CASE REPORT

Leprosy acquired by inoculation from a knee injury

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Summary  This case study reports on the development of clinical leprosy in a young Caucasian female from a non-endemic country who contracted the disease while living in a leprosy endemic country. In the presentation and discussion, some relevant factors will be reviewed and discussed that may play a role in the transmission, susceptibility and clinical development of the disease.

Introduction

The entry point of Mycobacterium leprae into the human body has been debated for a long time.1–6 There have been conflicting statements in the literature about leprosy transmission such as ‘the majority of leprologists believe that the portal of entry is through the skin’ and ‘most leprologists no longer consider the skin to be important as the port of entry or exit of M. leprae’.4,7 Most leprologists now hold the view that the more common route of entry is the upper respiratory tract. There is no conclusive evidence that the skin could not be the portal of entry for M. leprae and that entry through the skin could not lead to clinical leprosy in susceptible individuals. There have been many case studies, pointing to a possible relationship between broken skin, giving entry to M. leprae, and subsequent development of clinical leprosy.8–12 This report presents a case of leprosy in a young Caucasian female and will briefly review and discuss some relevant factors in the transmission and pathogenesis of leprosy.

Case study

The patient was a Caucasian female born in Norway. At the age of 10, while living with her parents in Ethiopia, she sustained a severe gash on the anterior aspect of the right knee while
playing in the school playground. The injury was dressed in the leprosy hospital where her father was employed. In 1985, at 14 years of age, a skin lesion was noted on the front of the right knee at the site of the previous injury. There was still a visible scar from her previous fall. In this case, the incubation time could be calculated as being 4 years.

MEB noted that the area on the knee remained hypopigmented while the remainder of the skin tanned with frequent swimming and sunbathing in the hot and humid climate of another country in which she was living at that time. It was also noted there was no hair growth within the lesion and the area remained dry when she was sweating.

Clinical findings

There was a well-defined hypopigmented skin lesion approximately 2 cm in diameter on the anterior aspect of the right knee, with slight darkening at the centre of the lesion. There was loss of sweating and no hair growth within the lesion. Diminished light touch sensation was present in the skin lesion when tested with cotton wool and monofilaments as compared to surrounding skin and the left knee. There was also diminished sensation to pain tested with pinprick. There were no enlarged or tender nerves and no subjective symptoms of nerve function impairment, e.g. paraesthesia or numbness. Voluntary muscle testing and sensory testing also did not reveal any nerve function impairment. Similarly, electrodiagnostic testing also did not reveal any abnormalities.

Laboratory studies and histopathology

Skin smears in two sites from the right knee were 1+ and 2+ for acid-fast bacilli. All other routine skin smear sites were negative (ears, elbows, and left knee). A biopsy from the edge of the lesion showed mild hyperkeratosis in the epidermis. Within the superficial dermis there was a focal perineural mononuclear cell infiltration. Acid-fast stain showed a few granular organisms inside macrophages. With these findings, the pathology was reported as indeterminate leprosy. A skin test was done with lepromin A with 3 mm induration at 26 days. A biopsy of the lepromin skin test was reported as a borderline lepromatous lepromin reaction.

Treatment and follow-up

Though diagnosis of leprosy in this case was clear, the clinical findings and laboratory studies were not considered definitive for either tuberculoid or lepromatous disease. Therefore, the working diagnosis for treatment purposes was indeterminate leprosy, which might well progress toward lepromatous disease if left untreated. The patient was initially treated with dapsone 100 mg daily and rifampin 600 mg daily. She did not tolerate rifampicin well and this was changed to rifampicin 600 mg monthly. After approximately 6 months, the dapsone was changed to 50 mg daily. She took these medications for a total of about 18 months, at which time treatment was discontinued. Repeat skin smears 3 months and 9 months after start of treatment were negative at all sites. She was seen in follow-up 7 years later. At that time, the
skin lesion on the right knee appeared normal with return of hair growth. There was still slightly diminished sensation to light touch in the lesion. Skin smears at all sites were negative.

Discussion

The Michigan inoculation case studies remain one of the more interesting and earlier studies that reported on the possibility of transmission of *M. leprae* through abraded skin. Two young men in the US military were tattooed in Australia in 1943 at the same time by the same person. They both developed a leprosy lesion 3 years later at the site of the tattoo. Both young men came from a town from which no leprosy case was ever reported. This study concludes ‘...it would be difficult to entertain a suggestion that both men, coming from a strictly non-endemic region, had somehow acquired the latent infection elsewhere and at another time and that it was exteriorised or brought to a focus by the tattooing to become apparent nearly three years after that was done’. In our case study, the skin smears at the site of the lesion were positive but were negative at the routine smear sites. This would support the theory that abraded skin can be an entry point for *M. leprae*.

It is established that *M. leprae* can survive for periods of up to 5 months in the environment. *M. leprae* is discharged in large quantities from lepromatous leprosy patients from the upper respiratory tract and the skin. Household contacts of MB (multibacillary) patients have a higher risk of developing leprosy than household contacts of PB (paucibacillary) leprosy patients. Naturally, in the immediate environment of a multibacillary leprosy patient there would be an abundance of *M. leprae* in the environment as opposed to that of a paucibacillary leprosy patient. Given the possibility of skin entry of *M. leprae*, it is unlikely that in our case *M. leprae* was introduced in the wound from the school playground but, more likely, that it was introduced from the leprosy hospital environment. This could include the possibility of flies having introduced *M. leprae* into the wound during the healing time. It is common knowledge that wounds attract flies.

It is still argued by some that *M. leprae* could be drawn to the site of an injury from within the body. The primary route of infection is then still considered the upper respiratory tract and local factors, local temperature and possibly local factors intrinsic to a wound, may then draw *M. leprae* to the sites of predilection.

A recent case report also gives further evidence for inoculation leprosy, transmission of *M. leprae* through the broken skin. A surgeon in a leprosy non-endemic country cut himself on the dorsum of the left little finger whilst performing a muscle and nerve biopsy on a patient with undiagnosed neurological complaints. The biopsy result showed that this person, who had lived in a leprosy endemic country for 5 years, had multibacillary leprosy. Three years later, the surgeon noticed tenderness of the dorsal branches of the ulnar nerve. There were no skin lesions. Five years after the injury, the surgeon underwent local neurolysis. Histopathology of the nerve showed tuberculoid leprosy. It should also be noticed that the first lesion in artificially infected armadillos is at the site of inoculation.

Some studies have reported on the distribution of single lesions. A common finding of these studies is that single lesions were predominantly present on the extremities, specifically on the posterior aspect of the arm and the anterior aspect of the leg. In a study by Ponninhaus et al., it was also found that there was a large proportion of patients who had a single lesion on the posterior aspect of the arm (23%) as opposed to the anterior side (7%). This difference was not found for lesions on the lower extremity, but lesions on the lower
extremity were very few as compared to the upper extremity. These authors also found that a large proportion of their population had a single lesion on the face only (28%).

The anterior aspect of the lower limb and the posterior side of the upper limb are areas, that are more prone to injury and are cooler than the opposite surfaces of the limbs and so may favour entry of \textit{M. leprae} through the skin. These body areas are also generally uncovered especially in the younger age groups. In our case the only lesion was on the anterior aspect of the knee.

Another unpublished observation, which supports the possibility of entry through the skin, is the fact that in some single lesions in proximity to a nerve, only the nerve in proximity to the lesion is enlarged and shows nerve function impairment. (Brandsma, unpublished data). Such lesions are often not in the sensory distribution area of that nerve. In 11 of 25 cases in Ghorpade’s study who had a single lesion in connection with a tattoo, there was nerve trunk involvement, tenderness or thickening, of the nerve in proximity to the tattoo. However, not all tattoos were necessarily in proximity to a nerve.

In histopathological studies of single lesion leprosy, Job et al, found indications that the portal of entry of \textit{M. leprae} could be the skin. ‘It is reasonable to conclude that the indeterminate skin lesion may be a primary site of the infection’. They also stated that single lesion leprosy, and indeterminate leprosy, could be a manifestation of early lepromatous leprosy or that these could evolve into (subpolar) lepromatous leprosy.

For single lesion leprosy, single dose drug treatment has been advocated. This is being practised in certain programs. In integrated leprosy programs in which diagnosis and treatment of the disease are solely based on clinical observation, there is the risk that some cases could be undertreated as could have been the case in our patient.

There is evidence that BCG vaccination may prevent leprosy. Our patient had received BCG vaccination as a child. She may also have had genetic susceptibility to leprosy, since her father had been diagnosed some years previously with subclinical leprosy with only some enlarged nerves.

The source of infection and the route of entry remain important areas for research. Research findings from these areas may be important in the elimination and eventual eradication of leprosy.

References