# Pythiosis presenting with digital gangrene and subcutaneous nodules mimicking medium vessel vasculitis

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#### INTRODUCTION

Pythiosis is an infectious disease caused by microorganisms of the *Pythium* genus, a form of funguslike oomycetes living in water-rich habitats. Human pythiosis is an emerging disease with a high rate of morbidity and mortality. Early recognition and management is crucial for successful treatment and outcomes. Because identifying the pathogen can be challenging, pythiosis might be easily misdiagnosed. Here we report a case of vascular pythiosis, the manifestation of which mimics medium-sized vasculitis.

#### **CASE REPORT**

A 54-year-old man presented with severe pain on his right thumb. Two months earlier, he had several subcutaneous nodules on his right forearm and bluish discoloration of his right thumb (Fig 1, A). He had no significant medical history and was not taking any medicines or herbs. He worked as a rice farmer and often was in contact with soil and mud. He had a 2 pack-year smoking history and excessive alcohol consumption. Skin biopsy was performed on the subcutaneous nodule for histology and microbiological culture. Three weeks later, his right thumb became gangrenous and the skin of his right second, fourth, and fifth digits became purplish-blue in color (Fig 1, B). The first histopathology finding showed large oval-shaped aggregates of mixed inflammatory cells, histiocytes, and multiple giant cells with negative special stains for bacteria, mycobacteria, and fungi. Results of tissue cultures were also all normal. Granulomatous vasculitis was suspected.

Absent pulses of digital thumb arteries and radial artery of the right hand were found by digital palpation and confirmed with a Doppler probe. New fluctuant swelling of the right thenar eminence was also recognized. In addition, he appeared moderately pale and mildly jaundiced on examination. Angiogram of the right arm found total occlusion of radial and ulnar arteries with collateral flow to the hand. Balloon angioplasty of the ulnar artery was introduced. The abscess in the right thenar was drained and debrided; however, laboratory tests failed to identify microorganisms from the pus.

Histologic sections of the subcutaneous nodule were reviewed and favored an infectious process, as there was a large suppurative granuloma apart from the oval-shaped granuloma resembling an obliterated vessel (Fig 2, A-C). Special Verhoeffvan Gieson stains were requested to identify elastic fibers of vascular structures but could not be seen. The oval-shaped granuloma was identified as a nonvascular structure (Fig 2, D), and many broad ribbonlike hyphae were seen by Gomori methenamine silver (GMS) stain both in the suppurative and oval-shaped granulomas (Fig 2, E and F). At that time, members of the genus Pythium—an angioinvasive funguslike microorganism-were also considered possible pathogens in this susceptible patient, so a serologic test for *Pythium* antibody was performed with a weakly positive result. After the third debridement of the thenar area, Pythium insidiosum was eventually identified from debrided tissue and pus (Fig 3).

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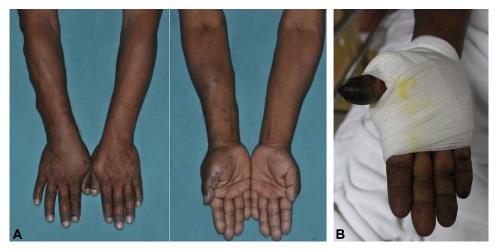


Fig 1. Clinical manifestations. A, Multiple subcutaneous nodules along the right forearm with blue-colored skin of the right thumb. B, Gangrenous right thumb with blue-colored skin of the right second, fourth, and fifth digits.

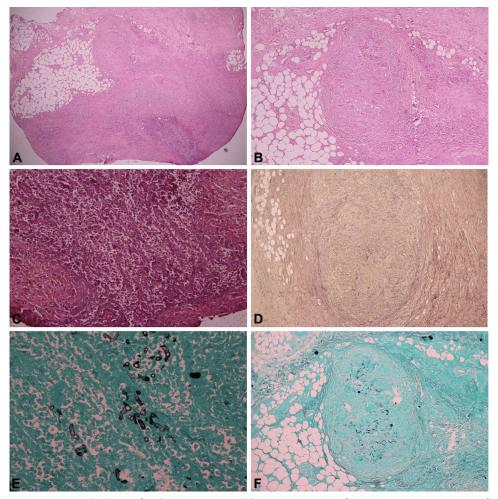


Fig 2. Histopathology of subcutaneous nodule. A, Low magnification. B, Aggregration of mixed inflammatory cells, histiocytes, and multiple giant cells. C, Large suppurative granuloma. D, Verhoff-Van Gieson stain shows negative results. E, GMS stain shows broad, ribbonlike hyphae in a high magnification. **F**, GMS stain show broad, ribbonlike hyphae in the granuloma.

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