CASE REPORT

A huge oral pyogenic granuloma with extensive alveolar bone loss and 'sun-ray' appearance mimicking a malignant tumour

Smitha Rani Thada,¹ Keerthilatha M Pai,¹ Pankaj Agarwal²

¹Department of Oral Medicine and Radiology, Manipal College of Dental Sciences, Manipal, Karnataka, India

²Department of Oral Pathology, KD Dental College, Mathura, Uttar Pradesh, India

Correspondence to

Dr Smitha Rani Thada, smitha.rani@rediffmail.com

Accepted 5 May 2014

SUMMARY

Pyogenic granuloma represents an exuberant connective tissue proliferation due to a chronic stimulus or injury. Aetiological factor for this lesion may sometimes be hard to identify, but the fact that it is usually located close to the gingival margin suggests that calculus, food materials and overhanging restorations are important irritants. It rarely grows more than 2 cm in diameter and usually does not cause any changes in the alveolar bone. We report an unusual case of pyogenic granuloma with unknown aetiology that presented as an extraordinarily large sessile bilobular lesion and showed a large radiographic defect in the alveolar bone with a sun-ray pattern. This kind of clinicoradiological appearance of pyogenic granuloma as presented in our case is rare and the first of its kind.

BACKGROUND

Pyogenic granuloma, a kind of inflammatory hyperplasia, arises in response to various stimuli such as low-grade local irritation, traumatic injury or hormonal factors.¹ It is usually small (<2 cm) in size and rarely presents with minimal bone changes, but here we report an unusual case of pyogenic granuloma presenting as an exceptionally large bilobular sessile growth of unknown origin. Radiographic picture was also unique as it presented with severe bone resorption resulting in loss of superior border of mandibular canal and also presenting with 'sun-ray' kind of periosteal reaction. The clinical features of this case were exceptionally different to fit into the histopathological diagnosis of pyogenic granuloma. Hence, we consider the need to highlight this unusual presentation of pyogenic granuloma.

CASE PRESENTATION

A woman in her early 20s presented to us with a swelling of 2 months duration on the right lower jaw. She gave a history of extraction of right lower first molar a year ago and after 8 months a small painless growth appeared at the extraction site. She underwent excision of the growth under general anaesthesia (histopathological reports were not available). Two months after surgery the growth reappeared at the same site and it gradually increased to attain the present size, causing interference with her speech and mastication. She had no associated symptoms of pain, bleeding, pus discharge, mobility of teeth or difficulty in mouth opening. No history of fever, paraesthesia and weight loss was reported. Her main concern was

aesthetic disfigurement due to facial swelling. Her medical and family histories were unremarkable and she was not on any medication.

On examination she had an obvious facial asymmetry with a diffuse, non-tender, firm swelling about 3×4 cm in size on the right lower third of the face (figure 1). A single right submandibular lymph node about 0.5 cm in diameter was palpable, which was non-tender, firm and mobile. Her oral hygiene status was satisfactory with mild gingival inflammation. All teeth were present except 46. A dumbbell-shaped, smooth-surfaced exophytic sessile growth was seen extending buccally and lingually. Buccal swelling (5×4 cm) extended from 45 to 47 and lingual swelling (5×5 cm) extended from 42 to 47. They were connected and fixed at the extraction site of 46 (figure 2). Some areas showed increased keratinisation, with upper teeth indentations but no surface ulcerations. The growth was soft to firm in consistency, non-tender, non-fluctuant, non-compressible and the overlying mucosa was smooth and shiny. The distance between 45 and 47 appeared to be increased due to the pressure effect from the growth. All teeth were vital. Deviation of the tongue to the left was noticed due to the extension of growth into the floor of mouth. Swelling could be lifted up from the floor of mouth. No pulsations or bruit were felt.

INVESTIGATIONS

Panoramic radiograph showed a well-demarcated radiolucency (2.5 cm in diameter) corresponding to

Figure 1 Diffuse swelling on the right side of the mandibular region. A diffuse swelling about 3×4 cm in size extending superoinferiorly from right middle third of the face to 1 cm below the lower border of the mandible, mediolaterally from the right corner of the mouth to 2 cm medial to the mandibular angle. There was no obliteration of the nasolabial fold. Overlying skin showed no abnormalities.

CrossMark

To cite: Thada SR, Pai KM, Agarwal P. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2013-202367

Figure 2 Sessile bilobular swelling. A dumbbell-shaped exophytic sessile growth was seen extending buccally and lingually. Buccal swelling (5×4 cm) extended from 45 to 47 and lingual swelling (5×5 cm) extended from 42 to 47. They were connected and fixed at extraction site of 46. Mucosa over the swelling was smooth and shiny with no ulcerations. At some areas there was increased keratinisation, with upper teeth indentations. The distance between 45 and 47 appeared to be increased. Tongue was deviated to the left due to the extension of growth into the floor of mouth.

the area between 45 and 47 with evidence of remnants of bone and scalloping of cortical plate. Superior border of mandibular canal could not be appreciated in the region of 46, 47 and 48. Distance between 45 and 47 appeared to be increased (figure 3). Lower occlusal radiograph showed 'sun-ray' appearance of the bone with small bony trabecular projections on the buccal aspect of cortical plate in the region of 46 and 47 (figure 4). The haemogram of the patient was within normal limits.

DIFFERENTIAL DIAGNOSIS

On the basis of the history and clinical presentation, the lesion was suspected to be a benign slow-growing neoplasm of mesenchymal origin. Since fibroma is the most common benign soft tissue tumour of the oral cavity, it was considered to be the most likely provisional diagnosis. Other clinical differential diagnoses that were considered included—peripheral giant cell granuloma, giant-cell fibroma, granular cell tumour, schwannoma/neurilemmoma, neurofibroma, rhabdomyoma and leiomyoma.

However, all of the aforementioned lesions are usually less than 2 cm in size and none of them show bone involvement

Figure 3 Panoramic radiograph—well-demarcated radiolucency seen corresponding to the area between 45 and 47 with evidence of remnants of bone and scalloping of cortical plate. Superior border of mandibular canal could not be appreciated in the region of 46, 47 and 48. Distance between 45 and 47 appeared to be increased.

Figure 4 Lower occlusal—sun-ray appearance. Lower occlusal radiograph showed 'sun-ray' appearance of the bone due to small bony trabecular projections seen on the buccal aspect of cortical plate in the region of 46, 47.

except for 'cupping' resorption of the alveolar bone that may occur in peripheral giant cell granuloma. The radiographic presentation in our case showed severe bone loss at the lesional site, with loss of superior border of alveolar canal and 'sun-ray' type of periosteal reaction similar to osteosarcoma, chondrosarcoma and/or haemangioma. The slow-growing nature and clinical presentation were not in favour of these malignant/vascular bony pathologies. Hence this case posed a great diagnostic challenge and we further proceeded for histopathological evaluation to establish the diagnosis.

FINAL DIAGNOSIS AND TREATMENT

The growth was excised under general anaesthesia. Histopathological examination showed the presence of parakeratinised stratified squamous epithelium which was ulcerated at places. Connective tissue adjacent to the ulcerated areas showed mixed inflammatory response mainly in the form of lymphocytes, plasma cells and few neutrophils. There were endothelial cell-lined small vascular channels which were engorged with red blood cells. Connective tissue showed areas of oedema with focal aggregation of inflammatory cells. There was no evidence of tumour cells in these sections. All these features were suggestive of 'pyogenic granuloma' (figure 5) which turned out to be a benign reactive process in spite of the masquerading clinical and radiological presentation.

OUTCOME AND FOLLOW-UP

The patient underwent complete surgical excision of the growth under general anaesthesia. The patient was followed up for 1 year and had no recurrence of the lesion (figures 6 and 7).

DISCUSSION

Oral pyogenic granuloma (synonyms—eruptive haemangioma, granulation tissue-type haemangioma, granuloma gravidarum, lobular capillary haemangioma and pregnancy tumour) is a non-neoplastic inflammatory hyperplasia originally thought to be caused by pyogenic organisms, but it is now believed to be unrelated to infection. Hence, it is a misnomer because the lesion does not contain pus and strictly speaking is not a granuloma. Its incidence has been described to be between 26.8% and 32% of all the reactive lesions.² It is a disorder of angiogenesis with various aetiological factors such as chronic low-grade local irritation from overhanging restorations or calculus, trauma, hormonal changes, certain drugs, bone marrow transplant and

Figure 5 Histopathological appearance. Parakeratinised stratified squamous epithelium was seen which was ulcerated at places. Connective tissue adjacent to ulcerated areas showed mixed inflammatory response mainly in the form of lymphocytes, plasma cells and few neutrophils. There are endothelial cell-lined small vascular channels which were engorged with red blood cells. Connective tissue showed areas of oedema with focal aggregation of inflammatory cells.

reactions to grafts.³ However, aetiology remains unknown even in our case. Oral pyogenic granuloma occurs over a wide age range of 4.5–93 years with highest incidence in the second and fifth decades.³ Women are more commonly affected due to the increased levels of circulating hormones, oestrogen and progesterone.⁴

Occurrence of pyogenic granuloma in man was first described in 1897 by Poncet and Dor.⁵ At that time, it was called botryomycosis hominis. Pyogenic granuloma has been referred to by a variety of other names such as granuloma pediculatum benignum, benign vascular tumour, pregnancy tumour, vascular epulis, Crocker and Hartzell's disease.³ Hullihen⁶ in 1844

Figure 6 Follow-up clinical photograph of the surgical site. Complete healing of the surgical site is evident.

Figure 7 Follow-up radiographic photograph of the surgical site. Evidence of bone formation seen on intraoral periapical radiograph at the site of the previous bony defect.

described this lesion but the term 'granuloma pyogenicum' was introduced by Crocker in 1903.⁷ However, some researchers believe that Hartzell⁸ in 1904 introduced the term 'pyogenic granuloma' that is widely used in the literature, although, it does not express accurately the clinical or histopathological features. There are two types of pyogenic granuloma: (1) lobular capillary haemangioma (LCH) (66%)—sessile form and (2) non-LCH (77%)—pedunculated form. The most common intraoral site is the interdental gingiva (75%) of maxillary anterior region, but it also affects the lips, mucosa and tongue.⁹ It is usually a painless small, smooth, lobulated, deep red to reddish-purple lesion which is either sessile or pedunculated. It may range in size from a few millimetres to centimetres. It rarely exceeds more than 2–2.5 cm. The surface may be ulcerated and shows a tendency for haemorrhage either spontaneously or on slight trauma. Our case presented with a pink large bilobular exophytic sessile growth in the edentulous ridge with each lobe measuring around 5 cm in diameter and firm in consistency, and bleeding tendency was surprisingly absent.

Pyogenic granuloma usually presents as an exophytic lesion without any bone involvement, however, this need not be always true.^{10–13} In our case, all the clinical and radiographic features presented by the patient did not fit into the classic presentation of pyogenic granuloma. A thorough literature search was performed and to the best of our knowledge this is the first reported case of pyogenic granuloma causing alveolar bone resorption to such a huge extent with appearance of 'sun-ray' pattern of the bone on the radiograph.

Pyogenic granuloma is treated by conservative surgical excision with the removal of potential traumatic factors. Careful monitoring during surgery is important because of its vascular nature leading to profuse bleeding. Recurrence occurs in approximately 16% of lesions excised, with gingival cases showing a much higher recurrence rate.^{1,3} Therefore, pyogenic granuloma of the gingiva, that is, epulis granulomatosa, should be excised and the surgical wound bed should be curetted and adjacent teeth should undergo scaling and root planing. If at all possible, removal in a pregnant woman should be postponed until after the birth. Lesional shrinkage postdelivery may make surgery unnecessary.¹⁴ Other conventional surgical modalities reported for the treatment of pyogenic granuloma are cryosurgery in the form of either liquid nitrogen spray or a cryoprobe and laser surgery using Nd: YAG (neodymium: yttrium-aluminum-garnet), CO and flash lamp pulsed dye lasers.¹⁵

Learning points

- ▶ Simple lesions such as pyogenic granuloma can also present with a masquerading clinical and radiological presentation.
- ▶ Pyogenic granuloma can present as a very small lesion <2 cm to a very huge lesion >5 cm in diameter (as in our case).
- ▶ Alveolar bone changes can be seen in huge lesions but 'sun-ray' kind of periosteal reaction as seen in our case is a rare radiographic presentation that has to be taken into consideration.
- ▶ Hence, pyogenic granuloma should be included in the differential diagnoses of even large smooth-surfaced, non-bleeding, soft tissue growth arising from the alveolar ridge causing considerable bone destruction.

Acknowledgements The authors would like to thank Dr Vineetha R, Associate Professor, Department of Oral Medicine and Radiology, Manipal College of Dental Sciences, Manipal.

Contributors SRT and KMP were involved in patient care and writing the manuscript. PA conducted histopathological diagnosis of the patient.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Chawla K, Lamba AK, Faraz F, *et al.* Treatment of pyogenic granuloma using Er, Cr:YSGG Laser. *Int J Laser Dent* 2013;3:29–32.
- 2 Kamala KA, Ashok L, Sujatha GP. Pyogenic granuloma on the upper labial mucosa: a case report. *J Clin Diagn Res* 2013;7:1244–6.
- 3 Reet K, Parveen D, Abhiney P. Oral pyogenic granuloma: various concepts of etiopathogenesis. *J Oral Maxillofac Pathol* 2012;16:79–82.
- 4 Sheiba RG, Qaid Johar S, Prarthana VT, *et al.* Tavadia. Pyogenic granuloma of the gingiva: a misnomer?—a case report and review of literature. *J Indian Soc Periodontol* 2013;17:514–19.
- 5 Poncet A, Dor L. De La botryomycose humaine : Identité' de nature de tumeurs d'apparence papillomateuse chez l'homme avec la botryomycose ou champignon de castration du cheval. *Lyon Med* 1897;86:213.
- 6 Hüllihen SP. Case of aneurism by anastomosis of the superior maxillae. *Am J Dent Sci* 1844;4:160–2.
- 7 Crocker HR. *Diseases of skin*. Vol 2. Philadelphia: P Blakiston's Son & Co., 1903:1081.
- 8 Hartzell MB. Granuloma pyogenicum (botryomycosis of French authors). *J Cutan Dis* 1904;22:520–5.
- 9 Regu P, Aatman S, Murali GV, *et al.* Pyogenic granuloma of the tongue—a rare clinical finding. *Int J Dent Case Rep* 2013;3:57–61.
- 10 Martins-Filho PRS, Piva MR, Da Silva LCF, *et al.* Aggressive pregnancy tumor (pyogenic granuloma) with extensive alveolar bone loss mimicking a malignant tumor: case report and review of literature. *Int J Morphol* 2011;29:164–7.
- 11 Singh VP, Nayak DG, Upoor AS. Pyogenic granuloma associated with bone loss: a case report. *J Nepal Dent Assoc* 2009;10:137–9.
- 12 Shenoy SS, Dinkar AD. Pyogenic granuloma associated with bone loss in an eight year old child: a case report. *J Indian Soc Pedod Prev Dent* 2006;24:201–3.
- 13 Lawoyin JO, Arotiba JT, Dosumu OO. Oral pyogenic granuloma: a review of 38 cases from Ibadan, Nigeria. *Br J Oral Maxillofac Surg* 1997;35:185–9.
- 14 Machuca G, Khoshfeiz O, Lacalle JR, *et al.* The influence of general health and socio-cultural variables on the periodontal condition of pregnant women. *J Periodontol* 1999;70:779–85.
- 15 Rizwanulla T, Bandana K, Shivalal S, *et al.* Pyogenic granuloma: a case report. *Health Renaissance* 2010;8:196–8.

Copyright 2014 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow