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KAPOSI SARCOMA DISGUISED AS PERICORONITIS: A DEFERRED DIAGNOSIS DUE TO COVID-19 PANDEMIC

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ABSTRACT

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Aim The current case report describes an uncommon presentation and subsequent diagnosis of Kaposi sarcoma caused by acquired immunodeficiency syndrome (AIDS).

Summary The COVID-19 pandemic led to global disruptions in healthcare services, sometimes resulting in postponed diagnoses of infectious diseases. Kaposi sarcoma (KS) is a malignant soft-tissue neoplasm commonly associated with human immunodeficiency virus (HIV) and AIDS, but it also occurs in other immune-compromised individuals. The oral manifestations of KS play a crucial role in its early diagnosis and may be a predictor of disease progression from HIV to AIDS. The current case report describes an unusual case involving a young male who presented to a dental clinic with persistent postoperative pain and delayed wound healing following extraction of his lower third molar. Clinical examination indicated a proliferative mucosal lesion with reddish-purple coloration in the vicinity of the surgical site that extended to the left retromolar pad, and cone-beam computed tomography scans showed marked osseous changes. Histopathological analysis confirmed a diagnosis of AIDS-related KS and an additional diagnosis of metastatic pulmonary KS.

Key learning points

1. The COVID-19 pandemic caused numerous disruptions in healthcare systems and services, which led to delayed healthcare visits.
2. Kaposi sarcoma is a malignant soft-tissue neoplasm commonly associated with HIV and AIDS.
3. Kaposi sarcoma may also occur in other immune-compromised individuals.
4. The oral manifestations of KS may play a crucial role in diagnosis of AIDS.
5. Kaposi sarcoma may present in uncommon ways, such as persistent postoperative pain and delayed wound healing.

KEYWORDS

Kaposi Sarcoma; Postoperative Pain; Delayed Wound Healing; Human Immunodeficiency Virus; Acquired Immune Deficiency Syndrome.

1. INTRODUCTION

Sarcomas are a rare mesenchymal cancer that grows in different types of connective tissue [1]. Kaposi sarcoma (KS) is one type of sarcoma that develops from the cells lining lymphatic or blood vessels [1]. It is classified as an intermediate neoplasm because it lacks the conventional features of a true malignancy [2]. Caused by human herpes virus-8 (HHV-8), KS is present in 1%–5% of the general population globally, but its seroprevalence is greater (20%–77%) among men who have sex with other men, particularly those diagnosed with human immunodeficiency virus (HIV) and acquired immunodeficiency syndrome (AIDS) [3]. Because HHV-8 is considered an opportunistic infection, it is often transmitted between individuals through saliva, sexual activity, blood, or organ transplant. However, it can also remain latent until the individual experiences an immunosuppression reaction, at which point the virus becomes associated with additional pathologies, such as KS or lymphomas. Therefore, it is crucial to understand these transmission routes and latency mechanisms for effective prevention and management strategies in susceptible populations.

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Case Reports

Oral KS occurs in approximately 22% of individuals diagnosed with HIV and is typically the first sign of undiagnosed HIV infection [4]. The most common sites of occurrence in the oral cavity are the hard palate, gingiva, and dorsum of the tongue [5,6]. Clinically, oral KS can present as solitary, multifocal, or multicentric macular patches, plaques, or nodules of varying dimension; colors can range from deep red to purplish blue [7,8]. Multifocal lesions may also coalesce into solitary exophytic masses. Oral KS can cause local tissue destruction, pain, spontaneous bleeding, masticatory difficulty, and interference with the wearing of oral prostheses. Although severe alveolar bone destruction and unexplained tooth mobility with underlying oral KS has been documented [9], oral KS presenting as a source of persistent dentoalveolar pain is uncommon. Therefore, the aim of the current case report was to describe an uncommon presentation and subsequent diagnosis of KS caused by AIDS.

2. CASE PRESENTATION

A 29-year-old White male presented to our Oral & Maxillofacial Surgery clinic with pain with accompanying intraoral swelling that emanated from the region around his lower left third molar. The only relevant medical history that the patient divulged is that he had been previously been diagnosed with HIV but had not followed with his physician for management and treatment. Patient did not provide any previous medical records or blood work. Clinical examination revealed erythematous, hyperplastic soft tissue partially covering the lower left third molar (tooth #17) and tenderness on palpation. A diagnosis of acute pericoronitis was established based on the clinical and radiological findings. Due to the acute nature of this event, a clinical decision to proceed with treatment was made. The tooth was considered to be not salvageable and was extracted. The socket was subsequently curetted to remove remnants of granulation tissue. The residual pericoronal tissue was rather prominent and noticeable. The patient was instructed to return for evaluation of the surgical site if his pain persisted or there was no resolution of the redundant pericoronal tissue. However, the patient did not follow up as instructed, likely because of the COVID-19 pandemic. His treatment coincided with increased public health restrictions designed to mitigate the effects of the pandemic, which meant that follow-up oral care services were severely limited. Four months later, the patient returned to the clinic with complaints of bleeding and constant, low-grade pain emanating from the surgical site. He reported that the bleeding started two weeks before and that his pain had intensified (5 of 10 on a numeric pain scale), leading him to seek care. At the time of this visit, his HIV disease was poorly controlled (CD4+ < 200 cells/mm3, viral load = 67,229 copies/mL), and he had stopped taking the highly active antiretroviral therapy over the past few months. He reported generalized weakness, insomnia, poor appetite, and weight loss during the past month.

Extraoral examination was significant for a nontender, nonmobile left submandibular lymph node that measured approximately 1 × 1 cm. Intraoral examination revealed a 3 × 3 cm bluish-purple proliferative lesion that originated from the extraction socket site of tooth #17, extended to the left retromolar pad, and involved the lingual gingiva and buccal vestibule (Fig. 1).

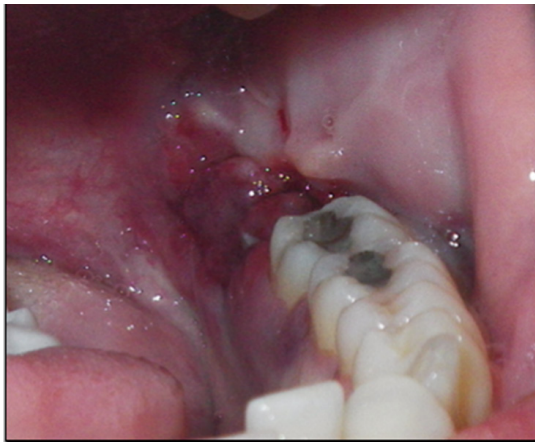

Figure 1. Irregular proliferative lesion at the healing surgical site of tooth #17 that extended toward the lingual and buccal gingiva and toward the faucial pillars of the oropharynx.

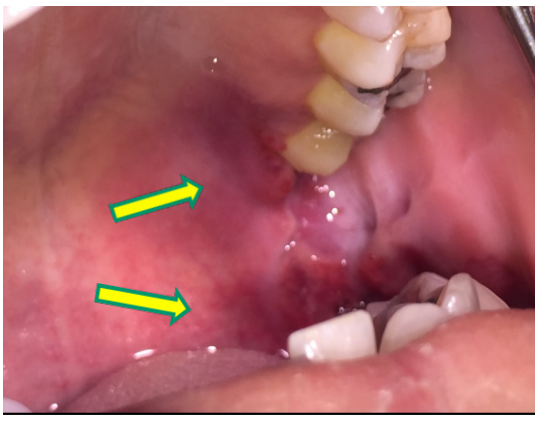

Figure 2. Reddish-purple hyperplastic discoloration (arrows) that extended superiorly around the attached palatal gingiva of tooth #16.

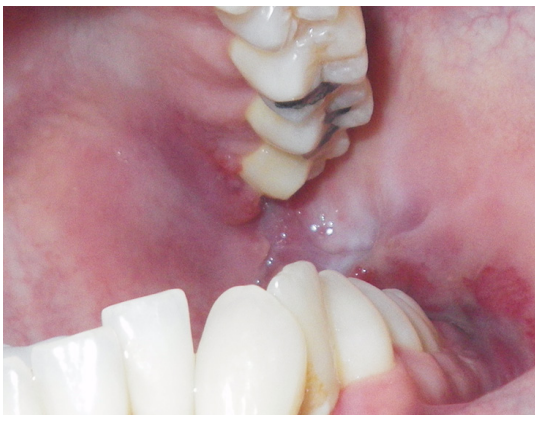

Figure 3. Reddish-purple coloration of palatal gingiva at tooth #16 and near the left maxillary tuberosity.

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A less intense, reddish-purple mucosal discoloration was also visible in the left maxillary tuberosity (Fig. 2). Palpation of the clinical sites elicited intense pain, which the patient indicated was the primary reason he returned for follow-up (Fig. 3).

At this visit, a panoramic radiograph was obtained and indicated a well-healed extraction socket for tooth #17 and intact cortical borders of the mandible (Fig. 4). The shadow of a dome-shaped soft tissue growth of approximately 9-mm wide × 3-mm high at the crest of the ridge posterior to tooth #18 was evident and likely represented the lesion.

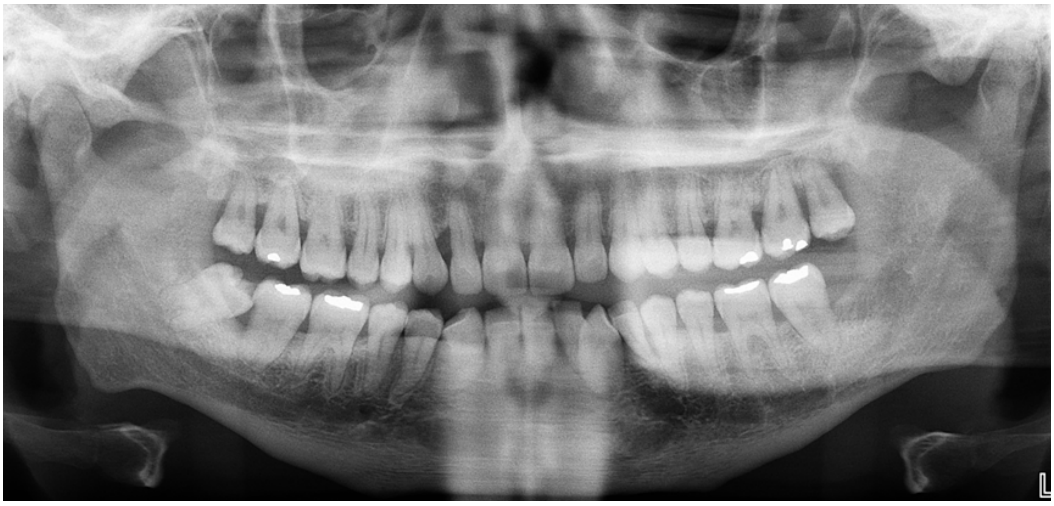

Figure 4. Panoramic radiograph showing intact cortical alveolar borders in the healed extraction socket (region of tooth #17), but marked alveolar bone loss around tooth #16.

Next, an incisional biopsy of the lesion growing from the extraction site of tooth #17 was performed. Histopathological examination with hematoxylin-eosin confirmed the presence of AIDS-associated oral KS. Low resolution (Fig. 5) and medium resolution (Fig. 6) photomicrographs showed a polypoid lesion composed of vascular connective tissue and pleomorphic spindle cells compressing the extravasations of erythrocytes.

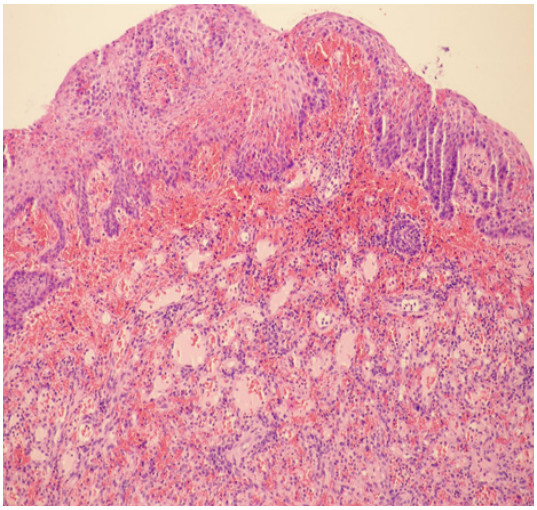

Figure 5. Low resolution histopathological photomicrograph showing a polypoid lesion composed of vascular connective tissue partially covered by stratified squamous (hematoxylin-eosin, magnification ×40).

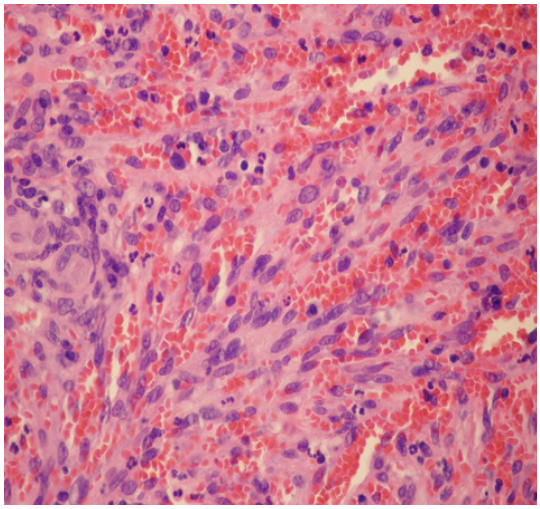

Figure 6. Medium resolution histopathological photomicrograph showing poorly differentiated vascular slits and fascicles of pleomorphic spindle cells compressing the extravasated erythrocytes (hematoxylin-eosin, magnification ×40).

Advanced imaging was then performed to determine whether there was any osseous change in the site of the lesion. Large volume cone-beam computed tomography scans revealed an irregularly shaped osseous defect in the left posterior mandible that extended from the ramus anteriorly to the second molar area (Fig. 7). Although the buccal cortical plate was intact, the lingual cortical plate and the alveolar crest showed signs of resorption. There was a furcation defect in tooth #18 and loss of the lamina dura, which likely represented the most anterior extent of the lesion. The lesion extended inferiorly to the level of the inferior alveolar nerve canal, perforating the cortical shelf of that canal (Fig. 8). Infiltration of the inferior alveolar nerve canal explained the patient's reported episodes of sharp pain in conjunction with persistent low-grade pain.

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The patient was informed of the diagnosis and promptly referred to his infectious disease specialist for further evaluation and management. Diagnostic workup confirmed the presence of pulmonary KS, and he was again started on highly active antiretroviral therapy that helped resolve the oral and systemic manifestations of KS throughout his body.

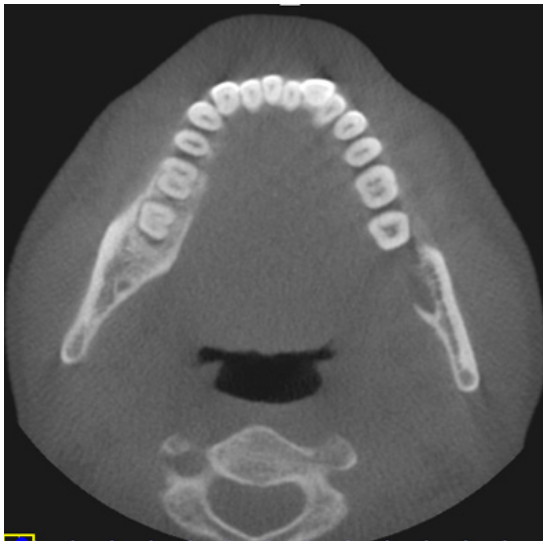

Figure 7. Cone-beam computed tomography scan showing irregularly shaped osseous defect in the left posterior mandible that extended from the ramus anteriorly to the second molar area. squamous (hematoxylin-eosin, magnification ×40).

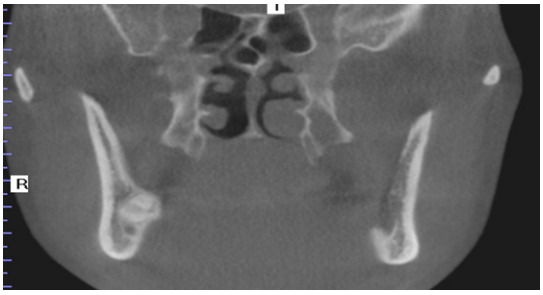

Figure 8. Large volume cone-beam computed tomography scan slices showing an irregular alveolar defect on the lingual cortex of the mandible that extended inferiorly to the level of the inferior alveolar nerve canal.

3. DISCUSSION

Manifestation of oral KS as pericoronitis or in conjunction with pain often serves as a warning sign for any form of injury sustained by the human body. In the current case report, persistent postextraction pain was the main reason the patient pursued follow-up evaluation. Although oral KS can occur at any intraoral site, it has a predilection for the hard palate and gingiva. Cases of oral KS resembling pericoronitis or local soft tissue inflammation with constant low-intensity pain have been rare. For patients living with HIV, unrelieved pain has been reported as a major problem. For example, patients with CD4+ T cell counts less than 200/mm^3 often have pain as a common, persistent symptom [10]. Given the outcomes of the current case, clinicians should consider HIV, AIDS, and their associated manifestations, such as oral KS, as potential sources of pulmonary involvement and was critical for effective and successful treatment of the patient. The disruption of healthcare services during and after the COVID-19 pandemic had a major effect on patient care and increased the number of complications for oral infectious diseases. Several studies reported that COVID-19 accelerated the incidence of KS in extraoral areas, especially the skin of the extremities. This increase was associated with reactivation of the HHV-8 virus, which was likely due to exposure to the SARS-CoV-2 virus. This type of surge in carcinomas was also observed in other oral malignancies that were not caused by HIV [28].

4. CONCLUSION

Manifestations of KS in the oral cavity can be an early sign of additional pathology in immunosuppressed individuals. Therefore, dental practitioners should purposefully investigate the origin of preoperative and postoperative intraoral pain in patients with HIV disease and effectively communicate any findings to medical providers to expedite systemic care of these patients. During the current post-pandemic period, dental practitioners should include an additional layer of screening during standard patient examinations for early detection of oral malignancies, such as oral KS.

AUTHOR CONTRIBUTIONS

PKH, SRS contributed to the concept, protocol, case documetation, data gathering and interpretation and making critical edits to the manuscript. AV, MV contributed to the protocol, case documentation, data gathering and interpretation and making critical edits to the manuscript.

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CONFLICT OF INTEREST

Authors declare that there is no conflict of interests.

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Questions

1. What is the primary cause of Kaposi Sarcoma (KS)?
❑a. Human Papillomavirus (HPV);
❑b. Epstein-Barr Virus (EBV);
❑c. Human Herpesvirus-8 (HHV-8);
❑d. Cytomegalovirus (CMV).

2. What was the initial diagnosis for the 29-year-old male in the case report?
❑a. Kaposi Sarcoma;
❑b. Acute Pericoronitis;
❑c. Osteomyelitis;
❑d. Oral Lichen Planus.

3. What delayed the follow-up care of the patient in the case report?
❑a. Personal negligence;
❑b. Financial constraints;
❑c. The COVID-19 pandemic;
❑d. Lack of transportation.

4. What was a significant clinical finding during the patient's follow-up visit?
❑a. A painless intraoral lesion;
❑b. A bluish-purple proliferative lesion;
❑c. Normal healing of the extraction site;
❑d. Discoloration limited to the hard palate.