DOI: 10.7860/JCDR/2022/53186.16318

Case Report

Dentistry Section

Recurrent Botryomycosis and Management Strategy with a Follow-up of Four Years: A Case Report

PS GOPINATH THILAK[1], SHARANYA MENON RADHAKRISHNAN[2]

ABSTRACT

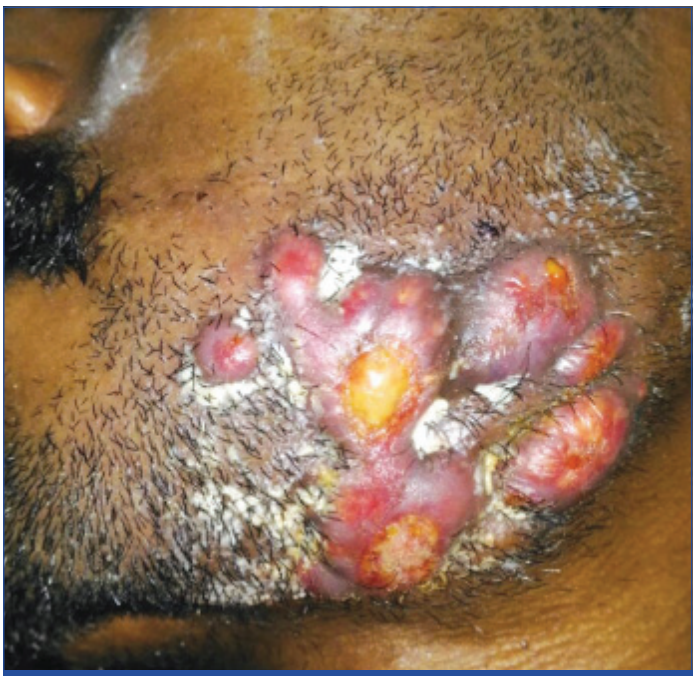
Cervicofacial botryomycosis is a chronic, granulomatous suppurative infection with varied presentations in the head and neck region. Since the discovery of this entity, treatment has been elusive without evidence of complete resolution of the lesion with a long period of follow-up. Here the authors report a case of cervicofacial botryomycosis with oral manifestations in a 37-year-old male patient with a history of trauma, for whom the authors devised a treatment plan tailored to the patient's microbiological profile after culture and sensitivity testing in addition to systemic and local antibiotic therapy and heat therapy which showed drastic regression of the lesion and an improved mouth opening. The patient has been under follow-up for the past four years without signs of recurrence. Botryomycosis is an aggressive lesion where a surgery-only approach is usually futile and requires further intervention due to recurrences. The present experience suggests surgical debridement along with antibiotic and local heat therapy to be effective in managing this usually recurring lesion.

Keywords: Bacterial pseudomycosis, Cutaneous botryomycosis, Granular bacteriosis, Granulomatous infections, *Streptococci viridans*

CASE REPORT

A 37-year-old male patient reported to the Outpatient Department (OPD) with complaints of persistent cutaneous eruptions on the left side of the cheek for the past several years. There was no associated history of pain in the region; however, constant ooze from the region which was colourless and odourless with a progressive decrease in the mouth opening and difficulty in eating was noticed by the patient. On anamnesis, the patient gave a history of trauma to the same site about seven years ago in 2011. It was a road traffic accident following which he skidded off and fell on the road and sustained soft tissue injuries to the face for which he was treated conservatively at a local hospital. A month after the incident, he noticed a single pus-filled lesion on his left cheek which progressively increased to multiple lesions with a gradual reduction in mouth opening. The patient with these complaints consulted a hospital where he underwent wound debridement under general anaesthesia in 2014 along with intravenous antibiotic therapy (Vancomycin and Metronidazole) and there seemed to be no regression of the lesion. He underwent a second surgery of the same lesion with resection of the lesion to gain healthy wound margins under general anaesthesia in May 2017 at the same hospital. A sample tissue was subjected to a histopathological examination which was revealed to be botryomycosis, following which he was started on Injection (Inj.). Amoxicillin and Clavulanic acid 1.2 gm TID, Inj. Amikacin 1 gm OD, Inj. Metronidazole 100 mL BD for a week. The lesion recurred after three months with advanced borders. The agony and pain of the unresolved infection brought him to the hospital in the year 2017.

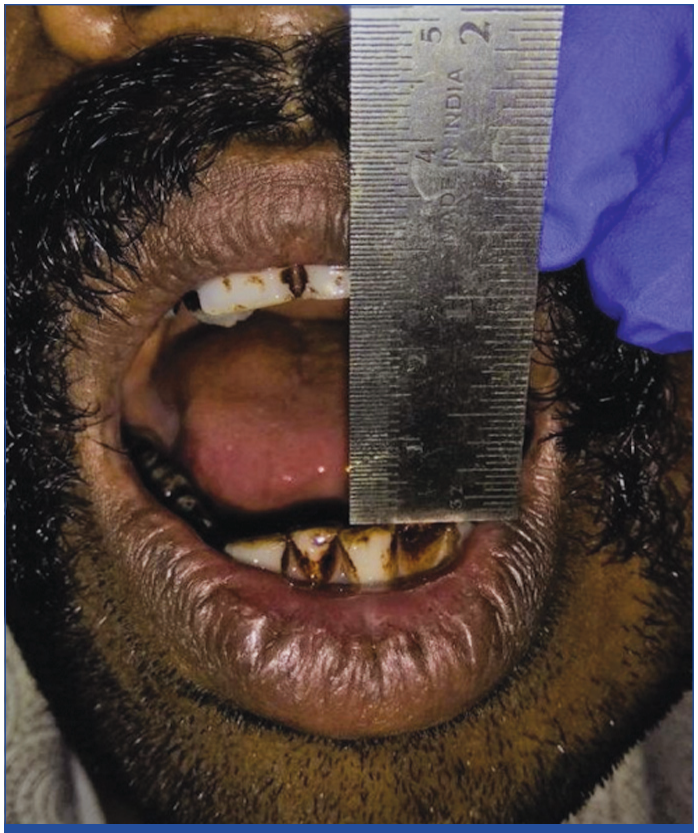
The patient's medical history, drug allergy, and family history were non contributory. Extraoral examination revealed facial asymmetry with diffuse swelling on the left side of the face measuring around 5×3 cm in size; the lesion was seen extending from the left commissure of the mouth up to the ascending border of the mandibular ramus. The swelling had multiple draining sinuses with a yellowish exudate along with the presence of crusting [Table/Fig-1]. Paraesthesia could not be elicited. Intra oral examination revealed an ill-defined swelling extending from the distal aspect of 35 to the distal aspect of 38, with obliteration of the buccal vestibule, multiple sinus tracts, and purulent material was noticed along with a reduced mouth opening of about two finger breadths [Table/Fig-2].



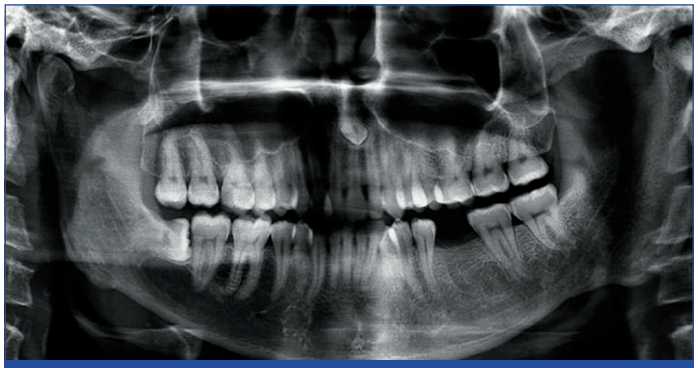
[Table/Fig-1]: Shows extra oral cutaneous eruptions over the left cheek.

The panoramic radiograph showed no abnormalities in the region of interest [Table/Fig-3]. The patient was referred for the evaluation of immunocompromised status, and the results were negative.

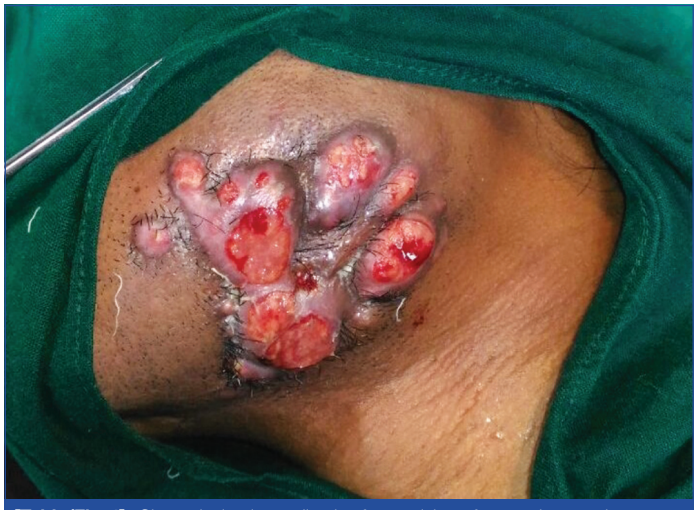
Avascular margins were excised until fresh bleeding was observed from the extraoral lesion [Table/Fig-4] which was sent for histopathological analysis which revealed a diagnosis of botryomycosis [Table/Fig-5]. To confirm this diagnosis, the surgically resected tissue [Table/Fig-6] from the hospital at which he initially



[Table/Fig-2]: Shows reduced mouth opening.



[Table/Fig-3]: Shows the panoramic radiograph of the patient.

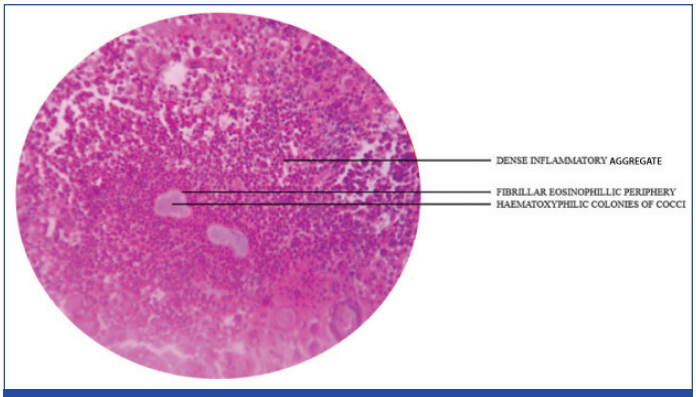


[Table/Fig-4]: Shows lesion immediately after excision of avascular margins to induce fresh bleed.

consulted was also obtained and re-examined histopathologically that evidenced the presence of botryomycosis.

The photomicrograph shows the specimen in 10x magnification with Haematoxylin and Eosin (H&E) staining [Table/Fig-5].

The histopathology reveals epidermis and dermis. The dermis shows the presence of dense inflammatory aggregates that is composed



[Table/Fig-5]: Shows the histopathological picture suggestive of botryomycosis (Haematoxylin & Eosin, 10x).

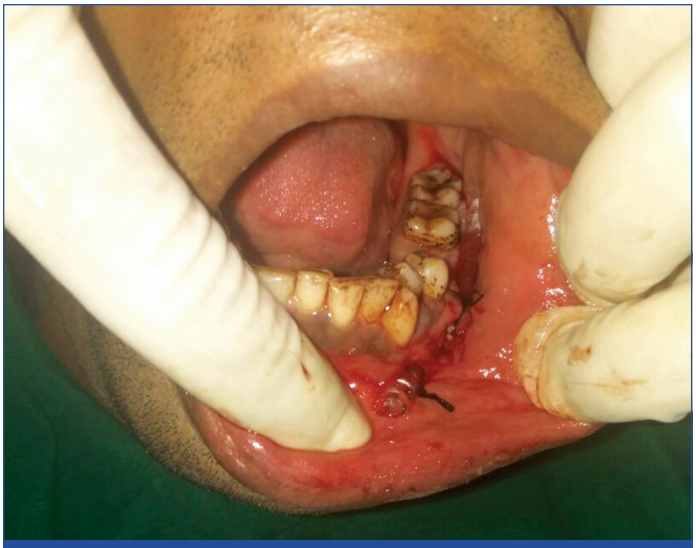
[Table/Fig-6]: Shows the specimen after initial surgical resection in 2017.

of neutrophils, lymphocytes, plasma cells, histiocytes and foamy macrophages. Few haematoxyphilic colonies of cocci are seen surrounded by eosinophilic material in a fibrillar stroma suggestive of botryomycosis.

Pus from the lesion was sent for culture and sensitivity testing. The cultured micro-organisms were *Streptococci viridans*. A strategic conservative treatment protocol was devised specifically for the patient's microbial profile as previous surgical intervention led to repeated aggressive recurrence.

Treatment was planned as a triple mode therapy with intravenous and intraoral antibiotic lavage with a modified drain in place along with extraoral skin treatment. An infant feeding tube with a diameter of 3.03 mm was modified with multiple small holes made throughout the surface and was secured in place with the help of sutures in the buccal vestibule [Table/Fig-7]. It was used in such a way that while irrigating the antibiotics in a saline medium, there was increased perfusion of this mixture into the intraoral lesion.

The patient was initiated on systemic antibiotic therapy based on culture sensitivity report [Table/Fig-8] along with local antibiotic therapy intraorally via irrigation through the modified drain this was followed by hot water fomentation for the skin lesion which



[Table/Fig-7]: Shows the modified drain used intraorally.

MICROBIOLOGY

Pus for C/S

Culture and Sensitivity

Sample: Pus

Microscopy:Gram stain shows numerous pus cells and few gram positive cocci in pairs.

Culture report: Viridans streptococcus

Antibiotic Sensitivity Report

Antibiotic	MIC	Result	Antibiotic	MIC	Result
Cefotaxime		R	Erythromycin		S
Cefepime		R	Clindamycin		
Cefotaxime		R	Linezolid		S
Ceftriaxone		R	Vancomycin		S
Levofloxacin		R	Azithromycin		S
Chloramphenicol		S	Trimethoprim/sulfamethoxazole		

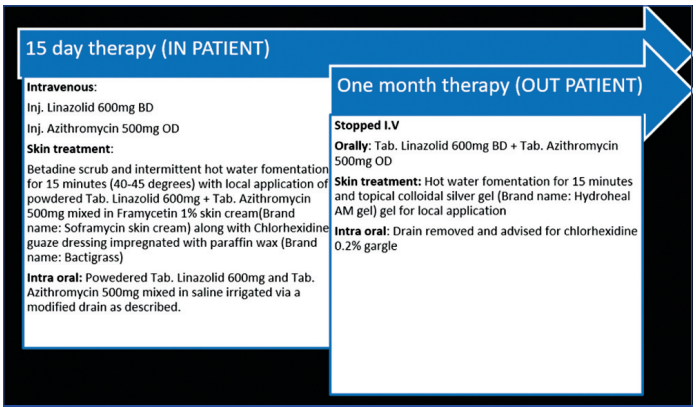
Comment : Significance of the isolate has to be correlate clinically.

****

[Table/Fig-8]: Shows the culture and sensitivity report (R=resistant, S=sensitive).

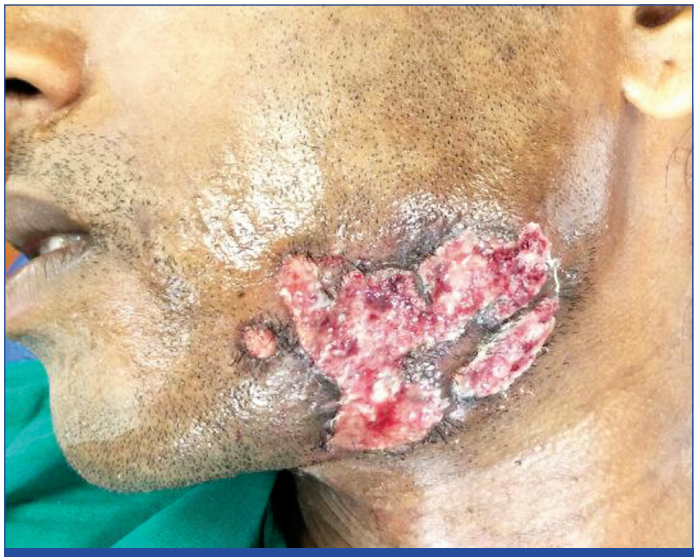
was carried out during the first week of admission, after which the patient was subjected to excision of the avascular margins under local anaesthesia after which the antibiotic and heat therapy was continued postoperatively.

The antibiotic regimen was tailored to the patient's microbial profile and the culture and sensitivity report. The treatment was devised by the author after the excision of the lesion. The protocol that was followed for the present case has been listed in [Table/Fig-9]. This treatment protocol was devised by the author for the management of this lesion.

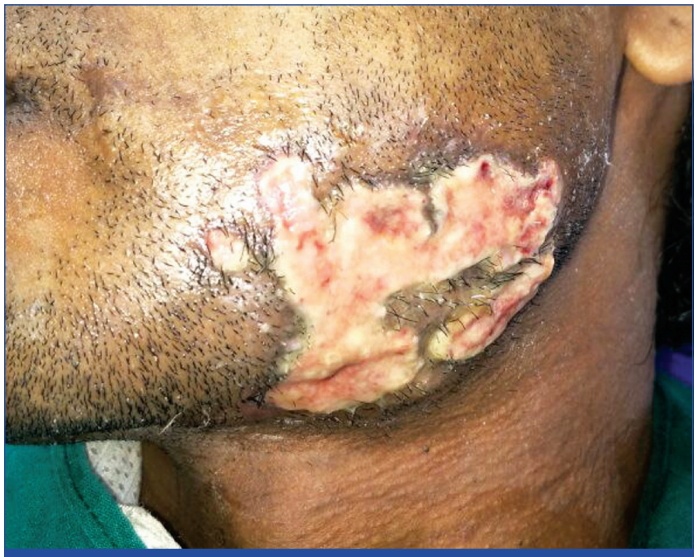


[Table/Fig-9]: Shows the treatment protocol devised by the author.

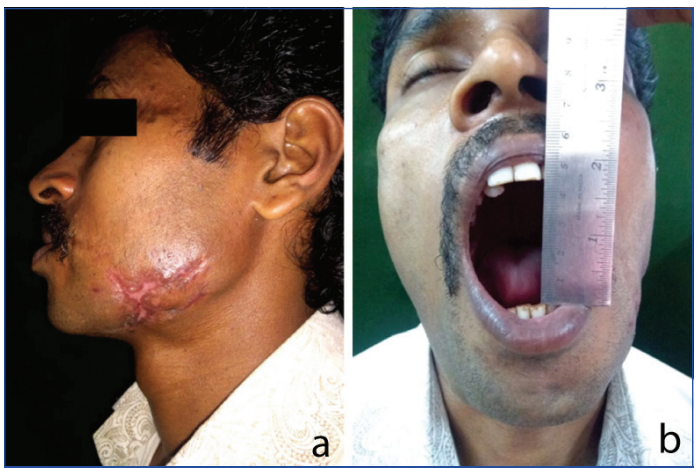
[Table/Fig-10] shows immediate postoperative results. The lesion visibly regressed within 15 days of rigorous local treatment [Table/Fig-11] with an increase in mouth opening. Our patient also reported better sensory function in the overlying skin during hot fomentation as treatment progressed. After 15 days of hospital stay and robust local management, the patient was discharged with oral antibiotics and was advised for topical colloidal silver gel (brand name: Hydroheal AM gel) application over the skin and was recalled for follow-up a month after discharge. On review at 3 months postoperative, there was a radical regression of the lesion with clear skin margins, and a mouth opening up to four fingers was recorded [Table/Fig-12]. [Table/Fig-13] shows the complete resolution of the lesion 1-year postoperatively.



[Table/Fig-10]: Shows immediate postoperative.

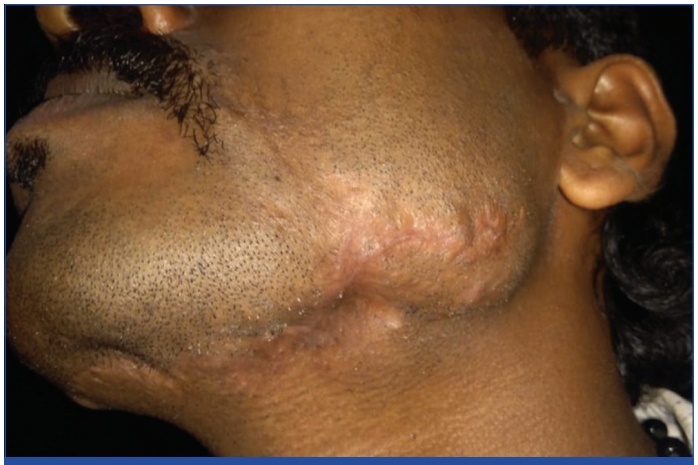


[Table/Fig-11]: Shows the lesion on the 15th postoperative day.

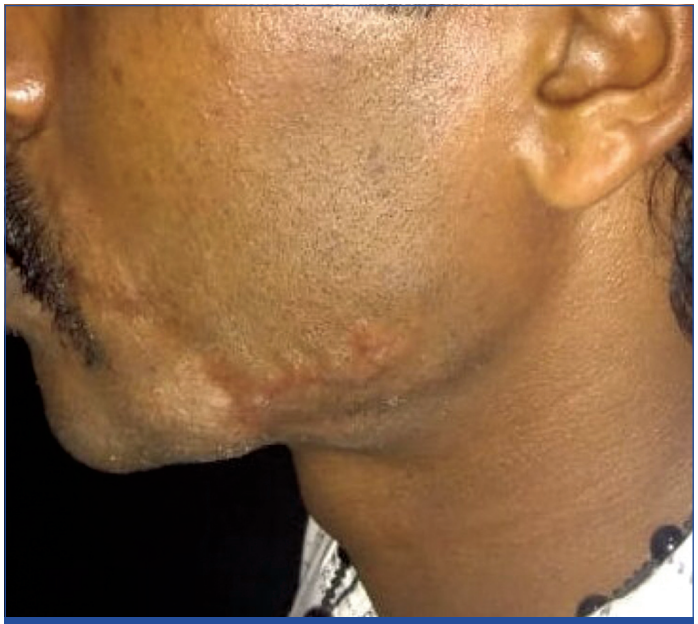


[Table/Fig-12: Showing recovery three months postoperatively a) regressed lesion b) Improved mouth opening.

The patient has been under a long-term follow-up period of more than four years with no remissions [Table/Fig-14].



[Table/Fig-13: Shows the complete resolution of the lesion 1-year postoperatively.



[Table/Fig-14: Shows no evidence of recurrence on follow-up after four years postoperatively.

DISCUSSION

Botryomycosis is a chronic, suppurative and granulomatous lesion with clinical presentation of fungus-like grains or granules within suppurative foci, caused by *Staphylococcus aureus* organisms in association with *Pseudomonas aeroginosa*, *Proteus mirabilis*, and *Escherichia coli* [1]. The cutaneous manifestations are highly variable as they appear as fungating masses, blisters, nodules, papules, ulcers, and verrucous plaques with or without a sinus tract formation. It may also present with surrounding cellulitis with a superficial abscess which depending on the size and location of the lesion may involve underlying muscle, tendon, or bone. The diagnosis of botryomycosis is arrived at, after careful histopathological examination as it closely resembles an actinomycosis infection in its clinical and histological presentation [2]. It was first reported by Bollinger O in 1870. There have been various treatment modalities that have been suggested for treating this lesion since its discovery [3-5]. For the treatment of botryomycosis, a long course of antibiotics is required, depending on the pus culture and sensitivity. However, the most effective treatment for botryomycosis is antibiotics combined with surgical excision [6].

According to a systematic literature review published by Bailey P et al., the mean duration for antibiotic therapy ranges from 4.2 days to 40 weeks for complete resolution of the lesion without any adjuvant procedures [7].

Ellerbe DM et al., reported a case of botryomycosis that presented as cellulitis over the right cheek in an eight-month-old male child who was initially treated with Amoxicillin and Clavulanic acid which had a recurrence and the child was subjected to 19 dives of hyperbaric oxygen therapy along with Nafcillin which was the sensitive antibiotic for two weeks. They reported that the lesion recurred more aggressively and finally, the treatment was switched to medical management using prolonged therapy using Clindamycin for 10 months after which the lesion completely resolved [8].

Devi B et al., reported a case of cutaneous botryomycosis which was treated with surgical resection in addition to antimicrobial therapy for resolution of the lesion with a follow-up period of two months [4].

Eyer-Silva WD et al., in their paper reported a case of cutaneous botryomycosis over the right malar region in a 28-year-old Human Immunodeficiency Virus (HIV) infected woman. The patient underwent several courses of antimicrobial therapy that provided only partial and temporary remission. Complete clinical remission was only achieved five years later when a novel antiretroviral regimen consisting of Darunavir and Raltegravir was initiated. The authors stated that successful treatment of botryomycosis depends on several factors such as the immune status of the host, the causative agents, and the location and extent of the lesion. They concluded that the mainstay of therapy is appropriate antibiotic therapy guided according to bacterial resistance patterns and surgery should be reserved for diagnostic purposes or recalcitrant disease [9].

Lefell DJ et al., reported a case of botryomycosis over the dorsal surface of the foot which was treated with Carbon Dioxide (CO_2) laser ablation therapy following failure of the use of broad-spectrum antibiotics in resolving the lesion with a follow-up period of five months [10]. Topical heat therapy was first introduced for the treatment of sporotrichosis over the right dorsum of the hand by Mackinnon JE and Conti-Diaz IA based on the thermosensitivity of *Sporothrix schenckii* [11]. There are also sporadic reports of its effectiveness for chromoblastomycosis in literature [12,13].

In addition to medical and surgical therapy, we inculcated the use of heat therapy as adjuvant therapy for the present patient after an extensive literature search on the effects of temperature for therapy-resistant cutaneous skin infections [12-14]. In previously reported literature, there has only been one case report of cutaneous botryomycosis over the right foot which was treated with topical heat therapy along with antibiotic therapy reported by Ishibashi M et al., [14]. In their study, the patient was initiated on antibiotic therapy using oral Minocycline at a dose of 200 mg once a day along with topical heat therapy using a commercially available pocket warmer which was constantly applied every day for as long as the patient could tolerate without causing a burn injury. The authors noted significant clinical improvement within four months and no signs of recurrence during an eight months follow-up period. They concluded the study by saying that heat therapy is an effective complementary therapy in the treatment of cutaneous botryomycosis.

The exact mechanism of how heat therapy works in botryomycosis is unknown because the germicidal effect of heat therapy on *Staphylococcus aureus* cannot be established as they are capable of growth at 48°C even after incubating for seven days [15]. The literature reports that heat induces a phagocytic reaction as part of the host defence mechanism and exerts a cytostatic effect [14].

In this case report, instead of a constant heating device to provide heat therapy, we used hot water fomentation over the lesion for the desired effect. Of the cases of botryomycosis reported thus far [2-4,16,17] none have a follow-up period of more than one year with reports of relapse or remission of the disease. The extensive follow-up period without any recurrence in this case report reinforces the need for a multi-modality approach in the treatment of this disease.

CONCLUSION(S)

Botryomycosis is a disease that requires aggressive management to prevent relapse in the long term. A surgery-only approach that was initially done for patient was futile and required further intervention. Hence, the authors would like to recommend adequate antibiotic therapy based on the patient's culture and sensitivity reports along with surgical debridement and heat therapy for acceptable regression of the lesion. It is also recommended that the same team of doctors should follow-up with the patient so that any changes that occur during the treatment of the lesion is immediately appreciable and further course of action can be determined accordingly.

REFERENCES

[1] Bonifaz A, Carrasco E. Botryomycosis. International Journal of Dermatology. 1996;35(6):381-88.
[2] Yencha MW, Walker CW, Karakla DW, Simko EJ. Cutaneous botryomycosis of the cervicofacial region. Head & Neck: Journal for the Sciences and Specialties of the Head and Neck. 2001;23(7):594-98.
[3] Devaiah AK, Hoffman HT, Robinson RA, Carter K. Oral botryomycosis: A case report. The Journal of Laryngology & Otology. 1997;111(1):77-79.
[4] Devi B, Behera B, Dash ML, Puhan MR, Pattnaik SS, Patro S. Botryomycosis. Indian Journal of Dermatology. 2013;58(5):406.
[5] DeWitt JP, Stetson CL, Thomas KL, Carroll BJ. Extensive cutaneous botryomycosis with subsequent development of Nocardia-positive wound cultures. Journal of Cutaneous Medicine and Surgery. 2018;22(3):344-46.
[6] Sirka CS, Dash G, Pradhan S, Naik S, Rout AN, Sahu K. Cutaneous botryomycosis in immunocompetent patients: A case series. Indian Dermatology Online Journal. 2019;10(3):311.
[7] Bailey P, Raybould JE, Sastry S, Bearman G. Cutaneous botryomycosis and staphylococcus aureus: Diagnosis, management, and a systemic literature review. Current Treatment Options in Infectious Diseases. 2018;10(3):347-61.
[8] Ellerbe DM, Parsons DS, Cook PR. Botryomycosis: Improved therapy for a difficult infection. International Journal of Pediatric Otorhinolaryngology. 1997;41(3):363-69.
[9] Eyer-Silva WD, Silva GA, Ferry FR, Pinto JF. Facial botryomycosis-like pyoderma in an HIV-infected patient: Remission after initiation of darunavir and raltegravir. Revista da Sociedade Brasileira de Medicina Tropical. 2017;50(02):277-79.
[10] Leffell DJ, Brown MD, Swanson NA. Laser vaporization: A novel treatment of botryomycosis. The Journal of Dermatologic Surgery and Oncology. 1989;15(7):703-05.
[11] Mackinnon JE, Conti-Diaz IA. The effect of temperature on sporotrichosis. Sabouraudia. 1963;2(2):56-59.
[12] Yanase K. Yamada M. Pocket-warmer therapy of chromomycosis [letter]. Arch Dermatol. 1978;114(7):1095.
[13] Tagami H, Ginoza M, Imaizumi S, Urano-Suehisa S. Successful treatment of chromoblastomycosis with topical heat therapy. Journal of the American Academy of Dermatology. 1984;10(4):615-19.
[14] Ishibashi M, Numata Y, Tagami H, Aiba S. Successful treatment of cutaneous botryomycosis with a combination of minocycline and topical heat therapy. Case Reports in Dermatology. 2012;4(2):114-18.
[15] Schmitt M, Schuler-Schmid U, Schmidt-Lorenz W. Temperature limits of growth, TNase and enterotoxin production of Staphylococcus aureus strains isolated from foods. International Journal of Food Microbiology. 1990;11(1):01-09.
[16] Razmi TM, Mahajan R, Agrawal P. Image gallery: Cutaneous botryomycosis at an unusual site in animmunocompetent patient. Br J Dermatol. 2017;176(1):e3.
[17] Aziz F, Ong F, Parikh RN, Hamilton AE. A case of cutaneous botryomycosis of the lower leg in a young adult male. Cureus. 2021;13(7):e16502. Doi: 10.7759/cureus.16502.

PARTICULARS OF CONTRIBUTORS:

1. Reader, Nitte (Deemed to be University), AB Shetty Memorial Institute of Dental Sciences (ABSMIDS), Department of Oral and Maxillofacial Surgery, Mangalore, Karnataka, India.
2. Postgraduate Student, Nitte (Deemed to be University), AB Shetty Memorial Institute of Dental Sciences (ABSMIDS), Department of Oral and Maxillofacial Surgery, Mangalore, Karnataka, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:

Dr. Sharanya Menon Radhakrishnan,
Postgraduate Student, Nitte (Deemed to be University), AB Shetty Memorial Institute of Dental Sciences (ABSMIDS), Department of Oral and Maxillofacial Surgery, Mangalore, Karnataka, India.
E-mail: menon.sharanya@gmail.com

PLAGIARISM CHECKING METHODS: [Jain H et al.]

- Plagiarism X-checker: Jan 13, 2022
- Manual Googling: Mar 09, 2022
- iThenticate Software: Mar 31, 2022 (8%)

ETYMOLOGY: Author Origin

AUTHOR DECLARATION:

- Financial or Other Competing Interests: None
- Was informed consent obtained from the subjects involved in the study? Yes
- For any images presented appropriate consent has been obtained from the subjects. Yes

Date of Submission: **Jan 01, 2022**
Date of Peer Review: **Feb 02, 2022**
Date of Acceptance: **Mar 11, 2022**
Date of Publishing: **May 01, 2022**