Schistosomiasis Workshop

Research priorities, progress and remaining gaps

Report

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Foreword

This report presents key findings from the Schistosomiasis Workshop held at Wellcome on 20 May 2024. Before discussing these findings, we would first like to take a moment to acknowledge the powerful engagement and collaboration that underpinned the event.

The inclusive and wide-ranging nature of the discussions between the researchers, funders, and other stakeholders at the workshop enabled us to substantially improve our knowledge of schistosomiasis research gaps and how they could best be addressed. Conversations were enriched by experiences and insights from a diversity of speakers, including leading researchers based in Africa, where disease burden remains greatest.

We would like to thank all the delegates, speakers and facilitators who ensured that we had a productive and meaningful workshop. This report is a testament to their efforts.

The need to fill research gaps for schistosomiasis has never been clearer. We are witnessing substantial progress towards global schistosomiasis goals, while also facing unprecedented challenges that include climate change and treatment failure.

With this in mind, we invite you to explore the following pages and consider the insights from our workshop. It is our hope that this report can be a catalyst for further research and investment in our collective effort to control and eliminate schistosomiasis.

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Summary

On 20 May 2024, Wellcome held a workshop on schistosomiasis at the Wellcome Collection in London, UK. The workshop catered for attendance both in-person and online, and had 60 participants representing researchers, policymakers, funders, and key organisations in the schistosomiasis space.

Topics highlighted in speaker presentations included World Health Organization (WHO) priorities for schistosomiasis research, and growth in knowledge around causes of treatment failure, how environmental change impacts schistosomiasis, and what is needed to reduce transmission. Speakers showcased new approaches for assessing impact, as well as the results of economic analyses demonstrating the value of mass drug administration (MDA) programmes.

In discussions, delegates identified research gaps across ten themes, detailed in **Table 1**.

Common challenges connected research gaps across themes, including:

- a limited understanding of schistosomiasis biology and epidemiology, including the morbidity burden associated with genital schistosomiasis, the drivers of treatment failure, and the ecology of snails.
- a lack of clarity around optimal intervention strategies – while some delegates emphasised a paucity of knowledge around best approaches to treatment and programme integration, others called for vaccine development or studies to assess the efficacy of praziquantel in animals.
- methodological research gaps, with uncertainty around how to measure morbidity, define transmission interruption, and design impact assessments.

Suggested approaches to address some of the research gaps recognised the importance of cross-cutting multidisciplinary approaches and decision-maker and community engagement in the schistosomiasis space.

The main themes from the workshop are summarised in this report. The information presented is suggested as a guide to inform future research and reduce the burden of this neglected tropical disease (NTD).

Table 1: Research gaps identified by delegates

Theme	Key research gaps
Burden	• Morbidity burden due to schistosomiasis including genital schistosomiasis • Most suitable approaches to measuring morbidity
Treatment and treatment failure	• Relative contributions of parasite, host, and environmental factors to treatment failure • Potential of drug resistance in field settings
Environmental change	• Ways in which parasites, snails, and people respond to climate and land-use change • Strategies for integrating schistosomiasis control into environmental programmes including land-use and climate change
End road	• Defining and achieving interruption of transmission • Impact of the release of a small number of eggs after interruption of transmission
Monitoring and evaluation	• Design of impact assessments • How to monitor and evaluate diagnostic tests
Implementation	• Optimal treatment strategy for different schistosome and hybrid species • Role of interventions that may be used in addition to praziquantel, such as snail control
Economic evaluation	• Economic costs of schistosomiasis • Cost-effectiveness of current, novel and integrated approaches to control
Snail intermediate hosts	• Distribution, diversity, and ecology of snails across settings • Approach needed to produce safe, sustainable snail control technology
Zoonotic reservoirs	• Contribution of animal reservoirs to transmission • Efficacy of praziquantel for treatment of animals
Immunity and vaccines	• Nature of acquired immunity • Composition of a vaccine that would prevent infection and reduce transmission

Scope and limitations

The content of this report is derived from the insights of delegates that were consulted during the workshop and should not be construed as incontrovertible facts or as reflections of Wellcome's opinions or stances. The insights presented are based exclusively on the information and perspectives that were shared at the time. It is important to acknowledge that, while efforts have been made to accurately convey the content of the discussions, these accounts are not exhaustive and might not encompass all viewpoints or all available data. Hence, the report serves as a summary of discussions rather than an expression of Wellcome's official position or a factual assertion.

Bob Shaban, 37, holds a handful of freshwater snails pulled from the banks of Lake Victoria in Uganda. Picture credit: RTI

Progress and research gaps

Burden

Discussions around the burden of schistosomiasis focused on understanding morbidity; in particular, knowledge gaps around the burden associated with genital schistosomiasis and improving morbidity measures. The key research questions surfaced are summarised below.

Key research questions that could fill knowledge gaps

Burden of genital schistosomiasis

- What is the scale of morbidity resulting from female and male genital schistosomiasis?
- How much mortality is associated with genital schistosomiasis?
- What is the impact of female genital schistosomiasis on fertility, pregnancy, and outcomes for infants?
- What is the longitudinal association between female genital schistosomiasis and human immunodeficiency virus (HIV) transmission across populations?
- Is there an association between male genital schistosomiasis and HIV transmission?
- Which screening and diagnostic strategies can be used to rapidly detect female genital schistosomiasis cost-effectively and at scale?
- Can a low-cost and scalable molecular test be developed to assist with diagnosis of male genital schistosomiasis?
- How effective is praziquantel for reducing morbidity associated with genital schistosomiasis?
- How do schistosomiasis prevalence measures relate to the burden of genital morbidity?

Measuring morbidity

- How can co-morbidity from conditions such as cancer, central nervous system disease, and mental illness be captured in measures of burden?
- How and why does morbidity vary geographically?
- How does nutrition status affect morbidity onset?
- Why are the most appropriate diagnostics for morbidity detection not used in all situations?
- Can reliable morbidity markers that move beyond intensity of infection be developed?

Children in infected water. Picture credit: Schistosomiasis Control Initiative

Treatment and treatment failure

Treatment failure was the topic of a presentation from Luc Coffeng (Erasmus MC, Netherlands) and Poppy Lamberton (University of Glasgow, UK). The pair defined treatment failure at the individual level as 'a less-than-expected reduction in the intensity of schistosome infection, two to three weeks after treatment' and at the community level as 'a less-than-expected reduction in average infection intensity, two to three weeks after treatment'. They explored reasons for treatment failure, citing parasite factors (including drug resistance and hybrid infections) and host factors (including drug absorption, immunity, co-infections, host genetics and nutrition) at the individual level and parasite factors (including zoonotic hosts and hybrid infections), host factors (including drug coverage, immunity, behaviour and host genetics) and environmental factors (including snail species diversity and infection prevalence) at the community level.

Looking to the future, Luc outlined the challenge of drug resistance, noting that it can be generated experimentally in the laboratory through changes in the $TRMP_{PZQ}$ receptor. He introduced the Resistance Evaluation and Surveillance Initiative for Schistosomiasis Treatment (RESIST) project, which is using whole genome sequencing to explore whether drug resistance has emerged in Zanzibar, alongside mathematical modelling to assess the impact of different monitoring and mitigation strategies. Poppy described a further project that aims to build upon RESIST, exploring the relative influence of a range of parasite, host, and environmental factors on treatment failure across multiple sites in mainland Africa. Delegates surfaced the following key research questions around treatment failure.

Key research questions that could fill knowledge gaps

Treatment failure

- What are the relative contributions of different parasite, host and environmental factors to treatment failure?
- How are these relative contributions affected by schistosomiasis endemicity and previous exposure to praziquantel?
- Has drug resistance emerged in field settings, and if so, is it spreading?
- How can the true burden of worms before and after treatment be best measured to assess treatment failure?
- What is the optimal follow-up time for measuring responses to treatment?
- Can better tools to understand non-compliance with MDA be developed?
- What is the effect of praziquantel on parasite fecundity across species and hybrids?
- How much standing variation is there in the $TRMP_{PZQ}$ receptor?
- How can we develop or repurpose drugs for use in the case of praziquantel resistance?
- Should any new drugs be used in combination with praziquantel or separately from praziquantel?

Children receive deworming drugs at a school in Tanzania. Picture credit: Louise Gubb/RTI Fights NTDs

Environmental change

The effects of environmental change, including climate change and changes to land-use, on schistosomiasis, were explored by presenters Giulio de Leo (Stanford University, CA, USA), Janelisa Musaya (Malawi-Liverpool Wellcome Programme, Malawi), and Mark Booth (Newcastle University, UK).

Giulio observed that, in an era of rapid change, the reference model for schistosomiasis dynamics is likely to be oversimplistic. He explained that missing elements may include decoy and competing snails, new hybrids, and food web interactions, and that because biodiversity can be influenced by climate change, climate change may, in turn, impact disease burden. Giulio therefore recommended a broader ecosystem approach to schistosomiasis modelling.

Giulio emphasised the importance of accurate data in models aiming to predict the effect of climate change on schistosomiasis. He noted that refined models based on new thermal optima estimates for *Schistosoma haematobium* (26.2°C) and *Schistosoma mansoni* (25.5°C) predict that more than half of the schistosomiasis-suitable regions with a mean annual temperature below the thermal optimum could face risk of increased transmission with future warming. This estimate is greater than those based on previous models. Giulio recommended researchers fully consider the assumptions and data that are fed into thermal sensitive process-based models, including the different thermal sensitivities of diverse snail species, the propensity for local adaptation within the same species, and behavioural and physiological responses to climatic extremes (such as overwintering and aestivation).

In terms of land-use change, Giulio acknowledged the importance of water management infrastructure for human survival but also highlighted links to increased schistosomiasis risk. He recommended incorporating schistosomiasis transmission risk into environmental impact assessments for water-management infrastructure and advocated for the integration of different approaches, both for sampling and for modelling, to allow a more complete understanding of schistosomiasis dynamics.

Janelisa and Mark provided two exemplar cases (**Case 1 and Case 2**):

Case 1: Schistosomiasis amidst current weather changes in Malawi

Janelisa introduced the Shire Valley Vector Control (Shire-Vec) study, which is measuring how the burden of schistosomiasis in Malawi is being affected by climate change. In Malawi, she explained, the prevalence of *Schistosoma haematobium* is 7.8% (1.0–25.1%) and of *Schistosoma mansoni* is 1.6% (0.0–5.7%), but infection prevalence can be much higher, with up to 83% of people at their study site in Chikwawa found to be infected.

Janelisa noted the recent increase in extreme weather events in Malawi, with three major cyclones over the last three years: Anna (2022), Freddy (2023), and Filipo (2024). She noted that resulting flooding and ecological changes have led to snails moving closer to people's homes, increased exposure to contaminated water, and the introduction of new invasive snail species. The Shire-Vec team are now completing a detailed assessment of the effect of these cyclones on schistosomiasis, alongside research into how the Shire River irrigation canal in Malawi is affecting vector-borne diseases.

Flooding in Malawi. Picture credit: Clinton Nkolokosa

Case 2: Climate change and schistosomiasis – a scoping review

Mark introduced a comprehensive scoping review of existing evidence carried out by the WHO's Task Team on Climate Change, NTDs and Malaria (formed in June 2023). He reported that 511 studies had met criteria for inclusion in the review (most of which relied upon modelling or statistical approaches), including 29 papers on schistosomiasis.

Focusing on the review's findings for schistosomiasis, Mark explained that a modelling analysis predicted increased prevalence and intensity of schistosomiasis infection in some areas of East Africa over the next few decades (particularly in Rwanda, Burundi, south-west Kenya and eastern Zambia), with concurrent substantial decreases in parts of Kenya, southern South Sudan and eastern Democratic Republic of Congo. Mark noted that the amplitude and direction of effects of climate change were likely to vary by location, be non-linear and evolve over time. He concluded that available analyses were not sufficient for confident prediction of the overall global impact of climate change on schistosomiasis.

Following the case study presentations, delegates surfaced the following research gaps relating to both modelling and data collection.

Key research questions that could fill knowledge gaps

Environmental modelling to inform policy

- Can models accurately predict the spatial distribution of snails and their food web interactions under climate change and land-use change?
- Can models accurately predict the effect of environmental change on parasite lifecycles?
- Can these models of snails and parasites be used to better understand how the burden of schistosomiasis might be altered by environmental change?
- How can sociodemographic and population density projections, and patterns of population movement be best incorporated into modelling of environmental change?
- How can multiple defined climate scenarios be best used in modelling of climate change for schistosomiasis?
- How can the multifaceted dimensions of climate change (as shown in the Shared Socioeconomic Pathways scenarios for climate change) be best considered in these models?
- How can the impact of mitigation and adaptation activities be best incorporated into modelling?
- How should schistosomiasis control be integrated into land-use change programmes and climate adaptation and mitigation interventions?

Biomphalaria snails in plastic pots. Picture credit: Trustees of the Natural History Museum

- How will thresholds for interventions and indicators for success be defined in a future with environmental change?
- Can suitable rapid assessment tools to map transmission risk for Environmental Impact Assessments be developed?

Data on environmental change

- How will snails adapt to changes in water conditions due to climate change?
- How will *Schistosoma* parasites adapt to changes in water conditions and snail density due to climate change?
- How will movement of people and zoonotic reservoir animals due to environmental change affect schistosomiasis transmission?
- Can current water contact behaviour, and the impact of environmental change upon it, be comprehensively described?
- How were past changes in burden linked to environmental change?
- Can more case studies on the impact of climate and land-use change on snail ecology be developed?

End road

Two case studies (**Case 3** and **Case 4**) relating to interruption of transmission were presented.

Case 3: Projects for schistosomiasis elimination in Zanzibar

Said Ali (Public Health Laboratory Ivo de Carneri**,** Tanzania) summarised the Zanzibar Elimination of Schistosomiasis Transmission (ZEST) project, which took place between 2012 and 2021. ZEST involved biannual MDA for the whole at-risk population in Zanzibar, which resulted in the elimination of schistosomiasis as a public health problem in 2017. However, Said explained that the project team encountered subsequent challenges, including rapid infection rebound in hotspots when MDA was paused.

Said explained that this had led to the SchistoBreak project, active from 2020 to 2024, which focused on strategies to achieve interruption of transmission in 20 shehias. The project provided combined MDA, snail control and behaviour change interventions to hotspots and took a surveillance-response approach (including a test-treat-track-test-treat programme) in low-prevalence areas. Said reported that early results from SchistoBreak highlight a key risk factor for infection: distance from waterbodies with Bulinus snails to households and schools.

Case 4: Geshiyaro project on interruption of transmission in Ethiopia

Birhan Abtew (Imperial College London, UK) provided an overview and update on the Geshiyaro project: an initiative that aims to develop a scalable and cost-effective model of interventions for the interruption of schistosomiasis transmission, leading to cessation of MDA, in the Wolaita zone, Ethiopia.

Birhan described the three-arm trial structure:

- Arm 1: community MDA, improvements in water, sanitation and hygiene (WASH) infrastructure and behaviour change communication.
- Arm 2: community MDA with the national standard WASH intervention.
- Arm 3: control, with school-based MDA and the national standard WASH intervention.

He summarised the mid-term trial results, which showed a decrease in prevalence across all arms, but with a relatively higher prevalence in Arm 3. He noted that while the Kato-Katz diagnostic method indicated a very low prevalence of schistosomiasis across all arms, point-of-care circulating cathodic antigen (POC-CCA) testing indicated a significantly higher prevalence, highlighting the importance of improved diagnostic methods.

These case studies sparked discussions around research gaps relating to interrupting transmission, summarised below.

Key research questions that could fill knowledge gaps

Reaching interruption of transmission

- How should interruption of transmission be defined in terms of intensity and prevalence?
- Can models to predict when MDA should be stopped be created and validated?
- Can goals for interruption of transmission be reached with current diagnostics and interventions?
- What are the optimal surveillance approaches for settings with prevalence below 10%?
- What sampling methods should be used for surveys that aim to verify interruption of transmission?
- Can improved, cost-effective diagnostic tests that are suitable for low-transmission areas and for verifying interruption of transmission be developed?
- How can health systems be used for monitoring outside surveys?
- How does acceptability of, and compliance with, MDA change as transmission falls to low levels?
- What is the optimal frequency and coverage for MDA in settings with prevalence lower than 10%?
- Should strategies for therapeutics in interruption of transmission include adaptive approaches, focal MDA, and test-and-treat?

- What is the role for social behaviour change communication, and WASH interventions in interruption of transmission?
- Can case studies of successful interruption of transmission be generated?

Maintaining interruption of transmission

- What would be the impact of the release of a small number of eggs after interruption of transmission?
- How should post-validation surveillance be performed to ensure resurgence is detected?
- How can the role of movement and migration in reintroduction after interruption of transmission be better understood?
- When would a therapeutic intervention programme be required after a reintroduction?

Hotspots

- How should hotspots be defined?
- What is the most effective way to identify hotspots?
- How can hotspots resulting from biological factors be distinguished from those resulting from programmatic factors?
- What role do hybrids have in driving heterogeneity of infection rates?
- How should hotspots be managed to reach interruption of transmission?
- How can rapid recrudescence be prevented in hotspots where MDA has been stopped?

Monitoring and evaluation

Fiona Fleming (Unlimit Health, UK) and Rachel Pullan (London School of Hygiene and Tropical Medicine, UK) introduced the topic of monitoring and evaluation. They focused on impact assessments to determine whether prevalence is above 10% in a community, which, according to WHO guidance, would imply the need for annual MDA for community members older than two years of age.

Fiona and Rachel highlighted the limited scalability of community-by-community sampling and decision-making for impact assessments, and the need for alternative approaches.

One such approach, introduced by Fiona, is implementation unit (IU)-level surveys. Fiona described the Schistosomiasis Practical and Precision Assessment (SPPA) strategy, which consists of an initial IU-level assessment, with subsequent community-level assessment only performed where there is evidence of variation in prevalence of around 10%. This aims to balance the need for accuracy at the community level with reducing the survey burden. Fiona explained that the SPPA was derived from the Schistosomiasis Oversampling Survey (SOS) study, in which high-quality, detailed survey data from four African countries and secondary data from three others were introduced into geostatistical models to generate high-resolution risk maps. These maps were used to simulate and test multiple survey approaches, before global experts and Ministry of Health representatives selected the SPPA

A person prepares to take praziquantel during MDA. Picture credit: World Concern

approach to carry forward. The SPPA strategy is now being piloted at scale.

Another alternative to community-by-community sampling, introduced by Rachel, is model-based approaches. Rachel noted that while these require more technical skill than IU-level survey approaches, they can be more efficient at scale. She outlined a model-based approach currently in use to predict schistosomiasis prevalence in Ethiopia, DRC, and Cote d'Ivoire. The model, she explained, relies upon spatially regulated sampling, with sampling density defined, in part, by the expected range of spatial autocorrelation and by oversampling in urban settings. She noted that sampling data were used in model fitting, with covariate selection based on preidentified domains, via a two-stage data-dredging method, and that interpretation of the results was standardised.

Subsequent discussions focused on research gaps linked to impact assessment, as well as diagnostics. The key questions that arose are summarised below.

Key research questions that could fill knowledge gaps

Impact assessment

- At what frequency should impact assessments be performed?
- How should "community" be defined programmatically?
- Which age groups should be sampled for impact assessments?

Are there suitable diagnostic alternatives to Kato-Katz and urine filtration for impact assessments?

Monitoring and evaluation of diagnostics

- How should we monitor and evaluate the performance and quality of diagnostic tests?
- Can more suitable diagnostic tests be developed for monitoring and evaluation across different prevalence settings and schistosomiasis species?

Implementation

Discussions around monitoring and evaluation tended to be linked to those around implementation. A wide range of research gaps were described in the implementation space, mostly relating to MDA, other interventions, and integration of health services. These are summarised below.

Key research questions that could fill knowledge gaps

MDA

- What is the optimal MDA strategy for the treatment of different schistosome species and hybrids?
- What are the optimal drug regimens for preschool children?
- What is the optimum treatment coverage by age group for morbidity control?
- What is the most suitable support for communities that can encourage uptake of MDA?
- How can people who have never been treated be reached with MDA?
- In which settings is it most important to include adults in MDA?

Primary school children line up to receive praziquantel. Picture credit: Schistosomiasis Control Initiative

- Why are pregnant and lactating women often excluded from MDA for schistosomiasis?
- What should be the treatment approach when schistosomiasis is comorbid with other conditions?
- How can we ensure drug supply and access?

Interventions provided in addition to MDA

- What should be the role of social behaviour change communication, WASH, vector ecology management, and veterinary public health in schistosomiasis control in different settings?
- How should new interventions, such as a vaccine, be deployed when available?
- What supports communities to take up and engage with interventions beyond MDA?

Integration

- What happens to safety and tolerability of praziquantel when delivered with treatments for other diseases during integrated MDA campaigns?
- Can surveillance for multiple diseases be integrated?
- How can schistosomiasis control be effectively and sustainably integrated into other health provision, such as primary health care, HIV programmes, and sexual and reproductive health services?

Economic evaluation

Hugo Turner (Imperial College London, UK) provided an overview of progress in economic evaluation related to schistosomiasis. He highlighted a recent review of the literature that found MDA for schistosomiasis control to be cost-effective, at between $5 and $692 per disability-adjusted life year (DALY) averted in moderate and high prevalence settings. Hugo explained that MDA is also likely to generate economic benefits not captured in the DALY metric.

Hugo acknowledged the growing number of studies evaluating alternative strategies to school-based MDA, such as community-wide treatment. He noted the varying results of cost-effectiveness analyses for these strategies, which depend heavily upon settings and methodology. He cited specific methodological parameters that can significantly impact model outcomes, including cost-effectiveness thresholds; approaches to incorporating age profiles of infection (a particular challenge, given the paucity of age-stratified data on pre-control intensity); and approaches to simulating morbidity (such as use of prevalent case-years versus intensity-based metrics). He emphasised that simulation of morbidity is often crude, with limited stratification.

Hugo highlighted the importance of understanding when shifting from the control of schistosomiasis to its elimination may be good value. He introduced an ‘All Disease – Whole Health System’ model (known as the ‘Thanzi La Onse’ model), which will be used to address this question in future. The model allows full consideration of the costs and benefits of transmission interruption, including impacts on other conditions, such as HIV and diarrheal diseases.

A field worker collecting snails. Picture credit: WHO/C. Lutandula

Discussions of the research gaps relating to economic evaluation are summarised below.

Key research questions that could fill knowledge gaps

Economic evaluation

- What is the age profile of infection in the absence of treatment?
- What are the full economic costs of schistosomiasis?
- What are the health impacts of treating different age and risk groups?
- What is the value of achieving elimination of schistosomiasis as a public health problem and of reaching transmission interruption?
- What are the costs and benefits of integration with other services?
- What are the costs and benefits of new treatment strategies?
- Can we develop investment cases for specific countries or settings?

Snail intermediate hosts

Discussion on snail intermediate hosts revealed several research gaps, summarised below. The most notable of these related to a lack of understanding of snail biology and the need for new interventions.

Key research questions that could fill knowledge gaps

Snail intermediate hosts

- Can the distribution, diversity, and ecology of the snails that are intermediate hosts for schistosomiasis be better described?
- Can robust protocols for mapping and surveillance of intermediate snail hosts be developed?
- Can new, environmentally friendly molluscicides be developed?
- Can safe, cost-effective, and sustainable snail control technology in general, be produced?
- How do snails respond to existing control measures?
- How would snails respond to new control measures?

Zoonotic reservoirs

Zoonotic reservoirs were an emerging theme from workshop discussions. Key research questions are summarised below.

Key research questions that could fill knowledge gaps

Zoonotic reservoirs

- What is the contribution of animal reservoirs to transmission of schistosomiasis?
- Can better maps of zoonotic schistosomiasis be developed?
- Can the diagnostics for monitoring infection in animal reservoirs be refined to increase sensitivity?
- How can quality control for diagnostics in animals be standardised and improved?
- What is the efficacy of praziquantel for treatment of schistosomiasis in animals?
- What doses and frequencies of treatment are best for animals?
- What effect would treatment of animals have on transmission in humans?
- How efficacious are alternative farming methods, such as stall feeding or preventing grazing in contaminated grassland, for reducing the burden of zoonotic schistosomiasis?
- Can exemplars of settings where the interface between humans and animals are being managed well be developed?

Immunity and vaccines

Several delegates noted progress and challenges related to immunity and vaccines. Discussions spanned topics including the effect of schistosomiasis on the human immune system and vaccine development pathways.

Key research questions that could fill knowledge gaps

Immunity and vaccines

- What is the nature of acquired immunity and what is its relation to past exposure?
- What is the effect of schistosomiasis on immunisation against other infections?
- Can a vaccine for humans and/or animals that prevents reinfection and reduces transmission be developed?
- Could a new vaccine for schistosomiasis be co-administered with vaccines for other infections?

Addressing research gaps

Delegates were asked to comment on useful ways of working for addressing the identified research gaps. Discussions focused on four key avenues that could strengthen research and boost impact (further explored in **Figure 1**):

1. **Cross-cutting approaches:** delegates described methods and strategies that could be valuable across the diverse disciplines involved in schistosomiasis research, such as creating standard processes and sharing outputs equitably.
2. **Policy engagement:** delegates emphasised the importance of understanding how policymakers use research, considering both regional and national policies, and engaging across the lifespan of a project.
3. **Community engagement:** discussions suggested that alignment with other actors and tailored messages could increase the odds of successful partnerships.
4. **Multidisciplinary working:** delegates frequently advocated for collaborations between researchers with differing expertise, citing the associated benefits.

Figure 1: Delegate suggestions for useful ways of working that can help to address identified research gaps.

Addressing research gaps with mathematical modelling

Deirdre Hollingsworth (University of Oxford, UK) spoke on the use of mathematical modelling to address research gaps for schistosomiasis. She explained that such models rely on real-world inputs (such as data on the biology of transmission and the impact of interventions) to produce outputs ranging from the distribution of infection to the cost-effectiveness of an intervention; these outputs can be crucial for informing policy.

Deirdre referenced multiple challenges facing schistosomiasis modellers, including difficulties in modelling disease elimination (which often requires individual-based models); the tendency to be supplied with aggregated data, despite extensive variability between settings; a lack of clarity over who is not treated across repeated rounds of MDA; and uncertainty around whether increases in the number of parasites are occurring in humans or in snails. She noted that validating models against epidemiological data can be challenging, especially when switches in diagnostic tools result in limited resources for cross-validation. Others noted that shortcomings included a lack of clarity around changes in transmission dynamics from year to year, the relative roles of immunity and ecology in driving age-intensity trends, and the likelihood that some parameters will never be measured. Despite this, there is great value in modelling, which was exemplified by a case from Kenya (**Case 5**).

> **"Modelling has been the best gateway from data to impact."**
>
> Quote from a workshop delegate

Case 5: Optimising delivery MDA to eliminate schistosomiasis in Kenya

Mutono Nyamai (University of Nairobi, Kenya) and Florence Wakesho (Kenya Ministry of Health, Kenya) described their use of the SCHISTOX model to explore the interventions needed to reach schistosomiasis goals in Kenya. These goals include elimination of schistosomiasis as a public health problem (defined as reducing the prevalence of heavy infections in school-aged children to less than 1%) and breaking transmission (defined as achieving a prevalence of less than 2% in school-age children by 2030).

The SCHISTOX model allowed Mutono, Florence and their team to consider different scenarios for treatment frequency (annual or biannual), target populations (school aged children or all community members aged above four years), and treatment coverage (75%, 80%, 85%, or 90%) across low, medium and high schistosomiasis prevalence levels, revealing the following:

- Schistosomiasis prevalence below 10%: goals achievable under all treatment scenarios.
- Schistosomiasis prevalence between 10% and 50%: goals achievable under all treatment scenarios except the scenario where school-aged children receive annual treatment at 75% coverage.
- Schistosomiasis prevalence above 50%: goals achievable only with biannual community treatment.

Mutono and Florence discussed potential next steps, including use of the SCHISTOX model to understand how the proportion of people who never receive treatment influences the 'breaking transmission' target.

A woman in infected water. Picture credit: Global Schistosomiasis Alliance

Conclusion

The Schistosomiasis Workshop brought together 60 stakeholders, including researchers, policymakers, and funders, to address research priorities, progress and remaining gaps in schistosomiasis research. The workshop provided a platform for engaging stakeholders from diverse sectors, fostering open dialogue. This collaborative environment was essential for identifying research gaps and considering the approaches needed to address them.

Presentations spanned a wide array of topics, emphasising progress in our understanding of environmental change, possible treatment failure, and how to manage control and evaluation programmes.

Through discussions, participants identified research gaps related to the biology and epidemiology of schistosomiasis, optimisation of intervention strategies, and research methodology. Key gaps included the lack of knowledge of the effect of environmental change and the drivers of treatment failure.

Delegates underscored the importance of ensuring that attempts to address these gaps use robust approaches. For example, both standardisation of processes and equitable sharing of outputs were highlighted as valuable. Building multidisciplinary collaboration, as well as engaging policy members and affected communities, were also highlighted as boosting impact.

Mathematical modelling was described as useful for informing policy, although challenges, such as low-quality data and difficulties in model validation, were acknowledged. Notably, a case study from Kenya demonstrated the role of modelling in determining treatment strategies that meet prevalence targets under different scenarios.

The insights gathered here are intended to guide future research in schistosomiasis. Progress is crucial for reducing the burden of the disease globally.

A person carries praziquantel. Picture credit: RTI Envision/Timothy La Rose

Appendix 1: attendee list

Delegates

Birhan Abtew, Imperial College London

Said Ali, Public Health Laboratory Ivo de Carneri

Roy Anderson, Imperial College London

Nebiyu Negussu Ayele, ELMA Philanthropies

Mark Booth, Newcastle University

Simon Brooker, Bill and Melinda Gates Foundation

Luc Coffeng, Erasmus MC

Giulio De Leo, Stanford University

Claudia Demarta Gatsi, Merck KGaA

Jennifer Downs, Weill Cornell Medicine

Julia Dunn, Clinton Health Access Initiative

Uwem Ekpo, Akwa Ibom State University

Darin Evans, US Agency for International Development

Fiona Fleming, Unlimit Health

Amadou Garba Djirmay, World Health Organization

Katie Gass, Task Force for Global Health

Mireille Gomes, Merck KGaA

Anouk Gouvras, Global Schistosomiasis Alliance

B.F. Lee Hall, National Institute of Allergy and Infectious Diseases

Timothy Hallett, Imperial College London

Deirdre Hollingsworth, University of Oxford

Carol Karutu, The END Fund

Kebede Kassaye, The Children's Investment Fund Foundation

Louise Kelly-Hope, University of Liverpool

Poppy Lamberton, University of Glasgow

Bridie Layden, The END Fund

June Lee, ELMA Philanthropies

Morgan Lemin, London School of Hygiene and Tropical Medicine

Chinwendu Emilian Madubueze, Joseph Sarwuan Tarka University Makurdi

Tara Mangal, Imperial College London

Roy Mayega, Makerere University School of Public Health

Janelisa Musaya, Malawi Liverpool-Wellcome Programme

Thumbi Mwangi, University of Nairobi

Pauline N Mwinzi, World Health Organization

Hasifa Nampala, Kyambogo University

Justin Komguep Nono, Institut de Recherches Medicales et D'Etudes des Plantes Medicinales

Mutono Nyamai, University of Nairobi

Collins Okoyo, Kenya Medical Research Institute

Rita Oliveira, Children's Investment Fund Foundation

Akinola Stephen Oluwole, Sightsavers

Derick Nii Mensah Osakunor, University of Glasgow

Alison Ower, The END Fund

Joaquin Prada, University of Surrey

Rachel Pullan, London School of Hygiene and Tropical Medicine

Jutta Reinhard-Rupp, Merck ATSA

David Rollinson, Global Schistosomiasis Alliance

Eugene Ruberanziza, The END Fund

Evan Secor, Centers for Disease Control and Prevention

Allison Shaffer, RTI International

Sheetal Silal, University of Cape Town

Anthony Solomon, World Health Organization

Thomas Spangenberg, Merck ATSA

Russell Stothard, Liverpool School of Tropical Medicine

Ploi Swatdisuk, United States Agency for International Development

Louis-Albert Tchuem Tchuenté, University of Yaoundé I

Hugo Turner, Imperial College London

Florence Wakesho, Kenya Ministry of Health

Martin Walker, Royal Veterinary College

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Emma Maynard, Infectious Diseases

Richard Muscat, Translation and Portfolio Integration

Megan Neary, Infectious Diseases

Sally Nicholas, Infectious Diseases

Alex Pym, Infectious Diseases

Jaspreet Turner, Infectious Diseases

Workshop participants.

Appendix 2: agenda

Time	Session	Speaker(s)
9:30am – 10:00am	Registration and refreshments	
10:00am – 10:10am	Welcome to workshop	Alex Pym & Sally Nicholas (Wellcome, UK)
10:10am – 10:30am	WHO targets and research priorities for schistosomiasis	Amadou Garba (WHO, Switzerland)
10:30am – 10:35am	Overview of schistosomiasis modelling research	Deirdre Hollingsworth (University of Oxford, UK)
10:35am – 10:45am	Optimising delivery of MDA to eliminate schistosomiasis in Kenya: insights from models	Mutono Nyamai (University of Nairobi, Kenya), Florence Wakesho (Ministry of Health, Kenya)
10:45am – 11:00am	Determining implementation and monitoring and evaluation (M&E) strategies and gaps for schistosomiasis	Fiona Fleming (Unlimit Health, UK), Rachel Pullan (London School of Hygiene and Tropical Medicine, UK)
11:00am – 11:15am	The drivers of treatment failure	Poppy Lamberton (University of Glasgow, UK), Luc Coffeng (Erasmus MC, Netherlands)
11:15am – 11:25am	Discussion	All
11:25am – 11:45am	Break and refreshments	
11:45am – 12:00pm	Current state of evidence of economic evaluation of elimination and future work plans	Hugo Turner (Imperial College London, UK)
12:00pm – 12:10pm	End road: country questions	Said Mohammed Ali (Public Health Laboratory Ivo de Carneri, Tanzania)
12:10pm – 12:25pm	Schistosomiasis in the era of global changes	Giulio de Leo (Stanford University, USA), Janelisa Musaya (Malawi-Liverpool Wellcome Programme, Malawi), Mark Booth (Newcastle University, UK)
12:25pm – 12:40pm	Overview and update on Geshiyaro project	Birhan Abtew (Imperial College London, UK)
12:40pm – 12:45pm	Discussion	All
12:45pm – 1:45pm	Lunch	
1:45pm – 1:55pm	Introduction to breakout group activity	Jaspreet Turner (Wellcome, UK)
1:55pm – 3:30pm	Breakout group activity	All
3:30pm – 3:50pm	Break and refreshments	
3:50pm – 4:40pm	Breakout group activity continues	All
4:40pm – 4:55pm	Feedback from breakout groups	Katherine Davis (Wellcome, UK)
4:55pm – 5:15pm	Open discussion	All
5:15pm – 5:30pm	Next steps and close	Jaspreet Turner (Wellcome, UK)
5:30pm onward	Drinks reception followed by dinner	

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