Pediatric Rheumatology

Poster presentation

Wegener's Granulomatosis: paediatric presentation with ischaemia of the feet and novel use of hyperbaric oxygen

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Wegener's Granulomatosis is a rare, chronic, multisystemic vasculitis, of unknown aetiology, affecting mainly the upper and lower respiratory tracts together with glomerulonephritis. However, the disease may involve any other organ. We report a 14-year-old previously healthy girl, who presented with severe ischaemic changes to both feet. 4 weeks prior to presentation she had an episode of epistaxis and conjunctivitis. She developed a blister on the distal dorsal aspect of her right foot, with bluish discoloration and pain. Within one week both feet had become ischaemic and swollen. The diagnosis of Wegener's was made on the basis of skin histology (leucocytoclastic vasculitis), strongly positive PR3 ANCA, focal segmental glomerulonephritis on kidney biopsy and a nodule on chest CT, with associated raised inflammatory markers.

She was treated with intravenous antibiotics, high dose steroids, and daily Iloprost infusions for 6 weeks, daily heparin, intravenous immunoglobulin, plasmapheresis, cyclophosphamide, rituximab and 30 sessions of hyperbaric oxygen. There was no further progression of the ischaemia of her feet and she now has islands of healthy tissue and granulation tissue visible in previously ischaemic areas. Currently, she has preservation of the left forefoot with digital ischaemia of her 4th and 5th toes. Her right toes have dry gangrene and are mummified. Presentation with peripheral ischaemia in Wegener's granulomatosis is extremely rare in the paediatric age group [1]. We report on a case that presented with peripheral ischaemia that responded to hyperbaric oxygen therapy as a novel adjunct to immunosuppressive therapy [2].

References

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