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Case Report

Bisphosphonate-related osteonecrosis of the jaw complicated by Ludwig's angina

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Abstract

Ludwig's angina is a life-threatening cellulitis that involves the submandibular and sublingual spaces. It often occurs after an infection of the roots of the teeth. However, modern dental care and use of antibiotics for oral infections have made Ludwig's angina rare. We present here a cancer patient exhibiting the sequential features of bisphosphonate related osteonecrosis of the jaw on bone scan complicating with Ludwig's angina. This report highlights the need for medical practitioners to be alert to these rare combinations in the compromised patient after bisphosphonate therapy. To the best of our knowledge, no case of Ludwig's angina secondary to osteonecrosis of the jaw has been reported.

Keywords: bisphosphonate; bone scan; jaw; Ludwig's angina; osteonecrosis

1. Introduction

Ludwig's angina is an infection of the submandibular and sublingual spaces. It can potentially lead to a fatal airway obstruction. It often occurs after an infection of the mandible. We present here a cancer patient exhibiting the sequential features of bisphosphonate-related osteonecrosis of the jaw (BRONJ) on bone scan and complicating with Ludwig's angina. This report highlights that practitioners should be alert to these rare combinations in the compromised patient after bisphosphonate therapy.

2. Case Report

An 83-year-old man, who had been diagnosed 2 years earlier with prostate cancer, presented with submandibular swelling and inability to swallow saliva for 1 week. With preliminary assessment as Ludwig's angina at a local hospital, and patient had undergone antibiotic prophylaxis and to abscess drainage prior to the referral. He was nonfebrile and not in respiratory distress. Computed tomography of his neck showed a significant inflammation with abscess and fistula formation in the left submandibular space. Presence of periosteal reaction, osteolytic and osteoblastic change, bone sequestration, and destruction of the mandible were noted (Fig. 1).

Tc-99m methylene diphosphonate bone scan (Fig. 2) exhibited intense bone lesion in the lower jaw (black arrow) and evidence of distant bone metastasis (open arrows). He was admitted to the dental ward for airway observation and further treatment in September 2010. The aerobic and anaerobic cultures of his blood yielded negative results. Empirical use of 1200 mg augmentin and 500 mg metronidazole intravenously q. 8 hours was given. Subsequently the patient received sequestectomy of the mandible and oral mucosa flap reconstruction. During the operation, an open wound over the left

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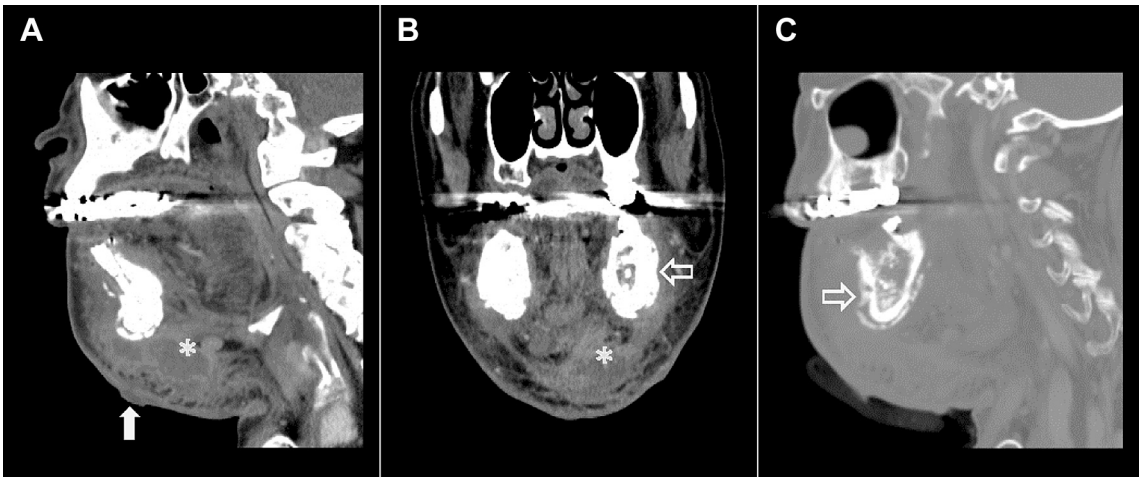

Fig. 1. Contrast-enhanced computed tomography (A, sagittal; B, coronal-reformatted) images show extensive subcutaneous swelling, abscess (asterisk) in the submandibular space and a fistula (arrow) noted. The airway is patent. (C) Bone window image revealing periosteal reaction, osteolytic and osteoblastic changes, and intramedullary sequestra within the mandibular body (open arrow). The computed tomography findings are consistent with osteonecrosis of the jaw.

chin, deep caries, and sequestrum formation in left mandible were found. Histologically, inflammatory cell infiltration, and granulation of soft-tissue were observed. Bone sequestration, necrosis, and ingrowth of squamous epithelium in the mandible were also noted (Fig. 3). A diagnosis of Stage 2 BRONJ, distinguished by bone exposure with soft-tissue and mandible infection, was confirmed. His pus culture yielded no bacterial growth due to the recent antibiotics exposure. The patient was then switched to 1 g augmentin tablet twice daily. Subsequent follow-up at 2 weeks and 3 months at the

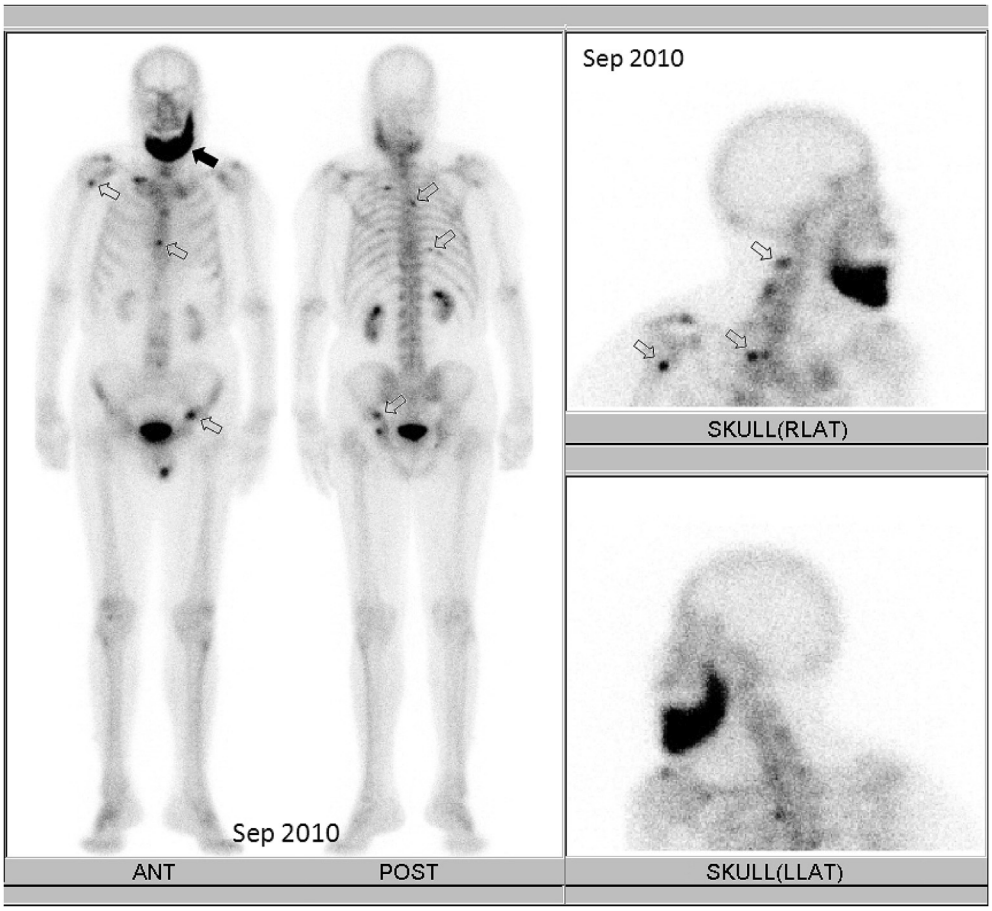

Fig. 2. Tc-99m methylene diphosphonate bone scan exhibited intense bone lesion in the lower jaw (black arrow) and evidence of distant bone metastasis (open arrows).