Background: *Candida viswanathii* is a rare Candida first isolated in 1959 from the cerebrospinal fluid of a fatal case of child meningitis. Since then, it has been isolated from non-human sources such as the gill of a fish in Indian Ocean and the soil in South Africa in 1967 and 1971 respectively. The second report of *Candida viswanathii* was published in 1976 describing yet another case of fatal meningitis in a teenager. There has been no further report of human infection thereafter. The pathogenicity in human and the spectrum of diseases which it may cause remain unknown.

Material/methods: Herein we describe a case of knee septic arthritis and osteomyelitis caused by *Candida viswanathii*.

Results: A 78-year-old lady presented with fever and acute worsening of her left knee pain one week after an intra-articular injection of lignocaine for her chronic osteoarthritis. Physical examination and laboratory findings from the joint aspiration were consistent with a septic arthritis. Gram stain revealed blastoconidia and the preliminary fungal microscopy and germ tube testing results suggested the growth of a non-*Candida albicans* species.

She was started on empirical intravenous fluconazole 400mg daily after the joint aspiration and on day six of treatment, she had undergone an arthrotomy but her fever and knee symptoms did not improve significantly. As such, her anti-fungal therapy was switched to oral voriconazole (400mg twice daily for 2 days then 200mg twice daily thereafter) on day sixteen of fluconazole while the identification of this Candida species remained unknown. After two weeks of voriconazole, an X-ray of the knee revealed osteomyelitis despite a significant improvement of her symptoms and the final report of the fungal culture revealed a rare species of Candida – *Candida viswanathii*. Its identification was confirmed by both phenotypical and molecular method. A second knee arthroscopy again grew the same Candida species and hence a repeat arthroscopy was performed on day twenty-one of voriconazole. The Sensititre testing for fluconazole had revealed the MIC had increased four times to 4mcg/ml after 16 days use. Similarly, voriconazole MIC had also increased four folds to 0.5 mcg/ml after three weeks use.

After two arthroscopies and 3 months into her voriconazole treatment, her knee symptoms had completely resolved. Her white blood cell count, C-reactive protein and erythrocyte sedimentation rate which were significantly raised at the beginning had normalised too. She went on to receive 3 more months of anti-fungal as per the guidelines by the Infectious Diseases Society of America for the management of Candida osteomyelitis.

Conclusions: We described the first case report of a knee septic arthritis caused by *Candida viswanathii* which was successfully treated with oral voriconazole. To our knowledge, this is also the first non-meningitis case caused by this rare Candida species.