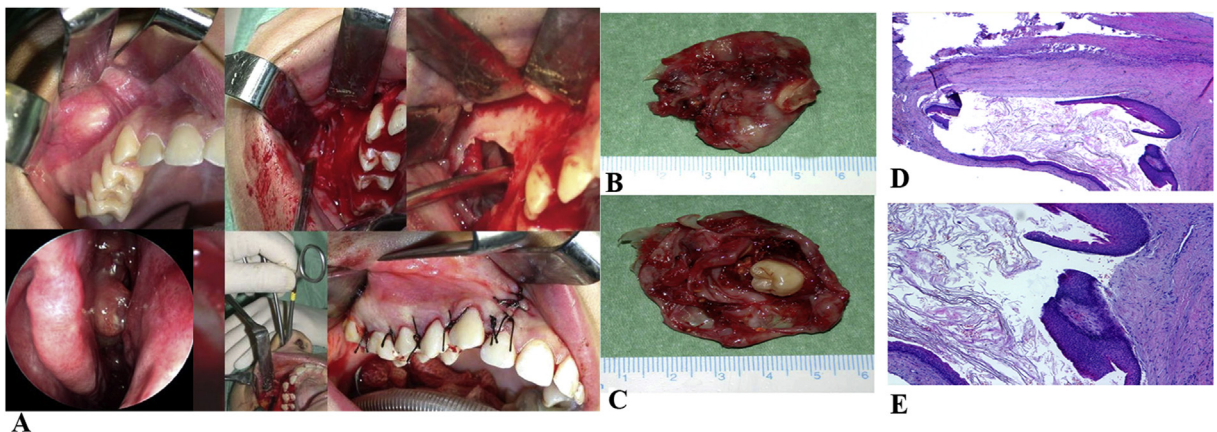


Fig. 2. a. Surgical steps of the cyst removal, also using of endoscope microscopy. b. Whole lesion, measuring 4 cm. c. opened lesion showing the bonding with the dental collar. d-e- histological pattern (5x and 10x magnifications Hematoxylin-Eosin stain). The epithelial layer appears stratified and squamous.

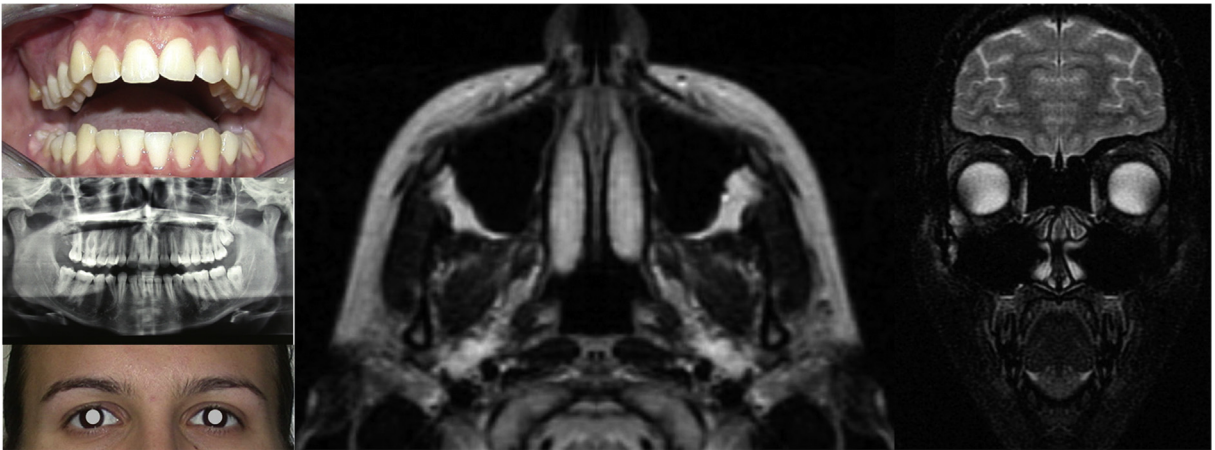


Fig. 3. The 5 years clinical and MRI follow-up, showing no signs of relapse.

the diagnostic study was completed by Magnetic Resonance imaging (MRI). The MRI showed the interface cyst-soft tissues of the pterygo-maxillary space and orbital structures, and the characteristics of the wall. These diagnostic information were fundamental for the surgical planning (Fig. 1c). Trans-antral surgical excision of the lesion was performed with approach to the pterygo-maxillary space and orbital structure, using the operating microscope (Fig. 2a). The cyst measured over 4 cm in diameter; the cyst opened measured were 7 cm and the lesion appeared inserted on the collar of 1.8 (Fig. 2a–b). Histological diagnosis was follicular cysts, characterized by the absence of inflammatory cells (Fig. 2c–d). Five years follow-up (Fig. 3)

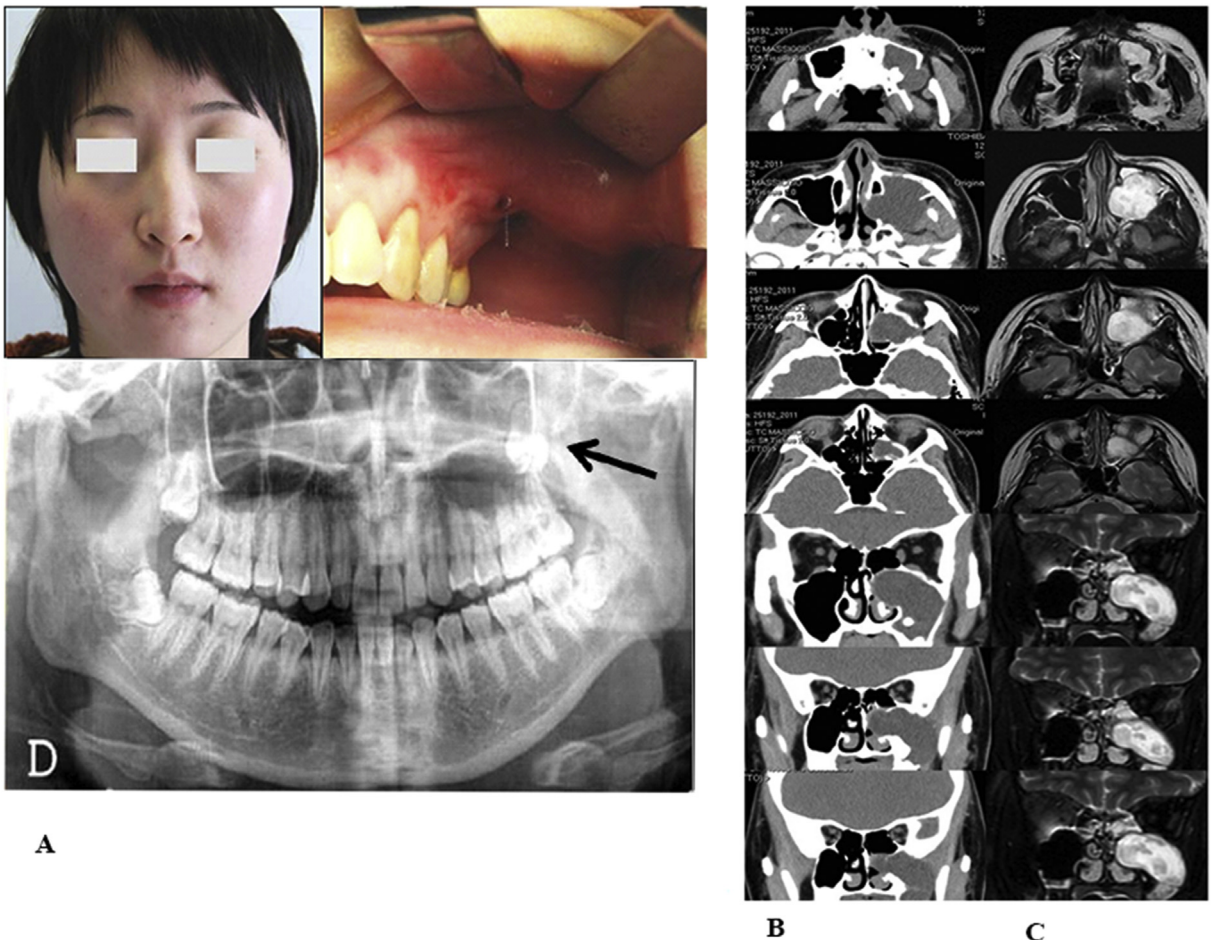


Fig. 4. a. Clinical aspect of the patient at the admission and orthopantomogram x-ray: the black arrow shows the impacted superior third molar. b. integrated imaging study showing by CT (b) and MR (c) the very large extension of the follicular cyst with the involvement of the hard palate, the pterygo-maxillary region and the ethmoidal-orbital right structures, both on axial and coronal scan.

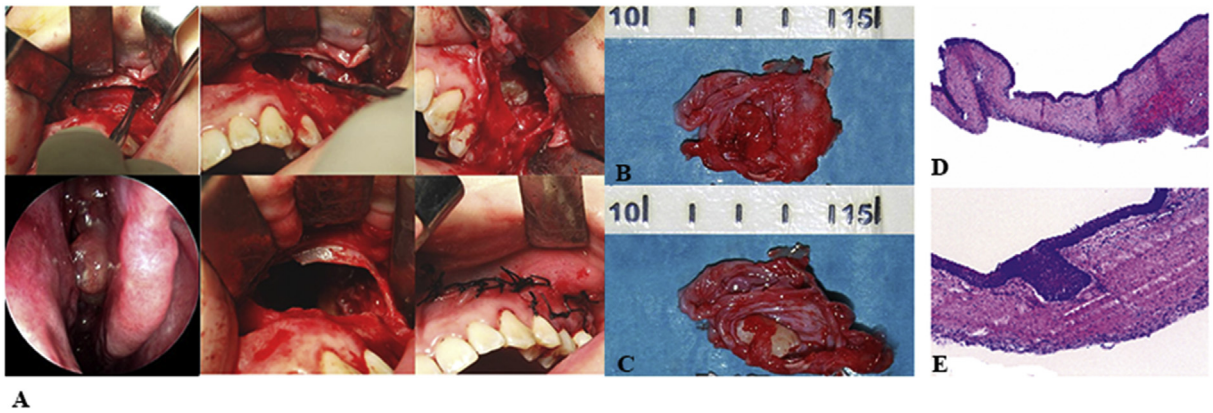


Fig. 5. a. Surgical steps of the cyst removal, also using of endoscope microscopy. b. Whole lesion, measuring 4 cm. c. opened lesion showing the bonding with the dental collar. d-e. histological pattern (5x and 10x magnifications Hematoxylin-Eosin stain). The epithelial layer appears stratified and squamous.

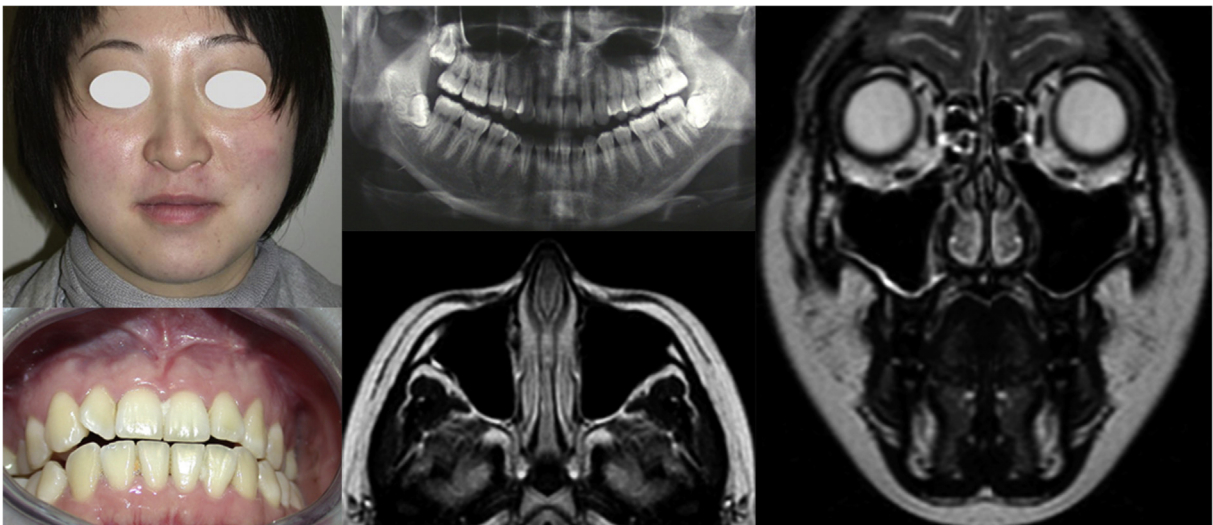


Fig. 6. The 5 years clinical and MRI follow-up, showing no signs of relapse.

clinical, OPG and MRI examination shows a good recovering of antro-pterygo-maxillary and orbital structures without any recurrence of the lesions and integrity of the anatomical structures.

2.2. Case 2

A Caucasian female 20 years old showed at the admission a swelling of the left cheek and oral vestibule, left nasal obstruction, orbital pain and diplopia (Fig. 4a). OPG showed very deep impacted left upper third molar and morphological alteration of the antral area (Fig. 4a). The integrated study Spiral CT-MRI allowed a very accurate preoperative study: the imaging analysis showed a lesion occupying the entire maxillary sinus, the pterygo-maxillary space, the adjacent left nasal cavity, ethmoidal structures with posterior orbital compression (Fig. 4b–c). Trans-antral surgical excision was performed with approach to the pterygo-maxillary space; nasal-ethmoidal and orbital structures were approached by endoscopic technique (Fig. 5a). The cyst body measured about 4 cm in diameter; the cyst opened was measuring 8 cm and appeared inserted on the collar of 2.8 (Fig. 5b–c). Histological examination showed a pattern of follicular dentigerous cyst (Fig. 5d–e). Long-term follow-up showed a good clinical, anatomical and functional conditions without any recurrence of the lesion (Fig. 6).

3. Discussion

Giant follicular cysts in the maxilla, particularly in young subjects, need accurate preoperative study, in relation to the tricky nature of the inner bio-morphology of the structures usually involved [3]. In the reported cases, the orbital structures represented a border of the lesions, with possibility of secondary involvement by means of the surgical intervention.

The follicular tissue around impacted third molars has a potential to develop pathologies. It is considered normal a radiolucency of follicular space <3 mm. The incidence of disease has been reported higher in the age group of 20–30 years, in men compared to women and in the mandible compared to the maxilla [4].

Pathological proliferation and cystic degeneration of the follicular tissue of the superior deep impacted maxillary third molars can cause particular clinical conditions especially when the lesion reaches very large dimensions. In these cases, usually vestibular and facial swelling appear after a latency time. In our cases, the nasal and mostly visual disturbances as exophthalmos and diplopia were uncommonly present.

These clinical signs led to assume a dentigerous pathology considering the young age of the patient and the non-eruption of the third upper molar. Authors point out that in both cases OPG shows the lesion as a radiopaque area (follicular space well above 3 mm) but a not accurate evaluation of position of the impacted third molar that seemed normal.

CT and MRI integrated study showed instead that tooth were dislocated by the lesions on the medial wall of the maxillary sinus. In particular the MRI analysis, showing in a more detailed way the soft tissues is a very useful tool for the diagnosis of jaws neof ormations and surgical planning [5–7]. As reported by Lenz et al. [8] the MRI offer an adjunctive aid in the diagnosis, due to its tissue contrast that allow to distinguish the lesions border from surrounding structures. In our cases, this morpho-clinical aspect was crucial to understand the disruptive behavior of the cysts. Indeed, the massive growth of the follicular lesions caused the progressive dislocation of the tooth towards the medial wall of the maxillary sinus in both cases.

Advanced imaging study, as we stated earlier [9–16], play a key role in providing an accurate overview of the clinical conditions of maxilla and paranasal sinuses: due to their inner nature, they constitutes a complex system of soft tissues, bone and airways, and the detection of the features of a mass in these area is fundamental for an effective and safe treatment planning.

In the reported cases, the pre-operative imaging analysis lead to the choice of the operative microscope in case 1 and the use of endoscopic technique in case 2.

In case 1 the choice of using the operative microscope was due to the position of the cyst wall near the orbital structures. Therefore the higher magnification and a complete visual field provided by the operative microscope [17,18] were required.

In case 2, the both orbital and ethmoidal structures were interested by the lesion. Therefore, as also widely reported in literature [19–21] the endoscopy technique resulted more indicated to approach the cyst.

4. Conclusions

The only effective treatment of follicular cyst is the surgical removal. Inflammatory complications make excision even more laborious due to pathological changes in the normal anatomy and the modification of the surgical planes. Consequently, the use of operating microscope and endoscopic procedures as support of the traditional surgery are necessary in determined surgical steps to perform the operation safely. Indeed, the lesion must be completely detached to avoid recurrence, in deep areas, not easily accessible [4].

The cases reported show how an early diagnosis is unlike due to the misleading position of the impacted tooth as appears on the OPG and to the potential space where the lesion can expand.

Conflict of interest statement

The authors have no conflict of interest to disclose.

Consent

Patients gave their consent to the publication of these data.

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