Diffuse sclerosing osteomyelitis of the mandible treated successfully with bisphosphonates. A case report.

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Introduction:

The diffuse sclerosing osteomyelitis (DSO) of the mandible is characterized by recurrent pain of the jaw and swelling of the surrounding soft tissues. The final understanding of the etiology of this disease is still unclear. In literature the therapy is described as difficult without sufficient response to the standard treatment methods like antibiotics, non-steroidal anti-inflammatory drugs and surgical procedures like decortication. The symptoms seem often to be recurrent. During the last years some promising reports and small series about effective therapy of the DSO with oral or intravenous bisphosphonates have been published.[1-5]

Case report:

We report about a 66 years old Patient suffering from DSO in the lower jaw.
In March 2012 he presented in our department with recurrent pain and swelling of the mandible for the first time. The panoramic x-ray and the cone beam computed tomography showed the typical findings of a DSO (Fig. 1 and 2).
In his medical history the patient reported from an extraction of the right third molar in 2007. Subsequently swelling and pain occurred in the right mandible for years. In the previous clinic a biopsy and mandibular decortication in combination with prolonged antibiotic treatment was performed without any lasting improvement of the symptoms since 2010.

During the primary examination in our department no dehiscence or exposed bone of the mandible was found. There were no signs for dental infection by positive sensivity test of all teeth. The medical history was free of pre-existing diseases or regularly prescribed medication.
Due to inconspicuous intraoral conditions we decided to choose a conservative treatment strategy first. We started with intravenous antibiotic therapy (Ampicillin 2g + Sulbactam 1g i.v. 3x per day) combined with overall 10 sessions of hyperbaric oxygen therapy from 23rd of April until the 11th of May 2012. During this therapy there was no reduction of the complaints or improvement of the radiological findings (Fig. 3).

We continued the treatment with i.v. antibiotics and hyperbaric oxygen therapy from the 18th of June until the 6th of July 2012. Again no significant improvement of the clinical findings and symptoms could be achieved.
With regard of the promising reports about effective therapy of DSO with bisphosphonates,[1-5] we decided to administer the patient a single shot of 4g zolendronate (Zometa®) intravenously after he has given his informed consent concerning an off-label use of the drug and the risk of developing a bisphosphonate related osteonecrosis of the jaw. Within 2 days the patient appeared completely free of pain and the long lasting pain medication could be discontinued.

Over the course of time the patient was free of complaints with complete absence of clinical findings. The panoramic x-rays five and ten month after administering 4g zolendronate showed a general bone homogenization with reduction of the osteolysis and a condensation of the bone especially in the mandible (Fig. 5 and 6).

In April 2014, 2 years after bisphosphonate infusion, the patient presented again with the symptoms of DSO in the lower jaw. At that time pain was the leading complaint. After a second single shot of 4g zolendronate (Zometa®) the patient immediately was free of pain und complaints again. Since this time the patient is still free of symptoms and the radiological findings are still constant without any worsening (Fig. 7).

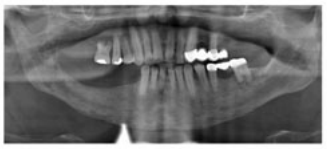

Fig. 1: panoramic x-ray with the primary findings 3/2012.

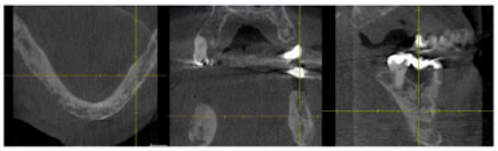

Fig. 2: cone beam computed tomography with the primary findings 03/2012.

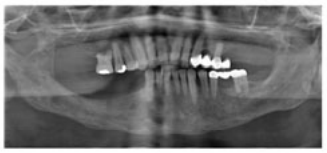

Fig. 3: panoramic x-ray with unchanged findings after antibiotic tretamend an 10 sessions of hyperbaric oxygen therapy 06/2012.

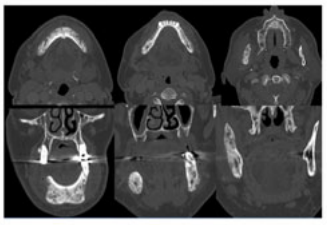

Fig. 4: computed tomography with unchanged findings after antibiotic tretamend an 10 sessions of hyperbaric oxygen therapy 06/2012.

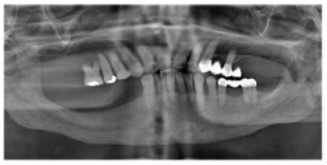

Fig. 5: panoramic x-ray from 11/2011 with general bone condensation and homogenization as well as reduction of the osteolysis.

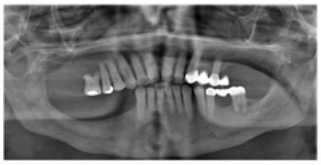

Fig. 6: panoramic x-ray from 4/2013.

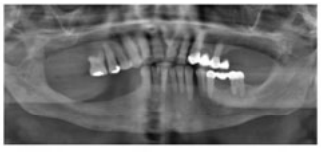

Fig. 7: panoramic x-ray from 08/2014 after the second single shot of zelendronate with constant homogenization of the bone.

Discussion:

Bisphosphonates seem to be a promising option to reduce the progress of DSO ard to control the clinical symptoms. However, longtime evaluation und follow up of larger case numbers are still necessary to proof the evidence of this treatmert approach. The therapeutic dosage protocol as well as the way of application (intravenous or oral) are further unclear and still just based on the experience of some single case reports or small case series.[1-5]

Apart from the promising positive aspects of bisphosphonate treatment in patients with DSO, the undesirable side effects, especially the risk of development a bisphosphonate related osteonecrosis of the jaw have to be taken under account. Therefore, intensive information of the patients about this possible risk and the off-label use of the drug are mandatory.

References:

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The authors disclose that they have no conflict of interest.