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Case Challenge
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Persistent Painful Ulcer of the Posterior Lingual Mandibular Mucosa

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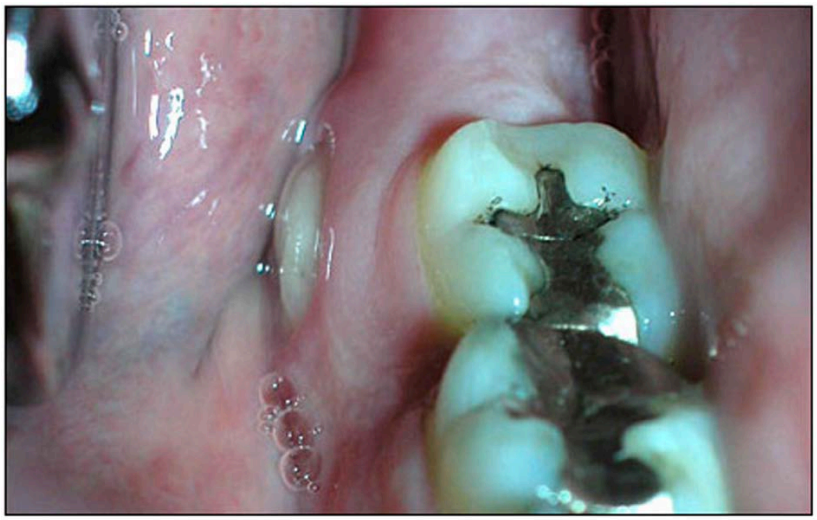


The following Case Challenge is provided in conjunction with the American Academy of Oral and Maxillofacial Pathology.

Case Summary

An ulcer involving the left posterior mandibular lingual mucosa was the chief complaint of this 55-year old white man. The ulcer was first noted about one month previously and no local eliciting factors could be identified. The ulcer had become progressively more painful in spite of antibiotic treatment (cephalexin, 500 mg, q.i.d, 8 days) and concomitant use of benzydamine hydrochloride rinse. The patient was healthy with no medical problems other than occasional nasal "stuffiness," which was treated, as required, with budesonide nasal spray.

After you have finished reviewing the available diagnostic information, make the diagnosis.

Diagnostic Information

Clinical Findings

A well-defined raised ulcer with a gray-white base, measuring about 7 mm by 3 mm, was evident on the left posterior lingual mandibular mucosa above the mylohyoid ridge. (Figure 1) Gentle manipulation of the ulcer base with a periodontal probe revealed the floor of the ulcer was hard, insensitive, and slightly mobile.

The teeth tested vital, and there was no evidence of periodontal involvement. The left second mandibular molar showed mild buccal inclination when compared to the first molar; the second molar also showed a prominent wear facet on the distal aspect of the occlusal surface. Other findings included buccal exostoses that were evident on the left maxillary alveolar bone (Figure 2) and to a lesser extent on the right maxilla (not shown). There was a mild left submandibular lymphadenopathy when compared to the right side.

Radiographic Findings

A periapical film of the left mandibular molars adjacent to the ulcer was not contributory. (Figure 3) An occlusal film showed a localized opacity contiguous to the lingual mandible in the area of the ulcer. (Figure 4)

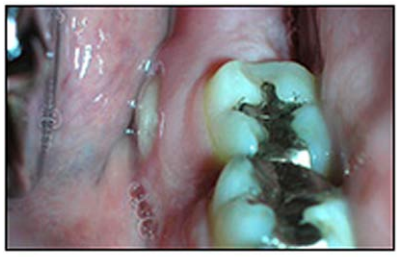


Figure 1. Ulcer with gray-white base involving posterior left lingual mandibular mucosa.

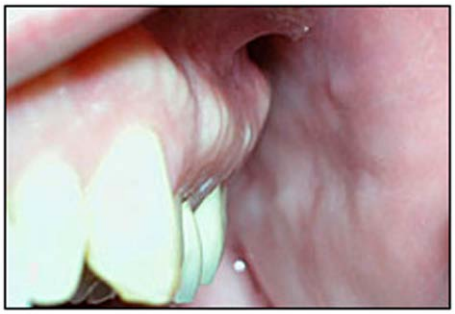


Figure 2. Buccal exostoses involving left maxilla.

Pathological Findings

The edge of the ulcer base was gently explored with a spoon curette. It was possible to find an edge under the hard base that could be engaged with the curette. Using minimal pressure, a hard irregular fragment (Figure 5) was lifted through the ulcer.

Microscopic examination showed this hard fragment was non-vital bone (with irregular zones of resorption involving the deep aspect. (Figure 6) Fragments of acutely inflamed granulation tissue adhered to the bone.

Incisional Biopsy and Microscopic Findings

Due to the significant size of the lesion, an initial incisional biopsy was performed under local

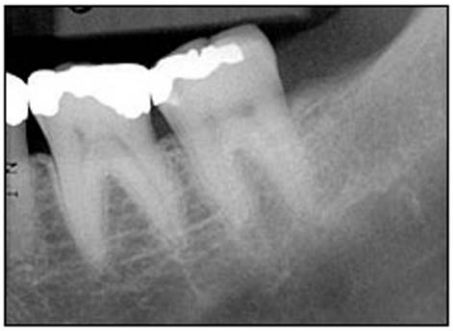


Figure 3. Periapical film of left posterior mandible; no bone anomalies are evident.

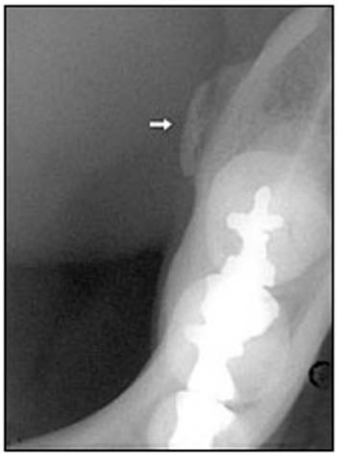


Figure 4. Occlusal film of the same region shown in Figure 3 shows a distinct lingual opacity (arrow).

anesthesia and conscious sedation in order to establish a definitive diagnosis. Unfortunately, the initial biopsy was inconclusive and the patient was referred to Eisenhower Army Medical Center, Ft. Gordon, GA, for an additional biopsy and treatment. During that procedure, abundant gelatinous material was obtained and the specimen was submitted in formalin for routine histopathologic examination. The gross specimen consisted of three fragments of soft tissue, which ranged from 1.5 x 1.5 x 0.4 cm to 0.6 x 0.4 x 0.3 cm.

The low power photomicrograph displayed variably dense fibrocollagenous connective tissue along the edge. The lesion itself dominated the image and was characterized by loose myxoid fibrocollagenous connective tissue that was hypocellular. (Figure 7) The medium power photomicrograph displayed loose and myxoid fibrous connective tissue with some small vascular channels interspersed as well as some extravasated erythrocytes. The nuclei were spindle-shaped to stellate and evenly dispersed throughout the specimen. Although the nuclei were somewhat hyperchromatic, they were uniform in appearance and without evidence of mitotic activity. (Figure 8) The high power photomicrograph showed similar findings. (Figure 9)

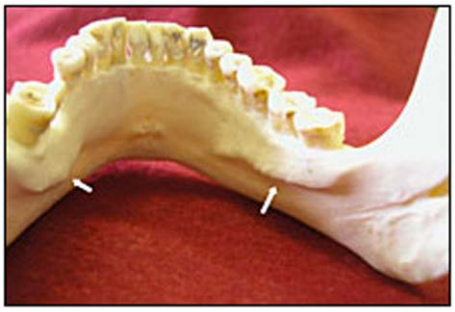


Figure 7. Medial view of the mandible showing prominence of the mylohyoid ridges (arrows).

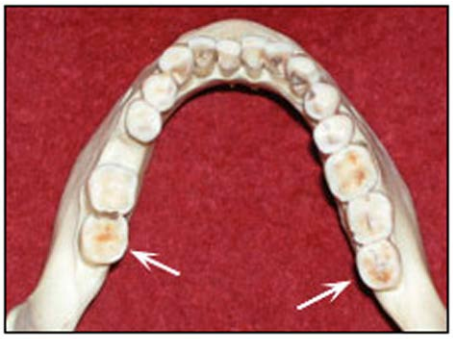


Figure 8. An occlusal view of the same mandible in Figure 7 showing the lingual inclination of the posterior molars (arrows) over the mylohyoid ridges.

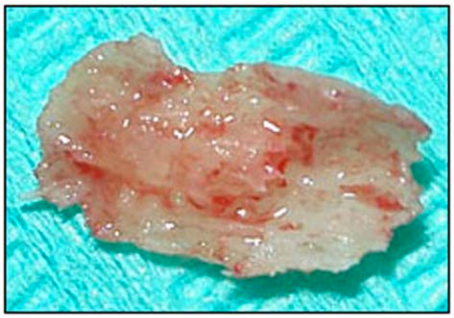


Figure 5. Irregular fragment removed through ulcer floor.

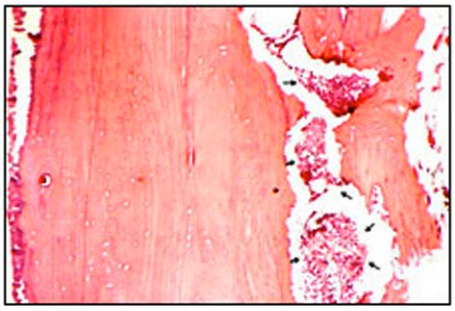


Figure 6. Microscopic view of a cross-section of the fragment after decalcification. There is a non-vital piece of bone showing irregular resorption, granulation tissue (arrows), and acute inflammatory cells.

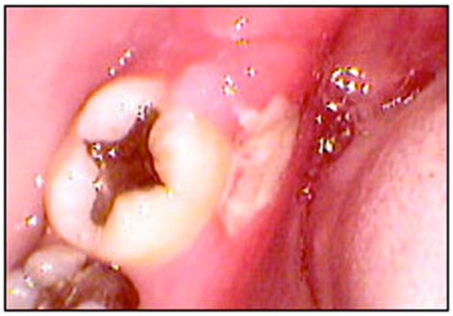
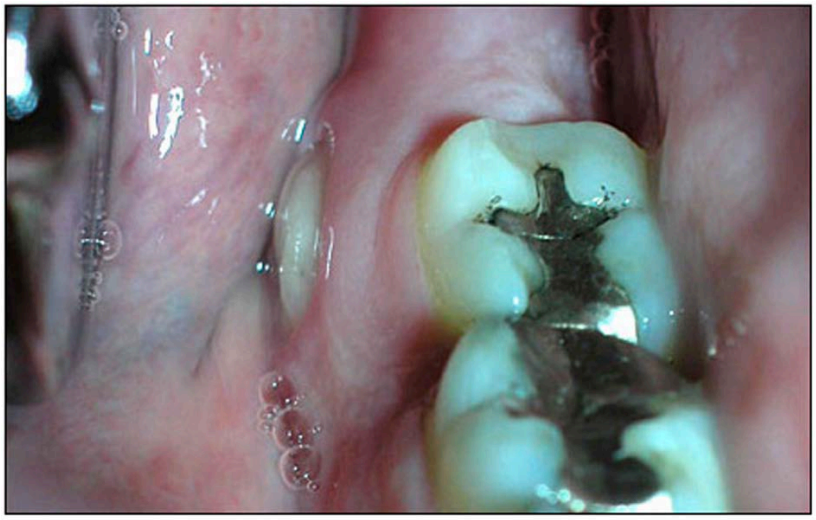


Figure 9. High power photomicrograph showing uniform nuclei within the myxoid stroma. These nuclei are spindle-shaped to stellate and evenly distributed across the field. (Hematoxylin and eosin, original magnification 400x).

Can you make the diagnosis?

An ulcer involving the left posterior mandibular lingual mucosa was the chief complaint of this 55-year old white man. The ulcer was first noted about one month previously and no local eliciting factors could be identified.



Select the Correct Diagnosis

A. Lingual Mandibular Sequestration and Ulceration
B. Acute Osteomyelitis
C. Aphthous Ulcer
D. Squamous Cell Carcinoma

Lingual Mandibular Sequestration and Ulceration

Choice A. Congratulations! You are correct!

Mucosal ulcers represent localized loss of the lining epithelium and possibly the subjacent connective tissue. This could occur in response to direct mechanical, chemical, or thermal trauma. Alternately, mucosal ulceration could occur in a variety of etiologically distinctive conditions that include malignancy, infarctions, infections, immune-mediated diseases, or as a feature of systemic diseases involving blood, skin, or the gastrointestinal tract. Diagnosis requires a careful history and identification of the presenting clinical stigmata. Sometimes, laboratory tests including biopsy are necessary. However, it is important to be aware of the range of clinical possibilities to permit the most appropriate interpretation.

The form of ulceration shown in the current case does not appear to be uncommon, but it has been discussed in only a limited number of reports.[1,2] The ulcer has been described in adults only, typically without predisposing systemic disease. Patients complain of symptoms, which range from local sensitivity and perception of a projection irritating the tongue to significant pain, possibly radiating as far as the ipsilateral ear.

The ulcer develops in the posterior lingual mandibular mucosa at the level of the mylohyoid ridge. It has a hard insensitive base that represents a superficial sequestrum.

Resolution does not occur until the sequestrum spontaneously exfoliates or is removed. This occurs over highly variable periods, which can range from a week to months.

Treatment usually involves conservative removal of the sequestrum and subsequent attempts to minimize sepsis in the healing area. A 0.12% chlorhexidine gluconate oral rinse is a useful supportive measure to minimize local sepsis during the healing phase; antibiotics might be indicated if there are concerns of infection. Occasionally, there are recurrences after initial healing.

The pathogenesis of such lesions is not well understood; however, the ulcer is thought to be the initial pathologic event with sequestration occurring as a result of the associated inflammation and secondary local infection. The sequestrum then maintains the ulcer.[1,2]

A variety of possible precipitating factors have been suggested. Of possible significance, many of these cases occur in patients who have lost posterior molars or have restorations that do not recapitulate the normal lingual inclination of the molars.[1] The relevance of the latter observation is illustrated in Figures 7 and 8. Figure 7 shows the prominence of the mylohyoid ridge relative to the superior lingual bone; while Figure 8 shows the protective lingual inclination of the molars over the mylohyoid ridge. If this anatomic relationship were altered, the mucosa over the mylohyoid ridge would not be shielded from repeated trauma during mastication. In the current case, the molar was developmentally positioned in a more buccal location that could have resulted in a similar exposure to masticatory trauma.

After the ulcer has formed, it would be susceptible to secondary infection because it is located in a relatively stagnant oral region. The subjacent bone could sequestrate in response to persistent irritation, infection, and inflammation. The sequestrum would delay healing and exacerbate the presentation. In this regard, it has been suggested that an aphthous ulcer could be the primary lesion.[2] Subsequently, the same pathologic sequence of events could occur. In our case, there was no history of aphthous stomatitis, but this could represent another initiating mechanism in some cases.

Two other predisposing factors might be implicated. The first is the blood supply to the superficial lingual cortical mandibular bone is thought to be relatively poor and could be significantly disrupted, if the periosteal layer, carrying blood vessels to cortex, becomes necrotic.[1,3-5] The second possible factor is suggested by the observation that many affected patients also have mandibular tori.[1]

Mandibular tori are thought to represent localized periosteal bone formation in response to multifactorial environmental and genetic factors.[6,7] Affected individuals might exhibit a more labile periosteal response to chronic

mucositis caused by irritation. The newly formed bone would protrude even further beyond the normal anatomic contour of the mylohyoid ridge, increasing the potential for further irritation and subsequent sequestration. The patient in this case did not show mandibular tori, but he did exhibit the less common buccal exostoses. In this regard, although the mylohyoid region appears particularly vulnerable to sequestration-associated ulceration, similar lesions have been reported occurring directly over exostoses in various locations.[1,8] Presumably, the contributing factors in these cases would include the relatively exposed and thinner mucosa over the exostosis and poor vascularity in the sclerotic bone that is often found in tori.[1]

Although this condition typically occurs in the absence of predisposing systemic factors, it is important to remember that because the mylohyoid area appears vulnerable to sequestration, it could also present as an early manifestation in patients who do have some form of systemic compromise.

Figure 9 shows a right posterior mandibular lingual ulcer, which occurred in a 41-year old, apparently healthy man. The ulcer developed following dental scaling and persisted for one month even after antibiotics and chlorhexidine oral rinses were prescribed. The presentation differed from the first case in that the ulcer extended to include the marginal gingiva. In addition, the gingiva was moderately hyperplastic at the superior posterior aspect. An occlusal

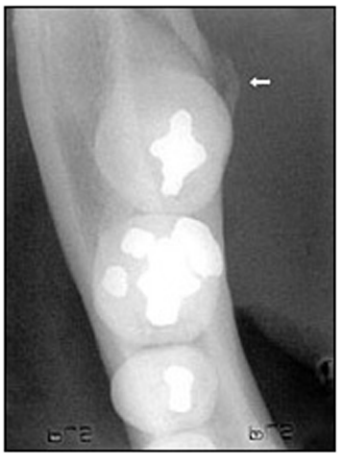


Figure 10. Occlusal film of the ulcerated area in Figure 9 showing sequestrum (arrow).

film (Figure 10) showed a sequestrum similar to the first case. Removal of the sequestrum in combination with a prescription of metronidazole and 0.12% chlorhexidine oral rinse resulted in rapid resolution of the ulcer. However, in view of the unusual presentation, the possibility of systemic disease was suspected and subsequent follow-up showed an acute leukemia.

In summary, ulceration of posterior lingual mandibular mucosa should prompt consideration that the ulcer is being maintained by an underlying sequestrum. This usually occurs in healthy individuals and conservative management is indicated as previously discussed. However, a predisposing debilitative systemic disease could be implicated in rare cases.

Acute Osteomyelitis

Choice B. Sorry, this is not the correct diagnosis.

The case does exhibit sequestration that is a feature of acute osteomyelitis, and it would be possible to argue the case could be characterized as a localized form of acute osteomyelitis restricted to superficial cortical bone. However, the diagnosis of acute osteomyelitis indicates a more aggressive inflammatory and infective process extending to include medullary bone and usually includes systemic stigmata such as fever, leukocytosis, and lymphadenopathy.[9] Treatment routinely includes antibiotic therapy and establishment of drainage. The current case illustrates a relatively indolent localized ulcerative process, which has a predilection for a specific site and without obvious local or systemic initiating factors.

Please re-evaluate the information about this case.

Aphthous Ulcer

Choice C. Sorry, this is not the correct diagnosis.

Aphthous ulcers probably represent focal epithelial necrosis caused by a cell mediated immune response, although the nature of the triggering stimulus is not well understood.[9] They can occur in a variety of contexts. These include stress, micronutrient deficiencies (B12, folate, and iron), food allergies (most commonly nuts, chocolate, or gluten), hormonal alterations, gastrointestinal problems (celiac disease, Crohn's disease) and, occasionally, blood dyscrasias such as neutropenia. Minor, major, and herpetiform varieties have been defined.

Minor aphthous ulcers are acutely painful and patients are well aware of their presence after development. The ulcers present on non-keratinized (lining) mucosa and resolve in about 7-14 days. This profile does not match the current case. However, it is possible to argue that a sequestrum developed in subjacent bone as a complication of an aphthous ulcer.[2] The sequestrum would prevent healing within the usualinterval.

A major aphthous ulcer is larger and lasts longer then the minor lesion. Multiple recurrent lesions can develop on any mucosal surface, although they usually occur on labial mucosa, soft palate, and tonsillar fauces. Herpetiform ulcers present as clusters of small lesions and are completely dissimilar to the current case. Both of these ulcer types are dissimilar to the current case.

Please re-evaluate the information about this case.

Squamous Cell Carcinoma

Choice D. Sorry, this is not the correct diagnosis.

Serious consideration should be given to this diagnosis any time there is a non-resolving ulcer.[9] In addition to presentation as an ulcer, oral mucosal squamous cell carcinomas can present as white patches (leukoplakia), red patches (erythroplakia), masses, or with combinations of these features. However, in this case, there was an apparent explanation for the ulcer (sequestration) in a patient without obvious carcinogenic factors such as tobacco use. In addition, the ulcer was located in a site that was not at high risk for the development of squamous cell carcinoma. Also important was the radiographic findings that did not indicate an invasive process. With this clinical information, the diagnosis of carcinoma would be unlikely.

Please re-evaluate the information about this case.

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About the Authors

Note: Bio information was provided at the time the case challenge was developed.

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