The false and fixed belief of being infected/infested by one or many different living organisms, such as insects or parasites, is not a common condition. Although several cases have been recorded since the end of the nineteenth century, it was the Swedish psychiatrist Karl-Axel Ekbom who first systematically studied the presenile syndrome of delusional dermatozoid parasitic infestation in 1938 (Ekbom, 2003). This disorder was named as delusional parasitosis (DP) in 1946 (Wilson and Miller, 1946). Several different names were used for DP over the years, but Ekbom’s name has become the eponym attached to the condition referred to as DP.

Patients who only experience fear or anxiety of being infected/infested are excluded from this diagnosis because they lack delusions, and the term parasitophobia is more appropriate for these patients. DP is also distinct from formication, where the patient has the cutaneous sensation of crawling, biting and stinging, but does not have a fixed notion that these sensations are induced by parasites. Even though many cases remain idiopathic, patients suffering from formication can accept that they do not have a fixed belief that these sensations are induced by parasites. Even though many cases remain idiopathic, patients suffering from formication can accept that they do not have a fixed belief that these sensations are induced by parasites.

The classification of DP is still debated in the literature. From a pathogenic point of view, the delusion of parasitosis can be divided into two major groups: induced and non-induced delusions. The induced delusion cases would be best explained from a psychosocial point of view similar to the transmission of political and religious beliefs. By contrast, the non-induced cases would be better explained as disturbances of brain function. Two physiopathological mechanisms are possible: a sensory mechanism and a cognitive one (De Leon et al, 1992; Berrios, 1985; Baker et al, 1995). Details of the debate about whether to classify DP as delusion (a cognitive approach) or as chronic hallucinosis (a sensory approach), can be found in the literature (De Leon et al, 1992; Baker et al, 1995).

Epidemiology
Although the prevalence of DP remains unknown, of 215 dermatologists (based in the United Kingdom and Ireland) who replied to a postal survey by Reilly and Batchelor in 1986, 144 reported having seen at least one case in the last five years (Reilly and Batchelor, 1986). DP affects married people more often than single persons, and is more prevalent in patients with less formal education and/or in those with a lower socioeconomic status. DP can occur at any time of life, but is more frequent in the elderly. DP is more commonly seen in females and is most frequent in post-menopausal women (Bhatia et al, 2000). It is not limited to a specific race or culture (Leel, 1983; Reilly and Batchelor, 1986; Koo and Gambia, 1996).

Clinical presentation
Most of the patients reported in the literature have delusional problems related to the skin; DP can be considered a ‘classical’ skin-related delusional disorder (Ait-Ameur et al, 2000). Patients often seek help from GPs or dermatologists and relatively few publications originate from psychiatric clinics (Nicolato et al, 2006).
The skin symptoms that have been reported are varied, ranging from itching to sensations of crawling animals, worms, insects or bugs on the skin or in the internal organs. Attempts at self-treatment, such as repeated washing and disinfecting of the skin, can cause secondary skin problems (Bhatia et al, 2000). DP usually has a high burden of psychosocial morbidity and sufferers may take extreme measures to rid themselves of parasites, including self-harming behaviour. Patients may try to pick the parasites out of the skin or body, causing cutaneous lesions and/or ulcerations (Wilson and Uslan, 2004). Suicide attempts have also been reported (Friedmann et al, 2006). Although patients are not typically violent towards others, there have been reports of patients attempting to murder the family doctor for refusing to treat them. The danger posed by some of these cases is reported by Bourgeois et al (1992).

Patients may present with clothing, lint, pieces of skin, or other debris contained in plastic wrap, on adhesive tape, or in matchboxes. They typically claim that these contain the parasites, however on examination no parasites can be found. This presentation is called the matchbox sign. Myerson (1921) suggested that the fear of parasites has been present for most of human history, and that the fear of parasites has been present as ‘folie à deux’ (i.e. different people), which trigger or exacerbate the disease. A seasonal pattern has been reported for DP (Goddard, 2003).

**Management and treatment**
Management of DP involves the initial exclusion of other diseases and underlying conditions, including psychiatric disorders, parasitological diseases, medical conditions with altered sensation, use of drugs (prescribed and illicit), or withdrawal from alcohol or cocaine. Mental state, including cognition, also needs to be assessed. Other investigations may include examination of skin scrapings and skin biopsies, when appropriate. Ideally, patients should be referred to a psychiatrist, but many resist this. Consequently, medication with an antipsychotic should be initiated by the doctor who makes the diagnosis. Treatments include pimozide (Bhatia et al, 2000) and the newer atypical antipsychotic, olanzapine, which has a safer adverse effect profile than its classic antipsychotic counterparts (Meehan et al, 2006). Corticosteroid creams and lotions may be helpful adjuncts to ‘alleviate’ skin symptoms. Medication compliance can be a problem. Many patients with DP refuse treatment and are lost to follow-up (Colombo et al, 2004). Even when treated with antipsychotics, prognosis of delusional disorders is often difficult. The assertion that antipsychotic treatment leads to a complete remission of symptoms in 54% of patients may seem optimistic (Bhatia et al, 2000); however, several small, non-controlled studies have reported recovery from DP in up to 90% of patients (Driscoll et al, 1993). Managing patients with DP is often time-consuming for the physician and can put a strain on the patient-physician relationship, at least during the initial stages (Bourgeois et al, 1992).

**Case report**

**Initial story**
In September 2000, a 55-year-old woman was sent to the Teaching, Research and Tertiary Referral Center for Bancroftian Filariasis (NEPAF, Hospital das Clinics, Federal University of Pernambuco, Brazil) by her primary care provider. She was referred for antifilarial treatment after having received two shots of benzathine penicillin (three days apart) to treat an acute episode in her lymphoedematous left lower limb, which had started seven days previously. She was accompanied by her 35-year-old daughter. A brief conversation with the patient revealed that she had cut herself in an attempt to extract the filarial worms that she believed were inside her affected leg. She had a two-year history of sensing that worms were crawling in her leg, and occasionally they were felt to crawl elsewhere in the body, mainly in the stomach and deep inside the head. She was convinced that the leg should be surgically opened to extract the worms. She believed that she was unsuccessful in extracting the worms because they were too deeply localised. Five years previously the patient had been diagnosed with filarial lymphoedema. Upon diagnosis the patient received one course of diethylcarbamazine (DEC) (6mg/kg/12 days), an antifilarial drug. Although the public health services tested her on five occasions using the thick blood smear, no microfilaria were detected. She reported that the size of her leg continued to increase, despite antifilarial treatment.

Three years after the antifilarial treatment, the patient began to persistently seek treatment at various health facilities complaining of worms crawling throughout her leg and body. On four occasions she sought antifilarial treatment from filariasis control clinics. She reported receiving a course of treatment every six months for the past two years and she brought with her the yellow envelopes that had contained the tablets, which had her name and the doses prescribed (6mg/kg/12 days) written on them. Her impression was that during DEC treatment the worms seemed to ‘calm down’ and were restricted, at least for a few days, to

**The onset of DP is sudden and generally associated with stressful life events or other psychosocial stressors (e.g. contact with animals, ingestion of material believed to be infected or wearing a garment belonging to someone else), which trigger or exacerbate the disease.**
her leg. She had attempted many other treatments to rid herself of the worms, including drinking numerous types of tea and applying aloe vera, herbs and various creams to the skin. She had been to see a local Shaman three times with no success. She said that sometimes she had spent so much money on attempts to rid herself of the worms that she had had no money left to buy food.

Her medical history indicated that she had arterial hypertension (and was currently under antihypertensive treatment with hydrochlorothiazide — 50mg/day — provided by the government), but she was otherwise healthy, except for at least one acute bacterial episode annually in the affected leg for the last four years. She denied any current or past history of other drugs (licit or illicit), and reported that she had only been a social drinker. She lived with her daughter and her son (28 years old) and she lived independently. There was no past history of psychiatric disorders or other dermatological disorders, nor did she have a known family history of psychiatric, neurological or endocrine illnesses.

The patient explained that the onset of the worms crawling through her body occurred two years ago on meeting an ‘old woman’ who had lymphoedema of a lower limb. She described the woman as having worms (possibly myiasis) coming out of a wound in her leg (possibly a varicose ulcer). The old woman had told her that the wounds were caused by worms that had burrowed out through the skin. The patient said she then saw worms flying from the leg of the old woman to her own leg and said: ‘I got the worms from her and that’s when they started crawling through my body’.

The lymphoedema was classified as stage 3 using Dreyer’s classification of seven stages (Dreyer et al, 2002). The leg exhibited a characteristic sub-acute phase of dermatologically lymphadenitis (ADLA) (Box 1); a skin lesion that could have served as the point of entry for the bacteria that probably caused the clinical episode could be identified. This condition, caused by pyogenic bacteria, is seen in Recife-Brazil, an endemic area for bancroftian filariasis (Dreyer et al, 1999). Residual exfoliative dermatitis was still present, mainly on the sole of the patient’s foot (Figure 1). There were mild interdigital lesions of the left foot. The patient received practical education regarding routine skin care, after which she indicated verbally that she understood that entry lesions, including interdigital lesions, worsen lymphoedema by increasing the risk of ADLA episodes (Dreyer et al, 2006).

It was also explained to the patient that the recent acute episode she had experienced was likely to be due to the self-inflicted injury. She was scheduled for filariasis tests, which included a nocturnal blood sample to detect microfilaria collected after 23.00h on the same day of the consultation. Brain magnetic resonance imaging (MRI), ultrasound and routine blood tests were also prescribed. Blood samples for active *Wuchereria bancrofti* infection examined the next day at the NEPAF were all negative (no microfilaria were detected in 16ml of nocturnal blood and the immunochromatographic test for circulating antigen was negative). Ultrasound of the breast, peripheral lymphatic vessels and lymph nodes did not reveal lymphangectasia or living *W. bancrofti* adult worms (Dreyer et al, 1996a; Dreyer et al, 1996b; Dreyer et al, 2008). The patient did not, however, show up for subsequent MRI or routine blood tests.

The authors concluded in principle that the patient presented with monothematic, lucid, organised, persistent delusions, based on the conviction that live filarial worms resided in and were crawling through her body.

**Initial social interview**

The social interview revealed an oriented and co-operative lady with primary level education (she knew how to read but not how to write), belonging to a low socioeconomic status. Neither she, her daughter, nor her son were in stable employment. They earned money by washing clothes.
working as a housekeeper and washing cars on the street, respectively. Their monthly per capita income ranged from approximately 45–55 US$. The patient reported that about one year prior to ‘becoming infected’ she had begun to retreat socially. She had come to avoid special social activities, including carnival. Her daughter confirmed that during the last three years the patient had begun refusing to participate in social events, even avoiding going to the church as time went by. The daughter expressed fear that she too might develop a swollen leg as a result of filarial worms.

Follow-up assessments

On the second day of consultation it was explained to the patient that her tests failed to demonstrate filarial infection and that her lymphoedema was due to recurrent acute bacterial episodes (Dreyer et al, 1999) — not to live filarial worms. At this point she became very distressed and emphatically repeated her belief that she had living worms inside her body.

All professionals at NEPAF dealing with the patient (physicians, social workers, laboratory technicians, nurses) tried to assure her that while her perception was real, there was no external evidence to corroborate her belief that she was infected. The patient was resistant to the idea that she might have a psychiatric illness and refused a referral to a psychiatrist. Several attempts were made to encourage the patient to accept further treatment. She was reassured that she was not alone and that the doctors to whom she was being referred would listen seriously to her and that they sincerely desired to help her to get better. Further appointments were scheduled for her at NEPAF in order to establish a trusting relationship and to pave the way for beginning suitable antipsychotic therapy. Attempts to contact the primary care provider to discuss the case and its implications for differential diagnosis, as the patient’s original referral to NEPAF had not identified the clinic of origin.

The authors have had no contact with the patient or her family since then. A visit was made to the patient’s former residence, and the neighbours confirmed that the family had indeed moved and they had no information about their new address. They also confirmed that a woman with ‘worms coming out through a wound’ had lived in the vicinity a couple of years ago. Two people said that the old lady had died of unknown causes some months ago before the patient’s first consultation at NEPAF. It was also not possible to contact the primary care provider to discuss the case and its implications for differential diagnosis, as the patient’s original referral to NEPAF had not identified the clinic of origin.

Discussion

To the authors’ knowledge, this is the first case report of delusional parasitosis in a patient with lymphoedema living in an area endemic for bancroftian filariasis. The patient shared many aspects with the classical cases described in the literature: she had a false and fixed belief that she was infected by live worms; there was no grossly disorganised behaviour or generalised thought disorder; she did not present with a precipitating medical condition or with a history or current use of a licit or illicit drug(s); a stressful situation seemed to have triggered the process; the tendency of social isolation, was present, even before the delusional state; she had attempted many types of treatment to rid herself of the worms; the psychological origin of the disorder was not accepted by the patient and consultation with a psychiatrist was refused; the patient tried to pick the parasites out of her body, causing cutaneous lesions; and family members supported the patient’s delusions.

The patient belonged to an even rarer group of DP patients, in which internal organs were believed to be involved (Faure et al, 1957). She also had an organic disease linked with her delusional parasite in contrast to the majority of DP patients, among whom only a small percentage have or have had previous infectious disorders (Reilly and Batchelor, 1986). Some patients consider the ‘source’ of the worms to have been a cat or a dog (Wenning et al, 2003). The patient in this study believed that she had acquired the worms from an old lady. Social isolation had begun one year before the development of DP. Wenning et al (2003) described social isolation as a pre-morbid feature, rather than a consequence of the delusional syndrome.
Although the most commonly recognised chronic sequelae of lymphatic filarial infection is lymphoedema of the leg (Ryan, 2004), affecting an estimated 15 million persons living in 83 endemic countries, as a rule, patients do not show active infection, even during the acute phase of the disease (Addiss et al, 1999). In 1994 ultrasonography was used for the first time to visualise living adult filarial worms in vivo in lymph vessels (Amaral et al, 1994; Dreyer et al, 1996a; Dreyer et al, 1996b; Dreyer et al, 2008) and lymph nodes (Dreyer et al, 2001). The active and continuous movement of the worms seen by ultrasonography is known as filaria dance sign or FDS (Figure 2). The W. bancrofti adult worms have a remarkable stability of location in a given patient, and it is believed that migration of living adult worms within the lymphatic vessels or lymph nodes does not occur (Dreyer et al, 1994). This stability has made it possible to evaluate with great accuracy the macrofilaricidal effect of different antifilarial drugs (Dreyer et al, 1996c; Norões et al, 1997; Dreyer et al, 1998).

It appears that healthcare providers seeing the patient during the two years prior to her presenting at NEPAF did not recognise that she was delusional. Instead, the patient seemed to have received positive feedback for her ‘filarial condition’. Thus, she had not suffered the additional stress of a healthcare professional acknowledging her delusional status. The authors raise the possibility that these healthcare providers accepted that a person with a disease caused by filarial worms could feel the parasites crawling in the swollen leg. In fact, the patient received antifilarial treatment twice a year as a result of pursuing treatment for a condition that was actually delusional, and not for her lymphoedematous leg. It is worth mentioning that antifilarial treatment is used to treat filarial lymphoedema in the city of Recife, Brazil, irrespective of the parasitological status of the patient (Dreyer et al, 1999; Dreyer and Mattos, 2007). In other words, the patient had a previous diagnosis of filarial lymphoedema, e.g. an organic disease believed to be caused by the filarial W. bancrofti parasite (Ryan, 2004; Dreyer and Mattos, 2007), which made her symptoms seem reasonable. In Recife, news of the discovery of the FDS quickly spread throughout the city through reports in newspapers, by word of mouth, and lectures and training courses for healthcare personnel involved with the filariasis control programme.

It is conceivable that the perception/complaints presented by the patient of having/feeling living filarial worms inside her swollen leg/body might not sound too strange to people who had incomplete or misleading information about FDS/filariasis. On the other hand, it is interesting to point out that there is no evidence in the scientific literature that infected individuals can feel a crawling sensation caused by filarial worms.

Patients with DP are usually not specific about the organism that is bothering them, referring to them generally as ‘insects’, ‘bugs’, ‘worms’, ‘chainsaw worms’, ‘shark bugs’. By contrast, this patient was specific that the pest causing her problem was the filarial worm.

It would be valuable to know whether positive feedback, as was attempted in the present case, could influence (negative, positive or neutral) the evolution of DP. Unfortunately, the patient’s condition was not recognised earlier, so a retrospective accurate analysis of her process/evolution was not possible, nor was the patient or her children able to give enough anecdotal information to provide further insights into her condition.

Losing the patient to follow-up not only deprived her of suitable treatment, but also deprived the medical team at NEPAF of the opportunity to learn more about DP in lymphatic filariasis endemic areas. The authors suggest that children of such patients may be at risk of being involved in a ‘folie a deux or trois’. Finally, although neuropsychiatric models to explain hallucinations are common, there is still a lack of brain models to explain the delusions (Arthur, 1964; De Leon et al, 1992).

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References

Figure 2. Ultrasound of the intrascrotal contents with a 7.5 MHz probe of a Brazilian microfilaraemic patient. In B-mode (left side) a spermatic cord-dilated tortuous lymphatic vessel (asterisks) is seen containing hyperechogenic linear structures (arrows) which in real-time examination revealed peculiar active movements representing living W. bancrofti adult worm(s), known as filaria dance sign or FDS. M-mode (right side) documents the predominantly transverse movements seen in real-time examination (arrows). Video clips of living adult worms can be seen at www.amaurycoutinho.org.br.

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Key points

- The false and fixed belief of being infected/infested by live animals (insects or parasites) is a rare condition known as delusional parasitosis.

- The symptoms experienced by the patients are variable ranging from itching to crawling of animals, worms, insects or bugs on the skin or in the internal organs.

- Patients may present with clothing, lint, pieces of skin, or other debris contained in plastic wrap, on adhesive tape, or in matchboxes. This presentation is called the matchbox sign.

- Although neuropsychiatric models to explain hallucinations are common, there is still a lack of brain models to explain delusions.