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Adult onset still's disease and pigment cast nephropathy triggered by Chikungunya virus

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Despite Chikungunya virus was considered a benign illness, it is very important to recall that severe atypical manifestations can occur generating high morbidity rates. Early detection of these complications may help clinicians to improve clinical outcomes.

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The adult-onset Still's disease (AOSD) is a rare inflammatory condition that involves many systems, with an etiology that has not been yet clearly established. Diagnosis is done by exclusion of infectious, autoimmune and malignant conditions and its clinical presentation may range from self-limited to clinical manifestations of chronic arthralgias and systemic symptoms associated with rare medical conditions that have been described in the literature (1).

In 2015 our group reported two cases of a hyperferritinemic syndromes; Still's disease and catastrophic antiphospholipid syndrome triggered by Chikungunya virus infection. This time an immunohistochemical study of a kidney biopsy was performed in one of the patients (2).

This case was about a 32-year old male patient with Chikungunya virus and renal function tests compatible with acute kidney injury network (AKIN) III acute renal injury with dialysis requirement.

Kidney biopsy showed normal glomeruli, diffuse tubular epithelial injury including tubulorrhexis, focal

regeneration and mild focal interstitial inflammation. There were numerous bright red granular casts (Figure 1A) that were positive with specific immunostaining for myoglobin (Figure 1B).

Renal involvement secondary to Chikungunya infection is a very atypical manifestation of this disease. Acute renal failure and nephritis with proteinuria and hematuria are the two main renal compromise described in the literature (3,4).

This rare case of pigment cast nephropathy evidenced in our patient has not been reported with Chikungunya infection and only one case report of this nephropathy have been associated with Dengue virus infection (5). Pigment cast nephropathy can occur in a setting of severe rhabdomyolysis that manifests in acute renal failure that even may need renal replacement therapy like in our patient. In a study conducted by Salgado et al, a direct viral invasion of skeletal fibers that could explain the pathogenesis of rhabdomyolysis in these patients was suggested but the definite mechanisms

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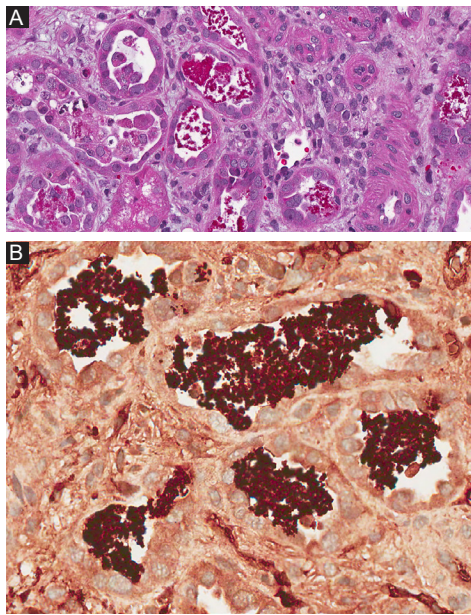


Figure 1. (A) Photomicrograph shows a section from the kidney with numerous bright red granular casts (H&E, $\times 40$ original). (B) Photomicrograph shows a section from the kidney with Myoglobin positive casts in the tubules (immunohistochemistry, $\times 40$ original, and myoglobin).

are still unknown (6). It has been estimated that factors associated with severity and chronicity of Chikungunya infection are age of more 60 years, previous joint disease, unusual host response and persistent titles of circulating specific Immunoglobulin G antibodies (7, 8). Despite Chikungunya virus was considered a benign illness, it is very important to recall that severe atypical manifestations can occur generating high morbidity rates. Early detection of these complications may help clinicians to improve clinical outcomes.

Authors' contribution

EPN: Initial draft, IPO: References, JNE: Preparing final manuscript, PAM: References, CAJ: Biopsy images, GJT: Preparing manuscript and final revision. All authors read and signed the final paper.

Conflicts of interest

There were no points of conflicts.

Ethical considerations

Ethical issues (including plagiarism, data fabrication, double publication) have been completely observed by the authors. The patient has given his informed consent regarding this case report.

Financial disclosures

None.

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