

PYODERMA VEGETANS. REPORT ON A CASE AND REVIEW OF DATA ON PYODERMA VEGETANS AND CUTANEOUS BOTRYOMYCOSIS

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ABSTRACT

A 47-year-old man is presented with a large verrucous, crusting purulent plaque on the dorsum of his hand. Bacteriological cultures revealed *Staphylococcus aureus*, Beta hemolytic *Streptococcus* and *Hemophilus influenzae*. Histology showed pseudoepitheliomatous hyperplasia and chronic inflammation. No grain formation was shown. Prolonged course of ampicillin treatment combined with x-ray resulted in complete clearing of the lesion.

The literature is reviewed with special emphasis on pyoderma vegetans and cutaneous botryomycosis. It is suggested that these two disorders are closely related and they should be classified in one group.

KEY WORDS

pyoderma vegetans, relation to botryomycosis

INTRODUCTION

Pyoderma vegetans or chronic vegetating dermatitis is an uncommon form of the pyodermas. The clinical picture is similar to a deep mycotic infection, therefore the terms blastomycosis-like pyoderma (1,2,3), mycosis-like pyoderma (4) are used as synonyms for pyoderma vegetans. The disease is characterized clinically by verrucous, solitary or multiple plaques with pustules and elevated border, histologically by pseudoepitheliomatous hyperplasia with abscess formation and the growth of pathogenic bacteria from the lesion. Cutaneous botryomycosis is a rare bacterial infection of the skin. Clinically the disease resembles a deep fungal infection. The histologic hallmark of the disease is the presence of basophilic, fungus-like

granules, the grains in a suppurative focus, with aggregates of causative bacteria. *Pyoderma vegetans* and cutaneous botryomycosis are similar diseases. The number of the reports on these diseases is relatively low. To the best of our knowledge there is no report discussing the two entities. Because of the similarities we feel that it would not be useless to summarize and compare the data of the two processes, while reporting a case.

CASE REPORT

A 47-year-old, alcoholic man presented to our clinic with a 1.5-year history of a slowly enlarging lesion on the dorsum on his left hand. There was a history of preceding trauma, by a rusty fence.

Before admission long term antimycotic, oral antituberculous and X-ray treatments were unsuccessful. On admission a verrucous plaque, 12 cm in diameter, with an elevated border, with purulent drainage and crusting was present on the back of his left hand. (Fig. 1) There was no regional lymphadenopathy. Otherwise the patient enjoyed an excellent health.



Fig 1. Large, verrucous, purulent plaque on the dorsum of the hand.

Physical examination showed a moderate hepatomegaly. Laboratory studies gave normal results, except an elevated GGT (133U/1). The differential diagnosis of this cutaneous lesion included a deep fungal infection, mycobacteriosis, atypical mycobacteriosis, and atypical large keratoacanthoma. Cultures and special stains for fungi and mycobacteria were negative. Multiple cultures isolated first *Staphylococcus aureus*, then *Streptococcus beta hemolyticus* and *Haemophilus influenzae*. A radiograph of the hand was negative. Two biopsy specimens were removed (hematoxylin-eosin and tissue Gram's stain). They showed marked pseudoepitheliomatous hyperplasia with chronic inflammation. (Fig. 2) Gram stain of the histological section did not result in the demonstration of any grain-like structure. The patient has been treated with a course of ampicillin (2 g daily) for 4 weeks, combined with 3x1 Gy X-ray. The lesion was completely healed after 2 months. We noted no recurrence after 10 months of follow-up period.

DISCUSSION

Azua and Pons in 1903 (5), Gay Prieto and Cascas in 1951 (6) reported cases with vegetating skin lesions similar to blastomycosis or tuberculosis. Histologic characteristic was pseudoepitheliomatous hyperplasia. They termed the condition pseudoepitheliomas cutanéus (5) and pyodermis

chronica vegetans (6). In 1957 Brown and Kligman described two patients with "mycosis-like pyoderma" (4). Clinical and histologic characteristics, the differential diagnosis and pathogenesis of the disease are summarized by Su et al (3). Botryomycosis was first described in humans in 1913 (7). The bacteriologic origin of the disease was established in 1919 (8).

Cutaneous disease appears as a chronic, suppurative, verrucous, often granulomatous lesion, it involves most frequently the hands, feet and head and may extend into the underlying muscle and bone (9,10,11,12). Clinically characteristic feature of the disease is the presence of granules in the pus. The histologic hallmark of botryomycosis is the presence of grains. Aggregates of bacteria are found in the centers of the grains. Around the area of central suppuration there is a nonspecific chronic inflammatory reaction.

Our patient had a skin lesion on his hand with a chronic course, lasting for 1,5 years. First the process was considered to be of mycotic or of tuberculous origin.



Fig 2. Histologic appearance of lesion from Fig. 1 Hyperkeratosis, parakeratosis, acanthosis and chronic inflammation. (Hematoxylin-eosin stain; x450)

Table I. Comparison of symptoms in pyoderma vegetans and cutaneous botryomycosis

Pyoderma vegetans	Cutaneous botryomycosis
Causative microorganisms:	
Staphylococcus aureus Streptococcus beta haemolyticus Diphtheroids Bacillus sp, Clostridium perfrigerans Proteus mirabilis E. coli	Staphylococcus aureus Streptococcus beta haemolyticus E. coli, Proteus vulgaris Moraxella nonliquefac Corynebacterium sp. Diplococcus pneumoniae Acinetobacter
Clinical features:	
purulent, hyperkeratotic, vegetating plaques ulceration elevated border, abscess, purulent exudate	purulent, hyperkeratotic, vegetating plaques ulceration elevated border abscess, purulent exudate nodules, draining sinuses
Histology:	
hyperkeratosis pseudoeplitheliomatous hyperplasia acute and/or chronic inflammation granulomatous reaction	hyperkeratosis pseudoeplitheliomatous hyperplasia acute and/or chronic inflammation granulomatous reaction
Contributing factors:	
immunosuppression leukemia, lymphoma, alcoholism poor nutrition diabetes mellitus cellular immunodeficiency	immunosuppression AIDS alcoholism poor nutrition diabetes mellitus cellular immunodeficiency
Grain -	Grain +
Visceral involvement -	Visceral involvement +

No fungus or mycobacterium were cultured and the histological examination failed to confirm the above mentioned diagnoses. The culture revealed Staphylococcus aureus, Haemophilus influenzae and Streptococcus beta hemolyticus. The histology showed epidermal hyperplasia and chronic inflammation.

The clinical picture - verrucous, vegetating, granulomatous lesion, with elevated border, with multiple abscess formation - and the result of the bacteriological examination was compatible with pyoderma vegetans or cutaneous botryomycosis, although grains were not proven macroscopically. Histology did not show

any grain formation, so the diagnosis of pyoderma vegetans was made.

The differential diagnosis included besides the deep fungal and mycobacterial infections, bromoderma, pyoderma gangrenosum, giant keratoacanthoma.

In lesional cultures in both disorders commonly grow *Staphylococcus aureus* and *Streptococcus B hemolyticus*. In the lesions of blastomycosis-like pyoderma often many other organisms are detected: *Pseudomonas aeruginosa* (2), diphtheroids, *Bacillus* species (3), *Clostridium perfringens* (2), *Proteus mirabilis* (3,13) and *E. coli* (3).

In botryomycosis the reported bacterial agents include *E. coli* (14), *Proteus vulgaris* (10), *Moraxella nonliquefaciens* (15), *Bacillus* species, streptococcus (11,12,14), *Neisseria* species, *Corynebacterium* species (11), *Diplococcus pneumoniae* (10), *Acinetobacter* (16).

In some cases multiple pathogenic bacteria were found on culture (2,14) as in our case.

The origin of these diseases is not completely clear. It is suggested that an unusual tissue reaction develops to a bacterial infection. The cause of this peculiar tissue reaction is not known.

Stone suggested that defects of the defense mechanisms might play a role in the histogenesis of the vegetations. The granulocytes-released peptides cause fibroblast proliferation and pseudoepitheliomatous hyperplasia in pyoderma vegetans (17).

The patients with pyoderma vegetans and with cutaneous botryomycosis may reveal a decreased resistance to bacterial infection. In patients with pyoderma vegetans chronic granulocytic leukemia, immunosuppressive therapy, local X-ray radiation, pulmonary granuloma, diabetes mellitus (3), poor nutrition, alcoholism (1,4), cellular immunodeficiency (18), T cell lymphoma (13), ulcerative colitis (19) can be associated.

In botryomycosis similar predisposing factors were shown: alcoholism, steroid therapy (20,21), diabetes mellitus (10,14,22), cellular immunodeficiency (14,23), and AIDS (24).

In our case the local tissue trauma and alcoholism promoted the development of the disease.

Treatment is generally difficult. Antibiotic treatment alone is often unsatisfactory. When antibiotic therapy fails, surgical resection and drainage is useful. Laser vaporization was successful in a case of cutaneous botryomycosis, in which antibiotic and surgical treatment was without result (25). In our case antibiotic treatment accompanied with X-ray (3x1 Gy) led to the disappearance of all symptoms.

The main characteristics of the two processes are summarized in the table 1.

The similar features between the two processes are: 1. clinical picture; 2. host reaction; 3. causative microorganisms; 4. contributing factors; 5. treatment modalities.

Differential diagnostic categories are the same in both diseases, but in such forms of cutaneous botryomycosis where single or multiple nodes, abscesses with multiple draining sinuses, with grossly visible grain formation dominate other granule associated processes i.e. maduramycosis, *Nocardia mycetoma* and actinomycosis also must be considered in the differential diagnosis. The most striking difference can be shown histologically: the presence of the grains in botryomycosis. A histopathologic analysis that proves the grains readily differentiates the two entities.

Another difference is that botryomycosis can occur in visceral organs, pyoderma vegetans can develop exclusively on the skin.

The questions: what is the role of the grains, why they develop, what specific role, if any, they play, are to be answered.

The development of the grains does not seem to influence the skin disease, as the clinical picture, the course of the disease, the response to treatment are very similar or the same in both conditions. We think that blastomycosis-like pyoderma and cutaneous botryomycosis are closely related processes. It is suggested that cutaneous botryomycosis should be classified as a special type of pyoderma vegetans.

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