

Recommendations from the International Paediatric Stroke Organization on pediatric neurointerventional best practices based on Delphi consensus

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ABSTRACT

Paediatric neurointervention (PNI) markedly differs from adult neuro-intervention, requiring highly subspecialized clinical skills, knowledge, and techniques. Minimum standards of practice are well established in adult neurointervention but are lacking in the field of PNI. We sought to develop expert consensus on best practices for neurointervention in children.

Using a two-stage Delphi consensus model we sought expert opinions from PNI practitioners worldwide regarding best practices. A two-stage online de-identified survey of PNI practitioners was undertaken assessing opinions on a range of topics including minimum recommended caseloads for PNI centres. Minimum agreement rates of >60% were set to determine consensus on any specific question. Consensus opinions on best practices were reviewed by the International Paediatric Stroke Organization Executive Committee. For the first-stage survey there were n=50 responses and for the second-stage n=45 responses, with practitioners from all inhabited continents represented. Consensus-based best practices included: i) Elective endovascular therapeutic neuro-interventions should be performed in high-volume paediatric centres with an established multi-disciplinary paediatric neurovascular team, and ii) High-volume centres are those that undertake at least 20 paediatric endovascular therapeutic neuro-interventions annually. Paediatric thrombectomy in large-vessel occlusion stroke, an area of increasing interest and attention, poses unique time-sensitive multidisciplinary logistical challenges meriting a dedicated analysis, and as such is not within the purview of this report.

Best practices for PNI reported here have been identified through expert consensus and are designed to enhance patient safety whilst providing appropriate clinical access to life-saving procedures.

INTRODUCTION

Pediatric neurointervention (PNI) differs from adult neurointervention in the clinical approach, patient age/size, therapeutic goals, underlying pathology, and endovascular treatment devices/techniques.^{1,2} For the purposes of this article, we consider PNI procedures to include embolization of congenital vascular malformations such as vein of Galen malformations (VoGM) and dural sinus malformations (DSM), intra-arterial chemotherapy (IAC) for retinoblastoma, embolization of extracranial head and neck tumors and arteriovenous malformations

(AVMs), embolization of intracranial tumors and non-Galenic AVMs (as used here, AVM is intended to include all forms of arteriovenous shunt lesions including arteriovenous fistulae regardless of angio-architecture), and embolization of spinal/paraspinal AVMs or tumors.^{1,3,4} Some of these procedures, in particular VoGM embolization and IAC, would form only a minute proportion of the caseload in adult practice. Other procedures commonly performed in adults like endovascular thrombectomy and intracranial aneurysm intervention may also be performed in children, but are not uniquely or primarily PNI procedures.

Specific clinical and technical skill sets necessary for advanced PNI have resulted in a trend towards subspecialized PNI practitioners at major pediatric hospitals. Such practitioners dedicate a significant proportion (or all) of their working week to pediatric practice.^{1,3} This trend is reflected in the progressive enlarging membership of PNI councils and committees within organizations such as the International Paediatric Stroke Organization (IPSO) and the Society for Neuro-Interventional Surgery (SNIS). The SNIS Paediatric Committee has published practice recommendations for pediatric diagnostic cerebral angiography,⁵ reflecting the additional precautions and skillsets required for safe PNI practice.

Neurointerventionalists with a predominantly adult patient practice may have limited pediatric exposure during training and a limited annual caseload involving children. This was demonstrated in a 2019 SNIS survey of its membership, in which approximately one quarter (26%) of practitioners were exposed to less than 10 pediatric cases during fellowship and just over half (53%) undertake fewer than 10 annual pediatric cases in their current practice.⁵ These results suggest that a large proportion of neurointerventionalists undertake progressively less pediatric work as their careers in adult neurointervention progress.

Minimum standards of practice in adult neurointervention are well established, with a clear example being minimum capabilities for centers undertaking thrombectomy for acute large vessel occlusion stroke. In 2018, multinational, multisociety international consensus standards of practice in acute ischemic stroke intervention were published, including recommendations on minimum caseloads.⁶ Similarly, multinational consensus training guidelines recommended that fellowship training for acute

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stroke intervention be undertaken at such a high-volume center.⁷ These recommendations were based on evidence that patient outcomes were improved by transfer to a high-volume thrombectomy center.⁸ While the exquisitely time-sensitive nature of arterial ischemic stroke often demands that endovascular therapy is undertaken at the nearest, often less experienced center due to geographic isolation, the important relationship of center experience with outcomes is well established even for neuroendovascular treatment of pathologies that are not considered time sensitive, such as endovascular repair of intracranial aneurysms⁹ and carotid artery stenting.¹⁰

At present, there are no formal recommendations for therapeutic PNI practice addressing the minimum capabilities and caseload of treatment centers. The absence of high-level evidence to guide such recommendations remains an obstacle. Nonetheless, given the progressively specialized nature of PNI practice and the potential for a large proportion of neurointerventionalists having infrequent exposure to pediatric patients and pathologies,⁵ the safety of this vulnerable patient population would benefit from expert consensus on best practices. While there are differing methods to form such recommendations, in the setting of rare pathologies and a relatively small number of dedicated practitioners, we chose a Delphi consensus approach. The Delphi process involves obtaining consensus from subject experts in a multistage process, with feedback of the summary results at each stage to assist responders in adapting future responses based on emerging consensus.¹¹ This allows later stages of the process to address more specific details within a topic based on initial results.

There are many geographically isolated regions in which patients have no access to high-volume PNI centers. In such regions, it may be the case that the nearest high-volume PNI center is 4–6 hours away by air travel. Similarly, some countries may not have a high-volume PNI center or a national health system that is resourced to assist with international transfer of patients for dedicated PNI care. Although less commonly, some conditions represent a hyperacute emergency with immediate concern for loss of life and critical neurologic function. In such instances, patient care is appropriately provided by an adult-practice neurointerventionalist with little to no prior pediatric experience. In this context, it would be inappropriate to deny treatment to a child by an established adult neurointerventional center. This survey process and best practice recommendations are not designed to prevent the timely treatment of children presenting with time-sensitive neurological emergencies or children without access to high-volume PNI centers. Rather, the intent was to focus on elective PNI procedures. We have also provided recommendations in the Discussion for geographically isolated regions which lack access to a high-volume PNI center.

METHODS

Ethics approval was obtained from a quaternary pediatric neurovascular center. The recommendations resulting from this Delphi process were reviewed and approved by the IPSO Executive Committee. The primary objective was to form best practice recommendations for PNI based on expert consensus from PNI practitioners. We defined PNI experts as neurointerventional practitioners who undertake a minimum of 20% of their caseload in pediatrics. The potential participants pool for the surveys was created with a view to ensuring representation from PNI practitioners across all inhabited continents and across a variety of health systems, as far as possible.

A two-stage survey was undertaken using the Delphi process with questions for each stage formulated by a multinational

working group of PNI practitioners (see online supplemental material containing both survey stages). The working group, which corresponds to the listed authors of this report, coalesced spontaneously during the pediatric session of the 20th annual meeting of SNIS in San Diego, California, USA in 2023. The survey responses were obtained online via RedCAP, overseen by the administration of IPSO, with links to the survey emailed to eligible participants. All responses were obtained in a deidentified fashion. Eligible survey participants who received the email link included the following:

1. Members of the IPSO INR Council (n=40).
2. Additional PNI practitioners worldwide (across all inhabited continents) who were known to members of the working group to have a significant pediatric caseload but were not members of IPSO (n=10).
3. A worldwide email distribution of over 1000 INR practitioners compiled from the INR distribution network NAN-Consortium (x.com/NANConsortium), with a caveat placed on the email survey invitation that responses should only be provided by practitioners who undertake 20% or more of their caseload in pediatrics.

Consensus was defined as >60% agreement across responses for a particular topic. The primary subset of questions designed by the working group was formed to identify consensus-based best practices, while a secondary subset was designed to identify the current variety of practices in various pathologies to better inform future research and surveys. Consensus was not sought on this secondary subset of topics.

Questions were designed to identify the practices of a center rather than an individual, recognizing that many PNI practitioners undertake complex procedures in pairs and noting that such team-based practice should be encouraged. The questions (and associated responses) were formed to identify the practices of single centers and were specifically not applicable to a network of centers working in conjunction. The exclusion of networks was based on the objective of ensuring any given center had appropriate multidisciplinary resources on site and an appropriate caseload for safe PNI practice, which may be diluted or difficult to assess across a network of centers. For questions seeking minimum numbers for recommended caseloads in PNI, a variety of ranges were provided in multiple choice form (including n<15, 5–10, 10–20, 20–50 etc) to assist respondents in selecting the range they thought was appropriate.

For the purposes of these surveys, diagnostic cerebral angiography refers to cases in which only a diagnostic angiogram is performed and does not apply to angiograms performed immediately preceding an endovascular therapeutic intervention. Endovascular therapeutic neurointervention refers to endovascular (trans-arterial or trans-venous) interventions performed in the intracranial circulation, skull base, or spine for therapeutic (not diagnostic) purposes. This would include AVM/VoGM embolization, IAC for retinoblastoma, aneurysm treatments, intra-arterial vasospasm treatments, mechanical thrombectomy for stroke, tumor embolization, dural venous sinus stenting, and so on. Sclerotherapy for low-flow vascular malformations of the head and neck was specifically excluded. These definitions and distinctions were clarified in a written statement at the beginning of each survey (see online supplemental material).

After the collation of deidentified summary results from the first stage of the survey, the results (including areas of identified consensus) were included at the beginning of the second stage survey to better inform respondents regarding emerging areas of consensus, consistent with the Delphi process. Results were analyzed using summary statistics.

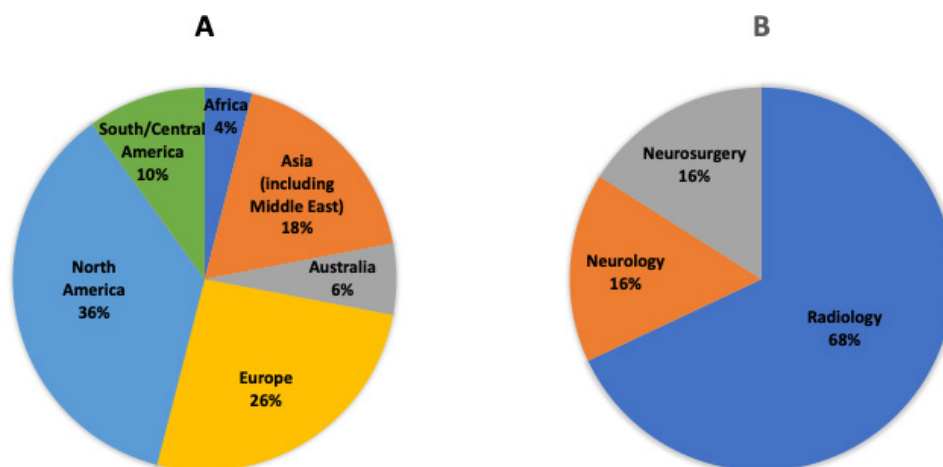


Figure 1 Distribution of pediatric neurointerventionalist respondents to the first-stage survey (total n=50) by continent (A) and medical training background (B).

RESULTS

For the first stage of the survey, there were n=50 individual responses, and for the second stage, n=45 responses. Responses for the first stage were received from PNI practitioners across all inhabited continents (Africa, n=2, 4%; Asia, including the Middle East, n=9, 18%; Australia/Oceania, n=3, 6%; Europe, n=13, 26%; North America, n=18, 36%; South/Central America, n=5, 10%). The medical training background of the respondents included radiology (n=34, 68%), neurology (n=8, 16%), and neurosurgery (n=8, 16%). These results are demonstrated in [figure 1](#).

Below we report areas of consensus (>60% response concordance) related to diagnostic cerebral angiography and endovascular therapeutic neurointerventions in the pediatric population.

Diagnostic cerebral angiography: areas of consensus (see [figure 2](#))

1. Diagnostic cerebral angiography in children <5 years of age should be performed in a high-volume pediatric center (first-stage survey; yes=41/50 responses, 82%).

2. The number of pediatric diagnostic cerebral angiograms a center should perform annually to be considered safe to undertake cerebral angiography in children <5 years of age is n>10 (second-stage survey; 42/45 responses, 93.3%).

Endovascular therapeutic neurointerventions: areas of consensus (see [figure 3](#))

1. All *elective* endovascular therapeutic neurointerventions in pediatric patients, excluding thrombectomy for acute pediatric stroke, should be performed in a high-volume pediatric center with an established pediatric PNI program and multidisciplinary team, including neurointerventionalists, neurosurgery, neurology, critical care, anesthesia, and developmental specialists—all with pediatric expertise (first-stage survey; yes=44/50 responses; 88%).
2. The minimum number of pediatric therapeutic neuroendovascular interventions that a center should perform annually to be considered safe to undertake *elective* pediatric neuroendovascular interventions is >20 procedures (first-stage survey; 37/50 responses; 74%; second-stage respondents

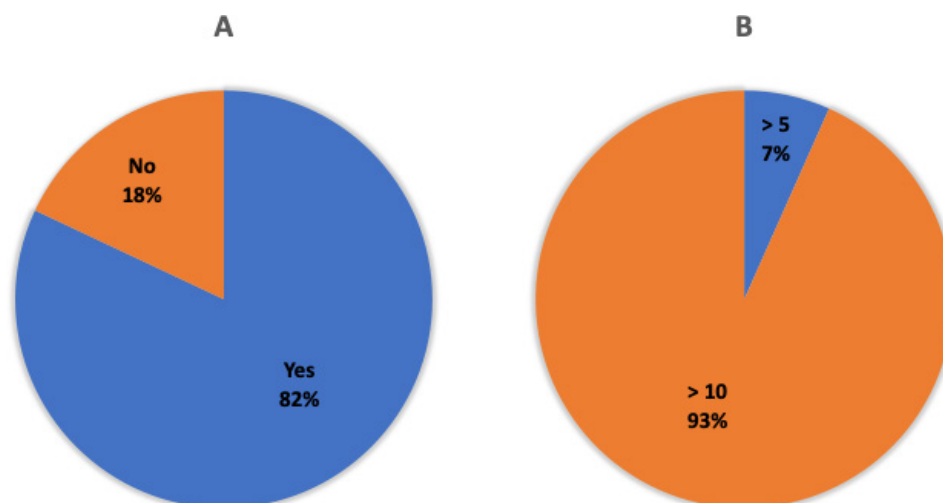


Figure 2 Diagnostic cerebral angiography in children <5 years of age. (A) In the first-stage survey, when asked if all diagnostic cerebral angiography in children <5 years of age should be performed in a high-volume pediatric center, 41 of 50 respondents (82%) responded 'Yes'. (B) In the second-stage survey, when asked what number of pediatric cerebral angiograms a center should perform annually to be considered safe to undertake cerebral angiography in children age <5 years, 42 of 45 respondents (93%) provided a response of >10 angiograms.

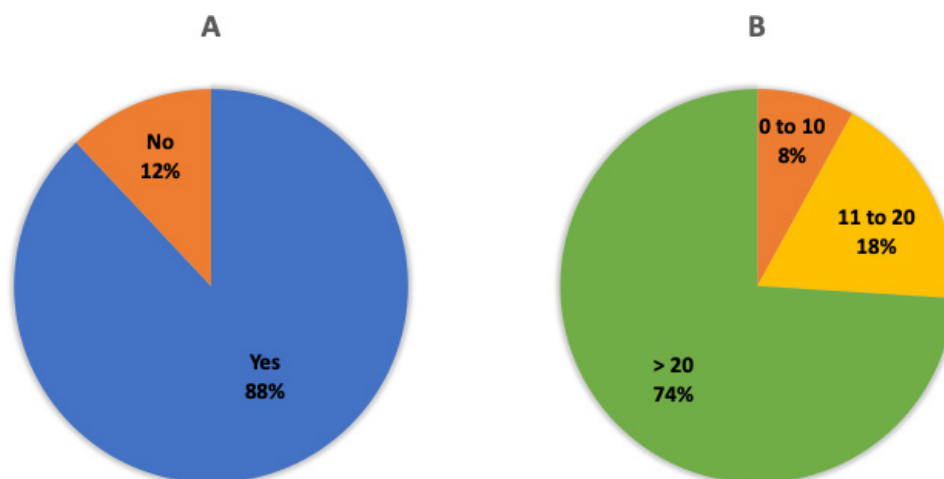


Figure 3 Elective pediatric endovascular therapeutic neurointerventions. (A) In the first-stage survey, when asked whether all elective pediatric endovascular therapeutic neurointerventions should be performed in a high-volume pediatric center with an established neurointerventional program and a multidisciplinary pediatric neurovascular team, 44 of 50 respondents (88%) responded 'Yes'. (B) In the first-stage survey, when asked the minimum number of annual pediatric endovascular neurointerventions a center should perform to be considered high volume, 37 of 50 respondents (74%) provided responses of >20 procedures.

asked to further detail these numbers, ie, >20 vs >30 vs >40 etc with 100% answering >20 procedures).

- For pediatric thrombectomy in large-vessel occlusion stroke, the decision to treat should be undertaken by a multidisciplinary team including pediatric neurologists (second-stage survey; yes=45/45 responses; 100%).

Survey results of existing practices: *not for expert consensus formation*

The results from the secondary subset of survey questions designed to identify the range of existing practices, rather than for expert consensus formation, are detailed in the online supplemental material. In summary, these results demonstrated moderate variability in existing practices across PNI centers in relation to:

- ▶ Heparin use in pediatric diagnostic cerebral angiography.
- ▶ Ipsilateral limb pulse oximetry at the arterial access site,
- ▶ Embolic agents used for VoGM embolization.
- ▶ Clinical and technical approaches to thrombectomy in pediatric stroke.

Key findings from these secondary survey questions include:

- The most common PNI therapeutic procedures performed are intracranial non-Galenic AVM embolization, IAC for retinoblastoma, and VoGM embolization.
- The majority (42/45 responses, 93.3%) of centers undertake five or fewer thrombectomy procedures annually for pediatric large vessel occlusion stroke.
- The majority (26/45 responses, 62.2%) would be willing to consider thrombectomy for infants (age 29 days to 12 months) in the appropriate clinical context.
- Approximately half (23/45 responses, 51.1%) of respondents do not apply a minimum pediatric NIHSS score for patient selection in thrombectomy for pediatric large vessel occlusion stroke, with clinical decisions not being based on this score alone.
- The majority (31/45, 68.9%) of centers employ an aspiration-first strategy for thrombectomy in pediatric stroke.

DISCUSSION

These are the first published consensus-based expert opinion recommendations for the practice of safe neurointervention in children. The formation of these recommendations was based on a multistage Delphi survey process assessing expert opinions from PNI practitioners worldwide to achieve consensus. The high number of responses (n=50) within the small field of PNI, as well as the presence of responses from practitioners on all inhabited continents (see figure 1), forms a strong basis to create these recommendations that are applicable across differing health systems. At present, several other surveys are underway related to PNI by a variety of research groups and societies that will hopefully further inform device usage and disease-specific workflow expertise.

Best practices for pediatric diagnostic cerebral angiography

It was recommended that diagnostic cerebral angiography in young children (<5 years of age) should be undertaken in centers that perform at least 10 pediatric diagnostic cerebral angiograms annually (see figure 2). We believe this recommendation best ensures the safety of young children who have smaller vessels,² higher rates of vasospasm,¹² and a potentially higher risk of procedure-related vessel occlusion.^{5 13} This minimum number represents a relatively low threshold for centers undertaking PNI procedures and intends to balance the clinical need for access to diagnostic cerebral angiography with the additional safety precautions necessary for safe practice in this vulnerable age group.

As members of the working group, we wish to clarify that prior to undertaking cerebral angiography in young children (age <5 years), the PNI practitioner and the treating center should have relevant neurointerventional experience in this young age group to reduce the risk of significant access complications. In our opinion, practitioners who undertake angiography in teenagers (via a predominantly adult practice) but have little experience with young children should not accept such young patients for elective cerebral angiography. Rather, we advise referring such young patients, whenever possible, to a center with relevant experience for young children.

We also note that the skill set for therapeutic PNI procedures is an extension of the skill set for diagnostic PNI angiography. Thus, in our opinion, adult centers that undertake limited or infrequent pediatric diagnostic angiography should not accept pediatric patients for elective therapeutic PNI procedures unless the child cannot reasonably access a center with relevant pediatric expertise. Angiography in young children (such as those age <5 years) is often combined with intervention, and it is progressively less common for standalone angiography to be performed in this age group in the setting of modern MRI techniques. Therefore, centers considering undertaking either angiography or intervention in young children should be equipped to undertake both.

Best practices for pediatric elective endovascular therapeutic neurointerventions

It was recommended that all *elective* endovascular therapeutic neurointerventions in pediatric patients (age <18 years) should be performed in a high-volume pediatric center with an established pediatric PNI program and multidisciplinary team, including PNI, neurosurgery, neurology, ICU, anesthesia, and developmental specialists—all with pediatric expertise. The minimum number of pediatric therapeutic endovascular interventions a center should undertake annually to be considered high volume, and therefore suitable to undertake elective PNI procedures, was agreed by consensus to be >20 procedures (figure 3). We stress again the focus here on elective neurointerventions; *emergent* situations may arise, to which this best practice recommendation does not apply.

Any major pediatric center regularly undertaking PNI procedures would likely meet this relatively low threshold, taking into account modern PNI practice inclusive of embolization for VoGM and intracranial non-Galenic AVM, IAC for retinoblastoma, and head/neck embolizations for tumors and AVM. This minimum threshold (equating to less than one therapeutic PNI procedure per fortnight) achieves a balance between providing appropriate access to care and patient safety in a vulnerable population. In our opinion, due to safety concerns, centers with minimal experience in PNI procedures should not undertake VoGM embolization or IAC for retinoblastoma, without an appropriate period of skill development in pediatric intervention and formation of appropriate pediatric multidisciplinary teams. Exceptions to this best practice would include children with no reasonable access to a center that has appropriate pediatric expertise. Under such exceptional circumstances, PNI should be guided by the consensus of a multidisciplinary team, and parents or guardians should participate in adequate informed consent discussions.

Exceptions to best practice recommendations

These best practice recommendations specifically apply to elective PNI procedures. Again, the authors recognize that in some clinical scenarios, endovascular treatment should be administered in adult centers. Such clinical scenarios include patients requiring emergent care and patients in geographically isolated regions that lack access to a PNI center. Adult centers anticipating the potential for such cases should have a referral pathway to, or channel of communication with, a major pediatric center that can aid with clinical decision making and technical advice.

Strategies for regions without a high-volume PNI center

During the IPSO Executive Committee review of these consensus best practice recommendations, specific concern was

expressed about geographically isolated and under-resourced environments in which there is no access to high-volume PNI centers. There is uniform consensus among the IPSO Executive Committee and authors of this report that it would be inappropriate to deny appropriately indicated PNI to a child within an established adult neurointerventional center in the context of such geographical isolation.

We recommend that such centers form clinical links or network-based referral pathways to a high-volume PNI center within their region. Any pediatric neurovascular cases being considered for local treatment should then be discussed with the linked high-volume center such that expert PNI input is provided for such cases. Neurointerventionists in such centers may also benefit from a period of observership in a high-volume center.

Limitations

As with any survey-based process, our methodology had several limitations. Inclusion of only practitioners who undertake at least 20% of their workload in pediatric patients created an obvious bias. However, this study design was intentional as our goal was to identify expert consensus on best practices among practitioners who regularly care for children. While responses were received from PNI practitioners in all inhabited continents, there was a dominance of North American and European centers in the results, potentially skewing responses toward better resourced health systems. The consensus rate for recommendation formation (>60%) could be considered relatively low, though we note that all five agreed best practice recommendations had a consensus rate of >70%. Although we requested that each center provide only one survey response, the responses were intentionally anonymous, so it is possible that some centers provided more than one response. Finally, given that there are no scientific results published in the literature for much of the clinical practice assessed in this survey, the level of evidence for these recommendations is low and is largely limited to expert opinion.

CONCLUSION

Pediatric neurointervention is a subspecialty with a unique knowledge base and technical skillset that serves an exceptionally vulnerable patient population. These best practice recommendations were formed through expert consensus via a two-stage Delphi process and are designed to enhance patient safety while providing appropriate clinical access to procedures that are critical for preservation of life and neurological function.

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Contributors KDB designed the study and wrote the first draft. KDB is also the guarantor. DBO helped design the study and edit the manuscript. TA helped design the study and edit the manuscript. AR helped design the study and edit the manuscript. CP-F assisted with study design and data analysis. KC assisted with administration and data collation. PBS assisted with editing the manuscript. AAD assisted with data collation and edited the manuscript. HF assisted with study design, supervision, and editing. PM assisted with study design, supervision, and editing.

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Provenance and peer review Not commissioned; externally peer reviewed.

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