

# Bilateral bisphosphonate-related osteonecrosis of the jaw with left chronic infection in an 82-year-old woman

Laure Gallay,<sup>1</sup> Anne-Gaëlle Bodard,<sup>2</sup> Christian Chidiac,<sup>1,3,4,5</sup> Tristan Ferry<sup>1,3,4,5</sup>

<sup>1</sup>Hospices Civils de Lyon, Lyon, France

<sup>2</sup>Unité d'Odontologie, Service de Chirurgie, Centre Régional de Lutte Contre le Cancer, Centre Léon-Bérard, Lyon, France

<sup>3</sup>Université Claude Bernard Lyon 1, Lyon, France

<sup>4</sup>Centre International de Recherche en Infectiologie, CIRI, Inserm U1111, CNRS UMR5308, ENS de Lyon, UCBL1, Lyon, France

<sup>5</sup>Centre Interrégional de Référence Rhône-Alpes Auvergne des Infections Ostéo-articulaires complexes, Hospices Civils de Lyon, France

## Correspondence to

Dr Tristan Ferry,  
tristan.ferry@univ-lyon1.fr

## DESCRIPTION

An 82-year-old woman, treated for severe rheumatoid polyarthritis by tumour necrosis factor inhibitors (2003–2008), rituximab (2008), abatacept (2008–2011) and long-term steroid therapy, also received prolonged bisphosphonate therapy to prevent bone mass loss. In 2009, a bilateral bisphosphonate severe osteonecrosis of the jaw (BONJ) was discovered along with evidence of left chronic mandible infection in early 2012, as mucosal disruption and suppurative sinus tract occurred (figure 1A,B). Bisphosphonate was stopped and mild surgical debridement was performed. Despite administration of antimicrobial therapy including high dose of amoxicillin and fluconazole, the suppurative sinus tract persisted. As a left mandibular fracture with significant mouth opening reduction was spontaneously carried out, a radical surgery, consisting of complete hemi-mandibular resection without reconstruction, was performed. Surgical bone sampling revealed *Actinomyces naeslundii* in cultures, and prolonged amoxicillin therapy was administered. During the follow-up, the outcome was favourable without recurrence of the suppuration (figure 1C,D).

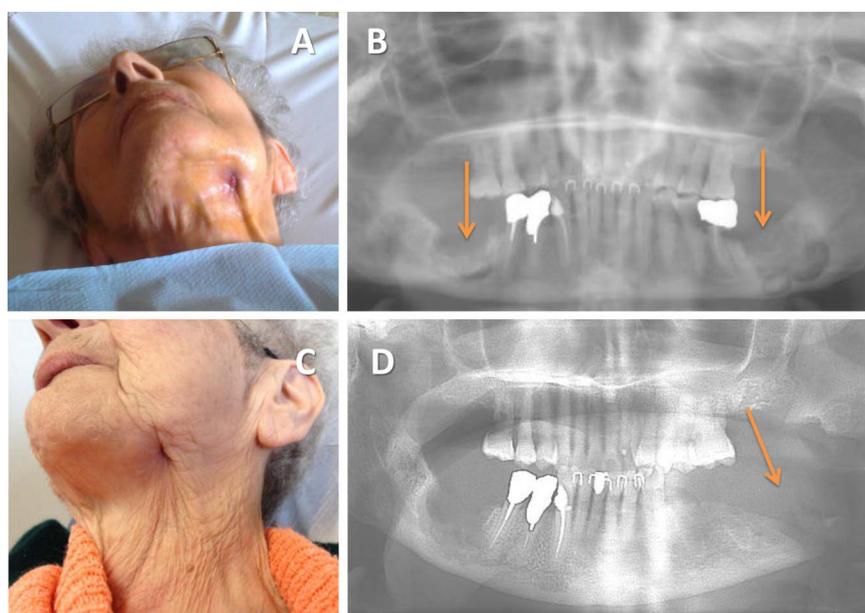
The pathogenesis of BONJ is not well known.<sup>1</sup> Some risk factors have been identified, such as the route of administration or the duration use of bisphosphonate, the concomitant administration of

corticosteroids and performance of dental invasive procedures.<sup>1</sup> BONJ may be owing to: (1) osteoclast inhibition and antiangiogenic effect of bisphosphonate, resulting in oversuppression of bone turnover and avascular necrosis; (2) bisphosphonate mucosal toxicity leading to mucosal disruption and (3) involvement of *Actinomyces* species in biofilm in bone.<sup>1–3</sup> BONJ with evidence of chronic infection is associated with severe morbidity, required adequate surgery (debridement or more aggressive surgery) and prolonged antimicrobial treatment targeting *Actinomyces*.<sup>1–3</sup>

## Learning points

Bilateral bisphosphonate severe osteonecrosis of the jaw:

- ▶ Occur mainly in patients with several risk factors, such as age and coadministration of corticosteroid therapy.
- ▶ Is due to bisphosphonate toxicity, but there is some evidence that bone colonisation by *Actinomyces* species in biofilm participate to the pathogenesis.
- ▶ Could be associated with evidence of infection, requiring surgery and prolonged antimicrobial therapy targeting *Actinomyces* species.



**Figure 1** Bilateral bisphosphonate severe osteonecrosis of the jaw with suppurative sinus tract (A). The orthopantomographic view revealed bilateral severe osteonecrosis (B, arrows). After radical surgery and prolonged amoxicillin therapy, the outcome of the infection was favourable without recurrence of the suppuration (C). The orthopantomographic view after complete hemi-mandibular resection with dental extractions and without reconstruction (D, arrow).

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## REFERENCES

- 1 Advisory Task Force on Bisphosphonate-Related Osteonecrosis of the Jaws, American Association of Oral and Maxillofacial Surgeons. American Association of Oral and Maxillofacial Surgeons position paper on bisphosphonate-related osteonecrosis of the jaws. *J Oral Maxillofac Surg* 2007;65:369–76.
- 2 Naik NH, Russo TA. Bisphosphonate-related osteonecrosis of the jaw: the role of actinomyces. *Clin Infect Dis* 2009;49:1729–32.
- 3 Magopoulos C, Karakinaris G, Telioudis Z, *et al.* Osteonecrosis of the jaws due to bisphosphonate use. A review of 60 cases and treatment proposals. *Am J Otolaryngol* 2007;28:158–63.

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