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Coexistence of solitary bone cyst of the jaw and submandibular sialolithiasis mimicking a supernumerary tooth: Case report

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Abstract :

The jaws may present different benign or malignant lesions of various origins. Diagnosis is generally determined based on their specific clinical and radiological characteristics and confirmed histologically. However, the coexistence of two lesions in the same region sometimes requires diagnostic caution and specific additional investigations in order to refine the diagnosis and determine the appropriate treatment. That's motivate this work which consisted to report a case of the association of submandibular sialolithiasis and solitary bone cyst in the same side of the jaw.

This case highlights the value of detailed clinical examination and the role of 3D X-Ray in the simultaneous diagnosis of salivary and osseous pathologies and emphasizes a rare association, underlining the necessity of adequate specific management of each lesion.

Key-words : sialolithiasis, solitary bone cyst, diagnostic, therapeutic, radiographies

Introduction:

Maxillary pathologies are a complex area of medical practice, often characterized by a diversity of clinical presentations and a multitude of differential diagnoses. These pathologies can affect soft, hard and glandular tissues. This diversity of lesions calls for a cautious diagnostic approach to confirm the type of pathology. This diagnostic precision is even more complicated when pathologies are combined. In fact, the coexistence of lesions can underline the inadequacy of 2D imaging in diagnostic accuracy because it can obscure crucial details due to overlapping anatomical structures. This limitation can lead to misdiagnoses and inaccurate treatment planning. Unlike 2D images, 3D imaging, such as cone beam computed tomography (CBCT), offers a detailed three-dimensional view of the anatomy, allowing for precise visualization of complex structures and their spatial relationships. This enhanced clarity improves diagnostic accuracy and supports more effective surgical planning, making 3D imaging essential for successful oral surgery.

Hence the interest of this work, which aimed to highlight the diagnostic challenges and the importance of clinical examination and the right choice of complementary explorations particularly the 3 D X-ray when faced with the association of different types of lesions in the maxilla, especially in the same region. These objectives will be optimized through the discussion of a clinical case report managed in the oral medicine and surgery department of the academic dental Clinic of Monastir, which was characterized by the coexistence of a solitary bone cyst and a submandibular lithiasis simulating at first sight a dentigerous cyst.

Case presentation:

An 18-year-old patient was referred to the oral medicine and oral surgery department for the management of a mandibular right dentigerous cyst of accidental discovery on a panoramic radiograph. His familial and past medical history were noncontributory.

The extraoral examination was non-significant (figure 1)

the intraoral examination revealed a palpable hard mass in the oral floor, which was covered by a normal mucosa, mobile in relation to the deep and superficial planes (figure 2)

Vitality test on 45, 46 and 47 was positive.

Panoramic radiograph showed a radiopaque image resembling the shape of a tooth in a horizontal position with a radiolucent image encompassing its anterior part at the level of the

right mandibular premolar-molar sector, suggestive of a dentigerous cyst related to a supernumerary tooth as it was concluded by his dentist. (Figure 3)

Regarding the palpable mass in the oral floor and the radiopaque lesion, further data need to be investigated such as clinical symptoms. Actually, patient reported the sensation of accidental pain during meals. This leads to suspected a sialolithiasis.

To confirm this proposal diagnosis, an occlusal bite was performed and it revealed that the radiopaque image was extraosseous at the level of the oral floor. (Figure 4)

In order to better explore these images especially the radiolucent image presented at the standardized x-ray, a cone beam was required and it showed a hyperdense image of the oral floor. Hypodense image in the horizontal branch of the mandible opposite the mandibular molars. (Figure 5)

According to clinical examination and complementary investigations, the notion of pain accentuated during meals, palpable mass in the floor of the mouth and the extraosseous radio-opaque image in the submandibular area , the diagnosis of submandibular lithiasis was proved.

For the radiolucent image in the horizontal branch of the mandible, the diagnosis of a non-inflammatory cyst or a maxillary begin tumor was suspected.

Surgical transmucosal approach was conducted in order to eliminate the salivary stone (figure 6) and a favorable mucosal healing was observed after 15 days (fig 7), followed two months later by the exploration of the osteolytic lesion(fig 8, fig 9).

Histological examination of the excised bone confirmed the diagnosis of solitary bone cyst for the intraosseous lesion.

Later the patient underwent surgical removal of mandibular wisdom teeth

Follow up of both lesions was uneventful for 18 months.(fig 10, 11)

Discussion:

Simple bone cyst (SBC) is an intraosseous cyst devoid of an epithelial lining, either empty or filled with serous or blood fluid, with a prevalence less than 1% (1). it can be referred to as traumatic bone cyst, hemorrhagic bone cyst, hemorrhagic cyst, unicameral bone cyst, idiopathic bone cyst. The WHO classification released in 2022 defines the SBC as an

intraosseous pseudocyst devoid of an epithelial lining, either or filled with serous or sanguinous fluid.(2) The sex and site distribution of SBC is quite controversial, in which female predominance and posterior region predilection of have been documented(3)

clinically, they are usually asymptomatic discovered accidentally in radiographs where they appear as a radiolucent image with irregular or scalloped but well-defined margins. Harnet et al. (6) described three theories on the formation of SBC:

1) local abnormality during bone growth due to an abnormality in cell differentiation during the jaw ossification and growth with local environmental factors that induce mechanical restrictions during osteogenesis and angiogenesis;

2) SBC as part of a tumor process or a central giant cell granuloma after surgery ;

3) the lesion is caused by a low intensity trauma, based on the appearance of intramedullary hemorrhage followed by hematoma after insufficient trauma to fracture healthy bone.

Histologically, the solitary bone cyst is remarkable by the fact that no cystic lining is identified. Generally, the submitted tissue consists of scant fragments of fibrovascular connective tissue, extravasated red blood cells and pieces of reactive vital bone.

Surgical intervention consisting on the exploration of the empty intraosseous lesion is the treatment option for this cyst, as it was the case of the reported patient.(9)

Sialolithiasis is the most common disorder of the salivary glands and it can go from small particles to stones that are several centimeters in length(7). Sialoliths are mainly composed of inorganic material, respectively carbonate apatite was identified in 99% of the stones, phosphate in 88%, calcium in 87%, magnesium in 68%, struvite in 44%, oxalate in 38% and carbonate in 35% (10). Sialolithiasis predominantly involves the submandibular gland (80% to 95%) followed by parotid (5% to 20%), sublingual gland and rarely the minor salivary glands in 1% to 2%(11). Although most salivary stones are asymptomatic or cause minimal discomfort, larger stones may interfere with the flow of saliva and may cause pain and swelling. As a rule, the onset of swelling is sudden and associated with salivation during a meal. If left untreated, salivary stones can result in chronic sialadenitis and glandular atrophy. Conservative treatment may consist of oral analgesics and antibiotics. Surgical management may include transmucosal incision, lithotripsy, basket retrieval, and sialendoscopy (7) In the reported case, surgical elimination of the lithiasis was performed since the mass was palpable.

The coexistence of these two lesions was an excellent opportunity to highlights the

the importance of the three D imaging. In oral surgery, reliance on 2D imaging techniques, such as conventional X-rays, can often lead to significant diagnostic errors due to the superposition of anatomical structures. These 2D images compress complex three-dimensional anatomy into a flat plane, resulting in overlapping tissues that can obscure critical details and lead to misinterpretations. For example, adjacent anatomical structures like teeth, roots, and bone can appear merged, making it challenging to accurately assess their spatial relationships and pathological conditions. Consequently, this limitation underscores the need for 3D imaging modalities, such as cone beam computed tomography (CBCT), CT scan…, which provide detailed, volumetric data. 3D imaging enables precise visualization of anatomical structures in their true spatial context, significantly enhancing diagnostic accuracy and planning for surgical interventions.

Actually, this reported case highlighted the importance of detailed clinical examination which should be guided by patient's history and complementary examination and showed the significant impact of 3D imaging in the management of associated lesions and prove the ability of CT and CBCT to provide cross-sectional images and three-dimensional reconstructions that enables detailed visualization of regional anatomy and lesions. In this case, the use of CBCT enabled us to confirm the diagnosis and effectively plan treatment, thus avoiding potential errors and subsequent complications.

Figures:

Figure 1 : Extra oral view: facial symmetry was preserved, no hard mass or swelling was observed

Figure 2 :intra oral view: papable hard mass in the oral floor covered by normal muccosa

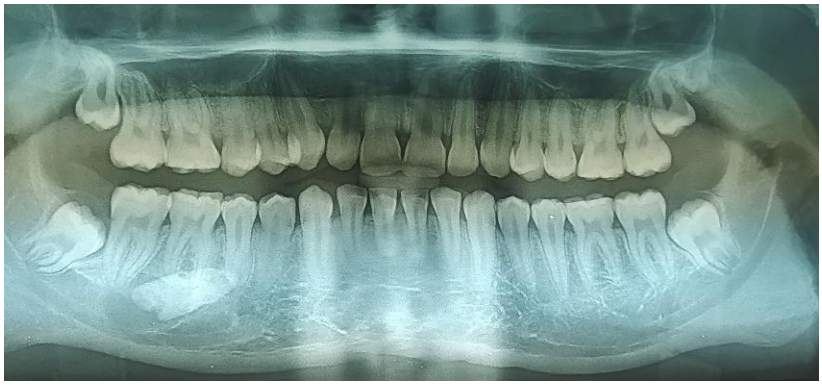

Figure 3 : panoramic radiograph : radiopaque image resembling the shape of a tooth in a horizontal position with a radiolucent image in the right mandibular area.

Figure 4 : occlusal bite : extraosseous radiopaque image

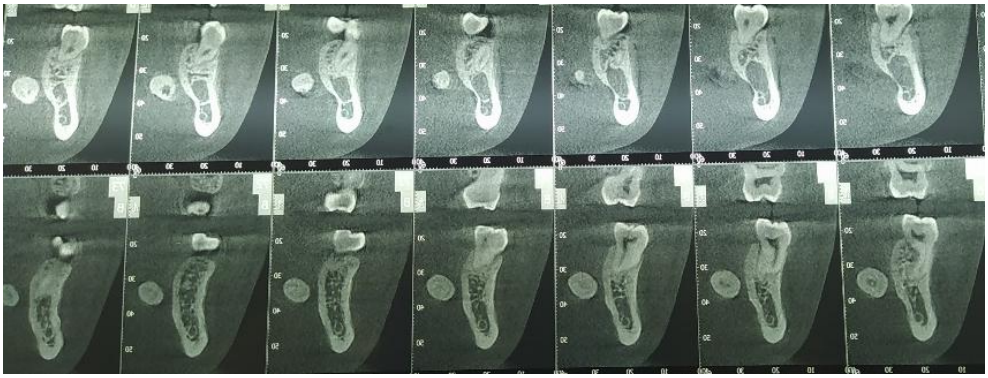

Figure 5 : CBCT : hyperdense image in the oral floor and a well circonscribed hypodense image in the mandibular right bone

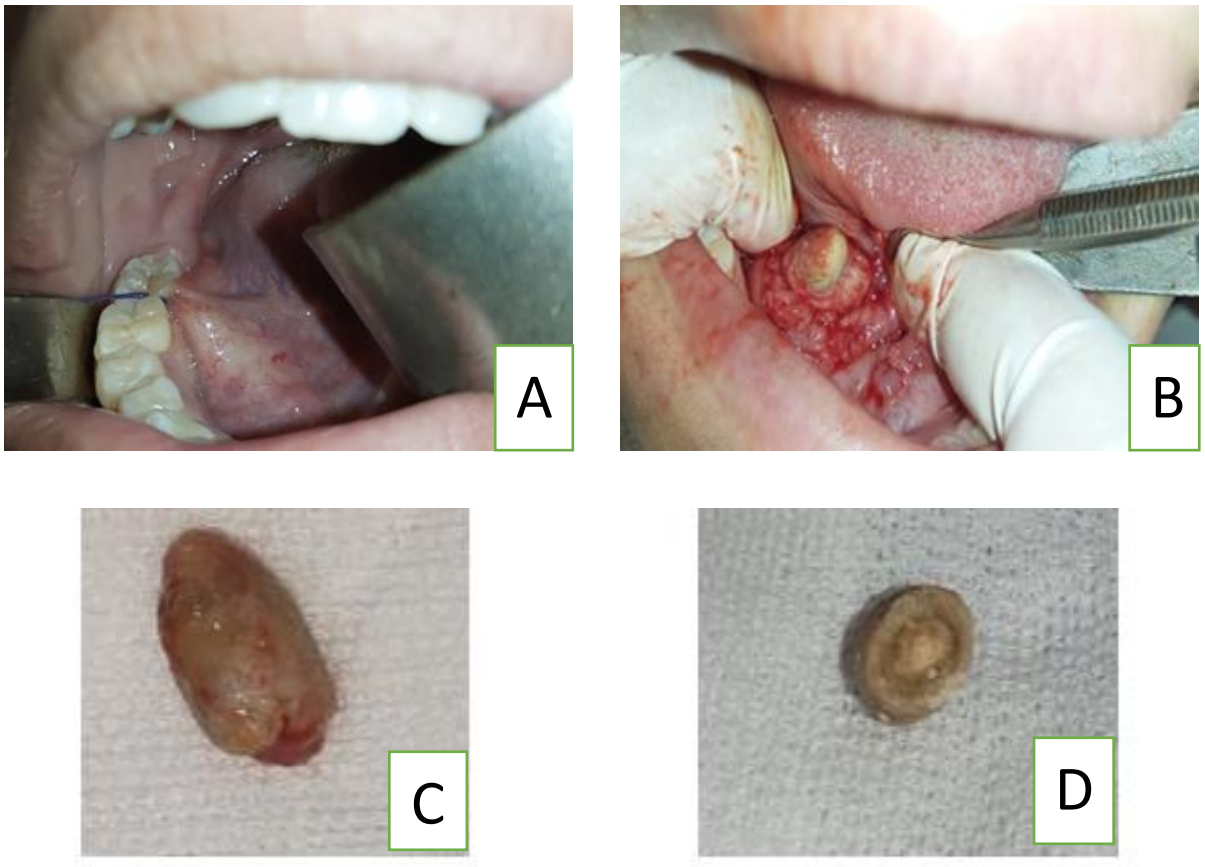

Figure 6 : A : spotting of sialolithiasis , B : transmucosal incision of the floor of the mouth., C: the removal specimen , D : The aspect of lithiasis separated into two parts

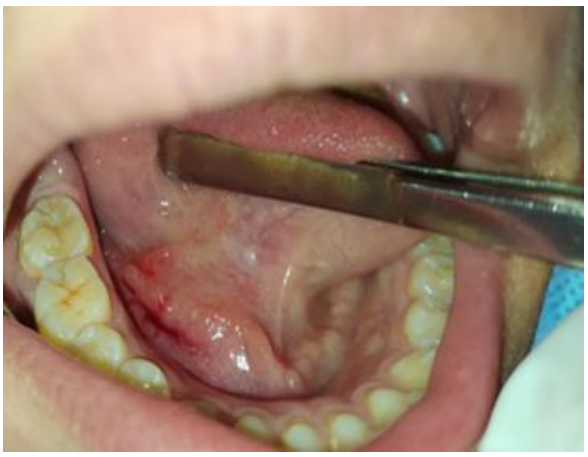

Figure 7 : Follow-up after 15 days : favorable healing

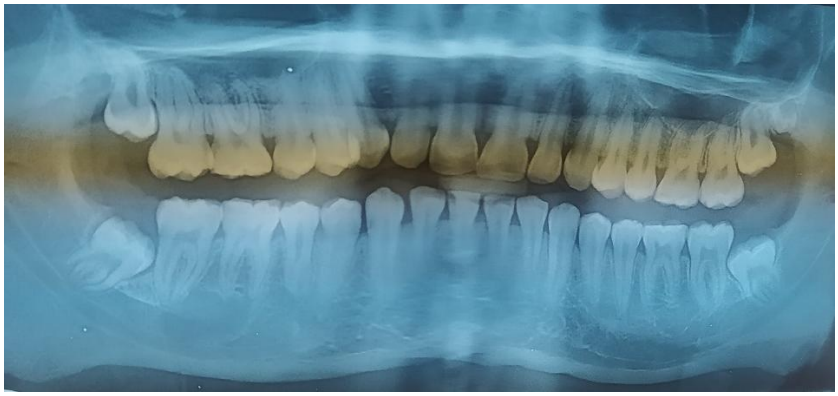

Figure 8 : new orthopantomography after two months: radiolucent image that extends from the apex of the 46th tooth to the 47th tooth

Figure 9 A,B,C : surgical exploration of the osteolytic lesion

Figure 10 :panoramic radiograph 6 months following surgical procedure : complete bone healing

Figure 11: aspect of the mucosa 18 month after the surgery

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