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Monoarthritis of the ankle revealing idiopathic granulomatous mastitis in a pregnant woman

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Abstract
Idiopathic granulomatous mastitis (IGM) is a very rare clinical-histological entity that affects young women of childbearing age and represents a real diagnostic and therapeutic challenge for clinicians.
Systemic manifestations during IGM are extremely rare. The most reported manifestations are: erythema nodosum, inflammatory arthralgia, fever, peripheral lymphadenopathies, and scleritis/episcleritis. Arthritis as extra-mammary manifestations of IGM remains exceptional, and revealing forms are unusual.
We report an original case of IGM occurring in a pregnant woman, whose inaugural symptom was an acute and isolated monoarthritis of the left ankle.

Keywords: idiopathic granulomatous mastitis, monoarthritis, ankle, pregnancy

Introduction

Idiopathic granulomatous mastitis (IGM) is a very rare clinical-histological entity that affects young women of childbearing age [1, 2].

It was described for the first time in 1972 by Kessler and Wolloch [3] and later characterized in 1977 by Cohen C [4], but remained controversial for a long time [5]. It still represents a real diagnostic and therapeutic challenge for clinicians [1, 2, 5], and the majority of patients are misdiagnosed by their primary care physicians because of non-specific presentations [5]. Indeed, the clinical and radiological presentation can mimic breast cancer (carcinomatous mastitis) and lead to mutilating and unjustified surgical procedures [2]. In addition to breast cancer, differential diagnosis of IGM may occur with mammary tuberculosis, sarcoidosis, fungal infections, atypical mycobacterioses, granulomatous systemic vasculitis, and foreign body reaction [1, 6, 7]. Thus this entity must always remain a diagnosis of exclusion [1, 5, 6].

Its etiopathogenesis is unknown until today [2, 5]. Several pathophysiological hypotheses have been suggested; the most plausible are: autoimmunity, infection, hormonal disruption, and autoinflammation [2, 5]. Certain trigger factors have also been mentioned to explain the genesis of these granulomatous lesions: pregnancy, lactation, hyperprolactinemia, oral contraception, smoking, local breast trauma, infection, alpha-1-antitrypsin deficiency, and immune disorders [2, 6].

IGM remains an exceptional and unusual cause of acute arthritis [1, 2, 8], especially in pregnant woman [2, 8, 9].

We report an original case of IGM occurring in a pregnant woman, whose inaugural symptom was an acute and isolated monoarthritis of the left ankle.

Case report

A 34-year-old Tunisian woman with no pathological medical history, pregnant at 29 weeks of amenorrhea, was hospitalized in the orthopedic department for acute pain and total impotence of the left ankle, without trauma or lesion of the adjacent soft tissues. The diagnosis of acute and isolated monoarthritis of the leftt ankle was discussed and an injectable antibiotic therapy was initiated as the patient was feverish at 39°C, leukocytes at 12000/mm^3, and C-reactive protein at 80mg/l.

The infectious investigation was negative and no improvement was noted leading to stopping antibiotics and prescribing a no steroidal anti-inflammatory drug. The evolution was rapidly favorable. One week later, the patient reported pain with left breast induration.

The somatic examination noted at the level of the infero-external quadrant of the left breast the existence of an indurated, sensitive lesion, 5cm in diameter, with inflammation of the overlying skin and ipsilateral axillary lymphadenopathies. Basic biology was without specific abnormalities. Mammary ultrasound showed an hypoechoic, heterogeneous, and poorly limited lesion of the infero-external quadrant of the left breast. This lesion contained anechoic focal zones evoking areas of necrosis (Fig.1).

Fine needle aspiration cytology resulted in granulomatous lobular mastitis without cancerous cells or caseous necrosis.

Subsequent investigations were negative (antinuclear antibodies, C and P antineutrophil cytoplasmic antibodies, serum calcium, angiotensin converting enzyme, liver tests, electrophoresis of serum proteins, blood and urine cultures, tuberculin skin test, Quanti FERON test, acid-fast bacilli stainings, polymerase chain reaction test, bacterial and fungal cultures of the breast tissue) excluding tuberculosis, sarcoidosis, atypical mycobacterioses,

fungal infections, and granulomatous systemic vasculitis. The diagnosis of IGM was retained. The evolution was marked by central ulceration of the breast lesion after one week (Fig. 1 & 2). The patient was treated with systemic corticosteroids and colchicine with a favorable outcome for both ankle arthritis and breast lesion

Figures

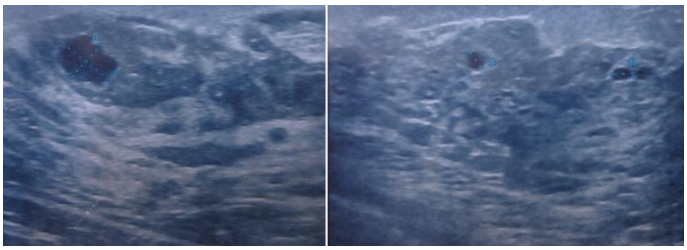

Fig 1: Mammary ultrasound: hypoechoic and heterogeneous lesion of the infero-external quadrant of the left breast containing focal zones of necrosis.

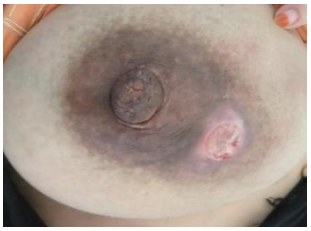

Fig 2: 2 cm ulcerated lesion of the left breast.

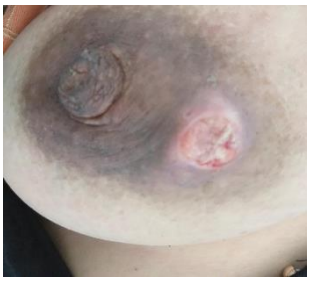

Fig 3: 5cm inflammatory induration of the infero-external quadrant of the left breast centered by an ulcerated lesion.

Discussion

Systemic manifestations during IGM are known to be extremely rare [1, 6, 10]. The most reported manifestations in the literature are: erythema nodosum, inflammatory arthralgia, fever, peripheral lymphadenopathies, and scleritis/episcleritis [1, 6-8].

Arthritis as extra-mammary manifestations of IGM remains rare, and revealing forms are exceptional and unusual [1, 2, 6, 7]. Indeed, Alungal J *et al*'s review of the literature done in 2016 found only ten patients with IGM associated with arthritis and erythema nodosum [6]. The occurrence of this entity in association with arthritis during pregnancy is still much rarer; the review made by Vural S *et al.* in 2017 noted only two patients with IGM and arthritis occurring during pregnancy [2]

Joint involvement in IGM may be monoarthritis [8, 11], oligoarthritis [1, 2, 7, 9], or polyarthritis [6], and arthritis may be uni-[1, 2, 8, 9] or more rarely bi-lateral [6, 7]. All joints can be affected; ankles, knees, and metacarpophalangeal appear to be the most affected joints during IGM [1, 2, 6-9].

The association with arthritis, and moreover if cutaneous lesions (erythema nodosum), as well as the occurrence during pregnancy reinforce once again the autoimmune hypothesis of IGM [5, 7].

In the majority of reported cases, arthritis occurs in previously known patients with IGM or synchronous with the breast lesion [1, 6, 8, 9].

Our observation is distinguished by the inaugural character of arthritis of the ankle; indeed, arthritis was the first symptom of the disease, having preceded by one week the appearance of the breast lesion. This chronology has not been reported before.

IGM forms associated with systemic manifestations (including arthritis) typically respond favorably to systemic corticosteroids [6, 7]. Nonsteroidal anti-inflammatory drugs, particularly indometacin, may also be effective [2]. Forms resistant to no steroidal anti-inflammatory drugs can be favorably treated with colchicine [2]. Otherwise, in the localized forms of IGM the treatment of choice remains surgery with extensive excision of lesions often preceded by systemic corticosteroids [1, 2, 6-9].

Conclusion

Idiopathic granulomatous mastitis remains an exceptional breast disease of young women, often neglected and misdiagnosed. It can rarely associate with extra mammary signs that indicate the systemic nature of this condition and its probable autoimmune origin.

Arthritis remains an exceptional and unusual extra mammary manifestation of this granulomatosis.

Our observation is characterized by its occurrence during pregnancy, and by the inaugural character of arthritis that revealed IGM.

Conflicts of interest: None

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