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Comparison between active surveillance and passive detection of zoonotic cutaneous leishmaniasis in endemic rural areas in Central Tunisia, 2009 to 2014

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ABSTRACT

Objective: To assess the contribution of active surveillance of zoonotic cutaneous leishmaniasis (ZCL) in rural areas in Sidi Bouzid from 2009 to 2014 in comparison with the passive case detection, and describe the pattern of the disease.

Methods: The monitoring was based on the notification of all new cases in primary health care facilities, among pupils in all schools and a community-based active ZCL surveillance was also performed. The medical staff of health care facilities reviewed and confirmed cases notified in schools.

Results: From July 2009 to June 2014, a total of 856 (51.1% male, 48.9% female) cases were enrolled; the average incidence rate of the disease was 2 514.4 per 100 000 inhabitants. The median age was 11 years (inter quartile range: 7-28) and 68.9% were aged less than 20 years. Most ZCL lesions arose between August and January and 46.1% of cases were reported between July 2013 and June 2014. Active surveillance reported more cases mainly in the delegation of West Sidi Bouzid and most cases (66.7%) were reported by the health care centers.

Conclusions: The present study showed ZCL was still endemic in rural areas in Sidi Bouzid Governorate. Active surveillance's cost-effectiveness is not certain thus, it would be rational to improve routine passive detection of ZCL in Sidi Bouzid than to continue active research of cases.

1. Introduction

Leishmaniasis is one of the neglected tropical diseases which are a group of parasitic diseases and related infections that represent the most common illnesses of the world's poorest people[1]. It is a zoonotic disease endemic in the Mediterranean basin, including Tunisia[2,3]. Zoonotic cutaneous leishmaniasis (ZCL), caused by *Leishmania major* (*L. major*), is widespread in the Middle East and North Africa region. The largest number of *L. major* cases occurs in the arid and Saharian bioclimatic stages[4-6]. Central Tunisia is the most endemic area; cases were reported mainly from the governorate of Sidi Bouzid, Kairouan and Gafsa[7-9].

Public health control measures in any country are strongly dependent on the information coming from the surveillance systems. The World Health Organization expert committee report 2010[2], stated that passive case detection and reporting of cutaneous leishmaniasis should be the basis for a control program. In Tunisia, control program of ZCL was implemented since 1990. The routine surveillance system in Tunisia relies on physicians and primary health care personal who observe ZCL patients and the data of notified cases are delivered from primary health care facilities to regional public health directorate, then to the central directorate of public health, Ministry of Health. Passive detection of communicable diseases suffers from severe underreporting mainly in developing countries and the public health impact of leishmaniasis is underestimated[10]. This study was part of an exhaustive population based survey using the ecohealth approach to improve the understanding of the various interacting climatic, ecological, epidemiological, agricultural and socio-economic factors that cause ZCL as well as to investigate possible options for decreasing the vulnerability of local populations.

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In this paper, we assessed the contribution of an active surveillance system to improve passive case detection and described characteristics of new cases in terms of date, place and persons.

2. Materials and methods

The surveillance network was implemented in three delegations in the governorate of Sidi Bouzid since July 2009. Ten rural districts (Bir Badr, Hichria, Zefzef, Souk Jedid, Rmilia, Sakdel, Gsaira, Garet Hadid, Friou and Zitouna) with a total population of around 34 044, were covered by this active detection of ZCL cases.

The monitoring was based on the notification of all new cases in people who came to primary health care facilities seeking for treatment, and the active research of other cases among their neighbors and families by the nursing staff. All schools in this area have been asked to seek for and notify all ZCL cases among students. Moreover, a community-based active ZCL surveillance was performed by the members of the research team. The medical staff of the health care facilities reviewed and confirmed cases notified in schools on the basis of clinical diagnosis of the lesion or scars. Because of the good knowledge of the disease by the medical staff and the population in this region and the high sensitivity and specificity of clinical diagnosis, parasitologic diagnosis of ZCL lesions was not carried out. We organized two conferences in 2010 and 2013 for public health care facilities and schools staff to inform and aware them of the importance of ZCL reporting. Information on age, sex, place of residence, date of diagnosis and date of onset of the lesion, the number and the location of the lesions and the treatment and evolution were collected using a standardized case record cart. Data on ZCL from mandatory notification of communicable diseases was used to compare incidence rates between active and passive detections. Estimated population data by districts for 2012 was obtained from the National Institute of Statistics.

3. Results

During the study period, from 2009 to 2014, 856 (51.1% male, 48.9% female) cases of ZCL were enrolled. With an average of 171 new cases occurring each year during 2009-2014 and a population at risk estimated to be 34 044, the average annual incidence rate of the disease in this foci was calculated to be approximately 502.9 per 100 000 people and the average total period incidence rate was calculated to be 2 514.4 cases per 100 000 inhabitants. Table 1 shows number of cases and incidence rate by districts and Figure 1 represents spatial distribution of ZCL cases. The median age was 11 years (inter quartile range: 7-28), ranging from 2 months to 87 years and most of the patients were assigned to the 0-9 and 10-19 years age groups with approximately 40% and 30% of the total recorded ZCL cases respectively for each group. Distribution of cases by sex and age was shown in Figure 2. In the area under study, the maximum number of cases (46.1% of total) was recorded between July 2013 and June 2014 and the least number of cases reported was in 2009-2010 season with 58 cases (6.8% of total). Recurrent ZCL was observed in 5.0% of subjects. The lower limb was the most frequent seat of ZCL lesion (48.0%) and 13.9% had reported face lesions. About 42.6% had a single lesion and the infection of the

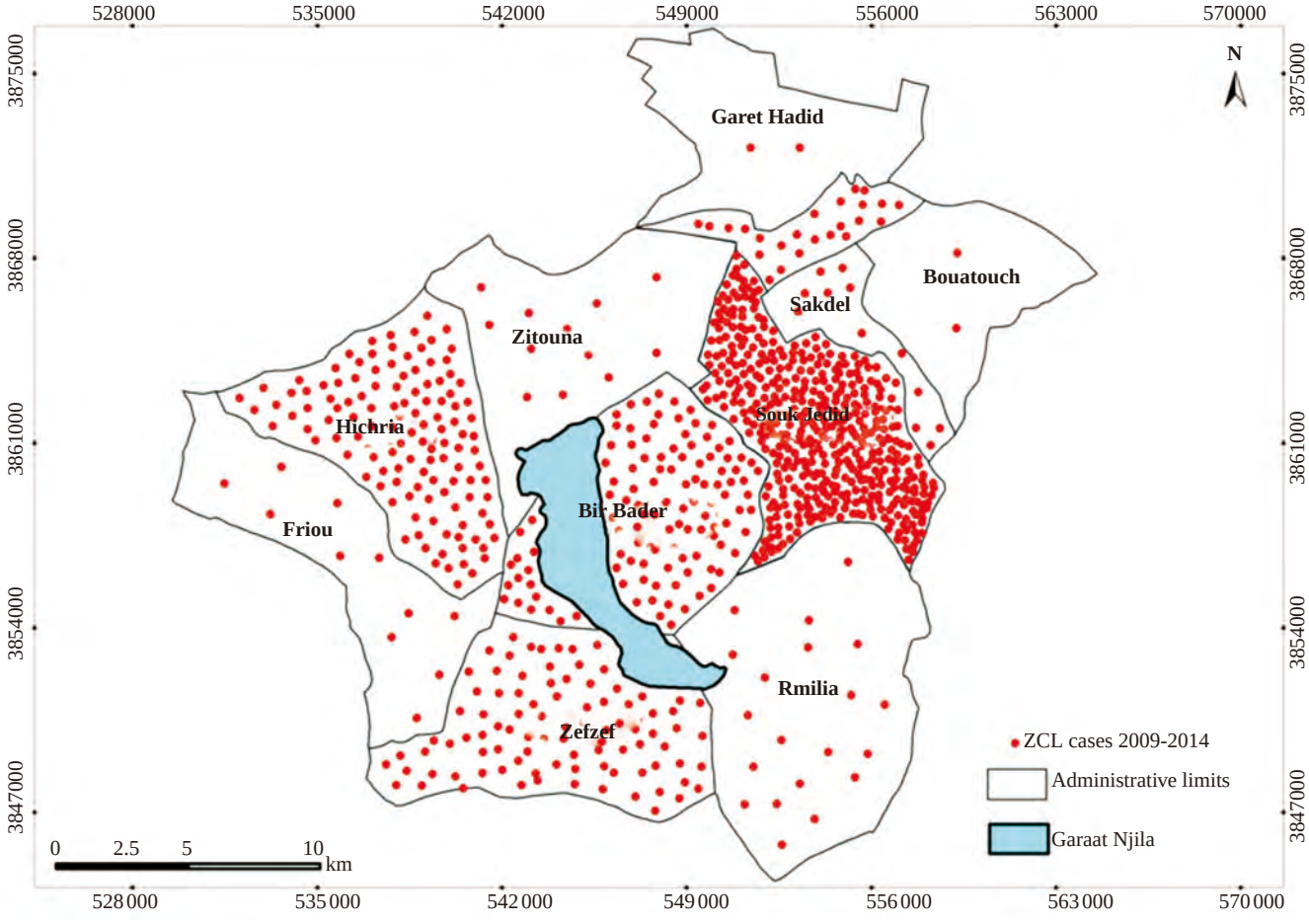

Figure 1. Spatial distribution of cases by area of residence, active surveillance of ZCL in Central Tunisia, July 2009-June 2014.

lesion occurred in 7.9% persons. Only 7 subjects were admitted to hospital, lesions treatment consisted in local disinfection (64.7%) and antibiotics (21.3%); the spontaneous healing was observed for 95.8% of individuals (Table 2).

Table 1

Number of ZCL and incidence rate by districts, active surveillance in Central Tunisia, July 2009-June 2014.

Locality	Number of cases	Population	Incidence rate per 100 000	Mean annual incidence rate per 100 000
Hichria	130	4 250	3 058.8	611.8
Friou	11	4 242	259.3	51.9
Zitouna	12	3 922	305.9	61.2
Garet Hadid	2	3 293	60.7	12.1
Souk Jedid	436	2 758	15 808.6	3 161.6
Gsaira	25	2 263	1 104.7	220.9
Sakdel	14	2 748	509.5	101.9
Rmilia	20	3 281	609.6	121.9
Zefzef	92	3 385	2 717.9	543.6
Bir Badr	114	3 902	2 921.6	584.3
Total	856	34 044	2 514.4	502.9

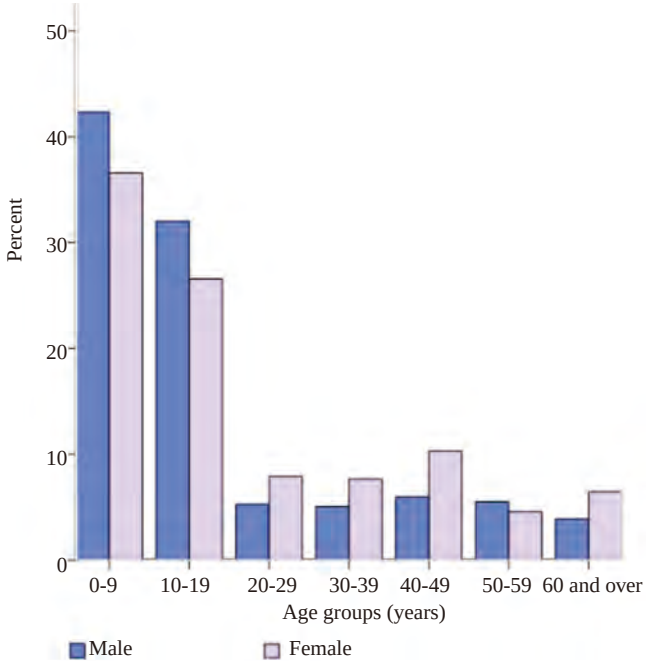

Figure 2. Distribution of cases by sex and age group, active surveillance of ZCL in Central Tunisia, July 2009-June 2014.

Table 2

Epidemiological characteristics of ZCL, active surveillance in Central Tunisia, July 2009-June 2014.

Characteristics		*N*	%
Sex	Male	437	51.1
	Female	419	48.9
Age in years	0-9	338	39.5
	10-19	251	29.3
	20-29	56	6.5
	30-39	54	6.3
	40-49	69	8.1
	50-59	43	5.0
	60 and over	44	5.1
Season of the lesion onset	July 2009-June 2010	58	6.8
	July 2010-June 2011	198	23.2
	July 2011-June 2012	129	15.1
	July 2012-June 2013	76	8.9
	July 2013-June 2014	394	46.1
Recurrent ZCL	Yes	43	5.0
	No	813	95.0
Seat of the lesion	Face	149	13.9
	Trunk	62	5.8
	Upper limbs	348	32.4
	Lower limbs	516	48.0
Number of lesions	1	364	42.6
	2	209	24.4
	3	118	13.8
	4 and more	164	19.2
Infected lesions	Yes	68	7.9
	No	788	92.1
Admitted to hospital	Yes	7	0.8
	No	849	99.2
Treatment	Local disinfection	827	64.7
	Antibiotic	272	21.3
	Local glucantime	174	13.6
	Systemic glucantime	5	0.4
Outcome	Spontaneous healing	820	95.8
	Infection	36	4.2

Active surveillance and passive detection reported respectively a mean incidence rate of 2 514.4 and 889.8 per 100 000 inhabitants for the total period. The mean incidence rates calculated from active surveillance were much higher than those given by passive detection mainly for the delegation of West Sidi Bouzid (Table 3).

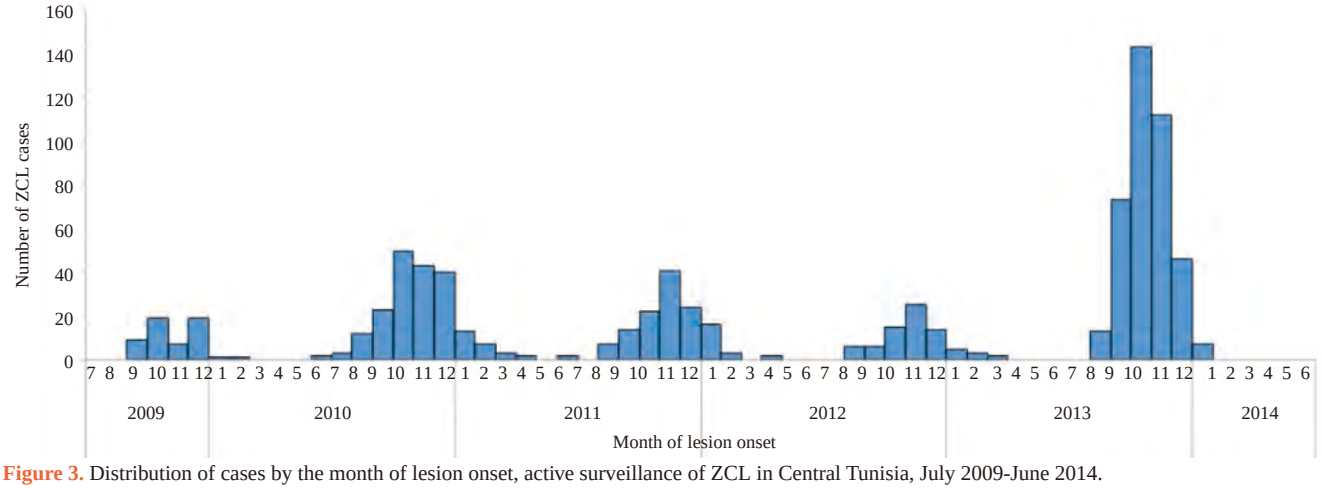

Figure 3. Distribution of cases by the month of lesion onset, active surveillance of ZCL in Central Tunisia, July 2009-June 2014.

Table 3
Incidence rate of ZCL reported from active surveillance and passive detection, 2009-2014.

Locality	Incidence rate per 100 000 inhabitants from routine notification	Incidence rate per 100 000 inhabitants from active surveillance
Sidi Bouzid Ouest	930.1	1 660.4
Sidi Bouzid Est	182.9	194.0
Souk Jedid	3 481.5	3 822.8
Total	889.8	2 514.4

The epidemic curve by date of lesion onset showed seasonality of ZCL occurance. Most ZCL lesions arose between August and January and the highest incidence was observed in October and November. An epidemic peak was observed between July 2013 and June 2014 (Figure 3).

A total of 571 (66.7%) cases were reported by the primary health care centers, 213 (24.9%) cases were reported by the school's staff and 72 (8.4%) cases were notified by the community-based surveillance.

4. Discussion

The existence of national health plans in many countries generates large databases that can be used for surveillance. In Tunisia, surveillance activities are supported and managed by a variety of vertical disease-control programmes such as tuberculosis, leishmaniasis, malaria and rabies. But, the surveillance function is far removed from any corresponding action such as disease control efforts and outbreak response.

ZCL is endemic with periodic outbreaks in Central Tunisia and outbreaks have an apparent tendency to occur at around 4-7 year intervals and cause disfiguring scars which lead to social stigmatization[8]. Effective control methods are not available and surveillance is necessary to study the disease, its patterns of occurrence and the population at risk. Moreover, there is a need for baseline data before the implementation of control measures to monitor and evaluate them, mainly when available data and alternative sources are not sufficient. Thus, this active surveillance was established in order to assess the burden of ZCL, analyze its pattern in terms of time, place and person, study possibilities of early outbreaks detection and have a baseline database before implementation of control strategies to monitor progress towards control or eradication.

We describe characteristics of individuals with new cases of ZCL and compare incidence rates between active and passive detection. This study confirmed the high endemicity of ZCL in this area, mainly in the locality of Souk Jedid and the seasonality of its patterns over time. ZCL occurs in males and females, but children were the most vulnerable. Lesions appear on exposed areas of body, mainly lower limbs, upper limbs and face.

The disease is not fatal, favorable issue and acquired definitive immunity were often observed. However, ZCL leaves a disfiguring scars that may cause social impact mainly when the lesion onset is in the face of women. The mean incidence rates reported from active surveillance were much higher than those given by passive detection and the primary health care facilities reported most cases.

This is the first active surveillance system for ZCL in Tunisia which has included many data sources and collecting procedures to provide the most accurate information collected in a practical and efficient manner and to satisfy the goals and objectives of the surveillance system. The collection of data is the most costly and difficult component of a surveillance system. The quality of a surveillance system is only as good as the quality of the data collected. Moreover, it is essential to establish denominator data for the target population. As they are paid, reporters and collectors were required to return the forms monthly. Data could be incomplete since we had not performed biological diagnosis so that not so typical lesions and scars should be missed. However, leishmaniasis occurs commonly in this area so that it is not necessary for all cases to be reported and this should not reduce the effectiveness of surveillance, since it is generally the trends of disease occurrence that are important for decision-making on control and preventive measures.

Incidence rates of ZCL were high attesting that this area is still a highly endemic region as it was observed in previous studies[7,8]. Souk Jedid reported most cases; however this apparent increase could not represent true increase, since actors of active surveillance were mainly medical staff of Souk Jedid health care facility. Moreover, there was not a big difference in the incidence rate compared with passive detection; as they are also the actors of mandatory notification and it is common that active surveillance enhances reporting from passive detection[11].

The seasonal distribution of ZCL shows that the highest rate of infection is in autumn, the lowest is in spring and winter and the peak of the disease occurs in October and November. In Central Tunisia, Toumi *et al.* demonstrated that the incidence of the disease was significantly higher during the group of months from October to March[8]. A similar seasonal pattern has been reported from other leishmaniasis endemic countries. In Libya and many provinces (Arsanjan, Sabzevar, Damghan and Kermanshah) of Iran[12-14], the highest percentages of patients with ZCL were reported in autumn and the lowest in winter. Faulde *et al.* described the seasonality of ZCL in Mazar-e Sharif, Afghanistan, where the maximum numbers of ZCL cases were recorded in September and October[15]. The transmission of leishmaniasis is highly dependent on climate conditions and the ecology of vector and reservoirs hosts[2]. Each species of sand fly has an annual cycle, and the highest transmission occurs at the end of this cycle[16].

The sex distribution was found to be approximately equal in the present study; this finding was also observed by Bettaieb *et al.*[9]. Some studies in Iran have shown an equal proportion of ZCL infection among the two sexes[17]. However in other endemic regions of Iran and Mazar-e Sharif, Afghanistan, the frequency of male patients with ZCL was higher than that of female patients[18]. Such observations seem to be related to different behavior patterns among men and women that increase or decrease vector exposure.

In this study, most cases were observed in children, suggesting exposure of non immune persons to sandfly bite in domestic or peridomestic places. In ZCL endemic regions of Iran, the highest risk group is often children aged less than 15 years[17,19,20]. In fact, in established endemic areas, people become immune against ZCL when they get older[2].

In this study, it was observed that most of the lesions appeared on the extremities, feet and hands and this pattern of lesion development on the limbs is common in ZCL. In a comparative study of skin lesions observed in three endemic varieties of cutaneous leishmaniasis in Tunisia, ZCL lesions were often observed on the limbs[21].

Almost 43% of the ZCL patients in this study had one skin lesion. Many studies in endemic regions of Iran and one report from Iraq observed that most patients presented a single lesion[6,13,14]. However, in ZCL due to *L. major*, multiple lesions might be seen. In the present study, about 14% of ZCL patients have three lesions and 19% of them have more than four lesions. Aoun *et al.* observed that 75% ZCL lesions in Tunisia were multiple[21]. Likewise, several workers in different provinces of Iran showed the high frequency of multiple lesions in ZCL patients[19,22]. A possible explanation is that sand flies usually have a discontinuous blood-sucking habit and may sting several times at every attack and cause the development of multiple lesions on the skin[2].

The primary health care facilities staff reported most cases compared to school staff and community-based surveillance. Indeed, this system was mainly implemented in health care facilities and schools; detection of cases in the community was made occasionally when the research team members are in the region and at the beginning of each season to inform, motivate and boost surveillance in schools especially. Our surveillance system reported more cases than passive detection. Active surveillance has been shown to increase the number and proportion of reported cases, and to promote closer personal ties between the providers and the collectors. However, active surveillance is relatively expensive, and its cost-effectiveness is not entirely clear[11]. In this study, most cases from active surveillance system were reported by the health care facilities staff members. The same staff should report some communicable diseases considered as mandatory notified, including ZCL cases, to the health authorities. However, this way of reporting suffers from a severe underreporting and it is decidedly due to the fact that the staff is not motivate enough to report new cases because of the absence of feedback information to the people who collect the data and their low level of awareness concerning the usefulness of the notified cases.

The present study shows that ZCL is endemic and causes a major health problem in Central Tunisia. In order to identify all epidemiological aspects, eco-health approach should be considered, including climate change and its impact on vector and reservoir behaviour, and socio-economic characteristics of the local population.

Recommendations for changes in the surveillance system need to consider these results so that it would be better to improve passive detection than to implement active surveillance. Some ways exist to improve a system such as the awareness of providers, the simplicity of reporting, the frequent feedback and dissemination of data back to the people who collected them, the use of multiple sources and methods, the motivation and the encouragement of providers by legal requirements, education, participation in projects, making important clinical and therapeutic information available to those who report, and by making specific drugs or biologics available to physicians on notification, and non-monetary or monetary rewards.

Conflict of interest statement

We declare that we have no conflict of interest

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