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Case Report Open Access

Acute polyarthritis revealing celiac disease of the elderly

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Abstract

Introduction

Celiac disease (CD) is an autoimmune disease that typically occurs in childhood and young age. Late-onset forms (after 60 years) are rare and often underestimated in current medical practice, as is joint damage during this disease. We report an original case of CD in an elderly patient revealed by acute polyarthritis.

Observation

A 62-year-old patient with no significant pathological history was explored for acute polyarthritis of both large and small joints. The physical examination noted synovitis of the hands and wrists without articular deformities, and a marked mucocutaneous pallor. The biology revealed a microcytic anemia at 8 g/dl with stigmas of malabsorption (low cholesterol and hypocalcemia). No inflammatory syndrome was noted. Joint X-rays were without abnormalities. Immunological tests for chronic inflammatory rheumatisms were negative. Gastroduodenal fibroscopy was performed and histological examination of duodenal biopsies showed diffuse villous atrophy with marked inflammatory infiltrate. Anti-endomysium and anti-transglutaminase antibodies were positive confirming the diagnosis of CD. Under gluten-free diet, the evolution was favorable with totally disappearance of articular signs.

Conclusions

Joint manifestations during CD are rare. The revealing forms are exceptional and can make a differential diagnosis with chronic inflammatory rheumatism, in particular that associations with CD was described. CD deserves to be evoked in front of any unexplained arthritis of the elderly, even without any specific digestive symptomatology of this disease.

Keywords

Celiac disease, arthritis, synovitis, joint, elderly.

Introduction

Celiac disease (CD) is a rare ubiquitous autoimmune disease; its global prevalence is estimated at 1-2% of the general population [1,2]. It is caused by a food antigen that is gliadin (one of principal protein groups of gluten), inducing the production of autoantibodies (anti-gliadin, anti-reticulin, anti-endomysium and anti-transglutaminase), and resulting, in genetically predisposed subjects, to intestinal villous atrophy [1,2]. It is currently considered not only a digestive disease, but rather a systemic one with multi-organ involvement and very polymorphic clinical manifestations [3,4]. This makes CD a real diagnostic challenge for clinicians, especially in the adult and elderly forms [2], and justifying its qualification as a "great simulator" [5].

It is a classically infantile disease, but can occur at any age [6,7]; late-onset forms in the elderly remain exceptional and unusual [6].

His clinic is highly variable, ranging from symptomatic forms with mainly diarrhea, malabsorption syndrome, and weight loss, to completely asymptomatic forms called "silent CD" [1,2]. Among the systemic manifestations of CD, osteoarticular involvement is rare and particularly noted in old and advanced forms of the disease [2,3].

Arthritis remains a rare manifestation of this disease, and described by the majority of authors as "atypical and unusual" [2,8].

We report an original case of CD of the elderly revealed by acute polyarthritis.

Case Report

A 62-year-old female with no significant pathological history was explored for acute inflammatory polyarthritis of both large and small joints. His joint symptomatology had been evolving for two weeks, with no obvious triggering factor, and not responding to analgesics and nonsteroidal anti-inflammatory drugs prescribed by his family doctor.

The examination noted swollen and painful joints (two wrists, two ankles, the proximal and distal metacarpophalangeal joints, and the carpometacarpal joints of both hands) without articular deformities. The mobilization of the joints was painful but without limitation of the mobility sectors. There were no periarticular cutaneous lesions, lymphangitis, or satellite adenopathies. The examination noted, moreover, a marked cutaneous and conjunctival pallor.

The biology revealed microcytic anemia at 8 g/dl with stigmas of malabsorption syndrome (low cholesterol and hypocalcemia). There was no inflammatory syndrome (erythrocyte sedimentation rate, C-reactive protein, and electrophoresis of serum proteins were normal).

The other basic biological tests were within normal limits: leukocytes, platelets, fast blood glucose, creatinine, ionogram, liver tests, muscle enzymes, alkaline phosphatase, TSH, fT4, T3, and urine analysis.

The joint X-rays were without abnormalities, as were the chest X-ray and the electrocardiogram. Immunological tests for chronic inflammatory rheumatism were negative (anti-nuclear antibodies, soluble anti-nuclear antigen antibodies, latex and waaler-rose reactions, and anti-CCP antibodies).

Gastroduodenal fibroscopy was performed in the presence of malabsorption syndrome (deficiency anemia, hypocholesterolemia and hypocalcemia) and histological examination of duodenal biopsies showed diffuse villous atrophy with a marked inflammatory infiltrate of lymphocytes and monocytes consistent with the diagnosis of CD.

Anti-endomysial and anti-transglutaminase autoantibodies were strongly positive, confirming the diagnosis of CD. Under gluten-free diet, the evolution was favorable with disappearance of the articular signs after one month and normalization of the biological balance after six months. No recurrence of joint complaints had been noted for three years now.

Discussion

The overall incidence of CD in the elderly is estimated to be around 7.2% [9], but it is mainly forms diagnosed since the pediatric age. Cases of late-onset CD, diagnosed after age 65, are exceptional and uncommon; indeed, in the large study of Tortora R et al, only 2.5% of CDs were diagnosed in subjects aged 65 years or older [6], and the incidence of CD in subjects over 55 was estimated at 0.08% in the Vilppula A et al population study [7]. However, these late forms are increasingly encountered in routine medical practice, mainly because of the accessibility to serological diagnosis of the disease [10], and it is nowadays admitted that, overall 34% of newly diagnosed CD patients are over the age of 60 [11].

Osteoarticular manifestations of CD include: inflammatory polyarthralgia, seronegative arthritis, sacroiliitis, seronegative spondylarthritis, osteoporosis, osteomalacia, and bone fractures [2,3,12,13]. Joint involvement can sometimes be major and prominent during CD [8], and may precede the diagnosis of the disease by a variable delay of up to 15 years [8,14].

Arthritis specific to CD is classically a seronegative polyarthritis of the large joints [15,16]; more rarely oligoarthritis [17,18] and monoarthritis [19] can be noted.

This arthritis may be axial or peripheral [12,13], sometimes erosive [3], and all joints may be affected with a predilection for the lumbar spine, hips, and knees [8,13,14]. This arthritis, during the course of the CD, can be isolated or sometimes be integrated within the framework of a serious multi-systemic attack of the disease [20].

The arthritis revealing the disease is exceptional. They can be seen in the child [14] and also in the adult and the elderly subject, inaugurating a CD until there "silent" [16,17,21,22].

The lack of knowledge by clinicians of joint involvement during CD means that these patients are often misdiagnosed and poorly managed, and the diagnostic delays is often important [23]. At the same time, the frequency of joint damage during CD is still under-estimated [12]; in fact, systematic prospective screening found arthritis in 31.1% of patients in Iagnocco A et al study (23/74 patients) [24], and in 37% of patients in Iqbal T et al study (131/356 patients) [13]. Similarly, systematic ultrasound evaluation noted the arthritis in 31.7% of newly diagnosed CD's children in the Grag K et al series [23].

The exact mechanism of arthritis during CD remains unclear. It is most likely immunological (immune-mediated synovitis) involving gluten itself as well as autoantibodies specific to CD [25].

The causality relationship between CD/gluten and arthritis was confirmed by the systematic ultrasound study of Iagnocco A et al, where synovitis was significantly more found in subjects with CD and gluten-containing diet compared to those with CD and well observant of gluten-free diet: 50% versus 11.1%, p = 0.007 [24].

These findings were also validated for adult CD: 41% versus 21.6%, p <0.005 [12]. Thus, the authors recommend that CD should be considered for any arthritis that is not proven at any age [19].

However, the occurrence of arthritis in a subject with suspected or diagnosed CD requires further investigation as it may be a sign of other chronic inflammatory rheumatism, often associated with CD itself, such as psoriatic arthritis [26], Familial Mediterranean Fever [27], ankylosing spondylitis, systemic lupus erythematosus, primary Sjögren's syndrome [13], and juvenile chronic arthritis [28].

The treatment of arthritis associated with CD is not specific. Usually the only gluten-free diet, especially if good compliance, allows the dramatic improvement of all osteoarticular involvement associated with CD [3,8,14,19,21].

Conclusion

Joint manifestations during CD are rare and those revealing the disease are exceptional. They can pose the differential diagnosis with several chronic inflammatory rheumatism (rheumatoid arthritis, juvenile chronic arthritis, psoriatic arthritis, ankylosing spondylitis, and systemic lupus erythematous) and represent a real diagnostic challenge for the clinician, especially that the associations of CD with these chronic rheumatism are described. The CD deserves to be evoked and screened in front of any unexplained arthritis of the elderly, even outside any specific digestive symptomatology of this disease.

Observation

Anti-CCP: anti-cyclic citrullinated peptide antibodies, fT4: free tetra-iodothyronine, T3: triiodothyronine, TSH: Thyroid Stimulating Hormone.

Conflicts of Interest

No conflicts

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