Radiological Evaluation Key to Diagnosis of Idiopathic Solitary Cyst

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Idiopathic solitary cysts have a predilection for long bones and the mandible. Although the origin of these cysts is unclear, the prognosis seems to be good, given proper diagnosis and surgical treatment.

A 14-year-old female patient with a bone lesion in the right mandibular ramus was referred to the Department of Pediatric Dentistry at the Medical University of Gdansk, Poland. Due to severe headache lasting two days, the patient had previously undergone magnetic resonance imaging (MRI) examination of the head. A unilocular bone cyst in the mandible was discovered incidentally. Comparison with a previous orthopantomogram (OPG) taken two years earlier for routine dental treatment revealed a clearly visible but smaller bone lesion at the same location. Surgery was carried out in order to achieve bone regeneration. This report documents the importance of careful evaluation and assessment of all radiographs taken, as the lesion could have been diagnosed and treated much earlier, thus lowering the risk of complications.

Keywords*: solitary bone cyst, bone lesion, mandible*

INTRODUCTION

Bone lesions are diagnosed very rarely in children. Most of dentists are not familiar with lesions such as cysts and tumors in young patients, mainly because they do not expect them to occur. This explains why anomalies are usually overlooked at the early stages of their development, when treatment would be relatively easy and uncomplicated.[1] Such anomalies develop in "peace" until they become too large to be ignored, or worse, give clinical symptoms or lead to secondary complications such as spontaneous fracture, pain, destruction of anatomical structures, or face asymmetry.[2] Radiographic examination is one of the most valuable diagnostic tools for dentists, since it can reveal many bone deformations without significant risk factors. The most popular and routinely taken are intra-oral radiographs and orthopantomograms (OPG), especially during orthodontic treatment. Every radiograph is performed for a particular medical reason, either to confirm or exclude a given pathology. The clinician evaluating that radiograph thus tends to concentrate exclusively on the very reason for which that particular radiograph was ordered. As a result, some practitioners make the mistake of omission evaluating bone structures, focusing strictly and exclusively on the teeth, and overlooking lesions that might be in their initial stages, not noticing them until they become much larger.[3]

The OPG allows a holistic assessment of the mineralized structures of the jaws. In children, OPGs are performed most frequently before and during orthodontic treatment. If a child does not require any orthodontic procedures, they are unlikely to have an OPG. In such cases, early diagnosis of bone cavities is highly unlikely.

Accurate diagnosis of the radiological image should include evaluation of bone tissues, joints, sinus space, and teeth structures. Indeed, focusing directly and mainly on teeth is the main reason why other lesions (not related to teeth but remaining in the area of interest for dental surgeons) are overlooked. As mentioned above, bone lesions are rare during childhood. They can be classified as odontogenic and non-odontogenic, with odontogenic lesions being predominant (25:3 ratio of cases).[1]

Non-odontogenic changes in bone structure include central giant cell granuloma, osteoblastoma, and fibrous dysplasia, all of which need distinguishing from each other.[1] Solitary bone cysts (SBC) are rarer than the aforementioned lesions, and are usually located within the mandible body in the premolar/molar area and surrounded by vital teeth.[2] However, the most common SBC location is the long bones (90%), with a predominance in the metaphyseal region, and 65% in humeral and 25% in femoral bones. Only 10% of all cases involve the jaws, with the mandibular body being affected in

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75%.[4] Matsuma *et al* reported three cysts in the maxilla and 50 in the mandible in a series of 51 patients[5] and Saito et al. reported two lesions in the maxilla and 17 in the mandible in a series of 15 patients.[6] The predilection of solitary bone cysts for the mandible therefore appears to be obvious.

Solitary bone cysts are referred to in the literature as simple bone cysts, traumatic bone cysts, hemorrhagic cysts, aneurysmal cysts, idiopathic bone cavities, unilocular bone cysts, and progressive bone cysts. All of them are synonyms. Although all of these names are correct, the World Health Organization and the international histological classification of tumors specifically recommends the term 'solitary bone cyst' (SBC).[7] The multitude of names for just one medical condition can lead to unnecessary chaos and disorder.

Solitary bone cysts do not seem to be real tumors, but rather to be reactive or dysplastic lesions. Their etiology remains unclear, although a few theories concerning empty bone cavities have been proposed. All are hypothetically possible, albeit none of them has been 100% proven.[8]

The most frequently described is the trauma theory, postulating the creation of a hematoma after an injury and post-traumatic medullar hemorrhage. Maintenance of proper bone density depends on continual bone apposition as well as resorption. Many cells and mediator substances participate in this process, known commonly as bone remodeling.[9] A good blood supply is a cardinal condition for appropriate nutrition of any tissue.[10] After an injury an abnormality may start at the hematoma organization stage, and as a result there may be insufficient or improper bone remodeling. The osteoblasts and bone marrow would remain destroyed, leading to the creation of an empty cavity. Although it is possible, the trauma theory is often challenged by the fact that many cases have no history or recall of any trauma (about 50%).[11]

The second theory involves the incorporation of the synovis layer intraosseously, which disables the bone metabolism, remodeling process and growth.[12] A third theory involves the blockage of lymphatic drainage, increased fluid pressure and bone atrophy.[13]

Although practitioners seem to agree that SBCs are not real tumors, it has also been postulated that such lesions arise following the degeneration of primary tumors, such as fibrous dysplasia and giant cell granuloma. This theory is based on clinical observations.[14]

Further studies of SBC etiology and pathological mechanisms are necessary, because without known marker factors, prophylaxis is impossible.

On OPG a solitary cyst is most frequently seen as a radiolucency with an irregular, but fairly well demarcated, outline. Often the lesion extends upward between the roots, producing the characteristic scalloped outline without root resorption and tilting of the teeth. Such a picture excludes odontogenic origin. More accurate evaluation requires cone beam computed tomography (CBCT), which shows much more than the standard two-dimensional OPG mentioned above.[15] However, because of the large radiation dose, CBCT should be performed only as a complement to OPG diagnosis. At present, despite the available options for visualizing various pathologies in the bone area, a definitive diagnosis is only possible after opening the bone cavity and taking material for further histopathological tests. As far as treatment is concerned, the method of choice remains surgical bone trepanation and cavity exposure, followed by gentle curettage and accurate closure with a mucoperiosteal flap.[16]

Case report

A 14-year-old female patient with a bone lesion in the right mandibular ramus was referred to the Department of Pediatric Dentistry at the Medical University of Gdansk, Poland in August 2017. One month earlier i.e. in July 2017, following severe headache lasting two days, the patient had undergone magnetic resonance imaging (MRI) examination of the head. A unilocular bone cyst in the mandible on the right side was discovered incidentally on the MRI scan (Fig. 1). The patient was generally healthy with no significant medical history. Routine hematological and biochemical tests were all within the normal lab reference limits. However, the girl was physically very active, and had been participating professionally in a traumatic sport (karate) for eight years. She did not recall any face injury in particular, but it was assumed to be highly probable, and thus definitely could not be excluded.

Comparison with a previous OPG taken routinely for dental treatment in October 2015 revealed a clearly visible but smaller bone lesion, at the same location (Fig. 2). However, the pathological lesion was not visible on an OPG dated October 2014.

The extra-oral and intra-oral examination did not reveal any anomalies such as face asymmetry, skin lesions, swellings, or lymphadenopathy. On palpation the lymph nodes were bilaterally non-tender. The alveolar process and mandible ramus displayed no swelling or pain during palpation. No expansion of cortical bone either buccally or lingually was found. The overlying mucosa in the region of interest also appeared to be normal.

The OPG showed a radiolucent lesion, situated in the ramus, near the distal bone edge and above the angle on right side of the mandible. To evaluate the lesion's proportions, CBCT was performed in August 2017. This gave a three-dimensional view of the lesion (Fig. 3). On the CBCT scan the bone cavity measured approximately 35 mm (height) x 20 mm (width) x 10 mm (depth). The cortical bone was thinner than normal in a few places but no perforation was found (Fig. 4, Fig. 5). The interface between the lesion and the cancellous bone was rough. This examination aroused suspicion of a tumor lesion, so further diagnosis appeared essential. The lesion itself appeared homogeneous and asymmetric.

Because the diagnosis in such cases is always uncertain, it was decided to perform a biopsy of the lesion. The lesion was assessed intraorally by approaching it via the anterior border of the ramus and preparing a mucoperiosteal flap while the patient was under general anesthesia. There was no evidence of cortical expansion. The cortical bone of the mandible ramus was exposed gently on limited surface. The bone was prepared with a piezoelectric saw (W&H Dentalwerk Bürmoos GmbH, Austria) and the window attached to the periosteum was elevated laterally. It revealed an empty cavity with a small amount of blood and no pathological mass (Fig. 6). Material for histopathological examination was quite scant (fluid content and cavity layer scraped using curette). Fortunately a brief intraoperative histopathological examination confirmed no neoplastic cells. Gentle curettage was subsequently performed to provoke bleeding. After that the bone defect was filled with autologous blood, the coagulation process began. The cavity and the bony window were closed with the mucoperiosteal flap. Six months after surgery an OPG examination was carried out (Fig. 7). Good bone remodeling was observed at the site at which the bone lesion had been diagnosed. At that point we concluded that the healing process had successfully cured the bone defect, but the patient remained under our care.

Figure 1: MRI scan with a well-defined mass lesion in the right mandibular ramus (July 2017).

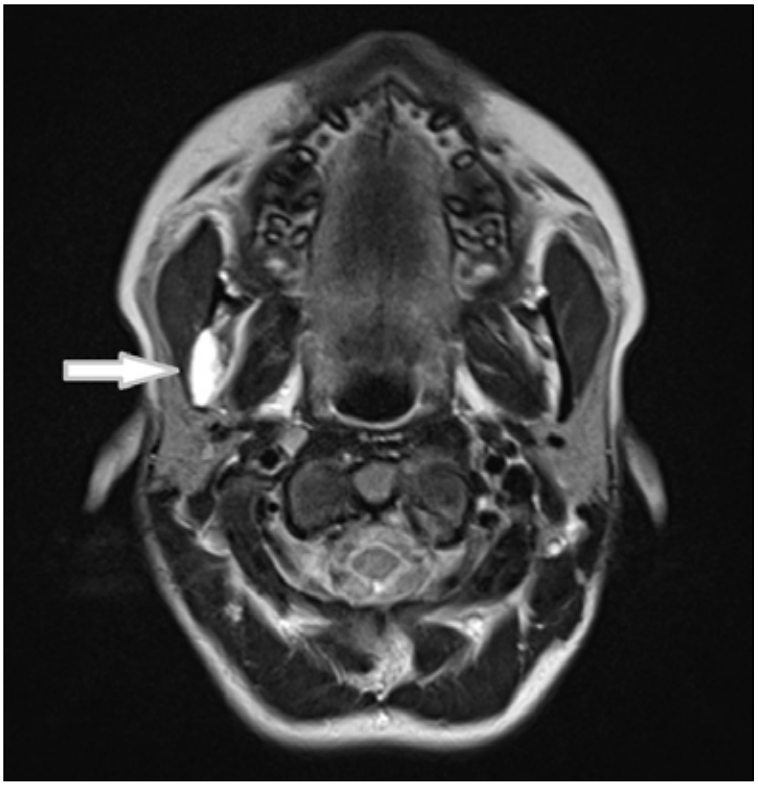


Figure 2: The same lesion displayed on an OPG from October 2015. The same location, but far smaller proportions. The ramus edge is still clearly visible.

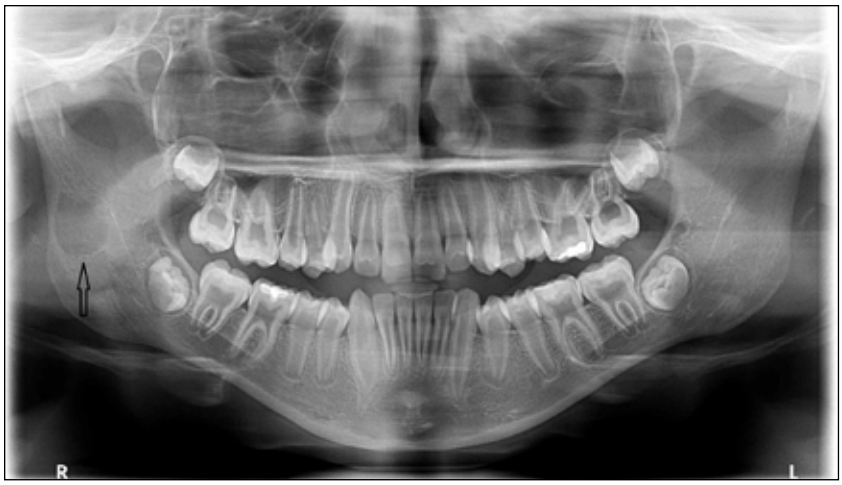


Figure 3: 3D dental reconstruction from CBCT data, August 2017.

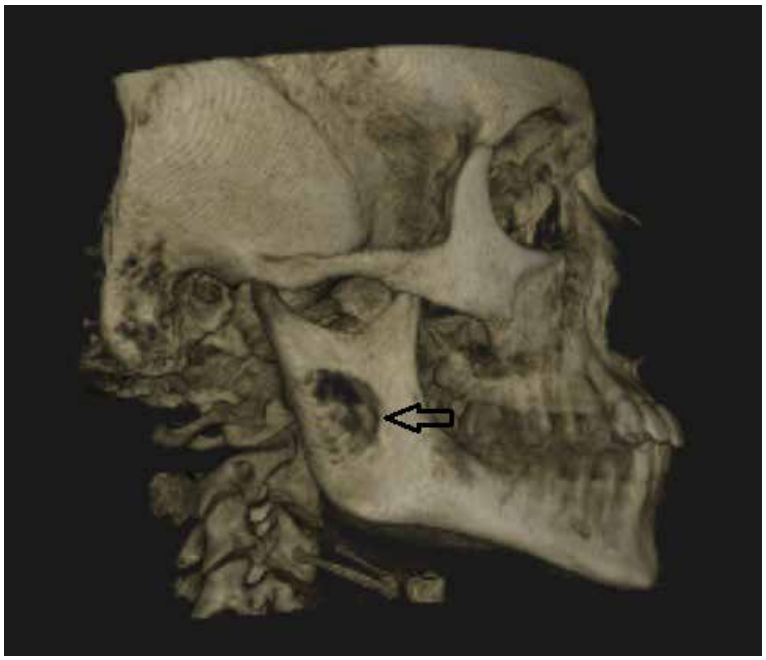


Figure 4: Panoramic view from CBCT scan (layer thickness 55.1 mm), August 2017. The arrow shows a lesion of significant dimensions, almost destroying the distal edge of mandibular ramus.

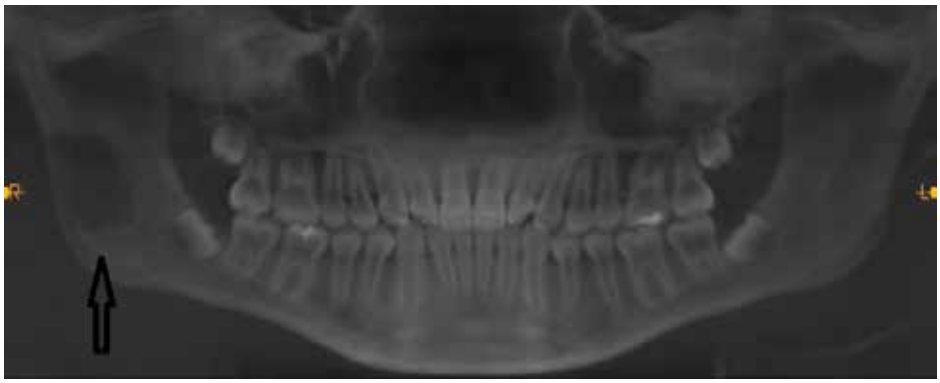


Figure 5: A cross-section of the right mandible ramus (layer thickness 300 µm) from CBCT scan, August 2017 demonstrating the thinned cortical outlines and the extent of the lesion.

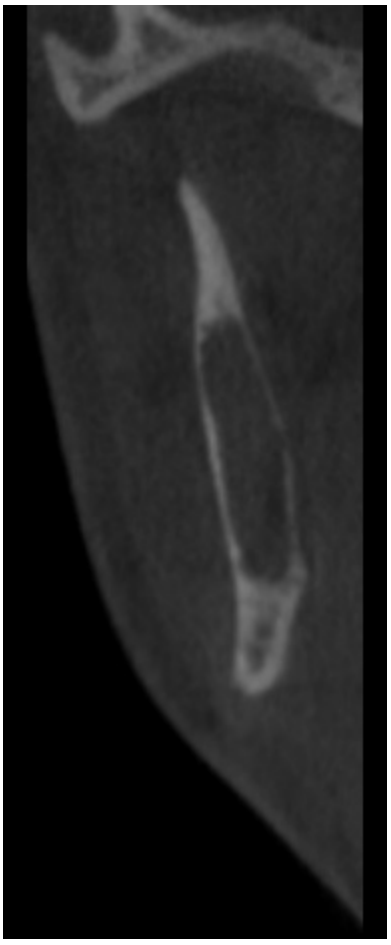


Figure 6: Intra-operative photograph demonstrating the intra-oral access. After removing the cortical bone layer, the almost- empty cavity was revealed, with a scant amount of blood. All the features pointed to a solitary bone cyst * – bone defect, R – ascending ramus of mandible, M – masseter

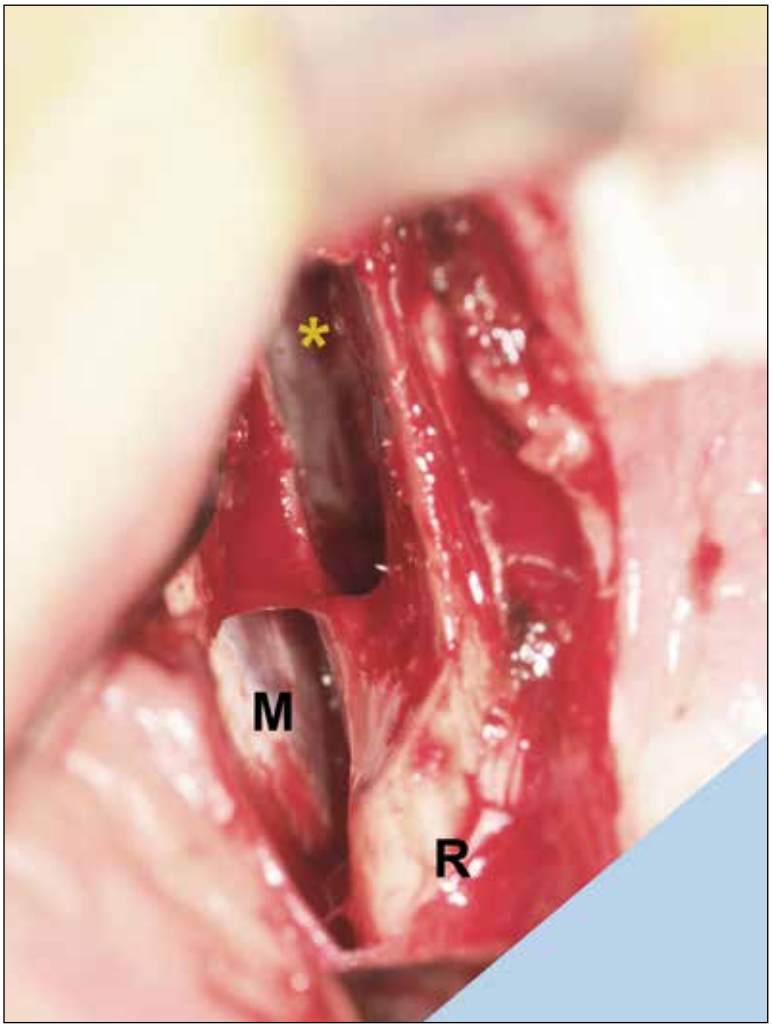
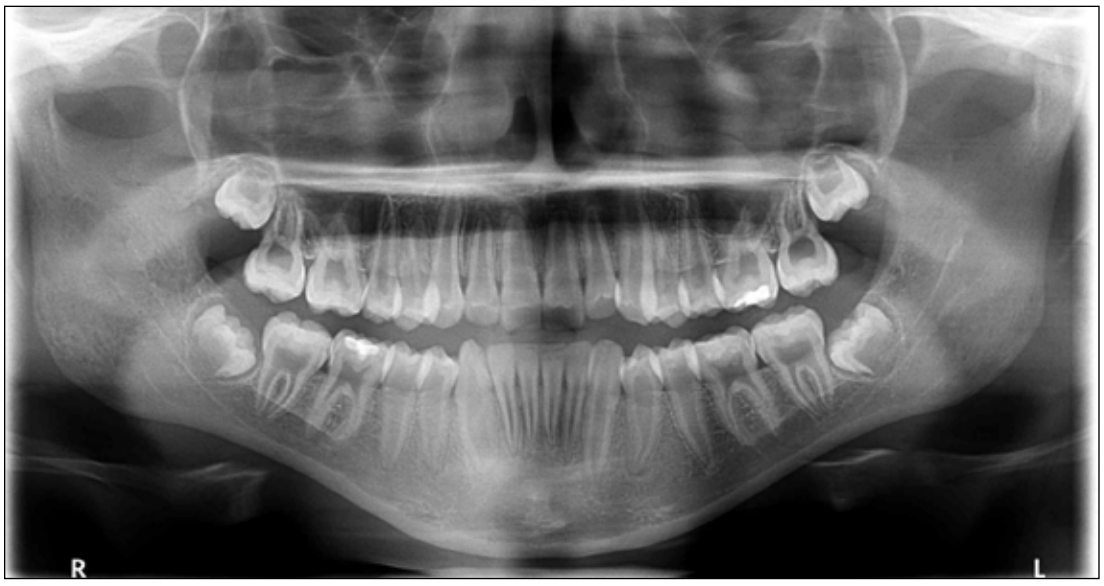


Figure 7: OPG examination 6 months after surgery (March 2018) showing the bone regeneration.



DISCUSSION

Solitary bone cysts occur mainly in young patients, frequently during the second decade of life, and mainly affect the proximal femur and humerus, although they may also occur in the mandible.[17] The distribution among males/females is relatively even.[18] They are tumor-like benign lytic bone lesions of an unknown cause attributed to a local disturbance of bone growth. Unfortunately, there are often no representative subjective symptoms. More than 70% of patients report no pain, swelling, face asymmetry, enlarged lymph nodes or skin lesions, which is the main reason for delayed treatment implementation.[19,20] However, pain is a symptom in 10% to 30% of patients.[21-23] Some researchers have reported more unusual symptoms, including tooth sensitivity,[22,24] paresthesia and displacement of the inferior dental canal,[24] fistulas[25] and pathologic fracture of the mandible and delayed eruption of permanent teeth.

In 1946 Rushon established the following criteria for solitary bone cyst diagnosis: single lesion, no epithelial lining, absence of infection, cavity filled with serous or hemorrhagic fluid or empty, bone walls.[26] Radiographic examination is the most important examination for making the initial diagnosis. An OPG usually reveals a single, radiolucent, homogeneous cavity, which is usually unilocular. When the cavity extends between the roots of the teeth, a scalloping effect can be seen on radiograph.[4] Such an appearance is highly typical of benign lesions, especially when accompanied by the lack of root resorption. The presence of an osteosclerotic layer indicates a non-infected lesion. This disappears when the lesion becomes infected and its contents become suppurative.[20,27,28]

Lesions of larger proportions tend to present themselves as more polymorphic images. They may appear multilocular and suggest other, potentially more aggressive lesions, such as ameloblastoma or myxoma. Occasionally, expansion or erosion of the cortical plate is noted.[21,22]

Diagnostic imaging should be supported by CBCT, which allows three-dimensional assessment[27] and advance planning for surgery. Because a SBC, among many other bone changes, is clearly visible on routinely taken radiographs in the early stages of development, the important role of each dentist in accurate assessment of radiographs cannot be underestimated. Basing the diagnosis exclusively on OPG/CBCT cannot be recommended. There are other abnormalities with a similar appearance. Distinguishing among them is virtually impossible. Central giant cell tumors, keratocystic odontogenic tumors, osteoblastomas, ameloblastomas, monostotic fibrous dysplasia, non-ossifying fibromas, eosinophilic granulomas, hemophilus intraosseous pseudotumors, enchondromas and brown tumors must be taken into consideration.[20,27] The only way to determine the exact diagnosis is to carry out an intra-operative exploration with histopathological evaluation. Histological features of SBC seen under the microscope are typical. There is a cavity containing a thin, sometimes incomplete, connective tissue layer, with spindle cells, hemosiderin pigment, and small numbers of chronic inflammatory cells (with no clinical importance). Numerous congested capillaries and cholesterol crystals, related to the osseous necrosis, may also be present.[29] The above-mentioned connective membrane is very fragile, is fairly difficult to remove in one piece and has a tendency to tear off.[30]

The cavity content may vary according to its location and stage of development. Solitary bone cysts found in the mandible body are usually literally empty. The cystic contents seem to change according to SBC evolution, from blood to serohematic, serous fluid to an empty cavity, which seems to be the final stage in its evolution[31].

For years, a wide spectrum of treatment modalities for SBC have been proposed; however, the appropriate treatment remains unclear. Documented options include: curettage, enucleation, embolization, block resection and even observation. More recently, percutaneous steroids and autogenous bone marrow injection have been introduced.[32] Nowadays, the standard treatment is enucleation with curettage. Surgical treatment involves three basic stages: evacuation of content, cochleation of the cavity to stimulate bleeding, and suture of the wound. The operation is performed under general anesthesia. After preparing the mucoperiosteal flap, surgeons must gain access to the lesion, and the external bone covering the cavity must be removed. During surgery an empty bone cavity is frequently revealed. Sometimes it is filled with scant blood or serous fluid, containing slightly more bilirubin than blood serum.[32] The epithelial sheath is never visible; if it were, the SBC diagnosis would be instantly ruled out. The wall of solitary bone cysts is covered with fibrous tissue, acting like a semi-permeable membrane. Intra-operative examination reveals no direct vascular or lymphatic connection with the cyst cavity.

The correct diagnosis is essential for successful treatment of every bone lesion. Solitary bone cysts can be mistaken for many other lesions (ameloblastoma, central giant cell tumor etc.) that may require far more extensive surgical treatment. The definitive diagnosis of pseudo cysts occurs during surgery after a brief histopathological examination, either confirming neoplastic cells, or, hopefully, the lack of them. If no neoplastic cells and no epithelium are apparent, the treatment remains conservative, focusing on gentle curettage to provoke bleeding. After covering the bone cyst with a mucoperiosteal flap, the blood clot is supposed to reorganize and eventually be replaced with new, osseous tissue.[33]

Borgonovo *et al* suggested that pediatric surgery should be minimally invasive, with careful clinical and radiographic evaluation before surgery.[34] The first signs of bone regeneration are usually displayed on regular radiographs after eight weeks.

The prognosis is usually good, and recurrence is rare. Precious and McFadden described a case of SBC recurrence after performing conventional curettage. Instead of a healing process on OPG, the lesion had increased in size and had become multilocular. Surgery was carried out again in order to achieve bone regeneration, this time by injecting autogenic blood into the cavity.[35] If conventional treatment fails, after confirmation that the lesion is free of neoplastic cells , the surgeon may implement additional procedures. The cavity, after additional curettage, may be filled with autogenic material blood or bone chips, or with allogenic material (lyophilized bone chips, hydroxyapatite or gel foam).[36] The literature also contains documented cases of spontaneous resolution of lesions and bone regeneration.[37] Although remission is possible, waiting for a miracle seems to be unreasonable, especially given that only surgery can distinguish a benign solitary bone cyst from other, more dangerous changes. Leaving any radiolucent lesion for observation appears to carry substantially more risk.

CONCLUSION

1. Solitary bone cysts are easy to overlook in the early stages of development, and can be difficult to distinguish from other, potentially malignant conditions.
2. In most cases the lesion is detected incidentally, during routine dental radiographs, so each dentist should be familiar with and well prepared to initially diagnose radiolucent areas in the jaws.

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