even greater long-term toxicity. A large well controlled prospective trial is needed.

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References

A case of familial Mediterranean fever, Behcet’s disease and polyarteritis nodosa complicated by perirenal haematoma

Sirs,

Familial Mediterranean fever (FMF) is characterized by attacks of fever, accompanied by abdominal, chest or joint pain (1). The most serious complication is the development of amyloidosis, causing chronic renal failure. There are also a number of reports of individuals who have vasculitis associated with FMF (2, 3).

We describe a case of FMF associated with the features of Behcet’s disease (BD) and probable polyarteritis nodosa complicated by spontaneous perirenal and retroperitoneal haematoma. A 37-year-old man was admitted to our hospital with a two-week history of malaise, high fever, severe myalgia and severe abdominal and right flank pain in the last four days. He had a history of recurrent abdominal attacks and fever since the age of 8. He also reported periodic arthritis independent of the abdominal attacks. He had been diagnosed as having FMF at the age of 20 and colchicine 1.5 mg/day, in addition to colchicine 1.5 mg/day, cyclophosphamide 2 mg/kg/day was administered orally this time. No further attack has been observed during the follow-up period since he was discharged from hospital. The patient was readmitted to hospital for recurrent oral ulcerations and uncontrolled hypertension (200/120 mm/Hg). Abdominal CT showed significant regression of the haematoma. His blood pressure stabilised with antihypertensive drugs and, in addition to colchicine 1.5 mg/day, cyclophosphamide 2 mg/kg/day was administered orally this time. No further attack has been observed during the follow-up period since he was discharged from hospital.

The presented patient fulfills the described criteria for FMF by Livneh et al. (4). BD was diagnosed on the basis of diagnostic criteria proposed by the International Study Group of Behcet’s disease (5).

Reports exist showing the association between FMF and PAN (3). FMF and Henoch-Schönlein purpura (2). FMF and

Fig. 1. Abdominal computed tomography revealing a right perirenal and retroperitoneal haematoma.
BD have also been described in the same patients (6). Whether there is an association between these two diseases or the occurrence is due to the high prevalence of these 2 diseases in the same area is not clear. Recently, Toutou et al. (7) screened MEFV mutations in a panel of BD patients and found that the M94V, V726A, and E148Q mutations tended to be more frequent in definite BD (2.6%, 2.6% and 5.2% respectively) than in controls (0%, 0% and 2.2% respectively), and suggested that they may act as an additional susceptibility factor in BD. Schwartz et al. (6) showed that the prevalence of BD was higher in FMF than in populations rich in BD (e.g., 16 per 4000 in FMF compared with 1 per 104 in Japan).

In our case, it may be difficult to make a diagnosis of PAN, as perirenal and retroperitoneal haematoma secondary to renal arterial aneurysm has been reported in two patients with BD to date (8, 9). Therefore, angiographic findings and retroperitoneal haematoma may be due to BD in this case. However, thrombophlebitis and venous thrombosis are often seen at or around the same time as arterial aneurysms in patients with BD (10, 11), and since our patient neither had any history of these nor was observed to have such manifestations during the follow-up period, we can conclude that the angiographic findings and retroperitoneal haematoma may also be due to PAN. Our case met the ACR criteria (12) and the angiographic findings and retroperitoneal haematoma may also be due to PAN. Further studies will enlighten the association of these diseases and shed light on the pathogenesis of inflammation.

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References

Letters to the Editor

Reporting a desensitization protocol for colchicine treatment

Sirs,

The development of toxicity and hypersensitivity to colchicine therapy is a well-known entity in patients with familial Mediterranean fever (FMF) and Behçet’s disease (BD) (1, 2). On the other hand amyloidosis is a known sequel of untreated patients affecting mainly patients with FMF, and therefore there is a need to treat such patients with colchicine to prevent this complication.

We recently examined a 47-year-old female with FMF in whom the diagnosis of BD manifested as recurrent oral and vaginal ulcers, pyoderma gangrenosum, erythema nodosum and arthralgias was established 20 years ago. The treatment regimen prescribed for BD as well as FMF included methotrexate, cyclophosphamide and colchicine which led to partial remission of both diseases.

The patient developed an allergic skin reaction to colchicine, proven by an oral challenge test. Despite the allergic reaction that erupted as a maculopapular rash evident on the lower limbs, it was necessary to continue the colchicine therapy because of its prominent role in preventing the development of amyloidosis. Desensitization was achieved with the oral administration of increasing doses of colchicine. Colchicine 0.5 mg was dissolved in 500 cc of glucose 5%. On the first day of treatment 0.5 cc was administered, and the dose was doubled each day until a dose of 250 cc (0.25 mg colchicine) was reached on the tenth and last day of treatment. No adverse effects were observed during the 10 day desensitization period. At the present time, two months after the desensitization process, the patient is receiving 1 mg colchicine/day, with no evidence of an allergic reaction.

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