

Unrecognised Peripheral Neuropathy and Septic Hand Ulcer as First Presentation of Diabetes Mellitus in a Nigerian Woman. A Case Report and Literature Review

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Abstract

Diabetes mellitus may for the first time present with its classical features or with long term complications which may or may not be recognised. Knowledge of diabetes mellitus and its complications, sociocultural beliefs and perception, patronage of traditional healers may influence time of presentation and hence, outcomes.

We report the case of a female Nigerian food vendor, who was admitted in coma and managed for septic left-hand ulcer and incidental diabetes mellitus. She had painless peripheral neuropathy, which gave her the "ability" to handle hot objects. This was misconstrued as some special gift derived from a benevolent spirit, until she sustained thermal injury and a non-healing wound on her left hand.

She presented to a traditional healer which delayed hospital presentation. She was eventually brought to hospital in coma and related diabetes mellitus was found. She was appropriately managed for hyperglycaemic coma and sepsis as well as wound care and was discharged to the out patients' services on appropriate drugs and life style measures.

Keywords: Hand; Infection; Unrecognised; Peripheral Neuropathy; Diabetes Mellitus.

Introduction

Diabetes mellitus, (DM), may present with characteristic symptoms such as thirst, polyuria, blurring of vision and weight loss¹. In its most severe acute form, ketoacidosis or non-ketotic hyperosmolar states may develop, leading to altered level of consciousness, coma and even death². Diabetes mellitus has many long-term complications that are mostly vascular, microvascular or macrovascular, some of which may be complicated by sepsis.

Type 2 diabetes, mellitus T2DM, is often initially asymptomatic, first becoming apparent due to one or more of the long-term complications which may or may not be recognized. Failure of recognition of the disease and its complications may be due to a

low index of suspicion, lack of or limited knowledge of the disease, or cultural beliefs that could have affected the perception of the disease³. Also, in resource-poor developing countries, patients may present late in the course of the disease because they are unable to afford the cost of care⁴ as well as patronizing alternative medicine practitioners and native doctors who may potentially worsen their outcome³.

Hand ulcers are less-well recognized, and not often referred to as diabetes-related complications,

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compared to diabetes foot ulcers². Available literature suggests that, diabetes-related hand complications occur at a ratio of 1:20 with diabetes-related foot complications⁵. Historically, diabetes hand infection and ulcers as complications of diabetes mellitus were first described in the 70s in the United States and subsequently in Africa and the Indian subcontinent⁶. In west Africa, the first cases of diabetes hand ulcer and infection were reported by Akintewe et al⁷ in the mid-80s, with a later follow-up report by Gill et al⁸, giving detailed description of this condition, which is currently known to show a typical clinical pattern and is now seen more often, if not exclusively, in patients from the tropics, and has been named tropical diabetic hand syndrome (TDHS)^{4,6,9}. The prevalence of TDHS in Nigeria ranges from 1.3% in Zaria, northern Nigeria¹⁰, through 1.6% in the Niger delta region¹¹ to 4% in a study done in the western part of the country⁷. TDHS is considered an acute complex hand complication affecting patients with diabetes in the tropics usually following very minor hand injury⁹. Pathophysiologically, once infection is established, a vicious cycle sets in with swelling of the surrounding tissues, resulting in stasis and adhesion of platelets and leukocytes in vessel walls, resulting in thrombosis and occlusion of the small vessels in the area of infection. These changes result in localised tissue ischaemia, and possibly gangrene (Meleney's gangrene)¹².

Pattern of clinical presentation of TDHS ranges from localised swelling and cellulitis, with or without ulceration of the hand, through to progressive fulminant hand sepsis, and gangrene affecting the entire hand and may be fatal^{10, 12-14}. TDHS differs from an entity referred to as Diabetic hand syndrome, DHS, is a collagen and rheumatologic disorder characterised by skin fibrosis with limitation of movements of the small joints of the hand in persons with T2DM⁹. While TDHS has been reported from some parts of Nigeria^{5, 7, 9, 10, 12}, the same cannot be said of Abuja, the Federal Capital territory of Nigeria. This case is here reported to sensitize clinicians to the existence of a potentially serious hand sepsis as a complication of diabetes mellitus.

Case Report

Mrs. KM is a 45-year-old left handed food vendor, who was admitted in coma with a septic left-hand ulcer. She is a Nigerian woman, who had become famous in her local community because of her said "unusual ability" to handle hot objects such as cooking pots from the fireplace, without feeling pain. In that community, it was believed that, she had some special benevolent powers which gave her the advantage of handling hot objects.

Unfortunately, she sustained burns and a resulting open wound in her left hand while trying to replace hot coal to keep the fire burning. In the following two weeks before presentation, the wound had expanded to cover most of her left little digit with a swollen hand. Believing that she needed native intervention, she presented to an herbalist who administered some treatment which was ineffective. In the days that followed, she was observed by relatives to be having polyuria and fever. The morning of the day of hospital presentation, she became restless and gradually lapsed into unconsciousness and was brought to hospital by relatives who gave part of the history, including their observation of polyuria and fever.

She did not smoke cigarettes or drink alcohol, was not a known hypertensive or diabetic patient. There was no history of seizure disorder or head injury. In addition she did not use illicit or recreational drugs, but ingests alternative medicines.

Examination at entry revealed; an unconscious middle-aged woman with recurrent focal seizures involving the left half of the face. She was pale, markedly dehydrated and febrile with a temperature of 38.9°C. She was anicteric with no pedal edema. Her pulse rate was 110 bpm with a blood pressure 130/90 mmHg. The patient weighed 67 kg with a height of 1.63 m, and BMI 25.2 kg/m², (checked after recovery). Apart from depressed ankle reflexes, the motor function of the limbs was apparently within normal limits. No lateralizing signs. Her peripheral pulses in both upper and lower limbs were palpable, no thickened vessels. Inspection of her hands revealed a linear left palmer ulcer measuring 2.5 cm across by 4.0 cm vertically with a scab and deep abscess involving the little finger (Fig. 1a). The dorsum of the left hand was swollen and fluctuant with desquamation of the skin (Fig. 1b). The patient

was examined further after she had regained consciousness; we elicited peripheral neuropathy of glove and stocking distribution in both upper and lower limbs. Laboratory investigations revealed the following significant findings: Random blood glucose of 36 mmol/l, leucocytosis of 21,000 per cm³; with neutrophilia of 80%, Urea, 15 mmol/l; Sodium, 151 mmol/l; Chloride, 109 mmol/l; Bicarbonate, 30 mmol/l; potassium, 4.0 mmol/l. Plasma osmolality was 361 mOsmol/l with an anion gap of 17. Wound swab microbiology revealed growth of *Staphylococcus aureus*, while blood was apparently sterile on culture. A plain radiograph of the left hand did not show evidence of osteomyelitis.

A diagnosis of hyperglycaemic emergency in coma, with sepsis syndrome focused on the left hand was made. After a treatment regimen including; intravenous fluid rehydration, insulin and antibiotic administration, she regained consciousness. The wound was debrided, abscess drained and followed by daily wound dressing. Tetanus toxoid was given and metformin added subsequently. She was also found to be hypertensive for which antihypertensive was added. With this treatment regime and acceptable blood glucose levels observed, the patient was discharged to the outpatients' services on insulin and metformin. She had diabetes-related counselling and commenced hand physiotherapy before leaving the ward. Wound healing with some deformity of the little digit was observed eight weeks after, (Fig. 2a, 2b).



Figure 1a and b: Shows left hand with a linear left palmer ulcer with a scab and deep abscess involving the little finger (1a), as well as swelling of the dorsum and desquamation of the skin (1b).



Figures 2a and b: Show post healing deformity of the little finger as viewed from the dorsal and plantar surfaces respectively.

Discussion



1a

Although diabetes mellitus, (DM), maybe asymptomatic, it may present with its classical symptoms, or for the first time in hyperglycaemic emergencies with or without coma. It often shows up with one or more of its known long-term complications, depending on the prevailing health seeking attitude^{1,2}. These complications that are mostly vascular, (microvascular or macrovascular), may be worsened by sepsis¹⁵. The case we present here, is one with an obvious peripheral neuropathy, involving the upper limb, which was misconstrued. Because of the lack of knowledge of diabetes and certain misconception and wrong belief, the patient failed to present early for standard medical care, hence the worsened morbidity and near-mortality characteristic of this condition, commonly referred to as TDHS^{10, 12-14}.

Hand infections in diabetic patients is less recognized than foot sepsis as a long-term complication², hence is not commonly mentioned amongst specific diabetic complications and so under-reported^{16,17}. Our patient had undiagnosed DM which went unrecognized; had already developed complications, (neuropathy), and then decompensated after infection had set in. Up until that point, because of their misconception and poor health seeking behaviour, her hospital presentation was still delayed. This may represent a pattern in which an undiagnosed, hence uncontrolled case of DM with unrecognized neuropathy, and had severe infection developing due to impaired immunological response¹⁴.

Studies have identified factors that may be responsible for failure to recognize a disease to include; a low index of suspicion, lack of knowledge of the disease, inappropriate cultural beliefs and perception of the disease state^{3, 10,12-14}. Furthermore, in developing economies, fear of hospital bills for services have also been earlier reported as a reason for late hospital presentation⁴. These factors may thus drive delay in hospital presentation.

Upper limb neuropathy has been variously affirmed as a risk factor for hand ulcers in persons with DM^{5, 9,10,13,14,16-20}. Benotmane et al¹⁸ however, reported upper limb neuropathy in 65.4% of DM subjects with infected hand ulcers, while a Nigerian study found 58.3% of diabetic patients with septic hand ulcer having

upper limb neuropathy¹⁰. This suggests that, there are other related factors. There may be concomitant lower-limb neuropathy as well in DM patients¹⁹, a finding mirrored by our patient, who had neuropathy of the glove and stocking distribution.

Interestingly, Gill et al⁸ opined that, hand sepsis in DM patients may not necessarily have associated upper limb neuropathy or ischemia, corroborating a previous finding by Archibald et al¹³, who reported an absence of clinical evidence of upper-limb neuropathy and/or peripheral vascular disease in DM patients with infected hand ulcer or TDHS in Tanzania, despite having demonstrable lower-limb neuropathy. This differential distribution in upper and lower limbs neuropathy may be related to the dimensions and class of affected nerve fibers²¹.

Apart from neuropathy, other independent associations identified for TDHS are poor glycaemic control, malnutrition and insulin treatment⁴. Insulin treatment may be a reflection of the degree of poor glycaemic control. At presentation, our patient definitely required insulin⁴, and subsequently oral hypoglycaemic agents. She showed no obvious features of malnutrition.

Some characteristics of DM patients with TDHS include a history of minor antecedent hand trauma and delayed hospital presentation, usually after seeking the help of a traditional healer first^{4, 9,10,12,20}. These were shown by the case reported here following a minor burn injury. Other characteristics are female gender predisposition, mostly type 2 DM, middle age and poor drug compliance in previously diagnosed cases of DM^{10,18}. Our patient displayed most of these characteristics. The importance of traditional healing in African setting cannot be overemphasized; when our patient sustained her burn injury, she first consulted a traditional healer, where some unknown substances and incantations were said to have been administered; our patient's ability to lift hot objects was perceived as some benevolent powers rather than an abnormality or disease. Visiting a traditional Healer first, delayed her hospital presentation as characteristic in low-income countries^{8-12, 20}.

Developing economies such as Africa's, exhibit a

wide range of socio-cultural perspectives on disease concepts that are often based on traditional belief systems^{22,23}. Some of these cultural perspectives, which may include sorcery, magic, and voodoo, are often at variance with current medical concepts²³. These wrong perceptions of disease may increase morbidity and mortality since patients may associate their clinical condition to spiritual forces requiring traditional rather than orthodox care. The care given by these traditional healers may be hazardous^{7,24}. This possibility has been corroborated by some studies in Nigeria that examined the possible role of traditional healers in the spread of viral diseases such as HIV infection²⁴, as well as complications that arise from the activities of traditional bonesetters²⁵.

Looking at the possible differential diagnosis, hand ulcers may be found in other diseases such as; leprosy, which is a cause of neuropathy²⁶. However, the short duration, absence of typical anaesthetic skin lesions and defining nodules largely eliminated that as a possibility. Malnutrition with various causes including alcoholism may present as a risk factor in limb ulcers, especially due to the associated deficiency in B-vitamins⁴. However, ulcers due to malnutrition are more frequent in lower limbs. A rare cause of hand ulcers with neuropathy as a risk factor is Bureau-Barriere syndrome, a condition characterized by acral ulcers complicated by osteomyelitis with sensory-motor polyneuropathy²⁷. Collagen disorders may also cause hand ulcers⁹.

The pattern of presentation is worthy of mention. Our patient who was not a previously known person with diabetes, presented in hyperglycaemic emergency for the first time, alongside the hand infection. In a Nigerian study, 33% of the patients with TDHS who were also previously known diabetic patients, presented with diabetic ketoacidosis¹⁰. T2DM can be asymptomatic and lead to complications, many of which may remain undetected especially if screening is inadequate. These complications may be minimized by education and aggressive metabolic control.

Assessment and definitive management of hand ulcers is vital. As with foot ulcers where about 67% of cases have been reported to have osteomyelitis in

one study, examination for osteomyelitis in upper limb ulcers is important, since it increases morbidity²⁸. A study in Zaria, northern Nigeria, reported an osteomyelitis prevalence of 25% among patients with TDHS. We did not find evidence of osteomyelitis in the single case reported here, using radiographic examination²⁹. Simple bone probing method could be useful in assessing for osteomyelitis especially in resource-poor setting²⁹. Bacteriological profiling is important as well, as it acts as a guide towards prescribing appropriate antimicrobial agents. The isolation of staphylococcus from our patient's ulcer was in keeping with a previously reported bacteriological pattern profile⁴.

Hospital data from centres with more experience in TDHS cases have emphasized the need for prompt and aggressive intervention due to the associated high morbidity and mortality. The prospective study by Benotmane et al.¹⁰ reported long durations of hospitalization, a mortality rate of 19.2% and a minor amputation rate of 23.1%. The study reported deformities and subjective dysfunction rate of 53.8%. Thus, prevention should be the watchword in regions lacking resources such as Africa, considering the disease burden on the patient, the family, the community and the nation at large.

With the rising prevalence of lifestyle diseases such as DM currently playing a major role in epidemiological transition in resource-poor countries³⁰ such as Nigeria, concerted effort must be made to educate the public on disease symptoms, complications and need for early presentation to hospital. The high morbidity and mortality associated with TDHS calls for prompt and aggressive care.

References

1. Expert Committee on the Diagnosis and Classification of Diabetes Mellitus. Report of the expert committee on the diagnosis and classification of diabetes mellitus. *Diabetes Care* 2003; Suppl 1:S5-20. doi: 10.2337/diacare.26.2007.s
2. Alberti KG, Zimmet PZ. Definition, diagnosis and classification of diabetes mellitus and its complications. Part 1: diagnosis and classification of diabetes provisional report of a WHO consultation.

3. Alberti KG, Zimmet PZ. Definition, diagnosis and classification of diabetes mellitus and its complications. Part 1: diagnosis and classification of diabetes mellitus provisional report of a WHO consultation. *Diabet Med* 1998; **15**:539-553.
4. Agodirin OS, Aremu I, Rahman GA, Olatoke SA, Akande HJ, Oguntola AS, et al. Prevalence of Themes Linked to Delayed Presentation of Breast Cancer in Africa: A Meta-Analysis of Patient-Reported Studies. *JCO Glob Oncol* 2020; 731 – 742.
5. Abbas ZG, Archibald LK. Tropical diabetic hand syndrome. Epidemiology, pathogenesis, and management. *Am J Clin Dermatol* 2005; **6**:21-28.
6. Lawal Y, Ogirima M, Salisu M, Dahiru I, Girei B. Tropical diabetic hand syndrome: Surgical management and proposed classification. *Archives of International Surgery* 2013; **3**:124.
7. Abbas ZG, Lutale J, Gill GV, Archibald LK. Tropical diabetic hand syndrome: risk factors in an adult diabetes population. *Int J Infect Dis* 2001; **5**:19-23.
8. Akintewe TA, Odusan O, Akanji O. The diabetic hand-5 illustrative case reports. *Br J ClinPract* 1984; **38**:368-371.
9. Gill GV, Famuyiwa OO, Rolfe M, Archibald LK. Serious hand sepsis and diabetes mellitus: specific tropical syndrome with western counterparts. *Diabet Med* 1998; **15**:858-862.
10. Akhuemokhan K., Echekwube P, Bakpa F, Okhodare P. Tropical Diabetic Hand Syndrome: Prevention through education. *S Afri J Clinical Nutr* 2011; **24**:205-206.
11. Okpe I, Amaefule K, Dahiru I, Lawal Y, Adeleye A, Bello-Ovosi B. Tropical diabetic hand syndrome among diabetic patients attending endocrine clinic of Ahmadu Bello University Teaching Hospital, Shika Zaria, North Central Nigeria. *Sub-Saharan African Journal of Medicine* 2016; **3**:106.
12. Unachukwu C, Anochie I. Hand ulcers/infections and diabetes mellitus in Port Harcourt, Rivers State, Nigeria. *Anil Aggrawal's Internet Journal of Forensic Medicine and Toxicology* 2005; **6**. Accessed 01/10/22
13. Okpara T, Ezeala-Adikaiibe B, Omire O, Nwonye E, Maluze J. Tropical diabetic hand Syndrome. *Ann Med Health Sci Res* 2015; **5**:473.
14. Archibald LK, Gill GV, Abbas Z. Fatal hand sepsis in Tanzanian diabetic patients. *Diabet Med* 1997; **14**:607-610.
15. Centers for Disease Control and Prevention (CDC). Tropical diabetic hand syndrome--Dar es Salaam, Tanzania, 1998-2002. *Morb Mortal Wkly Rep* 2002; **51**:969-970.
16. Cade WT. Diabetes-related microvascular and macrovascular diseases in the physical therapy setting. *Phys Ther* 2008; **88**:1322-1335.
17. Abbas ZG, Gill GV, Archibald LK. The epidemiology of diabetic limb sepsis: an African perspective. *Diabet Med* 2002; **19**:895-899.
18. Tiwari S, Chauhan A, Sethi NT. Tropical diabetic hand syndrome. *Int J Diabetes Dev Countries* 2008; **28**:130-131.
19. Benotmane A, Faraoun K, Mohammedi F, Benkhelifa T, Amani ME. Infections of the upper extremity in hospitalized diabetic patients: A prospective study. *Diabetes Metab* 2004; **30**:91-97.
20. Coppini DV, Best C. A case of hand ulceration in the diabetic foot clinic--a reminder of hand neuropathy in 'at risk' patients. *Diabet Med* 2000; **17**:682-683.
21. Yeika EV, Tchoumi-Tantchou JC, Foryoung JB, Tolefac PN, Efié DT, Choukem SP. Tropical diabetic hand syndrome: a case report. *BMC Res Notes* 2017; **10**:1-4.
22. Hovaguimian A, Gibbons CH. Diagnosis and treatment of pain in small-fiber neuropathy. *Curr Pain Headache Rep.* 2011; **15**(3):193-200.
23. Sanni A. Diagnosis through rosary and sand: Islamic elements in the healing custom of the Yoruba (Nigeria). *Med Law* 2002; **21**:295-306.
24. Pollanen MS. Alleged lethal sorcery in East Timor. *Forensic Sci Int* 2004; **139**:17-19.
25. Peters EJ, Immananagha KK, Essien OE, Ekott JU. Traditional healers' practices and the spread of HIV/AIDS in south eastern Nigeria. *Trop Doct* 2004; **34**:79-82.
26. Omololu B, Ogunlade SO, Alonge TO. The complications seen from the treatment by traditional bonesetters. *West Afr J Med* 2002; **21**:335-337.
27. Lema T, Woldeamanuel Y, Asrat D, et al. The pattern of bacterial isolates and drug

- Sensitivities of infected ulcers in patients with leprosy in ALERT, Kuyera and Gambo hospitals, Ethiopia. *Lepr Rev* 2012; **83**:40-51.
28. Dissemond J, Knab J, Goos M. [Bureau-Barrière syndrome of the hand. A case report on an unusual localization. *Hautarzt* 2004; **55**:371-375.
29. Newman LG, Waller J, Palestro CJ, et al. Unsuspected osteomyelitis in diabetic foot ulcers. Diagnosis and monitoring by leukocyte scanning with indium in 111 oxyquinoline. *JAMA* 1991; **266**:1246-1251.
30. Boulton AJM, Kirsner RS, Vileikyte L. Clinical practice. Neuropathic diabetic foot ulcers. *N Engl J Med* 2004; **351**:48-55.
31. Setel PW. Non-communicable diseases, political economy, and culture in Africa: anthropological applications in an emerging pandemic. *Ethn Dis* 2003; **13**(2 Suppl 2):S149-S157.