Case No. 18

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Patient: A 35-year-old Thai man from Chonburi

Chief complaint: Multiple exophytic erythematous nodules on the right knee for 1 year

Present illness:
The patient presented with multiple asymptomatic exophytic polypoid erythematous nodules on the right knee for 1 year. He was diagnosed as keloid without treatment because of financial problems. He denied any history of fever or arthralgia.

Past history: No history of previous trauma
No Current medication

Family history: No other family members had similar symptoms.

Physical examination:
Right knee: No sign of joint inflammation
Lymph node: No lymphadenopathy

Dermatological examination:
Skin: Localize multiple exophytic polypoid soft erythematous nodules with no sinus tract formations and no grains were seen at right knee.

Investigation:
Aerobic culture: Staphylococcus coagulase negative
Fungus culture: No growth
Mycobacterium culture: No growth
PCR for actinomycetes: Gordonia terrae
Film right knee: No bone involvement

Histopathology: Slide No. 58/0941 (Right knee)
Section displays irregular acanthosis with pseudoepitheliomatous hyperplasia. Focal aggregates of filamentous microorganism are identified and surrounded by abscess formation.

Diagnosis: Gordona terrae actinomycetoma

Treatment: Co-trimoxazole 160/800 mg/day and ciprofloxacin 1000 mg/day

Discussion:
Mycetomas are chronic subcutaneous infections caused by fungus or aerobic filamentous bacterium which characterized by a clinical triad of tumefaction, draining sinuses, and discharge of granules. It is endemic to tropical and subtropical climates such as Somalia, Mexico, India, Central and South America and Northern Africa.¹

The infection occurs after traumatic inoculation from contaminated soil and progresses to adjacent tissues or bone. The foot, hand, and lower-leg regions are the most commonly affected areas.²

The most common presentation is a painless lump or a swollen foot. Eventually, multiple draining sinuses will develop with the discharge of pus and granules which are composed of colonies of either actinomycotic bacteria or eumycotic fungi. With time, a cycle develops in which old sinuses heal and new sinuses form, causing the progressive development of granulation and scar tissue that can cause deformity or disfigurement.³

The diagnosis of mycetoma is made using clinical suspicion, radiologic studies, histologic examinations, and culture. Determination of the causative agent is necessary before treatment can be instituted.⁴ Culture methods are still considered to be the gold standard in species identification of the causal agents of mycetoma. However, some agents are difficult to identify by morphology alone. Moreover, precise identification of these organisms remains a challenge due to limitations including contaminations, the long period of time for positive cultures, and lack of experience of the pathogen.⁵ At the moment only molecular methods are able to identify the causative agent to the species level reliably. However, these methods usually have to be performed in large diagnostic centers and is too expensive for the endemic countries.⁵

Gordona terrae, previously known as Rhodococcus terrae, has been reported as a difficult-to-diagnose emerging pathogen that has been misdiagnosed by laboratories using conventional microbiological techniques.⁶ It is an aerobic, catalase-positive, Gram positive rod that is morphologically similar to other actinomycetes such as Nocardia or Rhodococcus. Accurately identifying these organisms requires sequencing data, most often using the 16S ribosomal RNA.⁷
G. terrae is predominantly an environmental pathogen that is rarely reported to cause human infection. When it does cause human disease, it is mostly in immunocompromised hosts. In immunocompetent hosts it has been rarely reported to cause soft tissue infection and is likely to be underreported due to misidentification.

We present the third known case of mycetoma with atypical presentation caused by Gordonia terrae. Currently, the patient was treated with Co-trimoxazole in addition to ciprofloxacin for 4 months, and improvements of the lesion is observed. We plan to continue this modality until all of skin lesions are completely healed.

Reference: