J Med Sci 2018;38(6):287-291
DOI: 10.4103/jmedsci.jmedsci_20_18

CASE REPORT

Botryomycosis of Orofacial Region: A Rare Case Report

Devika S. Pillai[1], Subhas Babu[1], Shruthi Hegde[1], Vidya Ajila[1], G. Sachin Ram[1]

[1]*Department of Oral Medicine and Radiology, A B Shetty Memorial Institute of Dental Sciences, NITTE (Deemed to be University), Mangalore, Karnataka, India*

Botryomycosis is a rare chronic granulomatous infection that usually involves the skin and rarely viscera. The main causative organism is *Staphylococcus aureus*, and rarely, Pseudomonas and *Escherichia coli*. Both cutaneous and visceral forms of the disease are recognized. Botryomycosis may develop in apparently healthy individuals as well as the immunologically compromised. The involvement of orofacial region is rare in the literature. Diagnosing botryomycosis is very challenging. Dental specialists require thorough knowledge regarding the features and management of this infrequent entity in their practice. Isolation of the causative agent and antibiotic susceptibility tests are essential to provide appropriate treatment. We hereby report a rare case of a 37-year-old healthy male with recurrent episodes of botryomycosis involving the orofacial region. The present report highlights the clinical and histopathologic features of botryomycosis with literature review.

Key words: Botryomycosis, granulomatous, infection

INTRODUCTION

Botryomycosis, also called as bacterial pseudomycosis is a chronic, granulomatous, suppurative infection caused by *Staphylococcus aureus*. Other bacterial species such as *Pseudomonas aeruginosa*, *Proteus mirabilis* species, and *Escherichia coli* have also been isolated from the lesions.[1] These organisms form granules which are composed of bacterial masses which are adhered together.[2] Skin is the most frequently affected site, aponeuroses, bones, tendons, and muscles can also be affected. The disease can also manifest with the involvement of visceral organs only.[2] The present report highlights clinical manifestations of a rare case of cutaneous botryomycosis involving the orofacial region.

CASE REPORT

A male patient aged 37 years old, a rice mill worker by occupation reported to the outpatient department with the chief complaint of swelling and pus discharge from the left side of the cheek for 3 months. The patient gave a history of trauma to the same site 4 years back by road traffic accident. On eliciting the history, he was apparently normal after trauma for 1 year and he noticed pus discharge and pain on the left side of the face 3 years back, following which he visited a private dental hospital and underwent surgical debridement. The patient was asymptomatic following the treatment for 2½ years. He had experienced similar symptoms 6 months ago for which he was treated again at the same institution and was given antibiotics. The patient again developed swelling in the same region with pain and pus discharge for 3 months. He did not experience any reduction in the symptoms following 15 days of tetracycline therapy and visited our hospital for further evaluation. The patient's medical history, drug allergy, and family history were noncontributory. Extraoral examination revealed facial asymmetry with diffuse swelling on the left side of the face measuring around 4 cm × 5 cm in size; lesion was extending from the left commissure of the mouth till the ramus of the mandible. The swelling had multiple draining sinuses with a yellowish exudate [Figure 1a and b]. No paresthesia was elicited. Intraoral examination revealed an ill-defined swelling extending from the distal aspect of 35 to the distal aspect of 38, with obliteration of the buccal

Received: February 19, 2018; Revised: April 28, 2018; Accepted: June 04, 2018
Corresponding Author: Dr. Shruthi Hegde, Department of Oral Medicine and Radiology, A B Shetty Memorial Institute of Dental Sciences, NITTE (Deemed to be University), Mangalore, Karnataka, India. Tel: 916362002363; Fax: 08242204776. E-mail: drshruthihegde@yahoo.co.in

How to cite this article: Pillai DS, Babu S, Hegde S, Ajila V, Ram GS. Botryomycosis of orofacial region: A rare case report. J Med Sci 2018;38:287-91.

vestibule, with multiple sinus tracts. No pus discharge was noticed [Figure 1c]. Considering the history and the clinical findings, a provisional diagnosis of chronic granulomatous infection of the left orofacial region was given. A differential diagnosis of actinomycosis, botryomycosis, fungal infection, and tuberculosis was considered.

Panoramic radiograph showed no abnormalities in the region of interest. A radiopacity resembling, a tooth-like structure was seen in the periapical region of the maxillary left central incisor, suggestive of an impacted supernumerary tooth. Horizontally impacted lower right third molar was also noticed, and the lower left first molar was missing [Figure 2]. The patient was referred for the evaluation of immunocompromised status, and the results were negative.

An incisional biopsy was done, one from the region of the intraoral sinus tract and another from the extraoral lesion. Pus from the lesion was sent for culture and sensitivity, but the results were nonspecific. The histopathological examination revealed the presence of dense inflammatory infiltrate with neutrophils, lymphocytes, plasma cells, histiocytes, and macrophages. Furthermore, colonies of cocci surrounded by eosinophilic material were seen in a fibrillar stroma, and the Periodic acid–Schiff staining showed the presence of bright pink granular areas surrounded by dense inflammatory foci, which were suggestive of botryomycosis [Figure 3]. The patient was treated by surgical debridement followed by antibiotic therapy with azithromycin 1 g IV for 2 weeks. The patient is currently under follow-up.

DISCUSSION

Botryomycosis is a chronic granulomatous infection, which was discovered 130 years ago, in horses. Otto Bollinger in 1870, first it described it in a horse lung and the term botryomycosis was coined by Sebastino Revolta in 1884. The first case affecting humans was reported by Spitz in 1903 and Opie in 1913 reported the first case of visceral involvement.[3] As the name suggests, "botryo" means bunch of grapes and mycosis is suggestive of fungal origin. The disease was thought to have a fungal origin till 1919 when Magrou described the bacterial etiology.[3] Small and Koebner[4] in 1967 reported the first case of involvement of the oral cavity. It is broadly categorized into cutaneous and visceral forms.[4] Multiple Gram-positive and negative, aerobic, and anaerobic organisms have also been isolated, including *S. aureus*, *P. aeruginosa*, *P. mirabilis* species, *Peptostreptococcus*, or mixed oral flora.[5] Defects of cellular immunity are seen in botryomycosis, particularly with low total lymphocyte counts, mainly of T lymphocytes. The most characteristic feature of the disease is that bacteria group together to form conglomerates

Figure 1: Clinical photograph of the patient showing extension of the lesion (a and b) Extraoral clinical photograph showing facial asymmetry, extension of the lesion with multiple nodules and draining sinus tracts (c) Intraoral photograph showing ill-defined swelling extending from the distal aspect of 35 to the distal aspect of 38, with multiple sinus tracts

Figure 2: Panoramic radiograph showing no bony changes in the region of interest

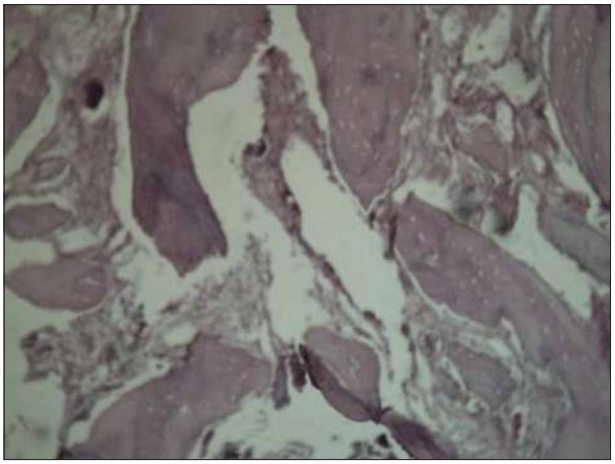

Figure 3: Histopathological photomicrograph showing the presence of dense inflammatory infiltrate with neutrophils, plasma cells, and macrophages with colonies of cocci surrounded by eosinophilic material

resembling the sulfur granules of actinomycotic infection.[5] The common predisposing factors include trauma to the skin and immunocompromised states.[6] The present case also has a history of trauma 1 year before the first episode of the disease. Botryomycosis occurs in all the age groups with a male predilection. It is clinically characterized by nodules, sinuses, fistulae, abscesses, and ulcers with seropurulent discharge.[7] The present case also had similar clinical presentation. Most frequently affected regions include skin of the hands, feet, gluteal and inguinal regions, rarely affecting the face and scalp. Bony involvement can be seen in the feet, legs, skull, spine, and mandible, due to direct spread of the infection from the skin, with clinical and radiographic features resembling osteomyelitis.[7] The visceral form frequently affects the lung, with lobar consolidation.

A literature search was carried out in PubMed using the search terminologies such as "botryomycosis" and "cutaneous" for 2001–2017. The total reported cases were 77, out of which only 19 cases had cutaneous involvement. The involvement of the orofacial region was reported only in four cases, out of which one case had associated mandibular osteomyelitis [Table 1].[8-25]

To the best of our knowledge, out of the three cases reported from India on botryomycosis,[8,12,15] this is the first case with involvement of the orofacial region which makes our case very rare.

Table 1: Literature search of case reports in PubMed during 2001-2017

Author and year	Age and gender of the patient	Site	Immune status	Treatment
Razmi *et al*., 2017[8]	60/male	Right orbito-temporal region	Immunocompetent	Cotrimoxazole and rifampicin
Mermin *et al*., 2017[9]	62/female	Right knee	History of Sezary syndrome	Gentamicin Gentamycin
Eyer-Silva *et al*., 2017[10]	28/female	Right malar region	Immunocompromised Seropositive	HAART therapy
Cataño and Posada, 2016[11]	65/male	Trunk and extremities	Immunocompromised Seropositive	Trimethoprim- sulfamethoxazole
Chintaginjala *et al*., 2016[12]	55/male	Left Leg	Immunocompetent History of trauma 8 years ago	Cotrimoxazole
Bashline *et al*., 2014[13]	83/female	Face and right arm	Immunocompromised, history of SCC of let lung	Surgical debridement
Mechow *et al*., 2014[14]	44/male	Right arm	Immunocompromised History of trauma 25 years ago	Clindamycin
Sreekanth *et al*., 2014[15]	17/male	Bilateral hands	Immunocompromised TB	Antitubercular therapy
Askari *et al*., 2014[16]	48/female	Cutaneous	History of diabetes mellitus	Trimethoprim - sulfamethoxazole
Ishibashi *et al*., 2012[17]	53/female	Right foot	Immunocompetent	Minocycline
Huang *et al*., 2012[18]	37/male	Nasal fistulas	Immunocompetent	Surgical debridement
Gosselin *et al*., 2011[19]	14 days	Inguinal region	Immunocompetent	Penicillin
Tomb *et al*., 2009[20]	23/male	Scalp	Immunocompetent	Penicillin Clindamycin Cefalexin
Coelho *et al*., 2009[21]	51/male	Right leg	Immunocompetent	Penicillin Cefalexin
Takata *et al*., 2009[22]	28/male	Left leg and right forearm	Immunocompetent	Cefazolin
Pielop *et al*., 2007[23]	52/male	Right hand	Immunocompetent History of trauma 8 years back	Fluoroquinolones
Meissner *et al*., 2007[24]	16/female	Left hand	Immunocompetent	Resistant to therapy
Templet and Straub, 2007[25]	46/female	Neck, trunk, and extremities	Immunocompromised, seropositive	HAART therapy
Yencha *et al*., 2001[3]	19/male	Cervicofacial region with mandibular osteomyelitis	Immunocompetent	Surgical debridement Clindamycin

SCC=Squamous cell carcinoma; HAART=Highly active antiretroviral therapy

The characteristic histologic feature of botryomycosis is the presence of eosinophilic fungus such as granules in a suppurative focus.[26] The center of the granules is basophilic and the periphery eosinophilic. They are embedded in a hyaline capsule which has club-like projections called Splendore-Hoeppli phenomenon.[26,27]

Combined antibiotic and surgical therapy are considered necessary to adequately treat the infection. The selection of antibiotics should be tailored to culture and susceptibility results. For Gram-positive infections, including *S. aureus*, oral trimethoprim-sulfamethoxazole, or oral clindamycin 30–40 mg/kg/day can be used. Depending on culture and susceptibility data, alternative agents such as doxycycline, minocycline, erythromycin, cephalexin, or dicloxacillin can be used.[17,27]

The present report highlights the clinical and histopathologic features of a rare case of botryomycosis in the orofacial region which adds valuable information to the literature. Dentists need to have thorough knowledge for the diagnosis and treatment of this infrequent entity in their practice. Future studies are required for the effective management of this rare condition.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. The patient understand that name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Deshmukh S, Saxena D, Sonarkar R, Saboo R. Botryomycosis in a young male: A case report and review of literature. Int J Curr Res 2016;8:33640-3.
2. Kinhal VA, Vishwakumar KS. A rare case of cutaneous botryomycosis – A case report and review of literature. J Evol Med Dent Sci 2017;6:3197-8.
3. Yencha MW, Walker CW, Karakla DW, Simko EJ. Cutaneous botryomycosis of the cervicofacial region. Head Neck 2001;23:594-8.
4. Small IA, Kobernick S. Botryomycosis of the tongue. Report of a case. Oral Surg Oral Med Oral Pathol 1967;24:503-9.
5. Desgarennes C, González DV, Bonifaz A. Botryomycosis. Clin Dermatol 2012;30:397-402.
6. Devi B, Behera B, Dash M, Puhan M, Pattnaik S, Patro S, *et al.* Botryomycosis. Indian J Dermatol 2013;58:406.
7. Fernandes NC, Maceira JP, Knackfuss IG, Fernandes N. Botriomicose cutânea: Relato de caso. Ann Bras Dermatol 2002;77:65-70.
8. Razmi TM, Mahajan R, Agrawal P. Image gallery: Cutaneous botryomycosis at an unusual site in an immunocompetent patient. Br J Dermatol 2017;176:e3.
9. Mermin D, Védie AL, Jullie ML, Fauconneau A, Beylot-Barry M, Pham-Ledard A, *et al.* Cutaneous granular bacteriosis occurring in *Staphylococcus aureus* septicaemia. Ann Dermatol Venereol 2017;144:275-8.
10. Eyer-Silva WA, Silva GA, Ferry FR, Pinto JF. Facial botryomycosis-like pyoderma in an HIV-infected patient: Remission after initiation of darunavir and raltegravir. Rev Soc Bras Med Trop 2017;50:277-9.
11. Cataño JC, Posada V. Disseminated cutaneous infection. Pan Afr Med J 2016;25:259.
12. Chintaginjala A, Harshavardhan K, Kumar AS. Cutaneous botryomycosis: A rare case report. Indian J Dermatol 2016;61:126.
13. Bashline B, Morrison M, Ramirez J, LaFond A. Disseminated botryomycosis: A rare presentation. J Drugs Dermatol 2014;13:976-8.
14. Mechow N, Göppner D, Franke I, Kolesnik M, Bonnekoh B, Gollnick HP, *et al.* Cutaneous botryomycosis diagnosed long after an arm injury. J Am Acad Dermatol 2014;71:e155-6.
15. Sreekanth R, Pallapati SC, Thomas BP. Tuberculous botryomycosis of the hand: Case report. J Hand Surg Am 2014;39:1810-2.
16. Askari K, Seyed Saadat SN, Seyed Saadat SM, Yousefi N, Ghorbani G, Zargari O, *et al.* Cutaneous botryomycosis caused by *Staphylococcus aureus* in a patient with diabetes. Int J Dermatol 2014;53:413-5.
17. Ishibashi M, Numata Y, Tagami H, Aiba S. Successful treatment of cutaneous botryomycosis with a combination of minocycline and topical heat therapy. Case Rep Dermatol 2012;4:114-8.
18. Huang CF, Liaw FY, Dai NT, Ou KW, Wang CC, Wang WM, *et al.* Botryomycosis presenting as nasal cutaneous fistulas caused by prevotella melaninogenica. Clin Exp Dermatol 2012;37:501-4.
19. Gosselin A, Bosdure E, Hesse S, Barlogis V, Chrestian MA, Dubus JC, *et al.* Neonatal botryomycosis. Arch Pediatr 2011;18:183-5.
20. Tomb RR, Stephan F, Haddad A, Choucair J. Cutaneous granular bacteriosis, a rarely diagnosed infection of the head and the neck. Clin Exp Dermatol 2009;34:887-9.
21. Coelho WS, Diniz LM, Souza FJ. Cutaneous botryomycosis: Case report. Ann Bras Dermatol 2009;84:396-9.

22. Takata T, Nakajima H, Ikeda M, Kodama H, Tamaki H, Sano S, *et al.* Cutaneous botryomycosis associated with eosinophilic cellulitis. J Dermatol 2009;36:551-4.
23. Pielop JA, Phillips R, Rosen T. Actinobacillus actinomycetemcomitans isolated from a case of cutaneous botryomycosis. Cutis 2007;79:293-6.
24. Meissner M, Spieth K, Wolter M, Gille J, Boehncke WH, Thaci D, *et al.* Cutaneous botryomycosis. A rarely diagnosed bacterial infection of the skin. Hautarzt 2007;58:966-8.
25. Templet JT, Straub R, Ko C. Botryomycosis presenting as pruritic papules in a human immunodeficiency virus-positive patient. Cutis 2007;80:45-7.
26. Vasishta RK, Gupta N, Kakkar N. Botryomycosis – A series of six integumentary or visceral cases from India. Ann Trop Med Parasitol 2004;98:623-9.
27. Eswari L, Revathi TN. Botryomycosis: A case report. Int J Sci Rep 2015;1:139.