Primitive breast localisation of Buruli ulcer in an endemic zone: a rare case

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Summary

A case of Buruli ulcer with primitive breast localisation with evident epidemiological and clinical aspects is reported. This localisation is exceptional; the differential diagnosis with breast cancer is essential. If diagnosed early, it can be cured with surgery, broad-spectrum antibiotherapy and thermotherapy.

Key words: Breast; Buruli ulcer; Mycobacterium ulcerans.

Introduction

Buruli ulcer is an infectious skin disease caused by *Mycobacterium ulcerans*. In many cases Buruli ulcer is probably the third most common mycobacterial disease after tuberculosis and leprosy among immunocompetent patients. The first case was described by MacCallum *et al*. in Australia in 1948 [1]. The disease is absent in Europe, and affects mainly Africa, Oceania and South America. The Ivory Coast is one of the countries in the world most affected by this disease. Between 1988 and 1997 over 10,000 cases were registered [2, 3]. The upper and lower limbs have always been the preferred location of the disease. In 2005 an Ivorian team reported two cases of Buruli ulcer with genital localization in humans [4]. A case of primitive breast localization in a woman is presented together with the clinic characteristics, diagnostic and therapeutic procedures.

Case Report

A 37-year-old illiterate woman living in Adzope, a highly endemic area, discovered a painless nodule in the left breast by self breast examination in June 2005. She had four children and no previous history of breast pathology. She did not consult a health centre for two years given the absence of symptoms. The patient was consulted in August 2007 by the Breast Pathology Service of CHU Cocody (Abidjan, Ivory Coast). Her second child had suffered from a Buruli ulcer of the left knee; he was cured after treatment in a specialised centre in 1999. The general condition of the patient remained unchanged. Examination of the left breast showed a necrotising ulcerative lesion (6 cm in diameter in the periareolar area with a portion of the areola). The edges were raised and palpation did not reveal any nodule under the lesion. There were no palpable axillary lymph nodes and other areas were normal.

Mammography and CA 15-3 analyses were carried out.

The biological samples revealed the presence of acid alcohol resistant bacillus. Mammography did not show suspicious lesions of malignancy. Polymerase chain reaction (PCR) was not carried out. Carcinoembryonic antigen and CA 15-3 were normal. Care of this patient required a multi-disciplinary team (Dermatology, Infectious diseases and Senology). Excision of the necrotic area under general anaesthesia was performed followed by local disinfectant solution for ten days, with dressings every other day. Broad-spectrum antibiotherapy and local heating to 40-50°C with serum helped heal the injury.

Discussion

According to the World Health Organization (WHO) at least 27 countries around the world have reported Buruli ulcers, mostly in tropical regions. In several of these countries, this disease is not considered as a public health threat [2]. The Ivory Coast is a country with a high endemicity of Buruli ulcers and the region of Adzope is one of five centers selected by the WHO for treatment-monitoring-evaluation of this ailment [2-4]. This region was where our patient resided. One of her children had had *Mycobacterium ulcerans*. The disease often occurs in the vicinity of ground water and aquatic insects belonging to the genus Naucoris (Naucoridae family) and Diplonychus (Belostomatidae family) which are responsible for this ailment. Some authors argue that the mode of transmission of this disease is not fully understood. They believe the lifestyle of people who have permanent contact with water is not the main element in the transmission chain. For these authors, a prior traumatic injury is the gateway to the germ [5, 6]. In our patient previous trauma in the breast was not found, nor a primitive location. The breast lesion was the first site of an outbreak of the disease; data from the literature does not note primitive breast localisation in women. Two genital localisation cases reported by Sica *et al*. [4] in the same region were all secondary sites and primitive injuries were on the lower limbs. The predominance of this infection

Revised manuscript accepted for publication March 25, 2009

Figure 1. — The breast lesion.

Figure 2. — The ulcer after treatment.

among children aged from 2-14 years and among women has been recognised, however no racial or socio-economic class is safe [7]. Female domestic chores, in an African context (especially in rural areas), and bathing children in rivers can explain this distribution in relation to the age and sex. Epidemiology has a key in establishing the diagnosis of Buruli ulcers. It should be considered with any nodule or ulcer in an endemic area until proven otherwise. Moreover, the clinical aspects outlined by Yamoussoukro [13] allow us to discuss the diagnosis of Buruli ulcers. The case definition used is as follows: infectious disease involving the skin caused by *Mycobacterium ulcerans*. It is characterised by a nodule, papule, plaque or painless swelling evolving into a painless ulcer, often leading to crippling after-effects [2]. In our case, although the patient did not present at the nodular stage of the disease there was slow ulceration, painless necrosis with a background and edges slightly off. In an epidemiological context symptoms were evocative enough to make the diagnosis. Bacteriological testing with the presence of acid alcohol resistant germs confirmed the diagnosis. In our health-developing countries, the germ culture has a long duration (10 weeks), and PCR is very expensive [8]. The differential diagnosis of a malignant breast lesion was easy. Mammography and the biopsy results did not allow diagnosis of a primitive malignant breast lesion. Currently, there is not any medicine for *Mycobacterium ulcerans*, and the use of conventional antibiotics (rifampicin and amikacin) alone or in combination have shown limitations. Surgical treatment remains the only recourse; blanket broad-spectrum antibiotics pre- and postoperatively for ten days are required [3-6]. In our case immediate suturing post excision was not performed because of the extensive necrosis. Cleaning and dressing with a daily solution of Dakin® and permanent thermal contacts provided some benefit in reducing the lesion. This therapeutic approach adopted by our team is confirmed by other authors [9, 10]. Some authors [11, 12] believe that rifampicin, amikacin and clarithromycin could promote healing of early pre-ulcerative or ulcerative lesions (< 2 cm), but they are ineffective for extended lesions (> 2 cm). Continuous local warming at 40°C promotes healing, even without excision, if applied consistently for four to six weeks. This therapy would promote blood circulation, penetration of antibiotics and phagocytosis. Hyperbaric oxygenation could also promote healing but it is expensive and not available in many endemic areas [11-13].

Conclusion

Localisation of a primitive breast Buruli ulcer is exceptional. The fear of breast cancer cannot exclude this possible diagnosis, especially in endemic areas. Early diagnosis and adequate treatment of this pathology can achieve healing.

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