Osteomyelitis is a recognized complication of pyomyositis in general [1] and ischial osteomyelitis has been reported in association with obturator pyomyositis [4]. Our patient has shown resolution of the abscesses with no evidence of osteomyelitis on follow-up MRI scans.

A new finding in our case is this patient’s bilateral obturator involvement, a phenomenon not reported before. We have also documented the importance of repeated imaging in such a scenario. Single imaging would have missed the development of bilateral changes. Obturator pyomyositis is an important differential diagnosis of acute pelvic pain.

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Rituximab inefficiency during type I cryoglobulinaemia

Sir, Rituximab, a chimeric antibody binding to the human B-cell antigen CD20, is effective and safe as adjuvant or primary therapy in several B-cell lymphomas. It has also been used in other haematological disorders sustained by B-cell lymphoproliferation and appears promising in some autoimmune diseases. Rituximab also has proved efficiency during type II mixed cryoglobulinaemia (Cg) [1, 2], reducing cryoglobulin levels and significantly improving clinical parameters in most cases. The same efficiency was recently reported in a patient with vasculitis-associated type I Cg [3]. On the contrary, here, we report two cases of patients with type I Cg who were resistant to rituximab and discuss these contrasting results.

Case 1 was a 31-yr-old woman who presented with necrotic cold-sensitive purpura and arthralgias. The symptoms had begun 5 yr previously and were related to an immunoglobulin (Ig) G cryoglobulin, resistant to steroids. She had no other medical or surgical history. The routine laboratory evaluation was normal. Quantification of cryoglobulin followed by immunofixation identified an IgGλ type I Cg (0.94 g/l) according to published methods [4]. Bone marrow biopsy revealed mild infiltration by monotypic λ CD20+ plasmacytes (9%) without morphological abnormalities. The thoracic and abdominal CT scan and skeletal radiography were normal. High-dose intravenous corticosteroids and cyclophosphamide did not improve the symptoms. The patient was stabilized only with repeated plasmapheresis. Rituximab was started (375 mg/m² intravenously, weekly for four courses). Unfortunately, cutaneous lesions progressed and cryoglobulin levels increased.

High-dose cyclophosphamide chemotherapy and autologous stem-cell transplantation were recently performed.

Case 2 was a 45-yr-old woman who presented with nephrotic syndrome (proteinuria 3.0 g/24h) and leg purpura. She had been partially thyroidectomized a few years ago for a goitre. Standard biology was normal except for hypoalbuminaemia. Further investigations revealed a low-level IgM cryoglobulin and hypocomplementaemia. According to the classification of Brouet and colleagues [4], the cryoprecipitate was made of a monoclonal IgMx type I Cg with no other component identified by immunofixation; the rheumatoid factor titre was negative (ELISA test and nephelometry). Bone marrow biopsy detected monoclonal IgMx CD20+ mature lymphocytosis (6%). The thoracic and abdominal CT scan was normal. Renal biopsy confirmed a membranoproliferative glomerulonephritis with IgMx and complement deposits. A regimen of steroids, azathioprine, chlorambucil and cyclophosphamide–vincristine–prednisone was unable to control the lymphoproliferation. Plasmapheresis partially improved cutaneous lesions and nephrotic syndrome. Rituximab was administered (375 mg/m², weekly, twice). Three days after the second infusion, a severe cutaneous flare occurred; 9 days later, the patient presented with an acute oliguric transient renal insufficiency (proteinuria 9.5 g/24h). The monoclonal IgMx level rose from 1 g/dl before rituximab to 4.9 g/dl. After additional steroids and plasmapheresis, chemotherapy intensification and stem-cell transplantation were then successfully performed.

Rituximab has already been successfully used in several cases of autoimmune-mediated manifestations related to a monoclonal Ig production [1, 2, 3, 5, 6]. Rosenthal and colleagues described a patient with polyneuropathy and vasculitis associated with a type I Cg whose evolution was controlled by rituximab [3]. Unfortunately, the same treatment failed in our two patients. Rituximab is thought to induce B-cell clearance through different mechanisms involving B-cell apoptosis, antibody-dependent cell-mediated cytotoxicity or phagocytosis, and complement-mediated cytotoxicity, the latter probably being a dominant mechanism depending on levels of expression of CD20 and complement regulatory proteins [7, 8]. Plasma cells are not supposed to be sensitive to rituximab as they do not express CD20. Yet in our first case we expected some effectiveness on plasma cell precursors, as seen in previous attempts at treatment [9]. However in this case, pathogenic plasma cells may belong to a long-lived population with few renewal capacities from CD20þ plasma cell precursors, and this could explain the short-term failure of rituximab. Alternatively, FcγRIIIa polymorphism has been shown to be associated with clinical and molecular responses to rituximab [8, 10]. In the second case, rituximab administration clearly induced a transient flare. We suggest that CD20 cross-linking might have provoked cryoglobulin release through massive B-cell apoptosis or activation, as has been described during Waldenström’s macroglobulinaemia [6]. In conclusion, although rituximab appears to be a serious alternative in several monoclonal Ig-induced autoimmune manifestations, its use during type I Cg remains questionable and can even exacerbate the disease.

Key messages

- The use of rituximab during type I cryoglobulinaemia is questionable.
- Rituximab can be ineffective and even exacerbate the symptoms.

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Eosinophilic fasciitis and eosinophilic colitis: a rare association

SIR, Although eosinophilic fasciitis (EF) was originally considered to be a disease predominantly localized to the fascia, several case reports have subsequently reported other manifestations [1–7]. Aplastic anaemia, haemolytic anaemia, thrombocytopenia, lymphoproliferative disorders, thyroiditis, pulmonary...