




# Consensus exercise identifying priorities for research in the field of general surgery of childhood in the UK

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## Abstract

**Background:** The evidence base underlying clinical practice in children's general surgery is poor and high-quality collaborative clinical research is required to address current treatment uncertainties. The aim of this study was, through a consensus process, to identify research priorities for clinical research in this field amongst surgeons who treat children.

**Methods:** Questions were invited in a scoping survey amongst general surgeons and specialist paediatric surgeons. These were refined by the study team and subsequently prioritized in a two-stage modified Delphi process.

**Results:** In the scoping survey, a total of 226 questions covering a broad scope of children's elective and emergency general surgery were submitted by 76 different clinicians. These were refined to 71 research questions for prioritization. A total of 168 clinicians took part in stage one of the prioritization process, and 157 in stage two. A 'top 10' list of priority research questions was generated for both elective and emergency general surgery of childhood. These cover a range of conditions and concepts, including inguinal hernia, undescended testis, appendicitis, abdominal trauma and enhanced recovery pathways.

**Conclusion:** Through consensus amongst surgeons who treat children, 10 priority research questions for each of the elective and emergency fields have been identified. These should provide a basis for the development of high-quality multicentre research projects to address these questions, and ultimately improve outcomes for children requiring surgical care.

## Introduction

In the UK, children with a problem requiring treatment by a surgeon are treated either by a specialist paediatric surgeon at a specialist paediatric centre (SPC) or by a general surgeon at a district general hospital (DGH). The majority of surgeons in this latter group are primarily adult surgeons who treat children as part of their practice. Most surgical procedures that come under the umbrella subspecialty of general surgery of childhood are carried out in the DGH setting by a general surgeon.

It is recognized that treatments offered to children and the outcomes achieved, in both emergency and elective settings, vary between units<sup>1,2</sup>. This may be due to differences of opinion, different but equally valid treatment pathways, or local considerations. However, as has been more generally acknowledged<sup>3</sup>, this variation may be attributable to a lack of evidence underpinning

current surgical practice. There exists a motivation amongst surgeons to generate this evidence by participating in and contributing to high-quality research<sup>4,5</sup>.

Applying a systematic approach to the identification of important research questions should make research more useful<sup>6</sup>. A particular challenge to performing research is the availability of funding and identification of priority areas for the limited funding that exists. The generation of priority areas may facilitate funding bodies to target resources effectively. A multicentre research programme within the high-volume subspecialty of general surgery of childhood, which has often been overlooked, has the potential to deliver benefit to a large number of children and their families across many units. Against this background, the aim was to generate a list of research priorities within the field of general surgery of childhood.

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## Methods

A UK-wide modified Delphi process was undertaken to identify and prioritize research questions within the field of general surgery of childhood; both specialist paediatric surgeons and general surgeons who treat children contributed. Although there is no standard process for research prioritization, the experience of other organizations and groups that have performed similar work was used in planning the process<sup>7-9</sup>.

The scope of the prioritization process was defined as general surgery of childhood, and it was intended to include any non-specialist condition treated by a surgeon that is regularly undertaken in the UK, in both SPC and DGH settings. Acknowledging that this practice has changed over time, conditions that have recently largely migrated from the DGH setting into SPCs, such as pyloric stenosis<sup>10</sup>, were excluded. Basic science research questions were also excluded. Conditions and procedures in both emergency and elective settings were included explicitly. From the outset, the intention was to generate a 'top 10' list for both elective and emergency fields.

The project was completed in accordance with a predefined protocol, and was overseen by a steering group containing general and specialist paediatric surgeons, a parent representative, and a surgical trainee.

### Stage 1A: scoping survey for identification of research questions

Participants were invited to nominate research questions that they believed were important to answer in the field of general surgery of childhood. A link to complete a questionnaire was distributed to members of the British Association of Paediatric Surgeons, the Association of Surgeons of Great Britain and Ireland, the Paediatric Surgery Trainees Research Network, the Paediatric Stoma Nurses Group, and directly to known personal contacts of the study team who work in the field of general surgery of childhood. The survey was also advertised via social media channels of individuals and these organizations. Through this broad approach, the aim was to engage a range of key stakeholders (specialist paediatric surgeons, general surgeons who treat children, surgical specialist nurses and other healthcare professionals involved in the surgical care of children) to ensure a breadth of opinion across different professional groups. Responses in a PICO (Population, Intervention, Comparator and Outcome) format were encouraged, explicitly inviting responses in elective and emergency fields, and not limiting the number of questions each individual could propose. The survey was administered online using the REDCap tool (<https://www.project-redcap.org>)<sup>11</sup> and was open for a period of 1 month, beginning in mid May 2019. Reminder invitations were sent 1 and 2 weeks after the survey opened.

### Stage 1B: refinement of research questions

Questions were reviewed by the study team to ensure that they were within the scope of general surgery of childhood, and separated into elective and emergency themes. Questions that were considered to be exclusively within the field of specialist paediatric surgery or basic science were excluded. Where possible, the study team merged similar or related research questions into a single question. Merging of questions took place only when there was consensus amongst the study team to do so, and care was taken not to lose the detail or meaning of questions during this process. Research questions were framed in a PICO format where possible, making it explicit that the term 'outcome' would

include a variety of domains: clinical outcomes, patient experience, process or resource use (in accordance with guidance from the Health Foundation<sup>12</sup>). It was made clear that when an appropriate core outcome set was available, it was anticipated that these outcomes would be used. In this way, a long list of research questions was generated.

### Stage 2A: modified Delphi stage one prioritization process (online)

In June 2019, invitations to complete the first prioritization stage were distributed through the same channels as for the scoping survey, as well as by e-mail to all those who had completed the scoping survey and provided their e-mail address. The prioritization exercise was again completed within the REDCap online survey tool. Respondents were asked to score each research question in terms of importance to them on a 1-5 ordinal scale (1 being low and 5 being high priority). Comments on how to refine the research questions were invited. Questions were grouped into themes based on mode of presentation (elective versus emergency), and then by disease area. It was made clear to respondents that they were not being asked to rank the questions, but to score each one independently. This first stage of the Delphi process was open for completion over a 3-week period, with reminders sent after 1 and 2 weeks.

At the end of the first stage of the Delphi process the study team reviewed all proposed refinements to research questions and implemented those that were considered to be an improvement, as long as the meaning of the question was not altered. The mean score for each question was calculated, and the questions were ranked in order of priority. The top scoring 20 questions, in each of the elective and emergency categories, were forwarded to stage two of the modified Delphi process (making 40 in total).

### Stage 2B: modified Delphi stage two prioritization process (online or in person)

Stage two of the prioritization process took place over a 36-h period during July 2019 that included a dedicated session at the annual congress of the British Association of Paediatric Surgeons held in Nottingham, UK. Attendees at this dedicated session, and all those who had either submitted a research question in the scoping survey or had completed stage one of the modified Delphi process were invited to take part in person or online (if they were not in attendance). Participants had been notified previously of the short time period during which the second prioritization stage would be open for completion, and were therefore actively encouraged to participate in stage two during this time window if they were not in attendance at the congress. Respondents were prevented from participating both online and in person.

The online version was again administered using REDCap, whereas the in-person session was administered by the study team and used a commercially available online voting tool (Slido; <https://www.sli.do>). Whether online or in person, participants were shown the top 20 research questions from each category (20 emergency, 20 elective). These were presented in random order along with the mean score assigned in stage one, together with the distribution of scores for stage one, given as a bar chart. Once again, respondents were asked to assign a priority score using the same 1-5 priority scale as in stage one, taking into account, if they wished, the score assigned in stage one. Scores from all respondents were analysed, and a mean score for each

research question was calculated. Questions were then ranked in order of priority.

## Results

In the scoping survey 226 questions were submitted by 76 different individuals (54 consultants, 16 trainees, 6 specialist nurses). Of these 76 participants, 63 treated children only and 13 treated both adults and children. All 226 questions (134 elective and 92 emergency) were considered by the study team; 53 were considered outside the scope of the study either because they were not research questions ( $n=4$ ) or because they related to specialized paediatric surgical care ( $n=49$ ). The remaining 173 questions were refined by the study team into 71 unique research questions, of which 38 related to elective and 33 to emergency conditions. A broad range of topics were covered, as classified into clinical themes by the study team (Table 1). All 71 research questions were included in stage one of the modified Delphi process. The questions submitted and details of how they were used are shown in Appendices S1 and S2.

A total of 168 individuals took part in phase one of the prioritization process (102 consultants, 40 trainees, 3 specialist nurses, 1 researcher, 22 unknown). Of the 145 known to be involved in clinical care, 132 treated children only whereas 13 treated both adults and children. The mean score for each question ranged from 2.3 to 2.9 (the individual scores are shown in Appendices S1 and S2). The top scoring 20 questions in each of the elective and emergency categories progressed to stage two of the prioritization process. This was completed by 49 people online and by 115 in person at the congress (total 164: 90 consultants, 66 trainees, 1 researcher, 1 specialist nurse, 6 patient representatives who were attending the conference as representatives of invited charities). Of the 157 known to be involved in clinical care, 123 treated children only and 34 treated both adults and children.

The top 10 priority research questions in elective and emergency general surgery of childhood are shown in Tables 2 and 3, with the mean score assigned to each in stage two. Scores for the remaining questions are shown in Appendices S1 and S2.

## Discussion

This study identified research priorities within the field of elective and emergency general surgery of childhood. This is the first time any such project has been undertaken within this clinical field. Clarifying the knowledge gaps that should be addressed may be useful for both researchers and funders, to

facilitate planning and allocate resources. Ultimately, the hope is that addressing these questions will improve outcomes for children.

Despite most children who require general surgical care in the UK being treated by a general surgeon in a DGH, paediatric surgical research tends to focus on specialist settings. Importantly, the research priorities identified in this study were based on the views of surgical teams delivering care in both SPCs and DGHs. Collaboration across both specialist and non-specialist centres is likely further to enhance clinical and research networks. Indeed, a wide range of professionals have had the opportunity to inform these priorities, which should enhance relevance, engagement and uptake.

The prioritized questions typically relate to common conditions or procedures that, with appropriate collaboration, are likely feasible to pursue. It is possible that some of the prioritized questions are not necessarily easy to answer, and there is a risk they may become redundant, but their inclusion in a top 10 list arising from this process may itself be adequate justification for them to be pursued.

Although the current methodology is based on that used by others previously<sup>7,8</sup>, there are limitations with this approach, and the list of priorities may not be comprehensive. Important stakeholders such as service users (patients, parents, siblings) and service funders (managers, commissioners and charities) have not yet been included within this prioritization process. It is possible that these other stakeholder groups may have alternative priorities for research in this field that are not represented here. From the outset, it was considered whether to engage with these groups within the same prioritization process as surgeons, and specifically how it would be possible to involve representative patients and carers. However, bearing in mind their completely different knowledge base, together with the psychosocial aspects of their involvement, it was felt that a meaningful prioritization would need quite different tools and methodologies (which may themselves need development). For instance, most patients and families will not have adequate knowledge of all the conditions included here to be able to make an informed prioritization. It is possible, therefore, that the process may become skewed towards those conditions represented most within any patient representative group. The issue of priority should be explored with these other stakeholders in the future.

Alternative methodologies exist for identification of research questions (such as systematic review of the literature), although these are typically labour and resource intensive. There may be important research questions not included here because they were not proposed in the scoping survey. Respondents to the scoping survey would be less likely to be aware of 'discovery phase' projects, or those addressing an important problem for a small group of patients. The inclusive approach of forwarding all proposed questions—no matter by whom they were proposed, to which topic they related, and how advanced they were in the research process—into stage one of the modified Delphi process ensured that the process itself did not generate additional discrimination. It is recommended that researchers and funders continue to consider the priorities of other stakeholders, the potential benefits of basic science research, the need to consider discovery-phase projects, and issues of importance to small patient groups.

The act of prioritizing research for general surgery of childhood may stimulate discussion about other conditions. Experience has been gained that will inform prioritization in other areas within the specialty, for example oesophageal atresia,

**Table 1 Clinical themes represented by questions proposed in the scoping survey**

Elective	Emergency
Cholecystectomy	Acute scrotum
Colorectal	Appendicitis
Elective miscellaneous	Emergency miscellaneous
Enhanced recovery after surgery (ERAS)	General emergency surgery
General elective surgery	Inguinal hernia (emergency)
Inguinal hernia/hydrocele (elective)	Trauma
Ingrowing toenail	
Phimosis	
Pilonidal disease	
Tongue tie	
Umbilical conditions	
Undescended testis	

Table 2 Elective questions: top 10

Rank	Question	Mean score (maximum 5)
1	In children having elective GI surgery, does the use of an enhanced recovery pathway result in better outcomes than current standard of care? Which parts of ERAS pathways are most beneficial in children?	4.06
2	In boys with palpable UDT, does orchidopexy at less than 13 months of age result in better outcomes than later orchidopexy? Does bilateral compared with unilateral UDT influence this?	3.78
3	In children with pilonidal disease, which surgical treatment results in the best outcomes?	3.73
4	In adult men, is a history of previous inguinal hernia or hydrocele repair associated with a higher risk of infertility than in men who have had no repair?	3.60
5	In children having inguinal hernia repair, does laparoscopic repair result in better outcomes than open repair? Are there factors such as age, sex, side of hernia that influence this?	3.59
6	In boys with palpable bilateral UDT, does bilateral synchronous orchidopexy result in better outcomes than two separate unilateral procedures?	3.52
7	In children having cholecystectomy, does treatment in a specialist children's hospital or in a general hospital result in better outcomes?	3.52
8	In boys with a congenital or acquired (not due to torsion) unilateral testis, does fixation of the solitary testis result in better outcomes than no fixation?	3.50
9	In girls with an asymptomatic inguinal hernia suspected to contain ovary, does urgent repair result in better outcomes than elective repair?	3.41
10	In children requiring general surgery of childhood, what are the patient and parent preferences about where they are treated – DGH versus specialist centre? What factors influence this?	3.38

GI, gastrointestinal; ERAS, enhanced recovery after surgery; UDT, undescended testis; DGH, district general hospital.

Table 3 Emergency questions: top 10

Rank	Question	Mean score (maximum 5)
1	In children undergoing appendicectomy, to what extent does duration, type and administration route of antimicrobial treatment affect outcomes? Do any specific organisms (e.g. <i>Streptococcus milleri</i> ) require variations in therapy (e.g. longer course of treatment)?	3.99
2	In children with blunt abdominal trauma and solid organ injury managed non-operatively, what duration of activity restriction results in the best outcomes?	3.90
3	In children with blunt abdominal trauma and solid organ injury managed non-operatively, does routine follow-up imaging result in better outcomes compared with none? And at what time interval should it be performed?	3.72
4	In children presenting with an ingested button battery that is seen radiologically within the stomach, which of urgent removal or selective removal or routine follow-up imaging or discharge with no follow-up results in the best outcomes?	3.69
5	In children presenting with an incarcerated inguinal hernia that is successfully reduced, does immediate (within 24 h) or urgent (within 1 week) or elective (longer than 1 week) repair result in the best outcomes? Are there clinical factors, such as age or sex, that influence this?	3.65
6	In children with uncomplicated acute appendicitis, does appendicectomy or non-operative treatment with antibiotics result in better outcomes?	3.63
7	In children with complicated appendicitis, does appendicectomy or non-operative treatment (with or without drain) result in better outcomes compared with no appendicectomy? Are there clinically distinct subgroups (e.g. mass, abscess, neither) that influence this?	3.61
8	In girls suspected to have ovarian torsion, to what extent might use of investigation (e.g. imaging) improve outcomes?	3.57
9	In children with a postappendicectomy collection, how might intervention (percutaneous drain or open drain) improve outcome compared with antibiotics alone? Are there factors, such as age or size of collection, that influence this?	3.47
10	In boys with testicular torsion confirmed at surgery, how might method of fixation (e.g. dartos pouch versus suture fixation, absorbable versus non-absorbable sutures, 2- versus 3-point fixation) influence outcomes?	3.40

Hirschsprung disease, and anorectal malformations. Such specialist areas of work are likely already to have better defined patient pathways and engaged parent/patient groups; it is likely that these may be approached in a way that allows service users' priorities to be captured more easily.

Disclosure. The authors declare no conflict of interest.

## Supplementary material

Supplementary material is available at BJS Open online.

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