Mandibular Osteolytic Lesion Associated with Exuberant Hyaline Ring Granuloma Reaction

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Key Words
Odontogenic cystic lesion ± Hyaline ring granuloma ± Pulse granuloma ± Mandible

Abstract
Objective: To report an unusual case of oral hyaline ring granuloma (HRG) that caused an extensive osteolytic lesion.
Clinical Presentation and Intervention: A 22-year-old female was referred to our hospital with a large expansile cystic lesion in the left mandibular ramus associated with a clinically visible, partially erupted third molar. A diagnosis of paradental cyst was made. After marsupialization of the lesion, histopathological analysis of the surgical specimen showed an unusual exuberant HRG reaction supported by scarce fibrous stroma.
Conclusion: This was a case of exuberant HRG reaction that caused extensive bone destruction.

Introduction
Hyaline ring granuloma (HRG) is a localized granulomatous inflammatory lesion considered to be a foreign body reaction to vegetable particles [1]. It occurs most frequently in the oral cavity associated with impacted lower third molar or in postextractive alveolar sockets [1]. However, HRG has also been reported in the lung, intestine, skin, gallbladder and uterine tube [2]. Microscopically, HRGs are characterized by hyaline rings associated with multinucleated giant cells [1–4]. Oral HRGs have been found in association with inflammatory fibrous hyperplasia [1], periapical granulomas [3], and odontogenic cysts [5, 6]. The purpose of this case was to report an exceptional case of exuberant HRG reaction that developed in the wall of a mandibular odontogenic cyst.

Case Report
A 22-year-old female patient was referred to our hospital for treatment because she complained about pain and swelling in her left cheek and presented with a clinically visible, partially erupted left inferior third molar with purulent discharge from its gingival
sulcus. On the panoramic radiograph, an extensive cystic-appearing lesion was visualized (fig. 1). After 7 days of oral antibiotic therapy and further evidence of a fluid-containing lesion, the tooth was surgically removed and the lesion marsupialized. The clinico-pathological correlation led to the diagnosis of a paradental cyst (fig. 2a). The patient discontinued her treatment and returned 2 years later with pain and without the device for draining the marsupialized cyst. The cyst was marsupialized again, and microscopically revealed an association with focal HRG reaction (fig. 2b). The lesion was fully enucleated, and this was followed by curettage. The histopathological analysis of the entire tissue blocks showed an exuberant HRG reaction (75% of the surgical specimen) supported by fibrous stroma (fig. 2c–f). Cyst-lining epithelial cells were not observed. On follow-up after 4 months, the panoramic radiograph showed new bone formation.

Discussion

We report herein an exceptional case of exuberant HRG reaction that developed in the wall of a mandibular odontogenic cyst. To our knowledge, similar to our case, there is only one published case showing an association of HRG reaction with a large radiolucent lesion in the mandible [7]; however, different from our findings, HRG reaction was observed only in the initial levels of the entire tissue block.

The paradental cyst is an inflammatory cyst that occurs most often on the buccal or distal aspect of partly erupted mandibular molars, commonly associated with a history of pericoronitis [8, 9]. A large osteolytic lesion involving the mandibular ramus is extremely unusual for paradental cysts; thus, we believe that the exuberant HRG reaction is involved in the pathogenesis of the current lesion.

In a review of 173 cases of oral HRG, Philipsen and Reichart [10] observed that more than two thirds of the lesions occurred in the posterior region of the mandible. Most patients presented pain, swelling, discharge or a chronic sinus tract. HRGs were found as small structures, uncommonly being a significant component in the cyst wall [7]. In relation to HRG pathogenesis [10], the current case supports the exogenous theory. In fact, we have observed two distinct vegetable matters, hollow matrices
and dark organic material. The surgical procedures indicated that therapeutic substances in the surgical area were not used. The hollow matrices contained fibrovascular tissue cores admixed with multinucleated giant cells, whereas the dark organic material induced a granulomatous foreign body response.

Conclusions

This was a case of exuberant HRG reaction, which was initially associated with a paradental cyst, and showed extensive destruction of large bone. After surgical treatment, clear signs of healing were observed.

References