Title:
Coincidently oral lesions in Crohn’s disease

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Dear Editor,

This paper reported a 21-year-old male who presented with a gradually enlarging painful mass in his mouth for three months. He was diagnosed with Crohn’s Disease (CD) 4 years ago and underwent an ileostomy at a local hospital due to acute perforations. Despite receiving Infliximab every eight weeks since then, his disease control has been poor (Figure 1A). Upon admission, a thorough oral examination revealed an immobile bone mass in the left mandibular region that was palpable but not noticeable (Figure 1B). Conical beam computed tomography (CBCT) revealed a low-density shadow, about 0.95 x 0.70 cm in size, affecting 34-37 gums, with 34-35 sharp absorptions (Figure 1C). Whole-body bone imaging combined with Single-Photon Emission Computed Tomography/Positron Emission Tomography fusion imaging (SPECT/CT) revealed a low-density vesicle in the left mandible body with no evidence of bone metabolism abnormalities (Figure 1D). Contrast-enhanced
computed tomography (CECT) scan demonstrated irregular cystic lesions in the left mandible (Figure 1E). After confirming the absence of malignant cells through frozen sections, a complete excision of the infiltrated bone surrounding the lesion was performed. The histopathological findings confirmed the presence of a Central type giant cell granuloma (CGCG) with admixed multinucleated giant cells, extravasated erythrocytes, and osteoid cell formation (Figure 1F). The patient was followed up for 6 months without any evidence of recurrence.

CGCG is a rare non-cancerous bone lesion that can cause local damage and has the potential for long-term malignant transformation. The etiology of CGCG remains unclear, but possible factors include local trauma, inflammation, intraosseous bleeding and genetic abnormalities. However, in the case of our patient, no apparent correlation was found with any of these factors except for the poorly controlled CD inflammatory condition. Histopathologically, CGCG is characterized by lobular hyperplasia of mononuclear fusiform and polygonal cells mixed with multinuclear giant cells, extravasated red blood cells, and variable osteoid formation cells. This presentation is distinct from non-caseating granulomas of oral manifestation of CD and Infliximab-induced sarcoidosis-like reaction. Although internal corticosteroid injections and Denosumab are effective alternatives, surgical intervention remains the optimal approach.

To our knowledge, we are the first to report an association between CGCG and both CD and anti-tumor necrosis factor therapy treatment. Although it may be coincidental, CGCG should be considered in diagnosing oral granulomas, especially when CD patients have oral manifestation or are treated with TNF-α antagonists.

The authors declare no conflicts of interest.

References


[2] Papanicolaou P, Chrysomali E, Stylogianni E, et al. Increased TNF-α, IL-6 and decreased IL-1β immunohistochemical expression by the stromal spindle-shaped


Figure Legend

Figure 1. A The control of CD condition was not well, B Oral examination revealed the presence of a hard, immobile bone mass in the left mandibular region that was palpable but not noticeable (black arrow), C CBCT revealed a low-density shadow, about 0.95 x 0.70 cm in size, affecting 34-37 gums, with 34-35 sharp absorptions, D SPECT/CT fusion imaging revealed a low-density vesicle in the left mandible body with no evidence of bone metabolism abnormalities, E CECT demonstrated an irregular cystic lesion in the left mandible, F The histopathological findings confirmed
the presence of CGCG with admixed multinucleated giant cells (blue arrow), extravasated erythrocytes (red arrow), and osteoid cell formation cells (yellow arrow)(HE 200x). (these images are published with the informed consent from the patient).

**Abbreviations list:**

CBCT: Conical beam computed tomography
SPECT/CT: Whole-body bone imaging combined with Single-Photon Emission Computed Tomography/Positron Emission Tomography fusion imaging
CECT: Contrast-enhanced computed tomography
CGCG: Central type giant cell granuloma
H&E: hematoxylin-eosin staining