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REVIEW

Tele-leprology: A literature review of applications of telemedicine and tele-education to leprosy

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Summary Global efforts to eliminate leprosy have brought about a steep decline in prevalence; however, new cases are continually detected. Without early diagnosis and appropriate management, these individuals are at risk of disability, disfigurement, and social stigma. Telemedicine and tele-education are increasingly utilised strategies to maintain access to expert healthcare for leprosy patients scattered in low-accessibility areas. However, an overview of tele-leprology, the application of these strategies specifically to leprosy, is currently unavailable. This review provides such an overview and discusses future directions for research and implementation.

Introduction

Leprosy, a chronic infectious disease due to *Mycobacterium leprae*,[1] remains an important cause of disability, disfigurement, and social stigma.[2–5] At the end of the first quarter of 2013, the World Health Organisation (WHO) reported 189,018 registered cases of leprosy in 115 countries or territories. The prevalence rate, 0·33 per 10,000 population,[6] demonstrates the epidemiological shift that has taken place since 1985, when the prevalence rate stood at

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12 per 10,000 population.[7] The elimination target, defined by the World Health Assembly as the reduction of prevalence to a level below one case per 10,000 population,[8] was achieved at the global level by 2000.[7] However, leprosy continues to pose a threat to public health. During the year 2012, 232,857 new cases were reported to the WHO. The number increased in the African, South-East Asian, and Western Pacific Regions, with altogether 6,231 more new cases reported in 2012 than in 2011. The rate of new cases with grade-2 disabilities per 100,000 population also increased from 0·22 in 2011 to 0·25 in 2012.[6]

A challenge in the 'post-elimination era' is maintaining access to high quality healthcare for all leprosy patients in order to ensure early detection and appropriate management. Barriers to access in rural areas include distance, poverty, and low health literacy. The latter two, along with poor coordination between healthcare providers, impede access in urban slums. Expertise is a key determinant of healthcare quality, but matching supply with demand is difficult when leprosy patients are scattered, including in low-accessibility areas.[1] Moreover, fear exists that the global pool of expertise will dry up if future healthcare providers rarely encounter leprosy patients in practice. A significant portion of the WHO's *Enhanced Global Strategy for Further Reducing the Disease Burden due to Leprosy (Plan Period: 2011–2015)* is devoted to this challenge. Specifically, it addresses the need to strengthen routine and referral services within the integrated health systems in all endemic countries and to develop sustainable training strategies at global and national levels. The strategy lists telemedicine as one of "a range of options for effective communication and transferring information."[9]

Tele is a Greek word meaning 'distance' and *mederi* is a Latin word meaning 'to heal.' Telemedicine has multiple definitions, but here we invoke the WHO definition of telemedicine as: "The delivery of health care services, where distance is a critical factor, by all health care professionals using information and communication technologies for the exchange of valid information for diagnosis, treatment and prevention of disease and injuries, research and evaluation, and for the continuing education of health care providers, all in the interests of advancing the health of individuals and their communities."[10] Telemedicine is divided into two primary forms. Store-and-forward, or asynchronous, telemedicine involves exchange of pre-recorded data between two or more individuals at different times (Figure 1).

Real time, or synchronous, telemedicine involves immediate exchange of data. This form often utilises videoconferencing, but both forms may utilise various media including text, audio, video, and still images.[11]

Tele-education is also relevant to the challenge of maintaining access to expert healthcare for all leprosy patients. The trend towards integration of leprosy services into general health systems has led to an increase in passive case-finding. This strategy relies to a greater extent than active case-finding on the capacity of primary care providers to diagnose, treat, and appropriately refer cases of leprosy.[9] The WHO defines tele-education as "a dynamic process by which change can be catalysed in attitudes, knowledge, information and skills, by means of information and communications technologies, by and for consumers, health professionals and communities, for the purpose of fostering improved health."[10] Tele-education is divided into asynchronous and synchronous forms and utilises a variety of media similar to telemedicine.[12]

In a 2007 editorial to this journal, Kumar *et al.* encouraged the 'development of tele-leprology and efficient networking.'[13] Research on tele-leprology, defined here as the application of telemedicine or tele-education specifically to leprosy, is gaining momentum.

Figure 1. A case of leprosy diagnosed by store-and-forward teledermatology in Botswana. This patient with ill-defined diffuse hypopigmented plaques was diagnosed with multibacillary leprosy through the African Teledermatology Project (africa.telederm.org) and was confirmed locally with slit-skin smear. (Courtesy of Campbell Stewart, MD.)

However, to our knowledge, a review is currently unavailable. Our objectives are to present the clinical and educational tele-leprology initiatives in the literature and to explore future avenues for research and implementation.

Methods

We searched the online databases PubMed and Scopus for publications between 1960 and April of 2014 using the following syntax: *(tele-leprology)* ***OR*** *(leprosy OR Hansen's disease)* ***AND*** *(telemedicine OR teledermatology OR tele-education)*. We subsequently repeated this search using French, Portuguese, and Spanish syntax. References from the retrieved publications were used to identify other relevant sources. Publications were eligible for review when they described an application of telemedicine or tele-education specific to leprosy. No language limits were applied and non-English sources were translated. The search yielded 71 results. These results were assessed for eligibility and 11 publications describing clinical tele-leprology initiatives and five publications describing educational tele-leprology initiatives were included in this review.

Results

TELE-LEPROLOGY: CLINICAL APPLICATIONS

Table 1 summarises the available literature on clinical tele-leprology initiatives.

The literature, published between 2001 and 2013, derives from Brazil, India, Indonesia, Italy, and Taiwan.[14–24] The initiatives utilised a variety of technologies to achieve aims ranging from diagnosis and treatment at the individual level to control and prevention at the population level. Notably, Trindade *et al.* in 2008 compared store-and-forward

Table 1. Clinical tele-leprology initiatives

Author(s)	Country	Description	Study design	Number of people assessed	Outcome
Mukherjee *et al.* (2001)[14]	India	Hybrid store-and-forward and real time telemedicine prototype focusing on leprosy enabled healthcare providers to annotate images and draw skin lesions using computer graphics	Activity report	Not specified	Not specified
Nunzi *et al.* (2003)[15]	Italy	Remote diagnostic support service	Case report	1	Transmission of clinical images via email and biological material via mail resulted in a remote diagnosis of lepromatous leprosy
Kundu *et al.* (2005),[16] Kundu *et al.* (2007)[17]	India	Skin Patch Viewer application enabled healthcare providers to view drawings of skin lesions on file for leprosy patients using PDAs	Activity report	Not specified	Not specified
Fadel *et al.* (2006),[18] Vigolo *et al.* (2008)[19]	Brazil	*Prontuário Eletrônico Para Pacientes de Hanseníase Via Web* (PEPHans) and Pocket-PEPHans prototypes enabled healthcare providers to collect and visualise leprosy patient data via web and PDAs, respectively, while providing monitoring and decision-support for treatment and discharge	Activity report	Not specified	Not specified
Trindade *et al.* (2008)[20]	Brazil	Store-and-forward teledermatology consultations for suspected cases of leprosy in public health clinics	Comparison of the diagnosis of the teledermatologist with the diagnosis made by a dermatologist via in-person consultation	106	The diagnostic agreement was 74%, the sensitivity was 78%, and the specificity was 31%
Ganapati (2009)[21]	India	Mobile phones and pagers for paramedical workers and community volunteers to contact physicians while conducting door-to-door surveys in rural areas and urban slums	Activity report	Not specified	Perceived improvement in speed of communication and patient care

Table 1. *continued*

Author(s)	Country	Description	Study design	Number of people assessed	Outcome
Cardoso *et al.* (2013)[22]	Brazil	Smart Reader® cell phone-based test reader platform for the new NDO-LID® quantitative rapid diagnostic test for MB leprosy designed to detect IgM and IgG antibodies to the PGL-1 mimetic, ND-O, and LID-1 in patient sera	Direct comparison of serologic responses between the NDO-LID® test assessed by Smart Reader® and a lab-based ELISA for the detection of IgM antibodies to PGL-1	441 (108 MB leprosy patients, 104 PB leprosy patients, 75 household contacts, 53 pulmonary tuberculosis patients, and 101 healthy endemic controls)	The NDO-LID® test assessed by Smart Reader® detected a greater proportion of leprosy patients than the PGL-1 ELISA and had a sensitivity of 87%, a specificity of 96.1%, a PPV of 94%, and a NPV of 91.4% for MB leprosy
Rachmani *et al.* (2013)[23]	Taiwan	Health information system to track leprosy patient home visits from direct treatment observers and public health nurses	Qualitative analysis of interviews	3 (public health officials)	Perceived improvement in treatment monitoring and compliance, contact tracing, and disease surveillance
Rachmani *et al.* (2013)[23,24]	Indonesia	Standardised digital format to compile reports on leprosy patient demographics and treatment from paper records at primary health centres	Qualitative analysis of interviews, focus group discussions, and observations at 3 primary health centres	3 (public health officials)[23] and 12 (6 public health officials, 3 primary health centre managers, and 3 leprosy health workers)[24]	Health information system expansion perceived as an opportunity to improve treatment monitoring and compliance, contact tracing, and disease surveillance

PDA = Personal digital assistant. MB = Multibacillary. IgM = Immune globulin M. IgG = Immune globulin G. PGL-1 = Phenolic glycolipid I. ND-O = Natural disaccharide with octyl linkage. LID-1 = Leprosy Infectious Disease Research Institute diagnostic 1. ELISA = Enzyme linked immunosorbent assay. PB = Paucibacillary. PPV = Positive predictive value. NPV = Negative predictive value.

teledermatology with in-person dermatology consultations for 108 suspected cases of leprosy near Sao Paulo, Brazil and demonstrated an overall diagnostic agreement of 74%, a sensitivity of 78%, and a specificity of 31%.[20]

Several initiatives incorporated mobile technologies including cell phones[21,22] and personal digital assistants.[16,17,19] Cardoso *et al.* in 2013 in Brazil evaluated a new quantitative rapid diagnostic test for multibacillary leprosy using a cell phone-based test reader platform, similar to that previously tested for the rapid diagnosis of malaria, tuberculosis, and the human immunodeficiency virus.[22] Weighing 65 grams, this reader attached to the camera unit of a cell phone, captured images of lateral flow immuno-chromatographic assays, and utilised a cell phone application to process the images for test validation and automated reading. Data transmission to a central server enabled visualisation of the results on a world map through geo-tagging.[25] Cardoso *et al.* demonstrated a sensitivity of 87%, a specificity of 96.1%, a positive predictive value of 94%, and a negative predictive value of 91.4%. In addition to consistency and objectivity, the authors identified record keeping, rapid solicitation of second opinions from off-site experts, and real time spatio-temporal analysis of disease prevalence and incidence as advantages of this technology over visual readings.[22]

TELE-LEPROLOGY: EDUCATIONAL APPLICATIONS

Table 2 summarises the available literature on educational tele-leprology initiatives.

The studies, based in Brazil and India between 2004 and 2009, utilised diverse technologies.[26–30] For example, a leprosy extension university course offered through the Sao Paulo Telehealth Portal for Brazilian Family Health Team members in the Amazon incorporated three-dimensional video animations and case simulation, classes via video-streaming, and other web-based resources.[29,30] All studies reported favourable learner satisfaction ratings ranging from 89% to 98%.[26–30] One tested medical students before and after an initiative and detected a statistically significant increase in leprosy-related knowledge.[27] Another assessed recall of the leprosy-related content of an initiative among non-dermatologist participants and found a mean correct response rate of 53%.[28]

Discussion

Limited conclusions may be drawn from the small number of tele-leprology initiatives published thus far. The reports derive primarily from India and Brazil.[14,16–22,26–30] Although these countries contributed the highest number of new cases to the global total in 2012 (58% and 14% respectively),[6] the data are not necessarily translatable to other contexts. Only two studies have quantitatively assessed clinical tele-leprology initiatives.[20,22] More data are available on educational tele-leprology initiatives; however, they are limited to learner satisfaction ratings[26–30] and assessment of knowledge[27,28] rather than competence or performance.

With these limitations in mind, however, the literature does support the value of tele-leprology. The 74% diagnostic agreement reported by Trindade *et al.*[20] is higher than the 65.3% weighted average aggregated diagnostic agreement rate between store-and-forward teledermatology and clinic dermatology reported by Warshaw *et al.* in a systematic review of controlled trials published in English between 1990 and June of 2009.[31] Trindade *et al.* partly attributed the specificity of 31% to the teleconsultant's inability to palpate peripheral

Table 2. Educational tele-leprology initiatives

Author(s)	Country	Description	Study design	Number of people assessed	Outcome
Misra *et al.* (2004)[26]	India	Neurology educational video-conferences discussed rare movement disorders associated with leprosy	Participant feedback collected through an unspecified mechanism	Not specified	Participants expressed satisfaction with the quality and usefulness of the video-conferences
Miot *et al.* (2007)[27]	Brazil	Virtual reality animation to educate about leprosy	Questionnaires to evaluate the quality of the intervention and to assess impact on leprosy knowledge among medical students	44	Over 89% of participants fully or partially agreed that the intervention was a quality educational experience and the mean correct response rate to 9 knowledge-based questions increased from 5·5 + / − 1·7 to 7·6 + / − 1·2 before and after the intervention
Paixão *et al.* (2009)[28]	Brazil	*Dermatúnel*, an interactive virtual reality environment, included leprosy as one of eight educational modules	Questionnaires to assess the degree of participant satisfaction with *Dermatúnel* and recall of the leprosy module answered by participants selected at random after completing all modules	773 (103 dermatologists and 670 non-dermatologists)	90% of participants ranked *Dermatúnel* "very good" or "excellent" and the mean correct response rate to questions assessing recall of the leprosy module among non-dermatologists was 53%
Paixão *et al.* (2009)[29,30]	Brazil	Leprosy extension university course	Course evaluation completed by participants and assessed using the WebMAC Professional tool	48	More than 95% of answers under each WebMAC motivational dimension* were positive and, according to WebMAC's classification criteria, 98% of participants evaluated the course as "Awesome Course!"

WebMAC = Web Site Motivational Analysis Checklist. *Stimulating, meaningful, organised, easy-to-use.

nerves.[20] Recent data suggests, however, that alternative methods to detect nerve thickening such as sonography[32] may overcome this limitation. Cardoso *et al.* demonstrated the utility of a cell phone-based test reader platform for a new quantitative rapid diagnostic test for multibacillary leprosy.[22] Data revealing favourable learner satisfaction ratings,[26–30] knowledge gains,[27] and knowledge recall[28] with educational tele-leprology initiatives are also promising.

FUTURE DIRECTIONS FOR RESEARCH

Ongoing dialogue in the scientific community is essential to optimise the value of teledermatology for leprosy patients. Diagnostic accuracy and concordance should be assessed not only between teledermatology and clinic dermatology, but also between teledermatology and primary care. For acute complications, such as immunological reactions to *Mycobacterium leprae*, the reduced time to treatment observed with teledermatology may prevent disabilities.[31,33] For chronic complications, such as neuropathic ulcers, teledermatology may facilitate frequent follow-ups.[34] Studies should therefore assess management accuracy and concordance as well as clinical outcomes. Several randomised controlled trials have demonstrated comparable clinical outcomes between store-and-forward teledermatology and clinic dermatology.[35–37] These trials were conducted in the United States, however, and do not provide data specific to leprosy. Investigation into tertiary forms of teledermatology that facilitate communication among dermatologists is also warranted.[38]

Since leprosy patients often require the attention of a multi-disciplinary team, future research should extend beyond teledermatology to incorporate other disciplines. For example, leprosy patients remain at risk for potentially sight-threatening ocular complications even after completing multi-drug therapy.[39] Research on applications of tele-ophthalmology[40] to leprosy may therefore provide a valuable contribution to the literature. Other relevant disciplines include teleneurology, telepodiatry, teleradiology, telerehabilitation, and telesurgery. In addition to assessing impact on individual patients, studies should continue to explore the potential of telemedicine to impact leprosy control and prevention at the population level.

In the domain of education, both formal curricula and practice-based learning merit further investigation. Studies in the United Kingdom, the Netherlands, and Israel assessing the percentage of teleconsultations perceived by general practitioners to be of educational value have reported rates from 61% to 75%.[41–43] A study in the United States examining the self-perceived competency of medical trainees with clinical dermatology skills before and after educational teleconsultations revealed significant improvement in five of six competencies assessed (diagnosis, management, physical examination, knowledge, and differential diagnosis) and non-significant improvement in the sixth competency (dermatology history-taking). 88% were very satisfied with the teaching methodology and 86% were very likely to apply the information in their future practice.[44] Studies in Burkina Faso and South Africa have also detected significant improvement in diagnostic agreement between general practitioners and dermatologists participating in teledermatology over time.[45,46] This literature is encouraging, however, its applicability to leprosy remains unknown.

Tele-leprology could furthermore be explored as a modality to educate leprosy patients and their communities. Studies in an underserved area of the United States demonstrated comparable effectiveness between in-person education and tele-education for teaching self-management to adults with diabetes mellitus.[47,48] Self-care is a cornerstone of disability prevention and management in leprosy patients;[49] however, not all are aware of best practises. For example, only 26·9% of leprosy patients enrolled in a study in Tamil Nadu, India reported knowledge of how to take care of the eyes.[50] In addition to filling knowledge gaps, tele-education could address misconceptions, including those that perpetuate stigmatisation of leprosy patients.[3–5]

FUTURE DIRECTIONS FOR IMPLEMENTATION

The priority for tele-leprology should be to provide underserved patients with expert healthcare. Kanthraj in India advocates for a national teledermatology system linking primary care providers in community health centres to dermatologists in tertiary care centres.[51] An advantage of addressing leprosy in the context of other dermatological conditions is the potential to detect cases not initially suspected by the primary care provider. An alternative disease-targeted approach is exemplified by the Chunampet Rural Diabetes project, which trains unemployed youth in Tamil Nadu, India in retinal colour photography and disperses them in vans to local villages. The vans are equipped to screen not only for retinopathy, using mobile phones to transmit photographs to ophthalmologists in Chennai, but also nephropathy, neuropathy, and cardiovascular complications.[52] Leprosy likewise affects multiple organ systems, and tele-leprology could coordinate multi-disciplinary care. These approaches are not mutually exclusive and merely illustrate the myriad possibilities. In parallel with telemedicine, tele-education should be expanded to enhance the capacity of primary care providers to diagnose, treat, and appropriately refer cases of leprosy.

Recent technological advances, particularly in satellite and Internet communication, have revolutionised telemedicine and tele-education. Mobile devices are increasingly equipped with cameras and data transmission capabilities. Advances in digital imaging and computerised measurements are especially relevant to tele-leprology.[51] Implementation research is required, however, to determine the local availability and distribution of these technologies. Of note, tele-leprology may serve as a bridge connecting low-resource settings to outside resources. For example, histopathology is valuable to confirm certain diagnoses such as histoid leprosy,[53] but is often unavailable.[49] The African Teledermatology Project diagnosed histoid leprosy in a case clinically suspicious for Kaposi's sarcoma using teledermatopathology.[54] Alongside such approaches, it is important to design and field-test technologies in low-resource settings.

The cost required to establish and maintain tele-leprology initiatives is a potential challenge. One strategy, exemplified by the cell phone-based test reader platform evaluated by Cardoso *et al.*,[22] is to take advantage of existing infrastructure. A second example is the Indian Space Research Organisation network, which is using its geo-stationary satellites to connect tertiary hospitals with rural hospitals and to send mobile telemedicine units into villages.[55] Another strategy is to integrate tele-leprology into broader public health initiatives. The majority of studies analysed by Warshaw *et al.* found teledermatology to be cost-effective if certain critical assumptions were met, notably patient travel distance, teledermatology volume, and costs of clinic dermatology.[31] Sharing costs to maximise sustainability is particularly relevant to leprosy given its low prevalence and significant morbidity.

Tele-leprology faces several challenges besides cost. One challenge is guaranteeing patients access to in-person consultations when indicated. This challenge also exists in the absence of tele-leprology. Resistance to telemedicine and tele-education among certain patients and healthcare providers is a challenge that must be met with evidence-based research and emphasis on provider-patient and instructor-learner communication. Further effort is required to develop policies addressing pricing, medico-legal, privacy, confidentiality, and security issues. This process should be guided by input from relevant governmental and non-governmental organisations.

Conclusion

Renowned bioethicist Norman Daniels argues that healthcare has a special moral importance that derives from its protection of equal opportunity.[56] Without early detection and appropriate management, patients with leprosy suffer disability, disfigurement, and social stigma. Maintaining access to expert healthcare for these individuals must therefore be considered the moral obligation of any just society. The question in the 'post-elimination era' is how to fulfil this obligation in the face of limited resources and competing priorities. The WHO is calling for 'innovative and practical strategies involving mainly operational solutions.'[49] Although limited conclusions may be drawn from the literature to date, the data do support a role for tele-leprology in strengthening routine and referral services and developing sustainable training strategies. Continued research and enhanced investment in telemedicine and tele-education hold great promise for safeguarding the quality of healthcare available to leprosy patients.

Contributors

Caroline A. Nelson conducted the literature review and drafted this manuscript. Carrie L. Kovarik and Christiaan B. Morssink contributed to the further development of this manuscript. Christiaan B. Morssink, the guarantor, accepts full responsibility for the work, had full access to the material, and controlled the decision to publish.

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