

Case Report

Oral cyst-like nodule caused by a long standing unusual embedded foreign object: Report of a case

Shokoufeh Shahrabi-Farahani^{a,*}, Saede Atarbashi-Moghadam^{b,1}, Ali Lotfi^c, Fazele Atarbashi-Moghadam^d^a Department of Diagnostic Sciences, University of Tennessee Health Science Center, College of Dentistry, TN, USA^b Department of Oral and Maxillofacial Pathology, School of Dentistry, Shahid Beheshti University of Medical Sciences, Tehran, Iran^c Oral and Maxillofacial Pathologist, Private Practice, Iran^d Department of Periodontics, School of Dentistry, Shahid Beheshti University of Medical Sciences, Tehran, Iran

ARTICLE INFO

Keywords:

Oral mucosa
Foreign material
Cyst-like, shrapnel
Synovial metaplasia

ABSTRACT

Traumatic implantation of foreign material in the oral mucosa is unusual and may occur. In this article, we report the presence of an unusual foreign object associated with a cyst-like inflammatory response in a 50-year-old male. The patient presented with a painless recurrent soft tissue swelling in the anterior vestibule of maxilla of at least 2-year duration. The lesion was surgically excised and a retained shrapnel-like object within a cyst-like and thick-walled structure was obtained. Histopathologically, the cyst-like structure exhibited a lymphohistiocytic infiltrate in the wall and a luminal synovial metaplasia-like change, predominantly composed of histiocytes. Immunohistochemical stain of CD163 highlighted the histiocytes. The patient stated a history of traumatic implantation of shrapnel fragments in the area that occurred 30 years ago due to an explosion in the Iran-Iraq war.

Introduction

Presence of foreign bodies in oral mucosal biopsies is unusual and the implantation process may occur either traumatically or iatrogenically. Any site of the oral cavity may be affected and dental restorative material such as amalgam, dental hygiene products, impression material, and cosmetic dermal fillers are accounted as the common sources of the foreign material [1–3]. In this article, a case of unusual foreign body lodged in the oral cavity is reported and the clinicopathological features is discussed.

Case report

A 50-year-old male presented with a painless and well-defined nodule covered by intact mucosa in the anterior maxillary vestibule, midline region; extending to the upper lip mucosa of at least 2-year duration (Fig. 1). The patient stated some episodes of rupture and recurrence of the lesion with discharge of some fluid. His past medical history was insignificant. A tooth-related inflammatory process, nasolabial cyst, and a benign salivary gland-related process such as salivary duct cyst and cystadenoma were considered as differential diagnosis. Through intra-oral examination, maxillary central incisors were found vital using vitality test and no tooth caries or filling was noted. Radiographically, a well-defined, and metal-like radiopaque object in the anterior maxilla was

* Corresponding author.

E-mail address: sfarahan@uthsc.edu (S. Shahrabi-Farahani).¹ These authors contributed equally to this work.



Fig. 1. The patient presents with a well-defined nodule covered by intact mucosa in the anterior maxillary vestibule; extending to the upper lip mucosa.

observed (Fig. 2). Therefore, a working diagnosis of foreign body reaction was made. Under local anesthesia, a muco-periosteal flap was performed and after reflection of the flap, the lesion was found completely in the soft tissue and the underlying bone was intact without involvement. The lesion appeared as a cyst-like structure grossly, containing amber-colored fluid. After drainage the fluid, the nodule was completely excised. The specimen was placed in 10% buffered formalin solution and submitted for histopathologic studies. Grossly, the specimen demonstrated partially ruptured, cyst-like features, with a thick wall measuring 1.2 cm. A dark silver-gray metal object measuring 0.5 cm with corroded surface was retrieved from the specimen (Fig. 3a).

Histopathologically, a cyst-like structure with an inflamed thick fibrous wall was noted. No epithelial lining was evident, and the luminal surface of the specimen exhibited synovial metaplasia-like change predominantly composed of eosinophilic epithelioid cells with distinct cytoplasmic border intermixed with fibroblast-like cells. A patchy lymphohistiocytic infiltrate and hemorrhage were also seen throughout the fibrous wall (Figs. 3b and 3c). Immunohistochemically, CD163 highlighted the histiocytes (Fig. 3d). Using polarized light, scattered refractile particulate foreign material was found both in the cystic space and in the fibrous wall. Collectively, a diagnosis of foreign material and associated cyst-like foreign body granulomatous reaction was made. Considering the patient's history of an injury caused by the explosion took place in the Iran-Iraq war in 30 years ago and history of some surgical attempts for removing some shrapnel from his body, the source of that foreign object was suggested. Post-operatively, in a 4-year clinical follow up, the patient showed no evidence of infection and recurrence.

Discussion

In the oral cavity, dental material such as amalgam, broken instruments, and impression material, and dental hygiene products are considered as the common sources for foreign body-related mucosal lesions. Cosmetic dermal fillers have also been focused recently in the literature and may cause mucosal changes through intra-oral migration [1–4]. Oral lesions caused by retained shrapnel or bullets are not commonly encountered in the oral practice and to our knowledge, the current case would be the first case report in the English literature. Some extra-oral examples have been previously reported in the parotid gland, and soft tissue or bones of the limbs and legs [5–7].

Clinical presentation of the oral mucosa with embedded foreign material depends on the nature of the foreign particle and local



Fig. 2. Panoradiograph showing a well-defined, and metal-like radiopaque object in the anterior maxillary region.

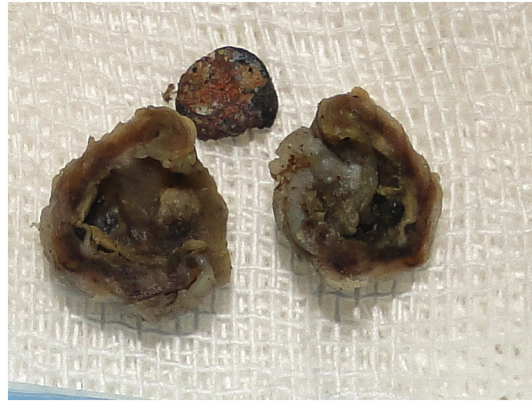


Fig. 3a. Gross image showing a cystic-like structure with the retrieved dark silver-gray shrapnel-like object.

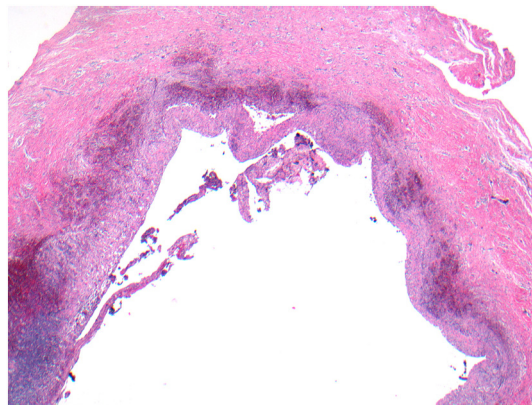


Fig. 3b. Photomicrograph showing a cystic space surrounded by an inflamed and fibrous connective tissue with hemorrhage (H & E, $\times 20$).

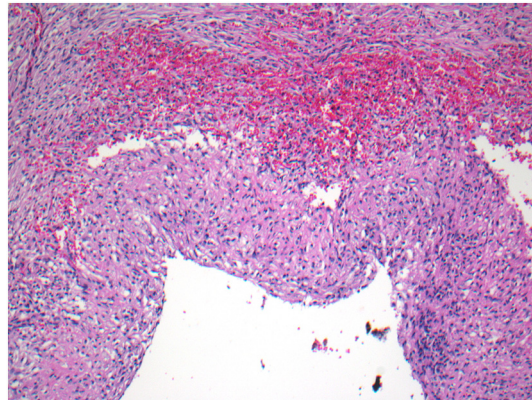


Fig. 3c. Photomicrograph showing a chronically inflamed fibrous wall showing luminal synovial-like lining (H & E, $\times 100$).

tissue response [8] and may be varied from a mucosal discoloration as seen in amalgam tattooing to a white and red lesion with or without lichenoid pattern likely seen in foreign body gingivitis, and also a nodule or tumor-like formation. Pain may be present or absent [1–4]. Our case clinically presented with a semi-fluctuant and cyst-like nodule in the anterior maxillary vestibule. The clinical differential diagnosis of such appearance would be inflammatory tooth-related lesions such as radicular cyst/abscess, developmental lesions such as nasolabial cyst, and salivary gland-related processes such as mucous retention cyst, and benign salivary gland tumors with predominantly cystic component such as cystadenoma. However, a benign soft tissue tumor like angioleiomyoma cannot be completely excluded. Radiographic examination and dental examination are essential to narrow the possibilities, as it was performed in our case and the diagnosis of foreign body-associated lesion was suggested.

Histopathologically, the most likely type of body reaction to foreign material would be granulomatous inflammation and

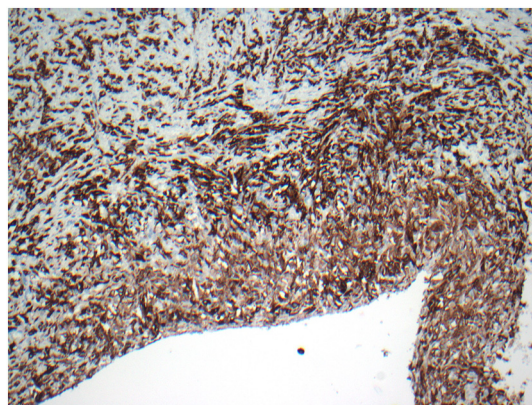


Fig. 3d. Photomicrograph showing CD163 immunoreactivity of the histiocytes in the luminal surface and the fibrous wall (immunohistochemistry, $\times 100$).

formation of multi-nucleated foreign body giant cells [8] as noted in response to some cosmetic dermal fillers [3]. In the oral mucosa other superimposed histopathologic features may be encountered. Foreign material embedded in the gingiva may induce a lichenoid pattern of inflammation as occurred in some cases of foreign body gingivitis [2] similar to the histopathologic features seen in amalgam-induced hypersensitivity reaction [1].

Soft tissue fluid-filled pseudocyst formation has been described in association with retained bullets in 4 cases by Ragsdale B et al. [7]. Two of which showed features of synovial-like surface lining [7]. Normal synovium is composed at least two types of cells including macrophages-like cells and fibroblast-like cells [9]. Formation of synovial-like tissue was initially described in the subcutaneous tissues as inflammatory reactions in response to injected foreign materials [10]. Synovial-like changes are most frequently found in the tissues surrounding silicone breast prostheses and may develop in oral mucocles in response to gliding trauma [11,12]. Therefore, the smooth gliding surface of some foreign material such as retained shrapnel in our case would elicit a foreign body reaction as a cyst-like structure and the luminal wall of the cyst exhibits synovial metaplasia-like change; this must be confirmed by histopathologic studies to exclude malignant growths or other inclusion bodies.

Surgical excision of the retained foreign material and inflamed surrounding tissue is the definite treatment and the recurrence is uncommon.

Conclusion

Retained shrapnel in the oral mucosa is rare and may cause a cyst-like nodule clinically which resembles to the other more common entities. Radiographic examination and patient's history are needed for diagnosis. Histopathologically, such foreign objects may induce a cyst-like tissue formation exhibiting luminal synovial metaplasia-like change.

Conflict of interest

None.

References

- [1] A. Buchner, L.S. Hansen, Amalgam pigmentation (amalgam tattoo) of the oral mucosa. A clinicopathologic study of 268 cases, *Oral Surg Oral Med Oral Pathol* 49 (1980) 139–147.
- [2] S.C. Gordon, T.D. Daley, Foreign body gingivitis: clinical and microscopic features of 61 cases, *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 83 (1997) 562–570.
- [3] S. Shahrabi-Farahani, M.A. Lerman, V. Noonan, S. Kabani, S.B. Woo, Granulomatous foreign body reaction to dermal cosmetic fillers with intraoral migration, *Oral Surg. Oral Med. Oral Pathol. Oral Radiol.* 117 (2014) 105–110.
- [4] D. Puliyl, A. Balouch, S. Ram, P.P. Sedghizadeh, Foreign body in the oral cavity mimicking a benign connective tissue tumor, *Case Rep Dent.* 2013 (2013) 369510.
- [5] B.G. Brogdon, W.C. Cottrell, P. Nimityongskul, D. Takhtani, A bullet-sired bone cyst, *Skelet. Radiol.* 35 (2006) 959–963.
- [6] J.L. Cembranos, J.A. Bua, J.A. Amezcaga, J.S. Zuazua, An unusual intraparotid foreign body simulating a tumour: shrapnel from the Spanish Civil War, *Int. J. Oral Maxillofac. Surg.* 40 (2011) 880–883.
- [7] B.D. Ragsdale, L.M. Lehmer, Soft tissue pseudocysts associated with retained bullets: a series of 4 cases, *Journal of Orthopedics and Trauma Research* (2018) 1.
- [8] . Kumar V, MBBS, MD, FRCPath; Abbas, Abul K., MBBS; Aster, Jon C. Robbins and Cotran Pathologic Basis of Disease, Ninth Edition. Philadelphia: Saunders; 2015:74.
- [9] M.R. Fowler, C.O. Nathan, F. Abreo, Synovial metaplasia, a specialized form of repair, *Arch Pathol Lab Med* 126 (2002) 727–730.
- [10] H. Selye, On the mechanism through which hydrocortisone affects the resistance of tissues to injury; an experimental study with the granuloma pouch technique, *J. Am. Med. Assoc.* 152 (1953) 1207–1213.
- [11] A.C. Chi, R.J. Haigney, D.B. Spagnoli, B.W. Neville, M.S. Richardson, Papillary synovial metaplasia-like change in oral mucocles: a rare and previously undescribed histopathologic variant of a common oral lesion, *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 109 (2010) 268–273.
- [12] M.R. Hameed, R. Erlandson, P.P. Rosen, Capsular synovial-like hyperplasia around mammary implants similar to detritic synovitis. A morphologic and immunohistochemical study of 15 cases, *Am. J. Surg. Pathol.* 19 (1995) 433–438.