

CASE FOR DIAGNOSIS

Submandibular Cutaneous Fistula[☆]

Fístula cutánea submandibular

Medical History

An 84-year-old patient consulted for a chronic suppurative, painless lesion over the left mandibular arch. The lesion had been present for 6 months. Of interest in her medical history were hypertension, type 2 diabetes mellitus, dyslipidemia, and osteoporosis. She had been receiving treatment with nimodipine, metformin, fluvastatin, and alendronate for the last 3 years, and had undergone a tooth extraction 2 years earlier.

Physical Examination

The left perimandibular region was swollen, erythematous, and indurated to palpation, with a submandibular, ulcerated, fistulous area of retracted skin, from which a seropurulent exudate drained. Within this area was a fleshy excrescent papule (Fig. 1). Examination of the oral cavity revealed teeth in poor condition.

Histopathology

Hematoxylin-eosin staining showed an area of ulceration and acute inflammation, and the presence of large multinucleated cells without any histologic signs of malignancy.

Additional Tests

Orthopantomography showed an area of osteolysis of the cortical bone in the left mandibular arch, as well as other radiolucent points indicative of general mandibular involvement (Fig. 2).



Figure 1



Figure 2

What Is Your Diagnosis?

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Diagnosis

Bisphosphonate-induced osteonecrosis of the jaw.

Course and Treatment

Alendronate was discontinued. After culture of the exudate, which was positive for *Staphylococcus aureus*, the patient was managed conservatively with antibiotic therapy with ciprofloxacin 750 mg every 12 hours for 3 weeks. The skin lesion improved markedly. The seropurulent exudate and inflammation were no longer present, but the fistula persisted (Fig. 3).

Comment

Bisphosphonates are potent inhibitors of osteoclastic bone resorption and angiogenesis. They are used in the treatment of diseases such as lytic bone metastases, malignant hypercalcemia, multiple myeloma, osteoporosis, and Paget disease.¹⁻³ Osteonecrosis of the jaw appears to be caused by poor blood supply and lack of bone remodeling and regeneration.⁴ This condition was first described in 2003.^{2,5} The incidence of this complication in oral bisphosphonate therapy is estimated to range approximately from 0.01% to 0.04%.^{3,5} Osteonecrosis of the jaw develops after chronic inflammation in an environment of deficient vascularization, leading to changes in bone microstructure and ultimately to collapse.² These events result from the inability of the bone to increase the regeneration process in response to chewing or dental manipulation or infections; in such situations a greater functional bone reserve is required.⁵ The risk of this complication is greater when the treatment duration exceeds 3 years, and 70% of cases are triggered by tooth extractions.^{1,3,5} Other factors such as corticosteroid use,

diabetes, advanced age, insufficient dental hygiene, smoking, and excessive alcohol consumption increase the risk of the disease and accelerate progression.^{3,5} Overall, 60% of cases are reported in women.³ The half-life the bisphosphonate that has accumulated in bone can be as long as 11 years.²

When a mandibular fistula is present, the differential diagnosis should include not only odontogenic cysts, foreign-body reaction, pyogenic granuloma, tumors, and infectious processes, but also bisphosphonate-induced osteonecrosis of the jaw if the patient is receiving bisphosphonates, particularly if multiple fistulas are present.

Diagnosis is based largely on the clinical manifestations. Imaging studies are the main additional tests and allow the severity and extent of mandibular involvement to be determined.⁵

No effective treatment has been established, although a conservative approach seems the most recommendable. Aggressive debridement should be avoided due to the risk of recurrence and sequelae, as bone involvement is generalized and there is a risk of increasing the size of the lesion.^{1,2} Systematic cultures are recommended.³ According to Marx et al.,⁶ the approach should consist of oral hygiene and rinsing with chlorhexidine 0.12%, long-term continuous or intermittent antibiotic treatment, and avoidance of surgery or extensive debridement.

References

1. Alonso Estellés R, Campo López C, Aguilar Jiménez J, Desco Agulló F. Fístulas mandibulares en una mujer de 75 años. *Rev Clin Esp.* 2008;208:165-7.
2. Joshi Otero J, Rollón Mayordomo A, Coello Suanzes J, Lledó Villar E, Lozano Rosado R, Sánchez Moliní M, et al. Osteonecrosis de los maxilares asociada al uso de bifosfonatos: revisión de ocho casos. *Rev Esp Cir Oral Maxilofac.* 2011;33:15-21.
3. Echeveste Inzagaray JM, Martínez Morentin M. Osteonecrosis mandibular relacionada con la toma de bifosfonatos por vía oral: a propósito de un caso. *Semergen.* 2011;37:430-2.
4. Truong SV, Chang LC, Berger TG. Bisphosphonate-related osteonecrosis of the jaw presenting as a cutaneous dental sinus tract: A case report and review of the literature. *J Am Acad Dermatol.* 2010;62:672-6.
5. Casal C, Someso E, Álvarez AM, Fariña J, Álvarez T. Osteonecrosis de maxilares relacionada con el uso de bifosfonatos. *FAP.* 2012;10:9-14.
6. Marx RE, Sawatari Y, Fortin M, Broumand V. Bisphosphonate-induced exposed bone (osteonecrosis/osteopetrosis) of the jaws: Risk factors, recognition, prevention, and treatment. *J Oral Maxillofac Surg.* 2005;63:1567-75.

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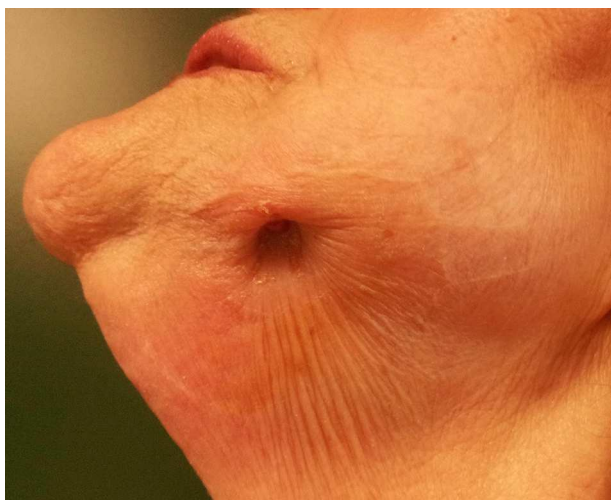


Figure 3