Alloprevotella rava isolated from a mixed infection of an elderly patient with chronic mandibular osteomyelitis mimicking oral squamous cell carcinoma

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Abstract

The anaerobic, Gram-negative bacillus Alloprevotella rava has recently been described in the human oral cavity. To our knowledge, this species has not been isolated from chronic osteomyleitis samples. We present the first case of *A. rava* infection in a 92-year-old woman with polymicrobial chronic mandibular osteomyleitis, mimicking oral squamous cell carcinoma. © 2021 The Author(s). Published by Elsevier Ltd.

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Introduction

Chronic mandibular osteomyelitis (CMO) is a serious condition requiring early and accurate diagnosis, antibiotics, and surgical management. Cases are usually mixed anaerobic infections, reinforcing the concept that osteomyelitis of the jaws is mainly related to microorganisms from the oral environment [1]. *Alloprevotella rava* is an obligatory anaerobic Gram-negative bacillus isolated from the human oral cavity, associated with oral dysbiotic infections such as dental caries and periodontitis [2–4]. We present the first case of *A. rava* infection in a patient with polymicrobial CMO initially thought to be oral squamous cell carcinoma from clinical and computed tomography findings.

Case report

A 92-year-old woman presented to our hospital with chronic, bloody discharge in the left mandibular body for 18 months,

since the extraction of teeth in the same region. According to her medical history, the woman's cervical lymph node had been removed and she had received radiotherapy with a diagnosis of lymphoma, 5 years ago. She also received medication for osteoporosis. Tissue specimens, obtained from defective bone and surrounding soft tissue, were examined histopathologically and microbiologically. Histopathology made a diagnosis of actinomycosis without any malignancy. However, microbiological examination revealed polymicrobial infection involving five bacteria. Four organisms were identified by matrix-assisted laser desorption/ionization time-of-flight mass spectrometry (MALDI-TOF MS; VITEK MS; bioMérieux, Marcy l'Étoile, France) as Veillonella parvula, Prevotella nigrescens, Klebsiella oxytoca and Corynebacterium durum. The fifth bacterium, mentioned as strain Marmara, for which MALDI-TOF MS was insufficient for identification, was identified by 16S rRNA gene sequencing as A. rava with 99% nucleotide identity to the strain isolated from the human oral cavity (GenBank Accession no. [Q039190) [2] (see Fig. 1). The actinomyces strain was not isolated despite the I4-day incubation period for anaerobic culture [5]. All isolated organisms were susceptible to amoxicillin-clavulanic acid. The patient was given empirical amoxicillin-clavulanic acid, and she was discharged on the fifth day of hospitalization with oral amoxicillin-clavulanic acid therapy $(2 \times I g \text{ daily})$ for 2 months. Her clinical condition



FIG. 1. Neighbour-joining phylogenetic tree based on 16S rRNA gene sequences, showing the relationships between strain Marmara and some related taxa. GenBank accession numbers are shown in parentheses. Numbers at nodes indicate bootstrap percentages (based on 1000 replicates). Bar represents 0.02 substitutions per nucleotide position.

improved considerably, and mandibular discharge and swelling disappeared.

Discussion

Three different anaerobic bacteria were isolated from the samples, consistent with the mixed anaerobic aetiology of CMO. Several fastidious strictly anaerobic bacteria are commonly present in the dental biofilm, which suggests that the source of infecting pathogens in osteomyelitis of the jaws is likely to be gingivitis, chronic periodontitis, previous dental extractions or endodontic treatments [1]. Alloprevotella rava was first isolated from dental plaque [2]. In our case, A. rava was isolated from a woman with CMO with a history of tooth extraction in the same area. This is the first case of CMO involving A. rava in the world literature. However, it may not reflect the actual result because phenotypic identification systems have insufficient databases for this microorganism. Alloprevotella rava has been detected in the oral microbiota of certain patients with oral dysbiosis by molecular techniques since 2013, when it was defined [2-4]. An epidemiological study, conducted among Chinese preschool children, revealed that the relative proportions of A. rava were significantly higher in the halitosis group compared with the control group [6]. A prospective cohort study showed a relationship between an increase in dental caries and the level of the genus Alloprevotella in the oral microbiome among Japanese university students [3]. Another report indicated that the proportion of A. rava was significantly increased in saliva samples of periodontitis patients [4]. On the other hand, Coit et al. [7] reported that the abundance of *A. rava* in the microbial community of saliva was significantly decreased in Behçet's disease compared with healthy controls. Recent research, focused on the role of bacteria in oral carcinogenesis, indicated that the genus *Alloprevotella* was among the bacteria that showed significantly higher abundances [8,9]. However, the features related to its pathogenesis have not been clarified.

Conclusion

Signs and symptoms of oral squamous cell carcinoma, which has a poor overall 5-year survival rate, and CMO can often be similar. The diagnosis of CMO is particularly important because appropriate treatment, culture-guided antibiotic therapy for the infection and surgical management provide successful cure. Defining the bacterial community composition associated with CMO can help in understanding the mechanisms of disease and the impact of the bacteria.

Conflicts of interest

The authors have declared that there are no conflicts of interest.

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