ORIGINAL ARTICLE



WILEY

Geographical distribution of mycetoma cases in senegal over a period of 18 years

Aïda Sadikh Badiane^{1,2} | Mouhamadou Ndiaye^{1,2} | Khadim Diongue^{1,2} | Mamadou Alpha Diallo² | Mame Cheikh Seck^{1,2} | Daouda Ndiaye^{1,2}

¹Cheikh Anta Diop University of Dakar, Dakar, Senegal ²Aristide Le Dantec Hospital, Dakar, Senegal

Correspondence

Aïda Sadikh Badiane, Cheikh Anta Diop University of Dakar, Dakar, Senegal. Email: asbadiane@gmail.com

Summarv

Background: Mycetoma is a pathological process in which fungal or actinomycotic agents of exogenous origin produce grains. In the absence of data on the global burden, it is important to map mycetoma cases, which are useful to implement control strategies.

Objective: The objective of this study was to map mycetoma cases diagnosed in Senegal over a period of eighteen years.

Methodology: The cases of mycetoma identified in the laboratory of Mycology at Aristide Le Dantec Hospital were extracted from the notebooks; information on the dates of collection, geographical origin and fungal agent identified was entered in Excel and analysed.

Results: Three hundred and thirty-seven cases of mycetoma were diagnosed from 1993 to 2016 at Aristide Le Dantec Hospital. Mapping shows that overall, the western zone presented the majority of cases 47% (120), followed, respectively, by the central zone 32% (80), the northern zone 18% (47) and the southern zone 2% (6). However, over the years, this distribution is different with a decrease in cases from the periods 1993-2000 and 2011-2016 of 19% in the western and a progressive increase of cases in northern and central zones of, respectively, 13% and 14%. In the 1990s, the cases were predominant in Dakar, Louga and Diourbel. During 2011-2016, Thies, Diourbel, Fouta and Louga presented more cases.

Conclusion: The spatial distribution of mycetoma in Senegal changed over the years, most frequent in the west of the country, and during 1993 to 2000, mycetoma is now more common in the north.

KEYWORDS

cartography, mycetoma, senegal

1 | INTRODUCTION

Mycetoma is a chronic condition caused by two groups of pathogens, namely the fungi determining eumycetoma and the bacteria causing actinomycetoma. The production of pus and grains or granules is the characteristic of mycetoma. Clinically, the lesions are much more inflammatory for actinomycetoma than for eumycetoma; however, biological confirmation is mandatory as the treatment is different. The evolution of the disease is chronic and very often the diagnosis is late, leading to spectacular lesions with a reduction in patient mobility, the foot location being most common.¹ The first case of African mycetoma was diagnosed in Senegal in the city of Saint Louis in 1894 by the French physician Aristide Le Dantec. Since then, several epidemiological studies have been conducted in the country describing the clinical and biological aspects.¹⁻⁹

Mycetoma mainly affects poor rural people, living in areas where local healthcare facilities are usually insufficient and/or lack medical specialists. The treatment is problematic and not codified. The treatment of fungal mycetoma was based mainly on surgery and for actinomycetoma antibiotics are used. Currently, several triazole antifungals including itraconazole, voriconazole and posaconazole are tested for the cure of eumycetoma¹⁰; however, these drugs are not available in most endemic countries in Africa.

Although mycetoma is endemic in several countries, its prevalence is not known. Several cases have been reported in endemic countries without being able to determine prevalence or incidence. All these problems have aroused the interest of mycetoma specialists and with advocacy, the disease has been listed since 2016 on the WHO list of neglected tropical diseases (NTDs). Despite this, so far, not much attention is devoted to mycetoma and very little is known about these conditions. Indeed, the mode of contamination is still not established with certainty.

It is admitted that it occurs by transcutaneous inoculation during an injury with plants, and cases have been reported finding debris or the patient remembering an injury.^{1,11,12} Mycetoma is reported in countries located between the 30°N and 15°S; this covers in Africa: Sudan, Nigeria, Mauritania, Ethiopia, Tchad, Kenya, Djibouti, Cameroon, Somalia, Tunisia, Niger and Senegal.¹³⁻¹⁹

In Senegal, the cases of mycetoma diagnosed in the country are mostly patients attending the specialised health structures, and the reported cases would be just the tip of the iceberg because most patients do not often visit clinics, due to economic reasons.

Although endemic countries are known, the distribution of cases within countries is not available for many countries. An estimation of the global burden of mycetoma was conducted in 2013²⁰ for endemic countries but it is important to map these cases within each country, to guide management. Mycetoma is a rural disease, and therefore, climate changes and the modernisation of African cities could change the geographical distribution within endemic countries. Indeed, in most African countries changes in the environment with the construction of new cities could drive to a reduction or a redistribution of the plants that seem to be responsible of the contamination. Therefore, a change in the geographical distribution of mycetoma within a country could occur overtime. In the absence of prevalence data, mapping the cases within countries is needed to guide strategies for controlling the disease.

All these factors should be taken into account to implement control strategies as this could have an impact in the prevalence of mycetoma. The first study that mapped mycetoma cases within a region was conducted in the 1950s in Sudan.²¹ But, in most endemic countries only census of mycetoma cases with clinical description of the lesions and the species responsible are reported.

The objectives of this study were to map mycetoma cases diagnosed in the laboratory of Parasitology-Mycology University Hospital (CHNU) Aristide Le Dantec in Senegal over a period of approximately 20 years.

2 | METHODOLOGY

This retrospective study was conducted at the laboratory of Parasitology and Mycology at Aristide Le Dantec Hospital of Dakar, Senegal. The registers consulted cover the period from 1993 to 2000 and from 2005 to 2016. The registers from 2001 to 2005 were not included because they could not be found. The records of myce-toma cases diagnosed in the laboratory were used to collect and map cases using the geographical origin of each patient. Other studies on these cases have already been published concerning the clinical and biological aspects.¹⁻⁹

Patients received during this period had come to the laboratory with a prescription of mycetoma diagnosis. These cases have been diagnosed using classical methods performed on the pus containing the grains.

From the registers, mycetoma cases were collected and the following data were recorded and entered into Excel: sampling date (day and year), geographical origin of the patient from which GPS coordinates were determined, the colour of the grains and/or the type of agent (fungal or actinomycotic). The GPS coordinates were not used to localise up to the neighbourhood level, only the region was determined.

Data were analysed using Excel.

2.1 | Ethics statement

The authors confirm that the ethical policies of the journal, as noted on the journal's author guidelines page, have been adhered to. No ethical approval was required as this study used routine data from the hospital.

3 | RESULTS

During this period of eighteen years, 337 cases of mycetoma were diagnosed, 39% (132 cases) during the period 1993 to 2000 (7 years), 30% (100 cases) between 2005 and 2010 (5 years), and 31% (105 cases) between 2011 and 2016 (5 years; Figure 1A).

The geographical origin was available for 275 of the patients allowing the determination of GPS coordinate used to map the cases. Among these mycetoma cases, 253 came from Senegal and 22 outside of the country; Mauritania (18 cases), Mali (three cases) and Cape Verde (one case). Only cases from Senegal were included in the analysis. To conduct the analysis, the country was divided into four zones according to the climate; the west (Dakar and Thies), the centre (Diourbel, Kaolack, Kaffrine, Fatick), the north (Saint Louis, Fouta, Louga, Matam), where the climate is Sudano-Sahelian, and the south (Sedhiou, Kolda, Tambacounda, Ziguinchor) with a

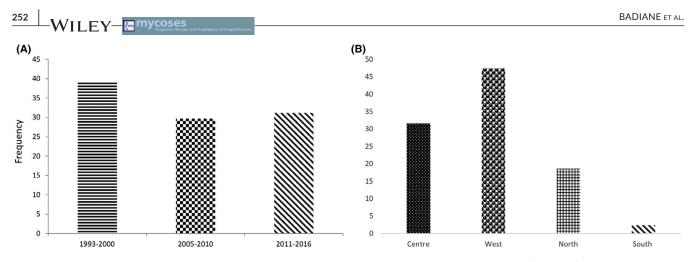


FIGURE 1 A, The frequency of mycetoma cases according to the period. 337 cases were collected, 39% (132 cases) during the period from 1993 to 2000 (7 y); 30% (100 cases) between 2005 and 2010 (5 y), 31% (105 cases) between 2011 and 2016 (5 y). B, The frequency of mycetoma cases according to geographic localization. Among the 337 cases collected, 253 were from Senegal with GPS coordinates available. 47% (120) of the cases were found in the West, 32% (80) in the Centre, 18% (47) in the North and 2% (6) in the South

Regions	Eumycetoma cases	Actinomycetoma cases	Undetermined cases
Dakar	53 (66.25%)	22 (27.5%)	5 (6.25%)
Thies	20 (50%)	15 (37.5%)	5 (12.5%)
Diourbel	16 (61.54%)	9 (34.61%)	1 (3.85%)
Saint-Louis	18 (66.67%)	8 (29.63%)	1 (3.70%)
Louga	24 (68.57%)	8 (22.86%)	3 (8.57%)
Matam	2 (33.33%)	3 (50%)	1 (16.67%)
Sedhiou	1 (100%)	0 (0%)	0%
Tambacounda	3 (100%)	0 (0%)	0 (0%)
Ziguinchor	0 (0%)	0 (0%)	1 (100%)
Kaffrine	0 (0%)	1 (100%)	0 (0%)
Kaolack	1 (25%)	3 (75%)	0 (0%)
Fatick	6 (42.86%)	4 (28.57%)	4 (28.57%)
Fouta	10 (71.43%)	0 (0%)	4 (28.57%)
Total	154 (61.11%)	73 (28.97%)	25 (9.92%)

TABLE 1Repartition of eumycetomaand actinomycetoma across the regions

Note: Eumycetomas were predominant in all the regions except in Kaolack, Kaffrine and Ziguinchor which are, respectively, located in the centre and the south, but the number of cases in these regions is too small.

Sudanian climate. In the Sudano-Sahelian climate, there is a difference of temperature between the regions and in Senegal; the highest temperatures are found, respectively, in the north and the centre. In the north, the temperatures could easily exceed 45°C during the dry season between April and May and reach 40°C during October and November. In general, during the dry season the temperatures in Senegal could exceed 40°C except in the west (Dakar, Thies). The Sudanian climate is characterised by a longer rainy season compared to the Sudano-Sahelian climate, during which the temperatures diminish. The temperatures are high during the dry season and could reach 40°C in Tambacounda where the heat is more accentuated than Kedougou, Kolda or Sedhiou.

Overall, 47% (120) of the cases were found in the west, 32% (80) in the centre, 18% (47) in the north and 2% (6) in the south

(Figure 1B), using a Kruskal-Wallis test, and the difference was not significative (*P* = .39).

In the west, 31.62% (80) of cases came from Dakar and 15.81% (40) from Thies (2A).

In the centre, 10.27% (26) of patients lived in Diourbel, 5.53% (14) in Fatick, 1.58% (04) in Kaolack and 0.40% (01) in Kaffrine (2A).

In the north, the cases were recorded 10.67% (27) in Saint Louis, 13.83% (35) for Louga, 5.53. % (14) in Fouta and 2.37% (6) for Matam (2A).

In the south, six cases were diagnosed, Sedhiou, Kolda and Ziguinchor recorded each 0.40% (1) of the cases and Tambacounda 1.18% (3) (2A).

Overall, for the cases diagnosed in Senegal, eumycetoma accounted for 61.11%, actinomycetoma for 28.97% and the type of

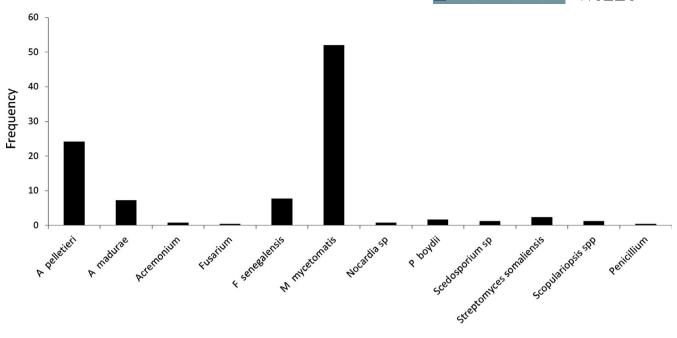


FIGURE 2 Frequency of mycetoma agents in Senegal. The most frequent agents were: *Madurella mycetomatis* (52%), *Actinomadura pelletieri* (24.19%), *Falciformispora senegalensis* (7.66%), *Actinomadura madurae* (7.25%). *Pseudallescheria boydii* known as *Scedosporium apiospermum* (1.61%), *Streptomyces somaliensis* (2.41%), *Scedosporium* sp (1.21%), *Scopulariopsis* spp (1.21%) were less frequent. Other species were noted such as *Acremonium* (0.80%), *Nocardia* (0.80%), *Fusarium* (0.40%) and *Penicillium* (0.40%)

agent could not be determined for 9.92% of the cases (Table 1). Despite the regions, eumycetomas were predominant except in Kaolack and Kaffrine where, respectively, 3 and 1 actinomycetes were recorded (Table 1). The most common species reported were *Madurella mycetomatis* (52%), *Actinomadura pelletieri* (24.19%), *Falciformispora senegalensis* (7.66%) and *Actinomadura madurae* (7.25%) (Figure 2). Others species were identified at lower frequency (Figure 2). There was no difference in the species distribution across the country.

98% of the mycetoma cases were recorded in the Sahelo-Sudanian climate (west, centre and north).

Between 1993 and 2000, 58% (59) of mycetoma cases were recorded in the west, 14% (14) in the centre, 26% (26) in the north and 2% (2) in the south (Figure 3A). During the period 2005 and 2010, 47% (38) of the patients were from the west zone, 35% (28) from the north zone, 14% (11) from the central zone and 4% (3) from the south zone (Figure 3A). Between 2011 and 2016, 39% (28) of the patients were from the north zone, 32% (23) from the west zone, 28% (20) from the

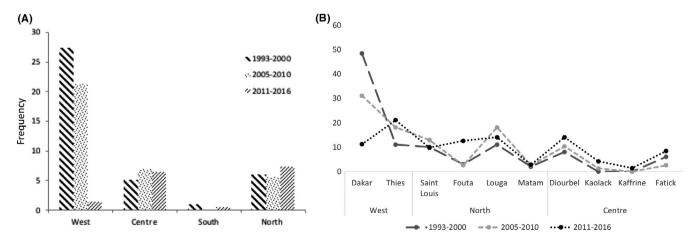


FIGURE 3 A, The frequency of mycetoma cases according to the period and the zones. The West presented more cases during 1993-2000 and 2005-2010. But between 2011 and 2016, the North presented more cases followed by the Centre while the south had less cases despite of the period. B, Temporal evolution of mycetoma cases by regions in Senegal. Between 1993 and 2010, Dakar and Thiès presented more cases than the other regions. Thiès, Fouta and Diouribel presented an increase of cases between me periods 2005-2010 and 2011-2016 while in Dakar a decrease was noted

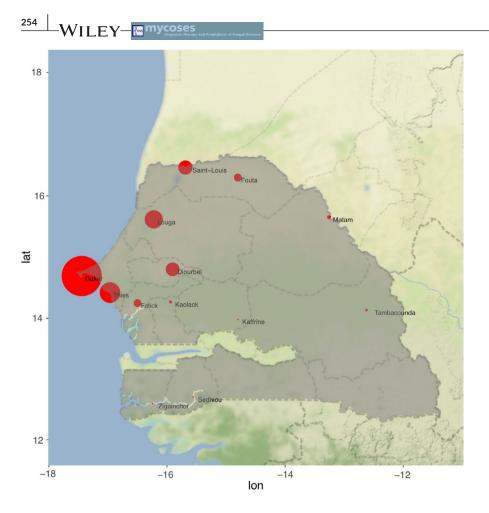


FIGURE 4 The frequency of mycetoma cases according to the regions. West: Dakar (31.62%) Thies (15.81%). Centre: Diourbel (10.28%), Fatick (5.53%), Kaolack (1.58%), Kaffrine (0.4%). North: Louga (13.83%), Saint-Louis (13.83%), Fouta (5.53%), Matam (2.37%). South: Sedhiou, Kolda and Ziguinchor (0.40% each) Tambacounda 1.18%

central zone and 1% (1) from the south zone (Figure 3A). The difference was not statistically different (Kruskal-Wallis, P = .98).

Between the periods 1993-2000 and 2011-2016, a decrease of 19% of the cases in the western and a progressive increase of cases in northern and central zones of, respectively, 13% and 14% were noted. In the Sahelo-Sudanian zone, 98% of mycetoma cases were recorded, and Diourbel, Thies and Fouta presented an increase overtime (Figure 3B).

Dakar in 1993-2000 (48.48%) and 2005-2010 (31.16%) presented more cases than the other regions. Matam, Kaffrine and Kaolack presented less cases regardless of the period (Figure 3B).

4 | DISCUSSION

Mycetoma is a worldwide infectious disease but its distribution is not even. Epidemic countries include Senegal where several cases had been notified.¹⁻⁹ This present study was conducted to map the cases of mycetoma from Senegal. All cases of mycetoma recorded in the laboratory notebooks and indicating the geographical origin of patients were included. Clinical and biological aspects of these cases were published elsewhere.¹⁻⁹ During this period, 337 cases of mycetoma were diagnosed, and for 275 patients, the geographical origin was available and 223 were from Senegal and 22 cases were from Mauritania, Mali and Cape Verde. 98% of the cases were found in the Sudano-Sahelian zone encompassing the north, the west and the centre of the country, where rainfall is between 300 and 700 mm/y, compared to the Sudanian zone represented by the south with a rainfall exceeding 700 mm/y.²² Thus, the distribution of mycetoma cases is in agreement with the rainfall distribution and follows a north-south gradient.

Despite the climate being more desert in the north, more cases were found in the west and the centre.

Overall, the west presented more cases followed by the centre, the north and the south. The north with a desert climate was expected to present more cases, but in addition to nature-related factors, the proximity of health services could explain the predominance of cases in the west and the centre. In fact, there are few mycology laboratories in Senegal and all are in the capital Dakar located in the west and also there are very few dermatologists out of Dakar. Nevertheless, mycetoma is a rural disease and most patients are not accustomed to go to healthcare facilities and prefer to use traditional medicine which underestimate the reported cases. Geographically, the centre is closer to Dakar and has a technical platform with more medical specialties, compared to the rest of the country with the exception of the regions of Dakar (67%) and Thies (33%).

There is variation in the distribution within each zone, and in the west, Dakar presented more cases than Thies. Within the central

zone, the majority of cases arose in Diourbel (58%) and Fatick (31%). In the north, Louga (43%) and Saint Louis (33%) recorded more cases than Fouta and Matam. It would be expected that within a zone, the most rural regions will present a higher prevalence, but the absence of healthcare facilities could underestimate the cases in rural areas compared to the cities. In addition, these rural populations cannot afford the diagnosis and treatment and do not usually have health insurance or universal health coverage.

However, the temporal evolution of mycetoma distribution in Senegal showed a change over the years, and indeed, from 1993 to 2000, the west concentrated the majority of cases but a decrease was noted from 2005, as well as a gradual increase in the northern and central areas.

The change in the distribution across the country could be due to the augmentation of the rainfall occurred starting at 2000 in all of the country with a series of inundation in the west and mostly in Dakar due to urbanization problems.²² In fact, this increase in rainfall could explain the reduction of cases in the west.

Despite the difference in rainfall between the west and the north, the interannual variation concerns all the country,²³ so the difference between the changes in the distribution is more likely due to the availability of the healthcare facilities and the changes in the urbanisation.

In fact, during the period 1993-2000, more than half of the cases (58%) came from the western part of Senegal represented here by two regions namely Dakar and Thies and in this area nearly 2/3 of the cases came from the Dakar region. This corresponds to a period during which Dakar presented large spaces with areas that were not inhabited and where some plants grew. In those spaces spiny plants such as *Faidherbia albida*, *Parinari macrophylla*, *Maytenus senegalensis*, *Ximenia americana*, *Eleais guineensis* (oil palm) bordering the Niayes, and *Casuarina equisetifolia* (Filao),²⁴ that could transmit the pathogens where found, and this could explain the frequency of this mycetoma in the peri-urban area of Dakar. Since then, the city of Dakar has undergone profound changes over the last 20 years with more built space at the expense of vegetation, explaining the decrease of the cases in 2011-2016.

In fact, it is recognised that the transmission of mycetoma agents usually occurs during an injury that introduces the infectious agent into the body, and plants have often been implicated in this mode of transmission.^{11,12}

Dakar, Thies, Diourbel and Louga presented more cases overall, but there is an increase of the cases for Thies and Fouta during 2011-2016, while in Louga and Diourbel, there is a decrease during the same period (Figure 3B).

Fungal agents were responsible for more mycetoma cases with 61.1% (Table 1), and this is in concordance with what has been found by other studies conducted in the country.⁵⁻⁹ The most common species were *Madurella mycetomatis* (52%), *Actinomadura pelletieri* (24.19%), *Leptosphaera senegalensis* (7.66%) and *Actinomadura madurae* (7.25%) (Figure 4).^{5,9} However, no difference was observed in the species distribution across the country; regardless of the zone, eumycetoma was predominant.

This study was retrospective and used the information available in the laboratory records; thus, several cases of mycetoma were excluded because the geographical origin of the patient was not mentioned. Moreover, concerning the identification of the mycetoma agents, for the most part only a group diagnosis could be obtained, that is fungal or actinomycotic, which prevented a mapping of the cases according to the infectious agents responsible.

5 | CONCLUSION

Mycetoma, although endemic in several countries and included in the list of NTDs, remains a disease of unknown prevalence and incidence, and without a control programme. In this study, it is clear that there is a redistribution of cases in Senegal due to geoclimatic changes and infrastructure evolution. There is a need for a census and mapping of mycetoma cases in endemic countries to better understand the epidemiology of these diseases and to guide control measures. For a good establishment of mycetoma mapping, it is crucial to implement case finding system in endemic countries.

ACKNOWLEDGMENTS

The authors thank Jules Gomis for the mapping using Google Earth and the patients.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

AUTHOR CONTRIBUTIONS

Badiane Aïda Sadikh wrote the article. Ndiaye Mouhamadou did the data collection and helped with the analysis. Diongue Khadim and Seck Mame Cheikh diagnose the cases at the laboratory and helped with data collection. Mamadou Alpha Diallo helped with the analysis. Daouda Ndiaye reviewed the article.

ORCID

Aïda Sadikh Badiane Dhttps://orcid.org/0000-0002-1118-4998 Khadim Diongue Dhttps://orcid.org/0000-0002-7033-1169

REFERENCES

- Develoux M, Dieng MT, Mycétomes NB. Encycl Méd Chir. Maladies Infect. 2002;8-606-A:10-11.
- 2. Develoux M, Dieng MT, Kane A, Ndiaye B. Prise en charge des mycétomes en Afrique de l'Ouest. *Bull Soc Pathol Exot*. 2003;96:376-382.
- Develoux M, Ndiaye B, Kane A, Diousse P, Huerre M. Les mycétomes du cuir chevelu et de la nuque à Dakar. J Mycol Med. 1999;9:65-67.
- Dieng MT, Niang SO, Diop B, Ndiaye B. Actinomycetomas in Senegal. Study of 90 cases. Bull Soc Pathol Exot. 2005;98:18-20.
- Dieng MT, Sy MH, Diop BM, Niang SO, Ndiaye B. Mycétomes: 130 cas. Ann Dermatol Venereol. 2003;130:16-19.
- Ndiaye B, Develoux M, Dieng MT, Ndiaye PD, Ndir O. Les mycétomes à la clinique dermatologique de Dakar (Sénégal) aspects épidémiologiques, à propos de 111 cas = Mycetoma in the

WILEY-

department of dermatology, Dakar, Senegal. Epidemiological aspects of 111 cases. *Med Afr Noire*. 1995;42:207-212.

- Ndiaye B, Develoux M, Langlade MA, Kane A. Les mycétomes actinomycosiques. À propos de 27 observations dakaroises. Traitement médical par le cotrimoxazole. Ann Dermatol Venereol. 1994;121:161-165.
- Sy MH, Diouf AG, Diakhate I, et al. Ostéites mycétomiques et mycétomes osseux. *e-mémoires de l'Académie nationale de chirurgie*. 2003;2:11-17.
- Ndiaye DM, Ndiaye PD, Sène MN, et al. Mycétomes diagnostiqués au Sénégal de 2008 à 2010. J Mycol Méd. 2011;21:173-181.
- Verma P, Jha A. Mycetoma: reviewing a neglected disease. Clin Exp Dermatol. 2019;44(2):123-129.
- Klokke AH. Mycetoma pedis due to Madurella mycetomi. J Indian Med Ass. 1965;44:379-380.
- Rey M. Les mycétomes dans l'Ouest africain. Thèse. Paris: Foulon édition; 1961:34.
- Zijlstra EE, van de Sande WW, Welsh O, el Mahgoub S, Goodfellow M, Fahal AH. Mycetoma: a unique neglected tropical disease. *Lancet Infect Dis.* 2016;16(1):100-112.
- 14. Fahal AH. Mycetoma: a thorn in the flesh. *Trans R Soc Trop Med Hyg.* 2004;98(1):3-11.
- Lichon V, Khachemoune A. Mycetoma: a review. Am J Clin Dermatol. 2006;7(5):315-321.
- Rattanavong S, Vongthongchit S, Bounphamala K, et al. Actinomycetoma in SE Asia: the first case from Laos and a review of the literature. *BMC Infect Dis.* 2012;12:349.

- Mathews S, Jadhav R, Reza A, Karim T. Actinomycetoma-The Welsh Regimen in a rural Indian scenario. *Indian J Surg.* 2012;74(6): 480-482.
- Welsh O, Al-Abdely HM, Salinas-Carmona MC, Fahal AH. Mycetoma medical therapy. *PLoS Negl Trop Dis.* 2014;8(10):e3218.
- 19. Fahal AH, Shaheen S, Jones DH. The orthopaedic aspects of mycetoma. *Bone Joint J.* 2014;96(b-3):420-425.
- van de Sande WWJ. Global burden of human mycetoma: a systematic review and meta-analysis. PLoS Negl Trop Dis. 2013;7(11):e2550.
- 21. Abbott P. Mycetoma in the Sudan. *Trans R Soc Trop Med Hyg.* 1956;50:11-24; discussion, 24-30.
- Cissé S. Etude de la variabilité intra saisonnière des précipitations au Sahel: impacts sur la végétation (cas du Ferlo au Sénégal). Climatologie. Université Pierre et Marie Curie - Paris VI; 2016.
- Dieng O, Roucou P, Louvet S. Variabilité intra-saisonnière des précipitations au Sénégal (1951–1996). Sécheresse. 2008;19(2):87-93.
- 24. Situation Economique et Sociale de la Région de Dakar, édition; 2014.

How to cite this article: Badiane AS, Ndiaye M, Diongue K, Diallo MA, Seck MC, Ndiaye D. Geographical distribution of mycetoma cases in senegal over a period of 18 years. *Mycoses*. 2020;63:250–256. https://doi.org/10.1111/myc.13037