CASE REPORT

Delays in diagnosis and treatment of extrapulmonary tuberculosis in Guatemala

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SUMMARY

A 23-year-old indigenous Guatemalan man presented in 2016 to our clinic in Sololá, Guatemala, with 10 months of recurrent neck swelling, fevers, night sweats and weight loss. Previously, he had sought care in three different medical settings, including a private physician-run clinic, a tertiary private cancer treatment centre and, finally, a rural government health post. With assistance from our institution's accompaniment staff, the patient was admitted to a public tertiary care hospital for work-up. Rifampin-susceptible tuberculosis was diagnosed, and appropriate treatment was begun. The case illustrates how low tuberculosis recognition among community health workers and health system segmentation creates obstacles to appropriate care, especially for patients with limited means. As a result, significant diagnostic and treatment delays can occur, increasing the public health burden of tuberculosis.

CASE PRESENTATION

A 23-year-old Guatemalan man of indigenous Maya ethnicity was referred to us for evaluation of recurrent neck swelling by a local community health worker from his town in Southwest Guatemala. The patient reported 10 months of painful recurrent neck swelling, fevers, weight loss, anorexia and night sweats.

Prior to our evaluation, the patient had sought care for his illness from three separate healthcare institutions. Shortly after onset of unilateral neck swelling in November 2015, he presented to a private general practitioner who conducted an incision and drainage of the area and prescribed a course of unknown antibiotics. Five months later, the patient made a 5-hour journey from his home to a tertiary private cancer treatment centre in Guatemala City for evaluation of three new intercurrent neck masses. There, the patient, who is a native Kaqchikel Maya speaker with limited Spanish language proficiency, was directed to complete laboratory and radiological tests to rule out malignancy. Due to embarrassment at not being able to afford the tests, and the anxiety of communicating in Spanish, the patient did not complete the work-up. Finally, 2 days before presentation to our clinic in August 2016, the patient visited a rural government health centre with worsening neck pain. There, clinicians incised and drained all three neck masses and prescribed unknown antibiotics. Specimens from the procedure were discarded without further testing.

Over the course of his illness, the patient reported that he was unable to continue his job as a storekeeper, and took largely to bed. He was also unable to continue his studies in computer technology, which he had been pursuing in hopes of becoming a teacher, and was unable to provide financial support to his siblings and single mother. He had no known exposure to tuberculosis (TB)-infected contacts. He had never travelled out of Guatemala, and denied any significant travel outside his own community. He denied animal bites, but lived in close contact with dogs, cats and chickens.

On examination, the patient appeared malnourished and in mild distress. Vital signs were within normal limits. Three 2–3 cm open wounds with gauze packing were present above the clavicles, one on each side and one at the midline, in addition to a 2 cm healed scar on the right neck. Several non-tender submandibular lymph nodes were palpated. The remainder of the physical examination was normal, with absence of hepatosplenomegaly, rash, pulmonary or cardiac abnormalities, or additional foci of lymphadenopathy.

Basic laboratory testing was performed. Significant results within normal limits included a leucocyte count of 8.1×10^9 cells/L (normal range 4.1×10^9 – 11.0×10^9 cells/L, normal differential), haemoglobin 11.4 g/dL (normal range 12.0–15.8 g/dL), lactate dehydrogenase 148 U/L (normal range <250 U/L) and negative rapid HIV test. A chest radiograph was within normal limits. In early September 2016—ten months after his initial presentation to a first medical provider—the patient was admitted to a public referral hospital in Guatemala City for lymph node biopsy and culture. A GeneXpert (Cepheid, Sunnyvale, CA) assay diagnosed rifampin-sensitive TB. He was started on standard short-course antibiotic therapy for TB. Two months after treatment initiation, he reported resolution of neck masses, improved energy and appetite, and 10 lb weight gain.

GLOBAL HEALTH PROBLEM LIST

1. Poor knowledge of TB presentation at the community level led to ineffective triage of a patient with evidence of extrapulmonary TB.
2. Segmentation of the Guatemalan health system produces challenges for coordinating care and resources necessary for TB control, particularly access to TB diagnostic testing.
3. Individuals who are indigenous Maya or from rural areas experience significant barriers accessing high-quality care, including the cost

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of care and language barriers for those who speak a Mayan language.

GLOBAL HEALTH PROBLEM ANALYSIS

Despite generally effective chemotherapy, TB is a leading cause of morbidity and mortality in the developing world.¹ Recently, the adoption of the United Nations Sustainable Development Goals,² along with new surveillance data indicating the TB epidemic is larger than previously estimated, has brought global TB control into renewed focus, with a new emphasis on improved identification of TB-infected individuals.³

However, improved TB detection requires that health systems coordinate resources for prompt diagnosis and appropriate, uninterrupted treatment. This case describes a patient with a classic presentation of extrapulmonary TB who experienced significant delays in diagnosis despite multiple interactions with medical institutions. Here we discuss limited TB awareness at the community level and features of the Guatemalan healthcare system which contributed to delayed diagnosis, along with a potential strategy for addressing these challenges.

TB knowledge among community practitioners

Guatemala has the fifth highest TB incidence in Latin America (25 cases per 100 000 people).³ Provider training includes education and exposure to the disease in its various manifestations, including extrapulmonary varieties. However, as evidenced by this patient's experience, gaps in health worker knowledge about TB presentation and management contribute to delays in care.

With declining TB incidence due to the success of treatment strategies such as directly observed therapy, timely management of TB cases may paradoxically worsen for atypical or advanced disease.⁴ TB control in Guatemala, as in various low and middle-income countries (LMIC), is primarily provided by frontline health workers in dedicated TB posts at government community health centres.^{5 6} While this framework is successful in improving access to TB care,⁷ the isolation of TB management from other care limits opportunities for general practitioners to maintain TB-related skills. This is especially concerning given the proliferation of the private healthcare sector in rural Guatemala, where private community-based practitioners are often the first point of contact for patients.⁸ In fact, data from other LMICs identify initially seeking care from the private sector as a predictor of prolonged delays in TB diagnosis, consistent with our patient's experience.^{9 10} In addition to possible gaps in knowledge due to limited TB caseload, this may also reflect weaker oversight in the private sector or financial incentives that drive treatment and testing,¹¹ such as the multiple non-diagnostic incisions and drainage our patient received.

As TB management becomes concentrated in specific health system units, TB control efforts must not fall from nations' priorities. Investment in TB training for all community health workers and providers, along with universal protocols and improved access to diagnostic testing, can improve TB case detection,^{12 13} and are imperative for containing TB.

Health system segmentation

Despite guaranteed universal healthcare in Guatemala, access remains especially limited for the rural and indigenous poor.⁸ This population relies on a mixture of services from Guatemala's chronically under-resourced public health system, private-for-profit providers and private non-profit providers such as non-governmental organisations (NGO).^{14 15}

The public health system consists of health posts and centres, which provide basic primary and emergency services, respectively. Tertiary care is provided by public hospitals located in larger cities. However, patient triage and referral to higher levels of care in the public system are made difficult by the geographic distribution of providers and facilities. Approximately 80% of Guatemalan physicians practise in the nation's capital, in which roughly 20% of the population resides.¹⁶ In contrast, nearly half of Guatemalans live in rural communities. For many patients, therefore, paying out of pocket to see a private sector provider is commonly viewed as a better strategy to attain care.¹⁴

In 1997, in an attempt to expand the capacity of rural public sector community clinics, the Guatemalan government implemented the Programa de Extensión de Cobertura (Expansion of Coverage Program, PEC), where the Ministry of Health contracted NGOs to provide healthcare to uncovered communities.⁸ The programme succeeded in providing basic primary healthcare services to over 2 million Guatemalans; however, in 2014, amidst suspected corruption charges, funding for PEC was eliminated.¹⁷

As a result of these changes, healthcare delivery in rural Guatemala is now even more highly segmented, consisting of a patchwork of providers and institutions, some public and some private, and all with differing integration into referral networks.¹⁸ This health system segmentation poses a barrier to TB control efforts, which depend on coordination between community and national level providers.¹⁹ In this case report, our patient's experience illustrates several manifestations of health system segmentation, including duplication of services, inappropriate triage and emphasis on acute episodic care instead of continuity. Two features of health system segmentation in Guatemala are noteworthy specifically for TB control: the predominance of vertical or siloed programmes, which insulate disease-specific care; and the organisation of laboratory facilities, which limit TB diagnostic capacity at the community level.

Vertical healthcare delivery, in contrast to integrated delivery, refers to a health delivery structure in which disease-specific interventions and care are provided by dedicated providers, in parallel to other care.²⁰ In Guatemala, for example, as illustrated in our case study, cancer care is largely restricted to stand-alone referral centres, which do not provide effective treatments for patients who present with non-malignant conditions. When patients present to the wrong 'vertical silo', their medical needs may not be addressed, and treatment delays and logistical, financial and time costs incur for both the patient and the institution.

Health system segmentation also impedes effective TB diagnosis by limiting access to necessary laboratory procedures.^{21 22} Guatemala's network of TB diagnostic laboratories, which includes 288 facilities that perform smear microscopy and 7 regional laboratories with culture capability, is situated at the referral level within the public sector. An increasing number of individuals in Guatemala access healthcare in an episodic fashion from private community providers who do not have privileges permitting direct referral to public tertiary care centres; thus care coordination is challenging. Indeed, a review of TB diagnosis in several high-incidence countries demonstrated that first seeking care from a private practitioner, as our patient did, predicted delays in diagnosis.²³

It is also notable that our patient was finally diagnosed by use of the GeneXpert assay, while admitted in a tertiary care hospital. Practically, this definitive technology is available in Guatemala largely for inpatients only—a logistical consideration which prompted us to recommend admission for expedited work-up. This patient's experience of failing to receive

sputum testing or culture, despite multiple encounters with both public and private providers, speaks to the need for a simpler and more robust system of coordinating TB laboratory testing at the community level. A potential solution is the utilisation of public–private partnerships (PPP), in which the public and private sectors both contribute to care. This mutual assistance, in which government programmes provide protocols and supplies, and the private sector provides human and economic resources, as well as ease of access by communities, has been successful in strengthening TB control in LMICs, and is recognised by the WHO as a useful strategy to harness the insufficient resources of each individual sector, towards an overall productive system.²⁴

While LMICs are limited by access to reliable testing—such as GeneXpert—due to financial and logistical restraints,^{25 26} and development of lower cost technologies should be a priority, TB diagnosis in developing countries will also benefit from a focus on strengthening laboratory referral networks via PPPs. The pooling of financial resources and technical expertise between the Ministry of Health and private entities in various countries has made this possible, specifically by making resources available for capacity building, guideline development, and planning and improvement of referral networks.²⁷

Barriers to health for indigenous and rural communities

Around the world, indigenous people suffer from worse TB morbidity and mortality than their non-indigenous counterparts,²⁸ and Guatemala—where some 40% of the population is indigenous Maya¹⁵—is no exception. Given the extensive healthcare segmentation discussed above, individuals who lack the economic and social resources to navigate the higher referral levels of the Guatemalan health system are often not able to access the necessary care.

TB prevalence in Guatemala is high among mobile worker populations, which are predominantly indigenous, owing to the historical precedents of seizure of indigenous land rights during Spanish colonisation, and subsequent emerging patterns of seasonal labour migration.^{29 30} More recently, political and social marginalisation of the Maya took on an extreme manifestation in the Guatemalan civil war, during which hundreds of thousands of indigenous people were killed and displaced. Following the end of the war in 1996, myriad challenges, including poverty, poor education, rural residence and mistrust in medical providers, have continued to limit access to and efficacy of health services for the Maya.³¹

As most indigenous Guatemalans live in rural areas, they bear the burden of transportation to cities, in which specialty care is concentrated. For example, during the course of his illness, our patient travelled twice from his rural community to Guatemala City for consultations—a journey of 5 hours on a public bus, at a cost to him of roughly 1 day's wages in bus fare per trip.

Language poses another significant barrier to healthcare for indigenous people. Although nearly half of the indigenous population is monolingual in Mayan languages, government facilities exclusively provide care in Spanish. This patient's experience at the urban cancer centre illustrates the difficulty of advocating for oneself and receiving quality care in the healthcare system as a non-primary Spanish speaker. Furthermore, mistreatment of indigenous patients in medical settings, ranging from professional neglect to outright racism, is a common experience in Guatemala.³²

Improving quality of care for rural and indigenous patients

A prominent theme in the WHO's framework for addressing TB as outlined in the End TB Strategy is patient-centred care, which invokes the notion that systems should be designed in ways that help patients overcome obstacles to achieving their

own care goals.⁷ One feature of our case study which stands out is the patient's report of how logistical barriers related to travel, language difficulties and unfamiliarity with the structure of referral institutions impeded his care. As the case illustrates, these barriers were surmounted with a patient navigator—a dedicated staff member who accompanied the patient to his appointments, to assist with interactions with staff and to provide language interpretation services.³³

While we recognise that improving TB control in rural and indigenous communities requires substantial investment in infrastructure, training and diagnostic capacity, we propose that the patient-centred care goals of the End TB Strategy can be furthered by more widespread implementation of a patient accompaniment model to, as in this case, alleviate the logistical, psychosocial and cultural barriers to high-priority medical care.

The underlying model of patient accompaniment, originally developed by Partners in Health in Haiti, has been implemented successfully in various settings, in LMICs as well as high-income countries.^{34–36} Indeed, the use of community health workers, or *accompagnateurs*, for medical visits and psychosocial support has improved treatment adherence, retention and outcomes for patients with HIV/AIDS and TB in several LMICs. Notably, analysis of costs for these programmes has demonstrated that patient accompaniment results in overall cost reduction, due to more efficient use

Patient's perspective

This all started when I was working as a storekeeper. I began to feel some strange symptoms, like the fever and the growths that appeared around my neck. I was worried, but I didn't know what to do. I didn't have a solution. I went to many doctors, and they asked for tests but I could not afford them. I am so thankful that when I came to this clinic, you fought for me. The doctors and the *compañera* [patient care navigator] did a great favor for me by getting me to the hospital in the capital. Then, in the hospital they did all the tests I needed and finally figured out what was happening. They told me I had tuberculosis. The truth is, when I went to the hospital, I was hopeless. But I thank God that it worked out. Now I am very satisfied with the treatment I received.

Learning points

- In a country with a medium incidence of tuberculosis (TB; 25 cases per 100 000 people), health system segmentation can lead to significant delays in diagnosis and treatment of a classic clinical presentation of extrapulmonary TB.
- Vertical healthcare programmes, in contrast to integrated care, can be highly inefficient, due to provision of redundant testing and incomplete preventative care.
- Public–private partnerships can be employed to overcome some of the inefficiencies in a highly segmented health system, including shortcomings in the laboratory referral network.
- Indigenous people in Guatemala experience poor healthcare access and health outcomes due to barriers of poverty, language and rural residence.
- Patient accompaniment provides a potential solution to the barriers of health system segmentation faced by marginalised populations.

of medical resources,³⁴ and prevention of infectious disease spread. The adoption of this model to streamline complex care in LMICs, particularly for indigenous people, warrants further thought, especially given the unique cultural, social and language barriers faced by these communities, in Guatemala and elsewhere.³³ Our organisation recently began deploying patient navigators in cases requiring coordination between various levels of care and providers. As illustrated in this case, one of our navigators accompanied this patient. The navigator, who is bilingual in Spanish and the regional Mayan language, alleviated barriers of language, literacy and provider mistrust. She also aided in transportation and communication between referring and referral providers during the patient's treatment, contributing to a successful outcome.

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