Supplementary Material 1: Stakeholder involvement

Contents

Report of Activity 1a

ACTIVITY 1 - Develop an intervention taxonomy and agree most important interventions. Establish which interventions, combinations and sequences, for treatment of CFC, are available and being used, which are considered most important, and to develop an intervention taxonomy **Activity 1a: Face to face team meeting.**	
1. Aim	
Task aim	Establish which interventions, combinations and sequences, for treatment of CFC, are available and being used, which are considered most important, and to develop an intervention taxonomy of terms using the identified treatments.
2. Methods	
Who was involved?	PPI group (lived experience and parents): Karen Jankulak, Margaret Ogden, Deb Smith, Clare Milligan. Health professional and charity group: Jonathan Sutcliffe, Brenda Cheer, Davina Richardson, June Rogers. Research group: Doreen McClurg, Lorna Booth, Alex Todhunter-Brown, Pauline Campbell, Claire Torrens, Andy Elders, Helen Mason, Suzanne Hagen. Note: Day 2 was planned as a training day for the PPI group, and not all SG members received invitations to this in advance, so some were unable to stay for this.
When was the involvement?	Within 1 month of the review project starting (28-29 January 2020).
What	Face to face (in-person meeting), held over 2 days.

happened?	Day 1 - Welcome and Introductions - Presentation of project background and aims - Role of the PPI group discussed: to ensure relevance and usefulness of the work being conducted. - Discussed & agreed meeting rules and methods of voting to reach consensus - Task 1 – discussion and agreement that management of CFC could be looked at in terms of levels (family, system etc.) in order to create the taxonomy. A draft taxonomy was presented to the PPI group based on the initial scoping review using the terms identified from the papers within the NICE guidelines (2017, 2012, 2010) and NASPGHAN guidelines (2014). During an extensive discussion using flip charts, whiteboard etc. substantial progress was made on agreeing the headings and sub-headings of the taxonomy (see below) with refinement completed on Day 2. The content of the taxonomy was agreed without the requirement to vote. Day 2 - Research Training – training provided on differences between primary research and secondary research, with explanations of how the SUCCESS project is secondary research. - Recap and refinement of taxonomy through discussion and agreement. - Combinations and Sequences of Treatments – The group discussed and agreed the treatments that should be prioritised when investigating effectiveness. It was suggested that self-management at home was seen as the key setting and first part of any timeline in relation to treatment. The group discussed and agreed a model of sequence for treatment (see below) - Forward Planning
Level of involvement	We consider that the SG had control over developing the intervention taxonomy and led the development of the pyramid.
3. Results	
Outcomes —Report	Consensus when voting. It was agreed that when voting was used to confirm if there was consensus:

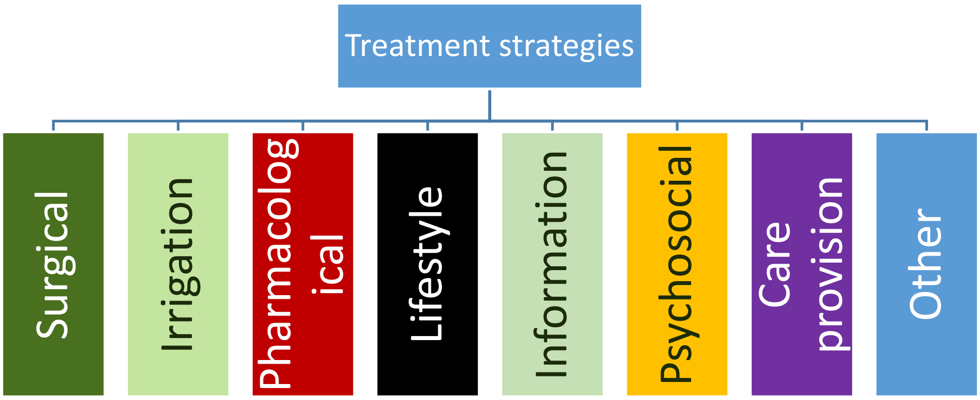
the results of SG in the study, including both positive and negative outcomes

- the importance of the decision may impact on how to determine consensus.
- Generally, it would be useful to combine strongly agree/ agree on one end and strongly disagree/ disagree (of a 5 point scale) to make a clear split.
- Comments and discussion including opposing views will be used to reach a conclusion.
- Votes of neither/nor (in the middle) will not be counted

<u>Key meeting outputs</u>

A draft taxonomy of treatment for childhood functional constipation and a draft sequence pyramid of treatments were developed by the SG:

Draft Taxonomy



Main heading	Sub-heading	Specifics
Surgical	Anorectal myectomy	
	Anal & pelvic floor interventions	
	Colon resection with anastomosis & rectal operations	
	Operations that provide antegrade colon irrigation (ACE)	Antegrade continence enemas
		Malone anterograde continence enema (MACE)
	Permanent or long term stoma	Cecostomy / cascostomy button
	Manual evacuation	

	Rectal biopsy	

Main heading	Sub-heading	Specifics
Irrigation	Colonic irrigation	
	Antegrade continence enemas (ACE)	
	Malone anterograde continence enema (MACE)	

Main heading	Sub-heading	Specifics
Pharmacological	Laxatives	Polyethylene glycol • *- PEG with electrolyte* • *- PEG without electrolyte*
		Senakot (Senna concentration)
		Oral biscodyl
		Glucomman
		Lactulose
		Macrogol (eg, Movicol)
		Cassia Fistula's Emulsion
		Sodium Picosulfate
		Ducosate Sodium
	Milk of Magnesia	
	Mineral Oil	
	Botox injection in the anal sphincter	
	Enema	Paraffin
		Saline
		sodium-dioctyl sulfosuccinate and sorbito
		Milk & molasses
		Soap enema
	Suppository	Glycerol Suppositories
	Demperdone	
	Lebriprostone	
	Osmotic bulk forming stimulants	
	Lubricating agents	

Main heading	Sub-heading	Specifics
Lifestyle	Exercise	Standing
		Yoga
		Strength Training
	Physical Therapies	?
	Diet	Tailored diet management
		Dietary fibre & whole grains
		Probiotics. - Lactobacillus GG - Bifidobacteria - Other micro-organisms
		Prebiotics
		Soy milk
		Diet restriction / diet replacement (e.g. removing cows milk from diet)

		Milk forumulas
		Goats yoghurt
		Supplements
	Fluids	
	Toileting Programmes	

Main heading	Sub-heading	Specifics
Information	Wider Children’s Workforce	
	Peer Support	
	Parental training and advice	
	Educational leaflets	
	Lifestyle advice	

Main heading	Sub-heading	Specifics
Psychosocial	Psychotherapy	
	Counselling and talking therapies	
	Incentives (eg, reward system or financial)	
	Interventions aimed at social issues	Social stories

Main heading	Sub-heading	Specifics
Care provision	Consistency of care	
	Continuity of care	
	Model of care	Nurse-led clinics
		Consultant-led clinics
		Bowel management clinics

Main heading	Sub-heading	Specifics
Other	Complementary & /or alternative therapies	Reflexology
		Connective tissue massage
		Acupuncture
		Mind-body therapy
		Homeopathy
		Musculoskeletal manipulations (e.g. osteopathy, chiropractic manipulation)
	Neuromodulation	Transcutaneous electrical stimulation
		Sacral modulation
		Tibial nerve stimulation
	Feedback	EMG biofeedback
		Biofeedback at home
		Biofeedback – video games controlled by external sphincter activity
		Manometry
		External anal sphincter EMG biofeedback
	Equipment	Continence containment products
		Toilet posture equipment
		Other?
	Other	Kinesio taping

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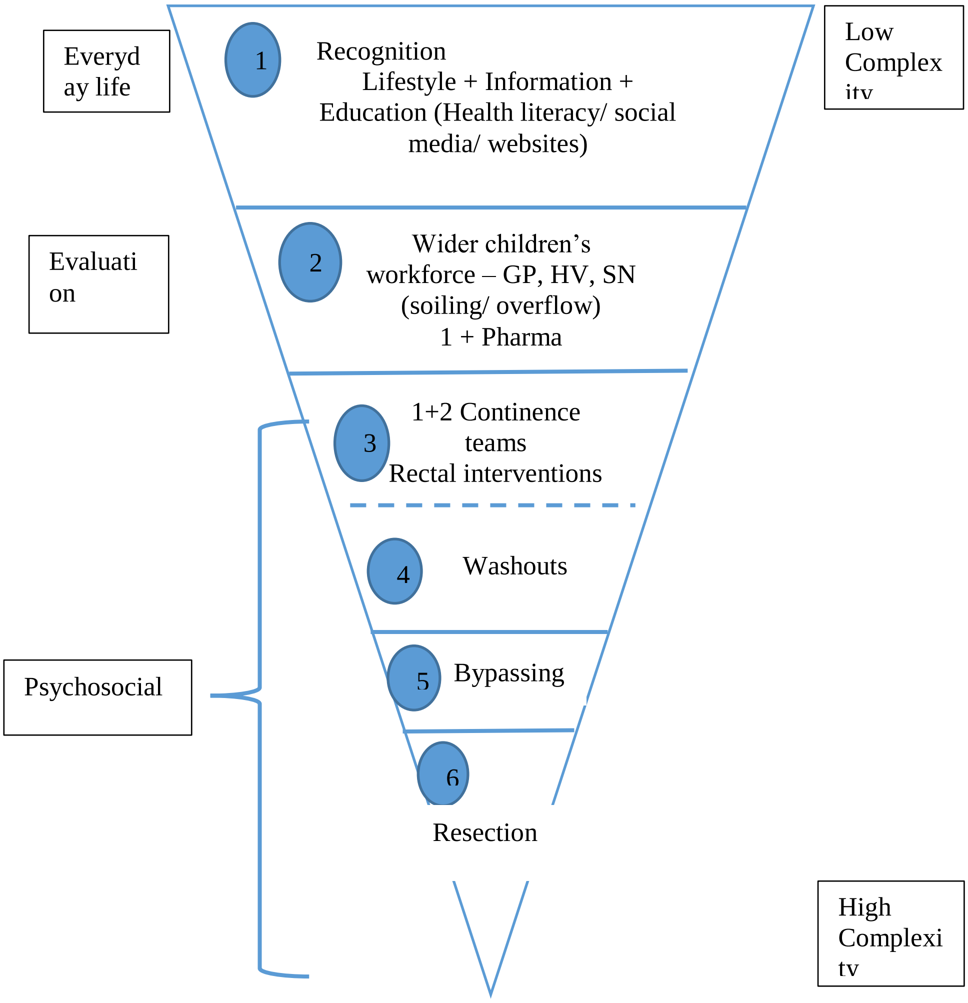
Draft ‘Pyramid’ Model



This initial draft Pyramid was refined in an iterative manner, by email and during subsequent meetings. This resulted in a reduction in the number of different levels (to four), labelling the first level as ‘level 0’, agreed terminology to describe each of the levels, and replacement of the straight lines with ‘wiggly’ lines to denote that there is often not a clear distinction between levels. The final version is presented in Chapter 2 of the report.

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4. Discussion & conclusions

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<tr><td>Outcomes
—
Comment</td><td>The taxonomy and sequence pyramid, created and agreed by the SG, influenced and directed the way in which the review identified and categorised different treatment types.</td></tr>
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on the extent to which SG influenced the study overall. Describe positive and negative effects	During discussion it was acknowledged that other ‘pyramids’, or hierarchies, summarising different levels of intervention exist. However, the inclusion of the “pre-clinical” level (referred to in this version of the Pyramid as ‘Recognition’, but later referred to as Interventions by family and carers) was proposed by the SG and considered to be unique. **Importance of prevention and diagnosis** During Day 1 there was discussion about the line between prevention and intervention. It was highlighted that diagnosis is very important, and that it is often delayed, leading to worsening symptoms and additional treatment. Prevention is also more important with, for example better education of parents of children who are more likely to develop constipation. There was a discussion to clarify the question we were trying to answer in this project which is ‘what management strategy/ies works best.’ It was concluded that diagnosis was not within the scope of this project, but that it was imperative that the importance of prevention and diagnosis should be highlighted, as this will – arguably - lead to better overall management. During Day 2 there was further discussion about including diagnostics (as per discussion previous day) and concerns that this was not within the remit of the project. After some discussion and debate, it was agreed that diagnostics should not be a main focus as the research was to identify combinations and sequences of TREATMENT strategies. However, as diagnosis was an important issue to all stakeholders and representatives, it was agreed that it should be highlighted at some point that accurate and timely diagnosis will affect treatment effectiveness. Looking at the full journey of constipation “management” could be a recommendation of the project. It was suggested that although diagnosis is not part of the remit of this project, it may be useful to capture some of the

	information in the extraction regarding the journey, early intervention, process of diagnosis, e.g. capturing early diagnosis, consider the context/ nature of diagnosis. This may have an impact for practitioners, accurate and timely diagnosis.
5. Reflections / critical perspective	
Comment critically on the study, reflecting on the things that went well and those that did not, so others can learn from this experience	***SG reflections on the developed Taxonomy and Pyramid*** ● “Think the treatment strategy list is excellent.” ● “*On reflection, not certain the term ‘care provision’ is clear – wonder if ‘organisation of care provision’ is a better term*” – other SG members later agreed with this reflection. ● ‘soiling / overflow’ was put under ‘Wider children’s workforce’: “*not sure why (soiling/overflow) are in this section 2. These are not the only conditions that would trigger the involvement of the wider children’s workforce. unresolved abdominal pain, constipation without soiling or overflow etc. would also result in involvement.*” ● **“**1 + 2” is not clear – what did we mean by this? ● ‘Continence teams’ – **“***shouldn't it just read bladder and bowel teams? The term children's bladder and bowel service is preferred by most services over continence now.”* ● *“there may be better terms for dissemination for 4, 5 and 6; 4= Irrigation. 5= Diversion (stoma formation). 6= complex surgical intervention (e.g. resection)….”. (some remained happy with the original terms, but others agreed with this view).* ***SG reflections on the meeting*** The SG were generally positive about the format of the meeting and the outcomes (taxonomy and pyramid) which were produced: “*The discussions were very interesting and informative, capturing different perspectives. The points raised and decisions made influenced the way the project progressed*.“ Some concerns were raised about the exclusion of diagnostics. There were some negative reflections relating to the meeting format and running: “*the rules not being observed and not reiterated around raising of*

hands before people spoke definitely left me overwhelmed and feeling powerless in relation to the clinicians who were present". These reflections were echoed in the reflections by research team members (see below).

Research team reflections:

The SG were provided the opportunity to contribute to the decisions made about strategies and the draft taxonomy. The researchers did not get involved in the decisions made. SG members discussed and shared clinical experience and personal experiences. It was explained that the researchers were interested in everyone's point of view and this session was about them leading the way that the review should be conducted - passing the power to the stakeholders.

The research team made some observations relating to the management / organisation of discussion:

- "*Some stakeholders did not always adhere to these rules (hand up to speak) and this may have left some of the stakeholders feeling overpowered at times."*

The research team reflected that they could have managed this better:

- "*Sometimes the discussions went off track, perhaps leaving it too long to bring the meeting back to the point of discussion.* "

Subsequent reflections (at the end of the project)

Later reflections relating to the fact that some people had not followed the 'hands up' rule and that others had felt overwhelmed included:

- "*I think the comments here are very pertinent and as someone who is prone to speaking, rather than hand raising, would like to add my apologies. I think that this is an important take away for clinicians and think there may be some lessons to be learnt for moderators of such a mixed group as well. It would be good to have feedback earlier in the project to prevent this from happening again, but also to hear the impact of the virtual world and if this continued to impact.*"
- *"I may, without meaning to, be someone that interrupted. I am very sorry if so. From my perspective, this was very different to a usual*

	discourse at work i.e. significantly more moderated. I’m therefore recognising the very different perspective on the same scenario. The intent was however benign (I hope). There were things that remained unsaid, perhaps for all of us.” ● *“There is always a risk of people overwhelming others when people are passionate about the topic”.*

Report of Activity 1b

ACTIVITY 1 - Develop an intervention taxonomy and agree most important interventions. Establish which interventions, combinations and sequences, for treatment of CFC, are available and being used, which are considered most important, and to develop an intervention taxonomy

Activity 1b: Online meetings and email prioritisation exercise.	
1. Aim	
Task aim	To reach consensus on which questions are of highest priority for a systematic review of effectiveness.
2. Methods	
Who was involved?	Meeting 31-03-20: **Attendees:** Doreen McClurg, Lorna Booth, Pauline Campbell, Alex Pollock, Suzanne Hagen, Andy Elders, Davina Richardson, June Rogers, Deb Smith, Clare Millington, Margaret Ogden, Claire Torrens, Jonathan Sutcliffe (attended part of meeting). **Apologies:** Katherine Barlow, Helen Mason, Brenda Cheer, Karen Jankulak. Meeting 13-05-20: Attendees: Doreen McClurg, Lorna Booth, Pauline Campbell, Alex Pollock, Deb Smith, Clare Millington, Margaret Ogden, Apologies: Karen Jankulak Meeting 06-07-20: Attendees: Doreen McClurg, Lorna Booth, Pauline Campbell, Alex Pollock, Andy Elders, Claire Torrens, Deb Smith, Clare Millington, Margaret Ogden, June Rogers, Davina Richardson Apologies: Karen Jankulak, Jonathan Sutcliffe, Tracey Barber, Brenda Cheer
When was the involvement?	Meeting 31-03-20 Presentation of written material – several emails and iterations of documents, including: ● Meeting 13-05-20 ● Voting (by email) 01-06-20 ● Meeting 06-07-20
What	**Online meeting 31-03-20.** This was discussed as one of several agenda items. Prior to the meeting a document summarising the challenges of

happened?	prioritising interventions for the systematic reviews of effectiveness was circulated. To inform their decision making, the SG asked the research team to draft research questions based on the intervention combinations recommended within the NICE guidelines. **Presentation of written material** - The research team completed the requested task of drafting research questions (as above) and presented the draft questions in a written format for the SG to consider. During this it was observed that a key limitation of basing the research questions on the NICE recommendations was that "level 0" from the pyramid were missed, as Level 0 interventions are selected and delivered by family/carers and are therefore not covered by clinical guidelines. Feedback from SG members led to a further iteration of the draft research questions, with 6 broad questions based on the pyramid, in which: ● Two questions applied across different levels of the pyramid ● Four questions each applied to one of the four levels of the pyramid (i.e. level 0, 1, 2 and 3 respectively) In addition, each of the 6 broad questions were 'broken down' into more specific questions. These more specific questions were based on (i) recommendations in the NICE guidelines and (ii) the 'intervention taxonomy' which was developed and agreed by the SG. These more specific questions were not designed to cover absolutely every intervention, and it was recognised that these could be added to (or amended) during the systematic reviews, but were included to provide the SG with examples of the sort of questions that could come under each of the broad questions. **Online meeting 13-05-20.** A meeting of the PPI group members only was held, as the research team had concerns that their voices were not being heard in full SG meetings. The prioritisation of questions for the

	effectiveness review was discussed as one agenda item, and all members given opportunity for questions and feedback. **Voting (by email) -** The SG members each independently ranked the 6 broad questions, with access to the example specific questions to inform their decision making. The instruction was to "RANK which you feel is of highest priority for a systematic review of effectiveness, from "1" (highest priority) to "6" (lowest priority)." The voting information sheet is at the foot of this document. **Online meeting 06-07-20.** The submitted rankings were combined statistically, and the combined rankings presented to the SG at the meeting. Based on these results, the SG reached consensus that there were 2 questions of shared "top" priority, three of "medium" priority and one of "low" priority for the systematic review of effectiveness.
Level of involvement	We consider that the SG had control over determining the questions that were prioritised and the priorities assigned to these questions.
3. Results	
Outcomes—Report the results of SG in the study, including both positive and negative outcomes	<u>Meetings and email discussion</u> It was agreed that the 'Pyramid' model should be used to guide prioritisation (rather than the Intervention Taxonomy). Subsequent development of questions for prioritisation were developed iteratively through a series of online discussions and email correspondence. During these discussions a number of further modifications were discussed and agreed for the Pyramid. <u>Final Priorities</u>

	The SG agreed that the six broad questions to be addressed by the systemic review of effectiveness, and their priorities (‘high’, ‘medium’ and ‘low’) were: ● What is the effectiveness of different models of service delivery? (High priority) ● What is the effectiveness of ‘everyday life’ interventions delivered by carers, without the involvement of healthcare professionals? (High priority) ● What is the effectiveness of interventions delivered/prescribed by the wider children’s workforce (primary care services – GP, HV, SN)? (moderate priority) ● What is the effectiveness of interventions delivered by continence teams (secondary care – specialist – services)? (moderate priority) ● What is the effectiveness of psychosocial and/or complementary interventions? (moderate priority) ● What is the effectiveness of interventions delivered by consultant-led teams (tertiary care services)? (low priority)
4. Discussion & conclusions	
Outcomes—Comment on the extent to which SG influenced the study overall. Describe positive and negative effects	During discussion on prioritisation, some members of the SG expressed that they would like to know more about what evidence is available before making any other priorities for evidence to be synthesised. The point was made that the purpose of this exercise was to agree what treatments are important to consider, and not necessarily the treatments where there is known evidence or evidence of effect; or the treatments which are available/accessible. This was repeated in any subsequent written documents which were sent out. In relation to the impact of the prioritisation, it was agreed that that all questions are important, but that to conduct the effectiveness review within our timescales, the review will concentrate on the high priority reviews providing statistical analyses. Results for medium priority reviews will be brought together but not in the same depth as for the high priority. A further

	step-down approach will be taken for the low priority review meaning that it will be unlikely to do any statistical analyses for this question.
5. Reflections / critical perspective	
Comment critically on the study, reflecting on the things that went well and those that did not, so others can learn from this experience	**Stakeholder reflections:** One of the documents that was sent round said “important to children with CFC, their families, and the health professionals providing care”. Stakeholders suggested that the word “carers” should also be used: “*And carers – important to say this. We’ve talked about family carers a lot. But in certain situations it may be professional carers working in social care*”, and “*I agree….the term carers should be used throughout*”. On reflection on the wording in written documents which stated: “Important note: What we do NOT want you to think about is whether you know that there is (or is not) research evidence to answer the question. It is the importance of the question that we want to know about, not the presence or absence of evidence to answer that question.”, one stakeholder reflected “*I think you got this message over to this very effectively – this was important as it’s not the usual approach we take in PPI but you communicated it with us well.*” On the instructions for the ranking exercise: “*this was a clear instruction and was an enjoyable task*”. On the wording of the questions: ● “*The term consultant-led teams is worth considering. Some of the treatments delivered by ‘continence teams (secondary care – specialist – services)’ are by nurses, surgeons, gastroenterologists. This section is in reality predominantly surgical team (including nurses) delivered.* ● “*Presumably for both E and F you are referring to interventions*

intended to help children who remain resistant to conventional management? And all parts need ongoing delivery and frequent review in parallel"

- *"We talked in one session about the potential to have a formal 're-evaluation' of care and opportunity for families to have questions answered at the beginning of this phase."*
- *"Most of the interventions in section E would only be offered by specialist teams working within secondary care (hospitals). Community bladder and bowel teams would not have access to anything other than possibly TENS, biofeedback at home (and that would be limited), containment products, toilet posture and other equipment and colonic irrigation. ACE and MACE would be formed usually in tertiary referral hospitals by paediatric surgeons and washouts would be started in hospital, although follow up support may come from the community bladder and bowel team."*
- *"Rectal biopsy is a diagnostic procedure, rather than an intervention to improve constipation, and should not have been listed as an intervention"*

Research team reflections:

"*This exercise did not go as we anticipated when we wrote our funding proposal. We envisaged that we would have a list of specific interventions, and that the stakeholders would prioritise these, leaving us with reviews of specific interventions to carry out. Prioritisation of the broad questions leaves the challenge of identifying the more focussed, intervention-specific questions which the reviews of evidence will address, and this will now need to be led by the evidence in order to synthesise evidence under the broad questions as requested by the SG."*

"*The identified questions, and prioritisation, highlights the influence (control) that stakeholders can have. It was challenging to work with the stakeholders, and the number of 'iterations' was high in order to reach something that the stakeholders were happy with. The input of the stakeholders has totally shaped the format of the reviews of effectiveness which will be done for this project.*"

"It is quite daunting to be tasked with doing reviews of effectiveness which

	address such broad questions, but from listening to the SG members I can understand why they want us to focus on bringing things together under these broad questions. This will hopefully make our reviews really useful to the end-user”.

Document sent out for email voting exercise

Prioritisation of effectiveness review questions

What information is in this document?

The Stakeholder Group have developed and agreed a "pyramid" which reflects who might be delivering interventions at different points in a child's care, and what those interventions might be.

We have written 6 broad questions based on this pyramid, and the discussion that the Stakeholder Group have had:

- Questions A. and B. are questions which apply across different levels of the pyramid
- Questions C, D, E and F each apply to one of the four levels of the pyramid (i.e. level 0, 1, 2 and 3 respectively).

We have also written a range of much more specific questions, 'breaking down' these 6 broad questions. These more specific questions are based on (i) recommendations in the NICE guidelines and (ii) the 'intervention taxonomy' which was developed and agreed by the Stakeholder Group at our first meeting. These more specific questions might not cover absolutely every intervention, and could be added to (or amended) during our systematic reviews, but we have included them here to let you see the sort of questions that would come under each of the broad questions.

The 6 broad questions are listed in the Table on Page 2.

The more specific questions relating to each of these are listed on the following pages (page 3-7).

What do you want me to do now?

We know that ALL of these questions are going to be important to children with CFC, their families, and the health professionals providing care.

However, we want to know **which you feel is of highest priority for a systematic review of effectiveness**.

To answer this, you could think about lots of different things, like:

- which question you currently feel most uncertain about answering
- what evidence (or answer to which question) you feel would provide the greatest benefit to children and their families
- other things which matter a lot to you, and to children with CFC and their families

Important note: What we do NOT want you to think about is whether you know that there is (or is not) research evidence to answer the question. It is the importance of the question that we want to know about, not the presence or absence of evidence to answer that question. The systematic review that we will do has been designed to deal with the different types of evidence that there might (or might not) be.

We want you to rank the 6 broad questions, from the one you think is of highest priority for a systematic review (number 1), to the one that you think is of the lowest priority for a systematic review (number 6).

We ask you to do this on Page 2.

<u>RANKING the 6 broad questions</u>

This table shows the 6 broad questions about effectiveness of interventions, and how these fit with the "pyramid".

We want you to **RANK which you feel is of highest priority for a systematic review of effectiveness**, from "1" (highest priority) to "6" (lowest priority).

Level	**PYRAMID HEADINGS**		**BROAD QUESTIONS**	Example specific questions	YOUR RANK
(Across different levels of the pyramid)			A. **What is the effectiveness of different models of service delivery?**	See page 3	
(Across different levels of the pyramid)			B. **What is the effectiveness of psychosocial and/or complementary interventions?**	See page 3	
0	Carer interventions	Lifestyle (diet, fluid, exercise) + Information (peer/social media/ websites)	C. **What is the effectiveness of 'everyday life' interventions delivered by carers, without the involvement of healthcare professionals?**	See page 4	
1	Wider children's workforce	Lifestyle (diet, fluid, exercise, toileting programmes) + Information (Education) + Pharmacological (laxatives)	D. **What is the effectiveness of interventions delivered/prescribed by the wider children's workforce (primary care services – GP, HV, SN)?**	See page 5	
2	Continence teams	As 1 + Feedback, equipment, physical therapies + Irrigation	E. **What is the effectiveness of interventions delivery by continence teams (secondary care – specialist – services)?**	See page 6	

3	Consultant-led teams	As 2 + Surgical	F. **What is the effectiveness of interventions delivered by consultant-led teams (tertiary care services)?**	See page 7	

Giving us your Ranking

Please send your ranking to us.

You can do this by putting your ranks in the right hand column of the table:

- Please do this by putting "1" beside to the broad question which you think is of the highest priority, "2" beside to the one you think is of next highest priority, and keep doing this until you get to number 6.

Or, you might find it easier to send your ranking in a different way. It's your ranking that we are interested in, and we don't mind how you send this to us. In our email to you we have included a table which you might prefer to use instead of this form.

Remember!

Remember – you are NOT saying which area of care provision you think is most important. You ARE saying what question you think is of highest priority for a systematic review of effectiveness.

Example Specific Questions for each of the 6 broad questions

A. What is the effectiveness of different models of service delivery?

1. What is the effect of different models of supporting / promoting consistency of care?
2. What is the effect of different models of supporting / promoting continuity of care?
3. What is the effect of different models of care, including nurse-led clinics, consultant-led clinics and bowel management clinics?

B. What is the effectiveness of psychosocial and/or complementary interventions??

1. What is the effect of talking therapies (e.g. psychotherapy and/or counselling on outcomes of children with CFC?
2. What are the effects of incentives (e.g. reward system, or financial) on outcomes of children with CFC?
3. What are the effects of interventions aimed at social issues (e.g. social stories) on outcomes of children with CFC?
4. What are the effects of complementary and/or alternative therapies on outcomes of children with CFC?
    - 4.1 What is the effect of reflexology on outcomes of children with CFC?
    - 4.2 What is the effect of connective tissue massage on outcomes of children with CFC?
    - 4.3 What is the effect of acupuncture on outcomes of children with CFC?
    - 4.4 What is the effect of mind-body therapy on outcomes of children with CFC?
    - 4.5 What is the effect of homeopathy on outcomes of children with CFC?

C. What is the effectiveness of ‘everyday life’ interventions delivered by carers, without the involvement of healthcare professionals?

	What is the effect of		**Compared to**
	INTERVENTION 1	**+ INTERVENTION 2**	**INTERVENTION 3**
1	**Lifestyle changes**		
1.1	Changes to ensure healthy balanced diet and adequate fluid		No treatment
1.2	Changes to ensure healthy balanced diet and adequate fluid	Exercise	No treatment
1.3	Dietary supplements – prebiotics (non prescribed)	Exercise	No treatment
1.4	Dietary supplements – probiotics (non prescribed)	Exercise	No treatment
2	**Information**		
2.1	Information from peers, or social media and/or websites		No treatment
3	**Pharmacological**		
3.1	Non-prescribed or pharmacy prescribed (over the counter) laxative		No treatment
3.2	Non-prescribed or pharmacy prescribed (over the counter) laxative	Lifestyle changes	Lifestyle changes

D. What is the effectiveness of interventions delivered/prescribed by the wider children's workforce (primary care services – GP, HV, SN)?

	What is the effect of		Compared to
	INTERVENTION 1	**+ INTERVENTION 2**	**INTERVENTION 3**
1	**Pharmacological interventions (maintenance therapy)** (NICE Recommendation 1.4.11)		
1.1	HP prescribed laxatives (polyethylene glycol 3350 + electrolytes)		No treatment
1.2	HP prescribed laxatives polyethylene glycol 3350 + electrolytes	stimulant laxative	HP prescribed laxatives
1.3	HP prescribed laxatives	Other pharmacological intervention (see taxonomy)	HP prescribed laxatives
2	**Diet and lifestyle** (NICE Recommendation 1.5)		
2.1	HP prescribed laxatives	Toileting programmes *(Negotiated and non-punitive behavioural interventions suited to the child or young person's stage of development. These could include scheduled toileting and support to establish a regular bowel habit, maintenance and discussion of a bowel diary, information on constipation, and use of encouragement and rewards systems. 1.5.2)*	Laxatives
2.2	HP prescribed laxatives	Diet. Fibre. *(Adequate fibre. Recommend including foods with a high fibre content (such as fruit, vegetables, high-fibre bread, baked beans and wholegrain breakfast cereals) (not applicable to exclusively breastfed infants). Do not recommend unprocessed bran, which can cause bloating and flatulence and reduce the absorption of micronutrients. 1.5.3)*	Laxatives
2.3	HP prescribed laxatives	Diet. Probiotics.	Laxatives

2.4	HP prescribed laxatives	Diet. Prebiotics.	Laxatives
2.5	HP prescribed laxatives	Fluid *(see Table 5. 1.5.3)*	Laxatives
2.6	HP prescribed laxatives	Exercise *(Advise daily physical activity that is tailored to the child or young person's stage of development and individual ability as part of ongoing maintenance in children and young people with idiopathic constipation. 1.5.6)*	Laxatives
3	**Information**		
3.1	HP prescribed laxatives	Educational leaflets / Lifestyle advice *(Provide children and young people with idiopathic constipation and their families with written information about diet and fluid intake. 1.5.4)*	Laxatives
3.2	HP prescribed laxatives	Parental training and advice *(Tailored follow-up. Could include:* ● *telephoning or face-to-face talks* ● *giving detailed evidence-based information about their condition and its management, using, for example, NICE's information for the public for this guideline* ● *giving verbal information supported by (but not replaced by) written or website information in several formats about how the bowels work, symptoms that might indicate a serious underlying problem, how to take their medication, what to expect when taking laxatives, how to poo, origins of constipation, criteria to recognise risk situations for relapse (such as worsening of any symptoms, soiling etc.) and the importance of continuing treatment until advised otherwise by the healthcare professional. 1.8.1)*	Laxatives
3.3	HP prescribed laxatives	Information. Wider children’s workforce *(point of contact for support 1.8.2)* *(school nurses raise awareness with young people and school staff. 1.8.3)*	Laxatives
3.4	HP prescribed laxatives	Information. Peer support.	Laxatives

E. What is the effectiveness of interventions delivery by continence teams (secondary care – specialist – services)?

	What is the effect of		**Compared to**
	INTERVENTION 1	**+ INTERVENTION 2**	**INTERVENTION 3**
1.	**Neuromodulation**		
1.1	Interventions delivered by wider children's workforce (see above)	Transcutaneous electrical stimulation	Interventions delivered by wider children's workforce (see above)
1.2	Interventions delivered by wider children's workforce (see above)	Sacral modulation	Interventions delivered by wider children's workforce (see above)
1.3	Interventions delivered by wider children's workforce (see above)	Tibial nerve stimulation	Interventions delivered by wider children's workforce (see above)
2.	**Feedback**		
2.1	Interventions delivered by wider children's workforce (see above)	EMG biofeedback	Interventions delivered by wider children's workforce (see above)
2.2	Interventions delivered by wider children's workforce (see above)	Biofeedback at home	Interventions delivered by wider children's workforce (see above)
2.3	Interventions delivered by wider children's workforce	Biofeedback – video games controlled by external sphincter activity	Interventions delivered by wider children's workforce

	(see above)		(see above)
2.4	Interventions delivered by wider children's workforce (see above)	Manometry	Interventions delivered by wider children's workforce (see above)
2.5	Interventions delivered by wider children's workforce (see above)	External anal sphincter EMG biofeedback	Interventions delivered by wider children's workforce (see above)
3.	**Equipment**		
3.1	Interventions delivered by wider children's workforce (see above)	Continence containment products	Interventions delivered by wider children's workforce (see above)
3.2	Interventions delivered by wider children's workforce (see above)	Toilet posture equipment	Interventions delivered by wider children's workforce (see above)
3.3	Interventions delivered by wider children's workforce (see above)	Other equipment	Interventions delivered by wider children's workforce (see above)
4.	**Irrigation**		
4.1	Interventions delivered by wider children's workforce (see above)	Colonic irrigation	Interventions delivered by wider children's workforce (see above)
4.2	Interventions delivered by wider	Antegrade continence enemas (ACE)	Interventions delivered by wider

	children’s workforce (see above)		children’s workforce (see above)
4.3	Interventions delivered by wider children’s workforce (see above)	Malone anterograde continence enema (MACE)	Interventions delivered by wider children’s workforce (see above)

F. What is the effectiveness of interventions delivered by consultant-led teams (tertiary care services)?

	What is the effect of		**Compared to**
	INTERVENTION 1	**+ INTERVENTION 2**	**INTERVENTION 3**
1.	**Surgical interventions**		
1.1	Interventions delivered by wider children’s workforce + continence teams (see above)	Anorectal myectomy	Interventions delivered by wider children’s workforce + continence teams (see above)
1.2	Interventions delivered by wider children’s workforce + continence teams (see above)	Anal & pelvic floor interventions	Interventions delivered by wider children’s workforce + continence teams (see above)
1.3	Interventions delivered by wider children’s workforce + continence teams (see above)	Colon resection with anastomosis & rectal operations	Interventions delivered by wider children’s workforce + continence teams (see above)
1.4	Interventions delivered by wider children’s workforce + continence teams (see above)	Operations that provide antegrade colon irrigation (ACE)	Interventions delivered by wider children’s workforce + continence teams (see above)
1.	Interventions delivered by wider children’s	Permanent or long term stoma	Interventions delivered by wider children’s workforce

5	workforce + continence teams (see above)		+ continence teams (see above)
1.6	Interventions delivered by wider children's workforce + continence teams (see above)	Manual evacuation	Interventions delivered by wider children's workforce + continence teams (see above)
1.7	Interventions delivered by wider children's workforce + continence teams (see above)	Rectal biopsy	Interventions delivered by wider children's workforce + continence teams (see above)

Report of Activity 2

ACTIVITY: Activity 2 Agree most important outcomes for the child, parents/carers/caregivers and health professionals, to inform the systematic review of effectiveness.	
1. Aim	
Task aim	To decide on the most important CFC outcomes which will guide the focus of the effectiveness review.
2. Methods	
Who was involved?	PPI group (lived experience and parents): Karen Jankulak, Margaret Ogden, Deb Smith, Clare Milligan. Health professional and charity group: Jonathan Sutcliffe, Brenda Cheer, Davina Richardson. Research group: Doreen McClurg, Lorna Booth, Alex Todhunter-Brown, Pauline Campbell, Andy Elders.
When was the involvement?	March 2020.
What happened?	Activities included email correspondence and an online meeting. Key activities / decisions involved: 1) The SG were sent a list of prioritised outcomes that had previously been identified in the literature (in a core outcome project). The SG voted to indicate agreement with whether “The 8 outcomes from the core outcome set project should be the outcomes considered for the systematic review of effectiveness” or whether they wanted to create their own list. The consensus was that there was that the list should be used. 2) The SG were asked to prioritise the list of outcomes from most important to least important and send these ratings to the research team. These ratings were then compiled to give an overall score. (see Table in Results). 3) The overall scores and prioritisations were then discussed and agreed at an online SG meeting.
Level of	It was the intent to allow the SG to make the overall decisions regarding

involvement?	important outcomes. The feedback about the meeting from the SG members indicate that they felt they had an “influencing” and “contributing” role at the meeting.
3. Results	
Outcomes—Report the results of SG in the study, including both positive and negative outcomes	1) Voting on whether the core outcome set should be used: consensus that this should be used (7 respondents; 6 agreed, 1 neither agreed or disagreed). 2) Results of ranking of importance of outcomes. (Ranks - 1 (most important) to 8 (least important)):

Outcomes	SG1	SG2	SG3	SG4	SG5	SG6	SG7	**Total***
►► Defecation frequency	2	7	3	1	4	6	7	**30**
►► Stool consistency	7	8	5	2	5	3	8	**38**
►► Painful defecation	3	2	1	4	1	1	5	**17**
►► Quality of life of parents and patients	1	1	4	3	7	2	2	**20**
►► Side effects of treatment	6	2	8	5	6	4	6	**37**
►► Faecal incontinence, if age appropriate	4	2	2	7	3	7	1	**26**
►► Abdominal pain, if age appropriate	5	5	7	6	2	5	3	**33**
►► School attendance, if	8	6	6	8	8	8	4	**48**

age appropriate								

SG = stakeholder group member/respondent. *Total = sum of individual rankings. The LOWER the total score the greatest the shared importance.

3) The SG considered the total scores, and the individual rankings (anonymised), and reached consensus that the systematic review of effectiveness should have:

TWO primary outcomes (considered of equal importance):

- *Painful Defecation*
- *Quality of life of parents/carers/caregivers and patients*

SIX secondary outcomes (considered of equal importance):

- *Defecation frequency*
- *Stool consistency*
- *Side effects of treatment*
- *Faecal incontinence, if age appropriate*
- *Abdominal pain, if age appropriate*
- *School attendance, if age appropriate*

4. Discussion & conclusions

Outcomes— Comment on the extent to which SG influenced the study overall. Describe positive and negative effects	The agreed prioritised list of outcomes decided by the SG guided the information extracted for the effectiveness review.

5. Reflections / critical perspective

Comment critically on the study, reflecting on the things that went well and those that did not, so others can learn from this experience	***Research Group reflections:*** The SG were provided the opportunity to contribute to the decisions made about prioritised outcomes. The researchers did not get involved in the decisions made other than provide a list previously identified in the literature. A challenge when synthesising evidence of effectiveness was that a common outcome reported in studies is "treatment success". Author definitions of this vary, and it is difficult to be certain how this outcome relates to our list of outcomes. Had we done this exercise again it might have been useful to discuss with the stakeholders what they thought about using an outcome of "treatment success". ***SG view:*** *"the list of prioritised outcomes very much reflects what is important to me"* *"I agree—these outcomes are entirely in keeping with my child's experiences"*

Report of Activity 3

ACTIVITY: Activity 3: Develop a logic model which describes the effect that interventions and intervention combinations have on important outcomes, and key factors relating to implementation.	
1. Aim	
Task aim	Develop a logic model which describes the effect that interventions and intervention combinations have on important outcomes, and key factors relating to implementation.
2. Methods	
Who was involved?	All members of the stakeholder group were involved in the iterative process of developing the logic model. People attending meetings at which the model was specifically discussed were: 09-01-2021 - Attendees: Doreen McClurg, Lorna Booth, Pauline Campbell, Alex Pollock, Andy Elders, Julie Cowie, Margaret Ogden, Brenda Cheer, Davina Richardson, Gemma Kierczuk, Suzanne Hagen, Helen Haywood 25-01-2022 - Attendees: Doreen McClurg, Lorna Booth, Pauline Campbell, Alex Todhunter-Brown, Margaret Ogden, Davina Richardson, Deb Smith, Jonathan Sutcliffe, Clare Millington, Karen Jankulak. 15-03-2022 - Attendees: Alex Todhunter-Brown, Margaret Ogden, Deb Smith, Jonathan Sutcliffe, Karen Jankulak, Brenda Cheer, Lorna Booth, Pauline Campbell. 20-04-2022- Attendees: Alex Todhunter-Brown, Margaret Ogden, Deb

	Smith, Karen Jankulak, Jonathan Sutcliffe, Clair Torrens, Brenda Cheer 24-05-2-22- Attendees: Alex Todhunter-Brown, Margaret Ogden, Karen Jankulak, Davina Richardson, Pauline Campbell, Jonathan Sutcliffe,
When was the involvement?	An initial (draft) logic model had been included in the funding application (See Logic model v1 – below). Stakeholders had had an opportunity to comment (by email) on this version. Further developments to the logic model occurred iteratively throughout the project. The iterative development of the logic model was closely linked to the development of the 'Pyramid', which was first drafted at the start of the project as part of Activity 1a, and continued to be refined throughout the project. (see Supplementary File Activity 1a) The logic model was explicitly discussed in meetings on: ● 9th February 2021 ● 25th January 2022 ● 15th March 2022 ● 20th April 2022 ● 24th May 2022
What happened?	All discussions were held via online Zoom meetings, with the logic model being one item on a larger meeting agenda. 9th February 2021 – the research team updated the initial draft logic model from the funding application to incorporate the Pyramid (see Logic model v2 – below). During the meeting, this updated logic model was presented. The group was asked for feedback, and also discussed if they thought whether it would be useful to have a logic model for each of the reviews and a final "umbrella" logic model. The group agreed that logic models are a useful way of presenting information as an adjunct to the final report and would prefer separate models for each review. A

	concern was raised regarding the terminology used within logic model and that parents may not understand their meaning. 25th January 2022 – Logic model v2 was presented again. The group discussed the fact that this model was "linear", and that a linear model failed to capture the complexity associated with treatments for constipation. The group suggested that "a traditional logic model may just not be possible there is not a single linear pathway". The new MRC framework for developing and evaluating complex interventions (doi: https://doi.org/10.1136/bmj.n2061) was discussed, and it was proposed that this may be useful for developing our logic model further. It was noted that the new MRC framework recognised the importance of context and implementation. The stakeholders asked the research team to come up with a new model which might reflect the complexity. February – March 2022. The research team initially built on the previous version of the Logic model (v2), drafting out Logic model v3 (see below). However, it was agreed that continuing in this 'linear' format was not addressing the comments of the stakeholders. The idea of reflecting the concept of the 'Pyramid' within a circular, "dart board" figure was raised, and the decision made to draft something based on this concept to share with and discuss with stakeholders. 15th March 2022 – A presentation was given, proposing the 'new' model as a circular model (Logic model v4 – below). This was discussed in detail. Key points included: ● There was agreement that this circular model was much more effective at demonstrating the complexity relating to treatments for constipation. There was positive feedback about positioning the child in the centre of the model: *"I think the concept is really good. I like the child at the centre".* ● Specific feedback about wording and layout was provided, e.g. *"what's left of the triangle [pyramid] seems to have been buried*

	in the detail of the rest of it….could the font be bigger on the former triangle as it is getting lost….I liked the numbers" and *"I appreciate that we want to get away from the triangle and the linear stuff but I want to have a little of that back……and put the numbers back in as it's our organising factor for a lot of this"*. ● There was detailed discussion on use of wording – e.g. use of Micro, Meso, Macro – some people were unfamiliar with these terms, while others highlighted that these were commonly used in academia. Proposal to replace the words Micro, Meso, Micro with "personal factors" ● It was proposed that it should be presented 'dynamically' – as a series of small figures building to the final version: *"series of small figures building up to this one"…… "making it simpler for people to understand…..doing it a bit at a time….do one bit and have your explanation and then build it up"* 20th April 2022 – prior to the meeting a narrated PowerPoint was circulated, providing a brief description of the "build up" of the new model (Logic model v5 – below). A word document version describing the 'build-up' of the model was also circulated (Logic model (build up) v6 – below). Key points from discussion were: ● "*the generated PowerPoint was a really neat way of bringing together all our thoughts….as a visual model it's got anything there, but it's very very busy, so for anyone who hasn't seen it before where do you start…but the PowerPoint builds it up*" ● "*I did find it easy to understand, I watched it and found it quite clear*" 24th May 2022 – prior to the meeting a draft of the NIHR Final Report chapter in which the Logic Model is presented was circulated. Stakeholders had submitted written comments on this draft. During the meeting the comments were discussed and actions agreed. There was lengthy discussion around the inclusion of the "outcomes" column within the model, and whether there should also be a "problems" column to the left. The "outcomes" that were represented on the model were those that had been prioritised by the SG, but these were priorities for outcomes to synthesise from research – not clinical priorities. Further, concerns were

	raised about the complexity of the logic model. The group concluded that there were too many "unknowns" to be able to complete a final logic model which encompassed all aspects of management of CFC, but that there was value in having the "central" part of the model as a representation of the complexity of current CFC management. Some key points raised relating to concerns about the model and its limitations include: • "*I know that outcomes are usually part of a logic model but I wonder if there needs to be some more integrated work with families"* • *"An observation would be that if you were coming into this cold and looking at this it's got to be not off-putting for somebody looking at it and in order to inform how its presented, I wonder whether or not we need to trial or test it with some different people"* • *"there are gaps in terms of knowing what the key outcomes are, we have a really complicated system, there is a massive evidence gap……we don't know…there are loads of gaps still….loads of questions…"* • *"this is not a logic model…..a logic model is a flow diagram…..we've agreed that it's not possible to represent this as a logic model….so removing the outcomes fits with that argument"*
Level of involvement?	The involvement of stakeholders has been 'controlling' the development of the logic model.
3. Results	
Outcomes—Report the results of SG in the study, including both	A final version of a logic model was developed through an iterative process. There was agreement that the word version, showing the 'build-up' should be integrated into the final report chapter - *"I thought it looked fantastic and I really liked the circular model with the child in the centre"*

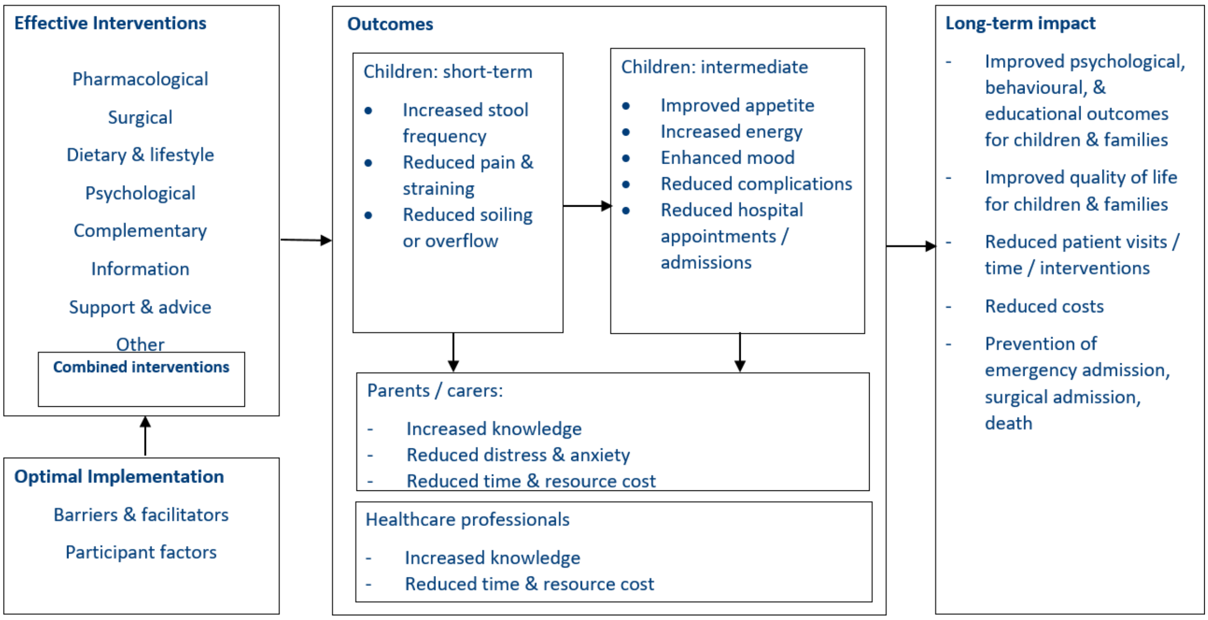
positive and negative outcomes	- *“the word version of the text was also very clear”* However, there were concerns about the complexity of the model, but uncertainty about the best way to deal with this: - *“someone who is not as invested and doesn’t know where it has come from and, let’s face it….they may look at the final model and go ‘cripes’….and that might make them disengage.”* - *“We maybe need a sanitised final model, explicitly trying to make it more palatable for the user - maybe need to ask for feedback from additional stakeholders to find out if they understand it, coming from it fresh”* - *Maybe we should “make a simplified version with the central and key parts……but everything on there is important so it’s difficult to know what to keep”* - *“We need our research to be consistent so we can pull things together and I think that that is one of the key things that I would like to have come out of this…..this is what we think is happening clinically, this is what we believe is important to families, this is what we believe may be important to clinicians…..but how do we pull all this together…? What we need to do is to identify where there are gaps. …..How we do that with the model I don’t know….”* - *“I am new to the concept of a logic model but it strikes me that if you put too many things in you are trying to simplify it so that it’s in one graphic representation….it can’t easily be summed up in this way….you don’t want to get carried away with the idea of a logic model encapsulating everything, because it won’t”* - - In response to these concerns it was agreed that it was important to continue to get feedback on the model, and to continue to refine it in response to feedback. - *“We should aim to engage with the audience prior to agreeing a final version”* - Stakeholders also raised a further important point about the limitations of the model: ● *“if the logic model is meant to represent in a visual format all our assumptions and how things feed into each other, underlying the entire logic model is the assumption that these are cases that are identified in the first place…….in explanations of that it might be good to make it very explicit briefly that these are cases that are*

	either identified or potentially identified and there's a whole other tranche of things that isn't even on this model YET because it's not yet identified……our issues with constipation, especially combined with learning disabilities, have mostly been on the 'trying to get onto the logic model pathway' rather than what happens once we are there". It was agreed that this was an important point which should be made within the chapter where the logic model is presented.
4. Discussion & conclusions	
Outcomes—Comment on the extent to which SG influenced the study overall. Describe positive and negative effects	The stakeholders had a substantial influence on the logic model which has been produced, leading a move away from the 'linear' (more traditional) logic model, to a circular model which illustrates the complexity of treatments for constipation. The positive effects of this is that the new model is considered to successfully represent the complexity in this field. The negative effects are that the model is highly complex and this may make it difficult for audiences to understand and engage with. Some solutions to this were proposed (use of narrated presentations, use of a version where the model is 'built up'), but a possible need for further refinement was acknowledged. In general, stakeholders have had control over the final model and what was produced. However, at some stages in the process, stakeholders were reacting to versions of the logic model produced by researchers meaning that, rather than there being true co-production, they were influencing researcher-led versions. But a level of control remained, as stakeholders would have been able to reject the versions produced by the researchers.
5. Reflections / critical perspective	

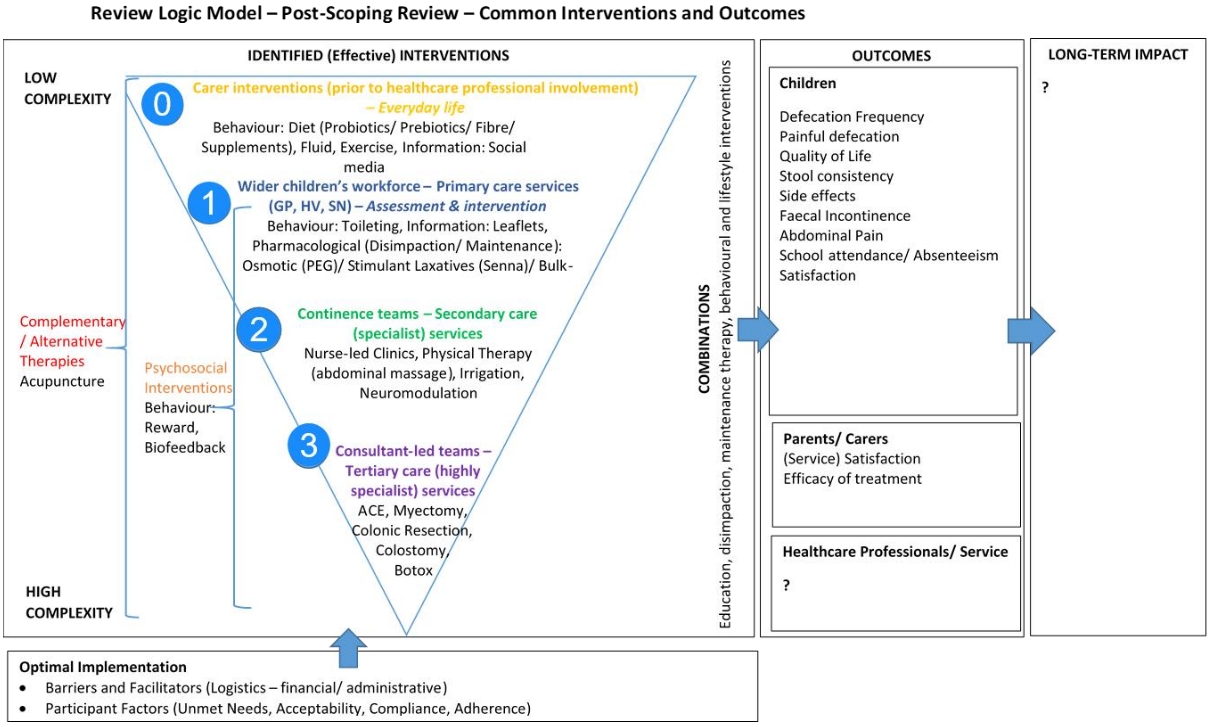
Comment critically on the study, reflecting on the things that went well and those that did not, so others can learn from this experience	**Stakeholder reflections:** • *"I think as stakeholders we had quite a lot of control over this but yes we were often reacting to what the researchers produced…."* • *"As a non-specialist stakeholder, I felt there was as reasonable as possible balance between reacting and co-producing. If we had been able to keep to our original plan of having more in-person meetings it might have simplified the process, but I suspect it would not at all have changed the ultimate result. Having the face-to-face meeting at the very beginning of this process was extremely helpful in many different aspects, and it set a positive tone for subsequent online meetings."* Sometimes there were practical difficulties in working on the logic model during an online meeting: *"Had to squint to see some of the text on the logic model (but managed to read it)"* "*it was not easy doing it online"* At the meeting on 09-01-2021 only one of the 4 PPI members was able to attend, meaning that there was only one PPI member, with a number of clinicians and researchers. This created an imbalance in the group and was not ideal: *"I missed the 3 PPI members today"* At the meeting on 25-01-22 there was not sufficient time to discuss the logic model adequately: *"A bit more time on the logic model"* Overall, the logic model was perceived as complex to develop and beyond the scope of the SG to conclude this: • *"we have said as a group what we think, but we are – with all due respect –we are a stakeholder group with a good range of perspective, but we are still a small group……."* • *"there is a huge amount more work to get this right. We need to know the problems and the outcomes, but it's beyond what we can do. This is one of our priorities for future research. We need to look at outcome measure and what matters to children and families"* • *"The logic model has been the most challenging for me to digest. However the research team have done their best to put explanations in to clear understandable English."* **Researcher reflections:** *"This model is complex, and further time and input is really required to*

	enhance the accessibility; but it does reflect the complexity which our stakeholders have been telling us about." *"The stakeholders had great ideas about how to develop and present the model, and the research team possibly did not have the skills or resources to bring them to life. A lesson to me is that, next time, we should apply for funding to bring in someone with creative expertise in order to get the most out of something like this".*

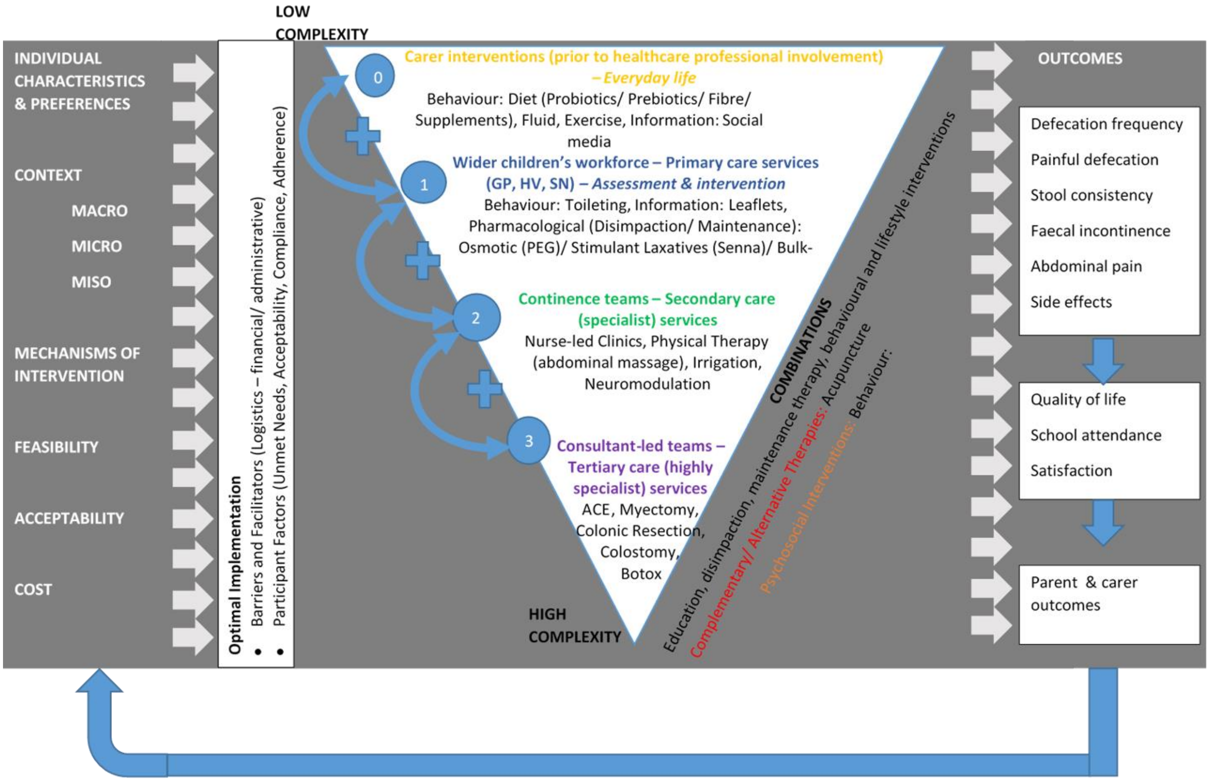
Logic model v1



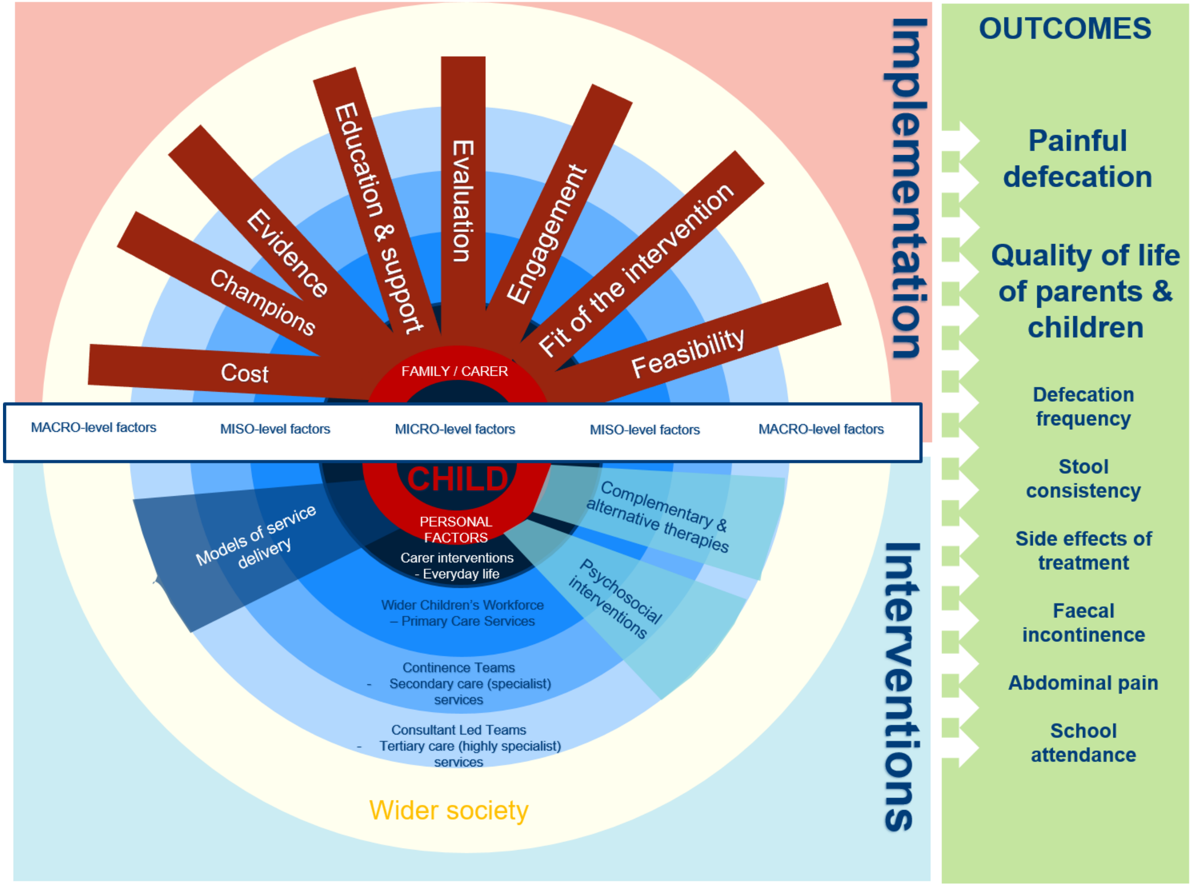
Logic model v2



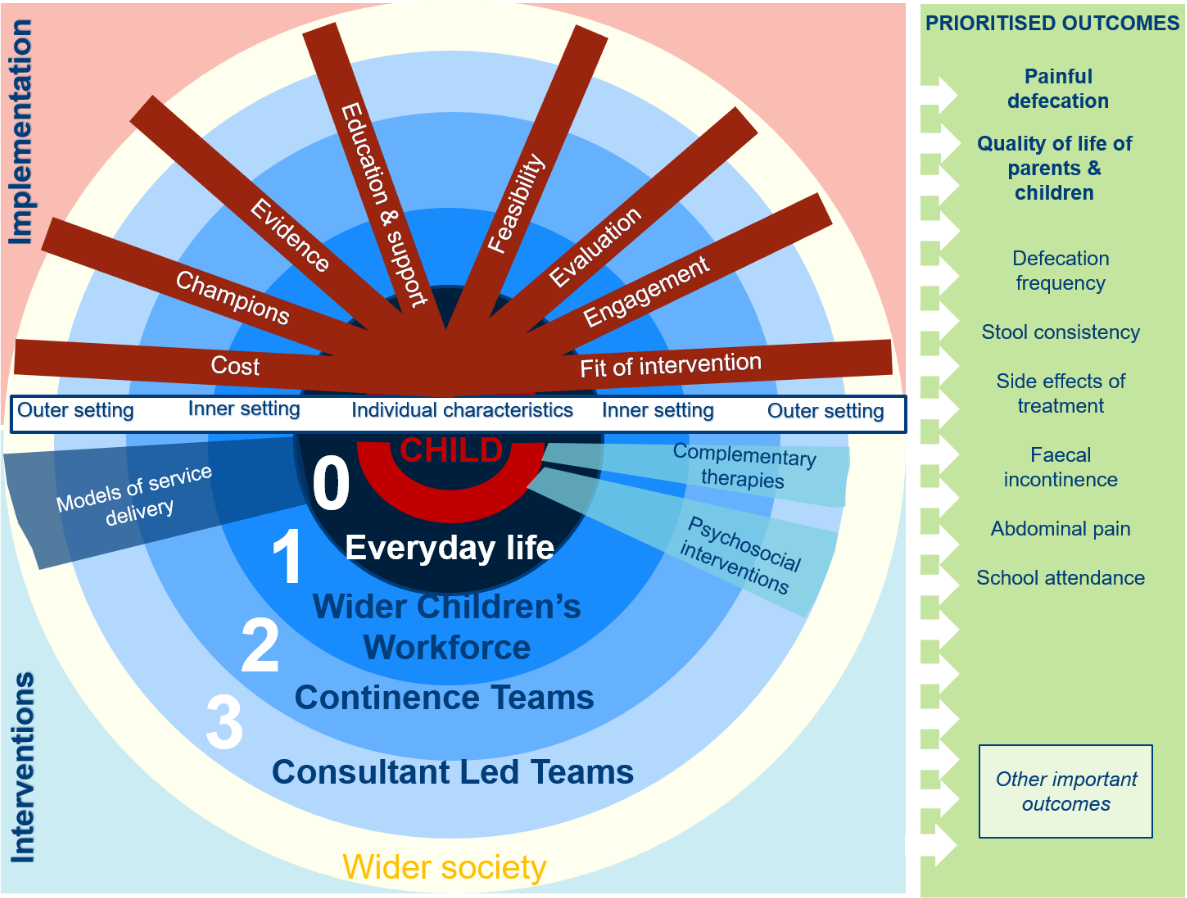
Logic model v3



Logic model v4

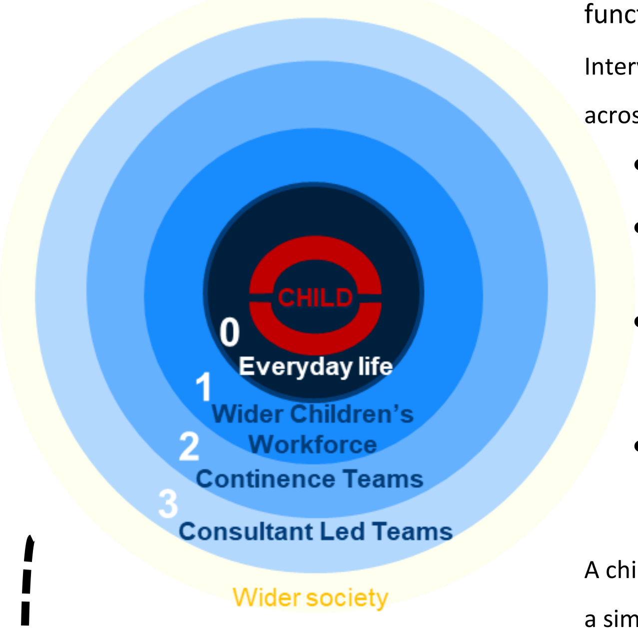


Logic model v5



A

Logic model v6 (‘build up’)



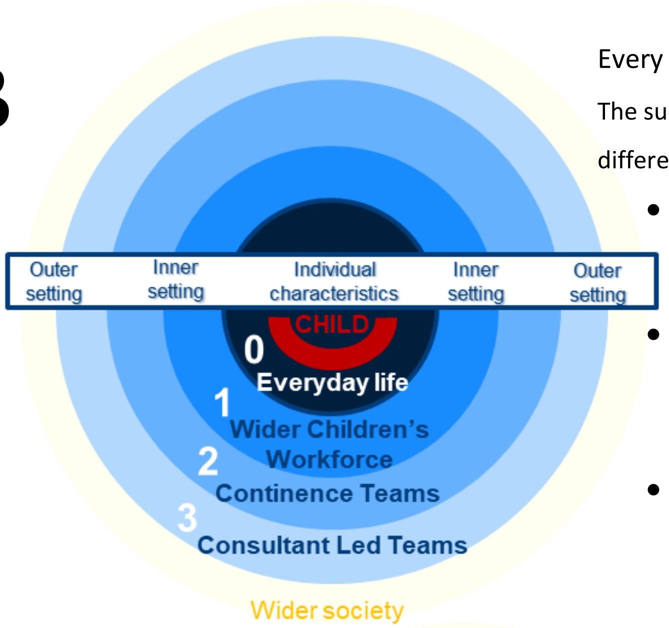
The child is at the centre of our model of management for childhood functional constipation (CFC).

Interventions for a child with CFC are delivered in a step-wise, or cumulative, way, across a number of different ‘levels’:

- **Level 0 – Everyday life**: interventions are delivered by carers, prior to healthcare professional involvement.
- **Level 1 – Wider children’s workforce**: in addition to any Level 0 interventions, children may be assessed and given interventions by primary care services (e.g. General practitioner, health visitor, school nurse).
- **Level 2 – Continence teams**: if CFC remains a problem, children may be referred to and given interventions by specialist secondary care services (e.g. nurse-led clinics, physical therapy, irrigation, neuromodulation). Interventions will be given in addition to Level 0 and 1 interventions.
- **Level 3 – Consultant-led teams**: if CFC remains a problem, children may be referred to and given interventions by highly specialist tertiary care services (e.g. surgery). Interventions will be given in addition to Level 0, 1 and 2 interventions.

A child’s journey through these Levels of interventions will be unique, and may not be a simple journey of 0 to 1 to 2 to 3, but may involve steps ‘down’ as well as ‘up’.

B

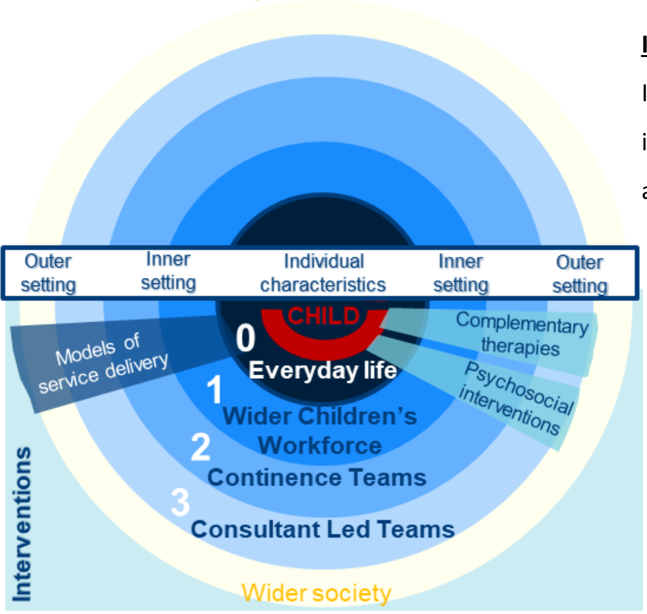


Every child is unique.

The success of CFC strategies will be affected by a number of different factors:

- **Individual characteristics** – the personal characteristics of that child, including personal, physical, physiological, genetic and behavioural factors, and individual experiences. (May also be described as *micro-level*).
- **Inner setting** – the characteristics of the community and organisation in which interventions are delivered, including the characteristics of their family, friends, school, healthcare system and society in which they live. (May also be described as *meso-level*).
- **Outer setting** – external context or environmental, including wider healthcare and social environment, including policies and strategies which can impact on the delivery of strategies for CFC. (May also be described as *macro-level*).

C

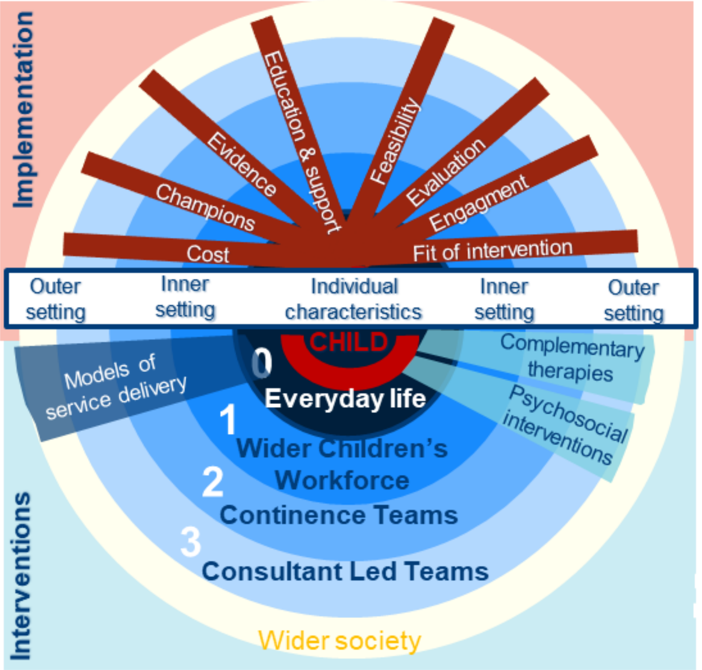


Interventions for CFC

In addition to the step-wise delivery of interventions at Level 0, 1, 2 and 3, the following are important ‘across’ all the Levels:

- **Models of service delivery** – i.e. service provision and how care is provided are important across all Levels. This could include the availability and accessibility of professional input at different levels, or communication and patterns of referral between levels.
- **Complementary therapies** and alternative therapies may be delivered to the child within any of the Levels.
- **Psychosocial interventions**, or behavioural interventions may be delivered to the child within any of the Levels.

D

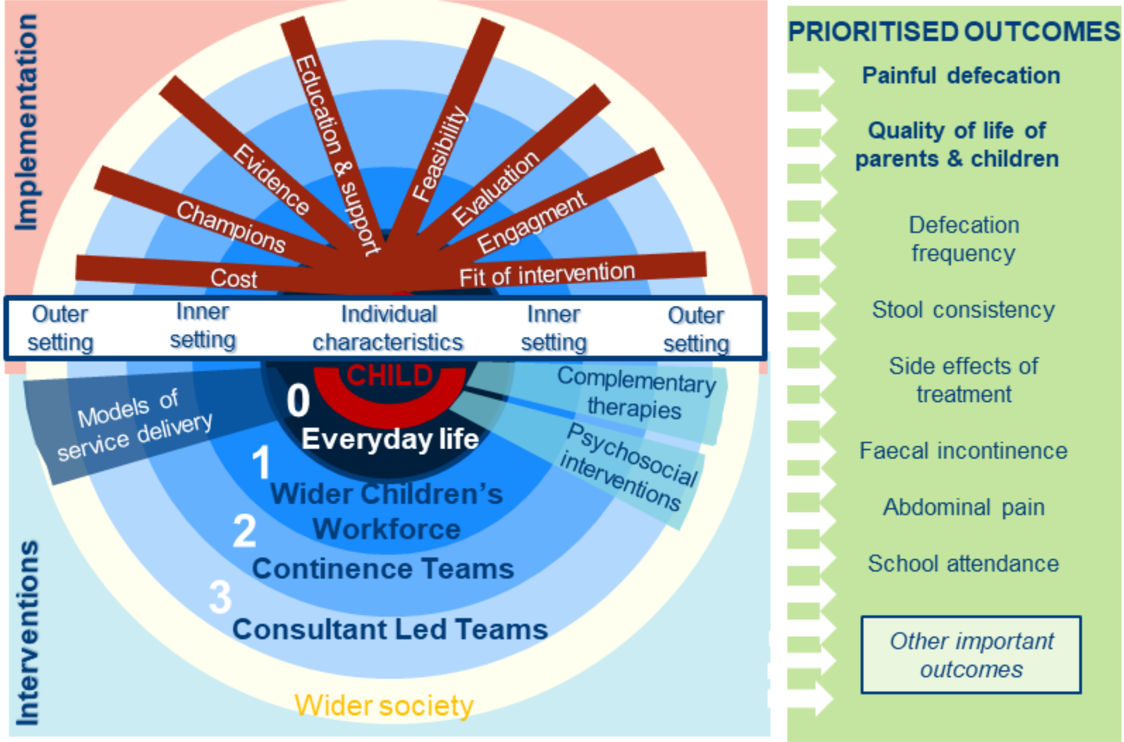


Implementation factors

Many factors affect implementation of interventions. These include:

- **Evidence** - the evidence-base (or lack of) behind the "successfulness" of the intervention
- **Fit of intervention** - whether the intervention was adaptable, flexible and offered an advantage over an alternative solution
- **Need for change** - understanding the tension for change (i.e. why clinicians and families felt that the changes were needed now)
- **Champions** - engaging champions to support children and young people was described as an important factor to enable successful implementation
- **Understanding** - A lack of understanding of what children, young people and their families need was a major obstacle to implementation
- **Addressing taboos** - The taboo nature of constipation and the reluctance of children, parents, healthcare professionals and wider society to openly engage in discussion about constipation was identified as a major obstacle to implementation
- **Self-efficacy** – self-efficacy was reported as a key component to the success of the implementation coupled with individual knowledge and beliefs were important facilitators

E: Logic model v5



Outcomes

Implementation of interventions will impact on outcomes.

Building on work to establish a core outcome set for CFC, stakeholders identified a number of top priority outcomes.

Other important outcomes

incorporate perspectives relating to healthcare organisations, as represented by the Balanced Scorecard:

- Patient perspectives, such as experience and satisfaction.
- Internal perspectives, such as processes, care delivery and clinical outcomes.
- Financial perspectives, including performance of healthcare organisation
- Learning and growth perspectives, including innovation.

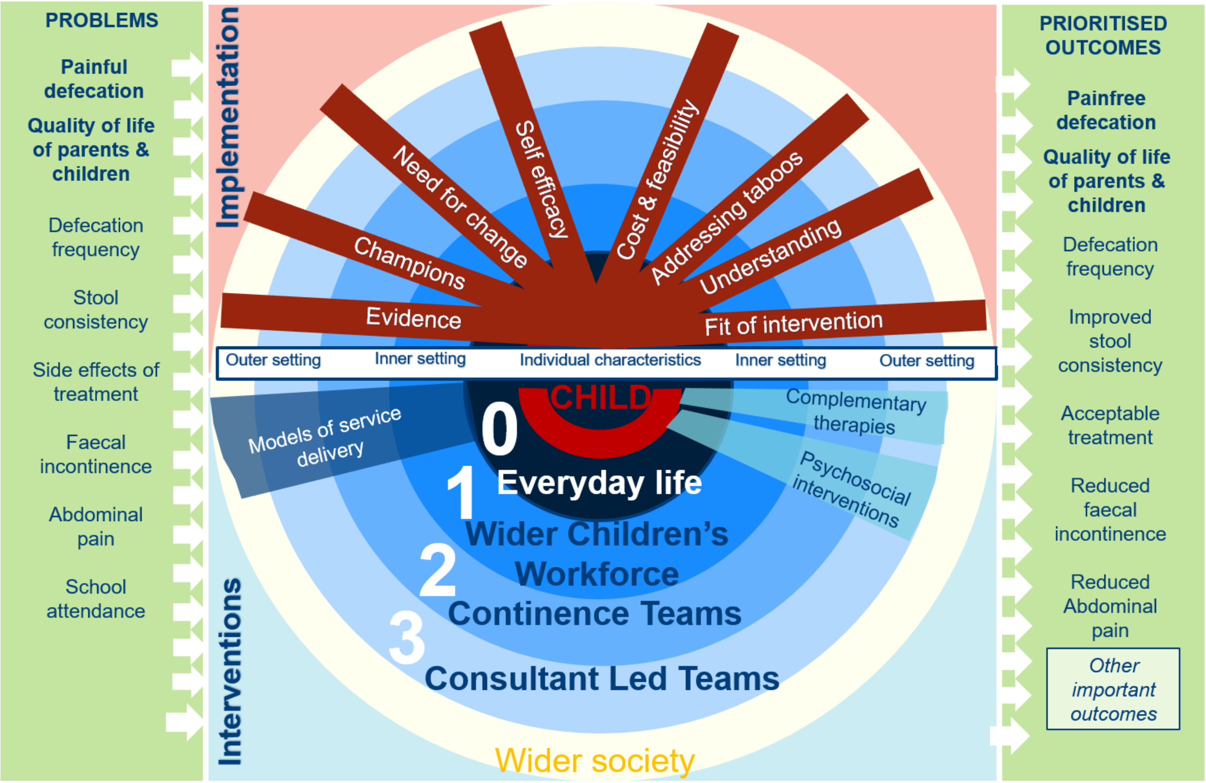
Logic model v7

Changes from v6:
The factors affecting implementation (in the red bars) were updated, following the results of the systematic review of evidence relating to implementation. These include:

- Evidence - the evidence-base (or lack of) behind the "successfulness" of the intervention
- Fit of intervention - whether the intervention was adaptable, flexible and offered an advantage over an alternative solution
- Need for change - understanding the tension for change (i.e. why clinicians and families felt that the changes were needed now)
- Champions - engaging champions to support children and young people was described as an important factor to enable successful implementation
- Understanding - A lack of understanding of what children, young people and their families need was a major obstacle to implementation
- Addressing taboos - The taboo nature of constipation and the reluctance of children, parents, healthcare professionals and wider society to openly engage in discussion about constipation was identified as a major obstacle to implementation
- Self-efficacy – self-efficacy was reported as a key component to the success of the implementation coupled with individual knowledge and beliefs were important facilitators

Heading relating to outcomes amended to reflect that these are outcomes relating to research.

Logic Model v8

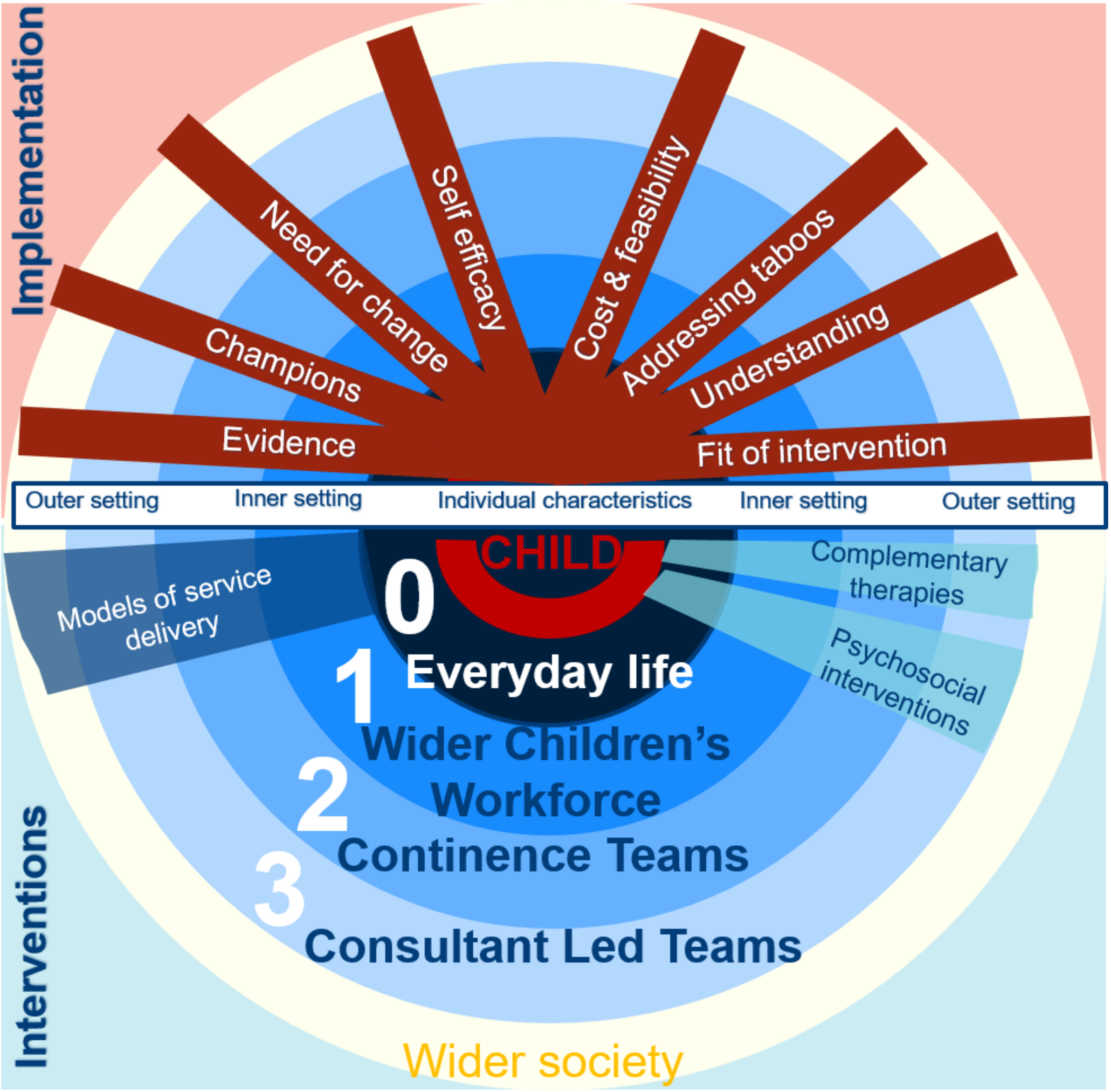


Changes from v7:

The ‘problems’ have been included.

Discussion on this version of the logic model led to the decision that the final version reported in the SUCCESS project should be simplified, and only comprise the central section (see Logic Model v9).

Logic Model v9



Report of Activity 4a

ACTIVITY: Activity 4: Reach consensus over clinical implications and guide knowledge translation activities. **Activity 4a: Reach consensus over clinical implications**	
1. Aim	
Task aim	To reach consensus over the implications arising from the results of the systematic reviews of evidence.
2. Methods	
Who was involved?	All members of the stakeholder group had opportunities to contribute to generating clinical implications arising from the reviews. People attending meetings at which implications arising from the results of the specifically discussed were: 01-12-2020 - Attendees: Doreen McClurg, Lorna Booth, Pauline Campbell, Alex Pollock, Andy Elders, Julie Cowie, Debs Smith, Clare Millington, Jonathan Sutcliffe, June Rogers, Margaret Ogden, Brenda Cheer, Tracey Barber. 08-03-2021 Attendees: Doreen McClurg, Lorna Booth, Pauline Campbell, Alex Pollock, Andy Elders, Julie Cowie, Margaret Ogden, Brenda Cheer, Davina Richardson, Gemma Kierczuk, Debs Smith, Jonathan Sutcliffe 29-06-2021 Attendees: Doreen McClurg, Pauline Campbell, Margaret Ogden, Davina Richardson, Suzanne Hagen, Andy Elders, Karen Jankulak

	13-09-2021 Attendees: Doreen McClurg, Pauline Campbell, Lorna Booth, Margaret Ogden, Debs Smith, Clare Millington, Karen Jankulak, Brenda Cheer 16-11-2021 Attendees: Pauline Campbell, Margaret Ogden, Debs Smith, Clare Millington, Doreen McClurg 30-11-2021 – Attendees: Pauline Campbell, Debs Smith, Margaret Ogden, Davina Richardson, Brenda Cheer, Doreen McClurg
When was the involvement?	Involvement occurred through a number of online meetings, at which implications relating to different evidence syntheses were discussed: - 01-12-2020 – Service delivery evidence synthesis - 08-03-2021 – Level 0 and Level 1 evidence synthesis - 29-06-2021 – Level 0 and Level 1 evidence synthesis - 13-09-2021 – Scoping review maps; Economic evaluation synthesis - 16-11-2021 – Level 3 evidence synthesis - 30-11-2021 – Level 2 evidence synthesis Involvement was also gained from reading and commenting on drafts of the chapters for the final report.
What happened?	Six meetings were held (online) at which a presentation of the draft results of one of the evidence syntheses were provided, and then meeting participants asked to discuss what they felt the clinical implications were: 01-12-2020 - the research team presented the findings from the care provision review. 7 questions were identified. Research team asked the SG members to think about implications of the findings for each question

	in regards to clinical practice and the patient perspective. Research team also asked RG members if they could identify any gaps from the findings (i.e. issues that were deemed important but were not covered within the 7 questions). 08-03-2021 – The research team provided an update on the reviews and explained that three identified systematic reviews were assessed as having low risk of bias, covering 3 interventions, probiotics, fibre and laxatives. These reviews will be updates as per the protocol. The SG were asked where they thought these interventions sit in the pyramid, level 0 or level 1. Draft results from Level 0 were discussed and implications and research gaps proposed by stakeholders. 29-06-2021 – Draft results from the Level 0 and Level 1 syntheses were presented and discussed. 13-09-2021 – Draft results and an example of the interactive map for the economic evaluation was presented. These were discussed. 16-11-2021 – A presentation was given summarising the findings from the Level 3 synthesis. Stakeholders discussed the implications of these findings. 30-11-2021 - A presentation was given summarising the findings from the Level 2 synthesis. Stakeholders discussed the implications of these findings. There were technical issues with very intermittent connectivity at times from the GCU end limiting discussion during this meeting. During the write up of the final report – all stakeholders had

	opportunities to read and comment on the evidence syntheses, and the implications and research gaps arising from these.
Level of involvement?	The stakeholders considered that their level of involvement was ‘influencing’.
3. Results	
Outcomes—Report the results of SG in the study, including both positive and negative outcomes	The following points were raised during the discussions: Service delivery synthesis • Concern that the wording “there is currently insufficient evidence to support this” could be taken out of context especially by commissioners. It was suggested that although there were was not any high quality evidence of effectiveness, the implementation review should capture facilitators and benefits of different services and models and that the information would be pulled together for the report. It was agreed that alternative wording should still be used that works for everyone (researchers, clinicians, commissioners, patients and parents). • Concern over what we mean by primary care as this is often understood to be GPs. It was agreed that clearer definitions would have to be used such as wider children’s workforce. • Concern over the use of acronym such as ASD. It was agreed that acronyms would not be used in final report. • It was highlighted that one of the papers included in question 5 (‘What are the effects of specialist (level 2) services and models of care?) should come under level 3 tertiary care. It was agreed that the research team would revisit this to check as decisions had been made using consensus. • There were questions and discussion around the nature of different models of service provision which were delivered in the synthesised studies. It was agreed that a more comprehensive description of the interventions would help answer these questions and be beneficial to readers of the review. • Regarding question 4 (effectiveness of follow-up web-based information) the group agreed that it would depend on what the web-based information was. All agreed it would be beneficial if it was targeted by directing people to robust information and that a big advantage is that people can return to the information when required which helps to tackle health literacy issues. • Missed opportunities for early intervention was discussed. A suggestion for this would be to make it standard practice for health visitors to ask parents about their child’s toileting habits. • Diagnosis highlighted as a research gap. This was considered

really important by the group. Explanation of why diagnosis was not a focus of the studies is that research studies will have an inclusion criteria including “children with diagnosed CFC” and therefore these studies are missing the children that have already slipped through the net. This issue should be made clear in our final report. Also children self-diagnosing was identified as another gap.

- “What makes a good service/effective team?” was also highlighted as an important gap. All agreed that the important thing is being seen appropriately and being given the right advice.

Level 0 and 1 synthesis

- It was suggested that it would be difficult to put laxatives /probiotics /fibre under a particular level – laxatives should be prescribed for children meaning it would sit with level 1 but pharmacists can suggest laxatives for over 12 year olds. Probiotics and fibre are not prescribable but parents will make a choice based on the information given at level 1 so would come under level 0. It was suggested that probiotics bought over the counter are not necessarily as effective as the prescribed probiotics, therefore is concerned if probiotics are put into level 0.
- It was suggested that writing the report as group of treatments rather than per level would be better. Some believed the levels in the pyramid are artificial.
- It was suggested that although parents may use some of the Level 0 interventions, they will be most likely to be based on professional input and advice.
- cow’s milk-free formula is prescribable and would be better in level 1
- Possible gaps identified by the SG for level 0 included:
    - Breast Milk
    - Baby massage (this may come under complementary)
    - Mobile/exercise/standing
    - Rebound therapy (this may come under physiotherapy – level 2)
    - Gluten (this may be more diagnostic rather than treatment)
- In relation to implication of the findings, the following were noted.
    a. Education is very important and should be prioritised.
    b. Probiotics, although the updated Harris review did not demonstrate significant benefit the probiotics used were diverse. It would be important to ask professionals how they advise on the use of probiotics and if they were available on the NHS, or had been identified on any care pathways of children with chronic constipation e.g. dietician, GP, consultant.
    c. It would be important that interventions such as a

	cow’s milk free diet and adding sugars (brown, Molasses etc.) should only be considered in the wider context of the child’s health d. Physical exercise, and potentially Pelvic floor muscle exercises may be used by e.g. physiotherapists, massage therapists, and use in practice should be explored - Stakeholders expressed surprise that there were few studies in which behavioural interventions were combined with pharmacology and it was suggested that more evidence of behavioural interventions may be identified within the review of laxatives that is being updated - Gaps identified include behavioural interventions especially in children with e.g. autism - It was agreed that although there is overlap Level 0 and 1 should still be separate but where possible we will highlight which interventions can be delivered by parents / carers (i.e. which do not require a prescription). Level 2 synthesis - There were discussions around the use of enemas for children with functional constipation and when these should be used. The response was that this was primarily in secondary care after laxative use has failed and following appropriate counselling and work up with the child and parents. - Digital disimpaction was discussed – this is usually used as a last resort and under general anaesthetic. - Suppositories – no research found on the use of suppositories or the type that should be used. Stakeholders raised that these can be used at home. Stakeholders reported that Bisacodyl rather than glycerine in children was commonly used – this should be noted as a research gap - Stakeholder proposed that there was now some evidence for the use of transcutaneous electrical stimulation. - Although being used more often there was still very limited good quality evidence on the use of transanal irrigation – really only Peristeen. The lack of evidence was why NICE did not recommend it - no good evidence. It was suggested that a review should be considered (Mosiello 2017). - Biofeedback was discussed and although the evidence is limited it should be included at Level 3. It was noted that Centre of excellence is the Royal London who are using it diagnostically as well as an intervention. - It was reported that there were some physios who use biofeedback using a balloon and or pressure.

	- There was a discussion around semantics between treatments that work on anal sphincter dysfunction and if this was really constipation? - Physical Therapy/Physiotherapy – some evidence that as part of a package some physio may be of benefit however the physio included in the 2 studies included different modalities. Tends to be used more in Australia and USA though increasingly some pelvic physios are seeing paediatrics. - The question around the psychological effects of all this especially soiling was raised. There are some papers on this and will be discussed further when reviewing the overview on psychosocial interventions <u>Level 3 synthesis</u> - Discussions from the group mainly focussed on the long-term implications, type of follow-up and help provided and the journey path to get to this stage. One member recounted how an ACE procedure was recommended for her twins without much information being provided, after finding out more about the implications she felt it was not for her children with complex needs. - It was also discussed how some treatments were life changing e.g. one boy could no longer play rugby, it was difficult to go on sleep overs/trips away etc. - Feedback from the clinicians would be helpful with potentially some case studies.
4. Discussion & conclusions	
Outcomes—Comment on the extent to which SG influenced the study overall. Describe positive and negative effects	Some key points were raised through discussion which influenced the discussion around the implications arising from the results. However, these points were limited. We would have benefited from a more systematic, comprehensive method to collect clinical implications and research gaps. Gaining feedback from stakeholders on the final written versions of chapters (particularly the Discussion chapter) has been a key way of gaining wider input into the statement of implications arising from the

	findings of these evidence syntheses.
5. Reflections / critical perspective	
Comment critically on the study, reflecting on the things that went well and those that did not, so others can learn from this experience	Stakeholder reflections Stakeholders reported that it was difficult to know whether the feedback / comments that were being given were being taken on board. They commented that they couldn’t tell this until they saw the drafts of the chapters. However, at times the stakeholders recognised that their input was valued / useful: • “[*I played a useful role in] validating information as true from a parent perspective*” • “*I made several minor suggestions, which were I think thought to be helpful.*” Clinicians reported that they felt they provided useful information in relation to where different interventions fitted within the pyramid: • “*[I] contributed thoughts on where different interventions fit on the pyramid, and overlaps with this*”, • “*I made a few suggestions around how best to present things*.” Stakeholders reported that they found the evidence synthesis useful: • *“It has been useful to clarify the diversity of papers and heterogeneous approach to research about CFC.”* Researcher reflections There was considerable positive feedback about the fact that the meetings were well chaired and people felt able to contribute. However, the feedback forms demonstrate that some meetings were perceived not to be as well chaired as others, meaning that in some meetings stakeholders reported being listened to and able to get their thoughts across, while in others they highlighted that there were problems with this. In some meetings stakeholders reported very different perceptions of involvement

	(e.g. from the same meeting – person 1: "*I didn't make a lot of points but feel what I said was taken on board*" and person 2: "*no one was watching then hands were raised and not going round people to get each person's input*"). "*The use of online meetings has limited our ability to collect implications from stakeholders. Had we been in the room, we could have used flip charts, post-it notes etc. This would have allowed us to collect clear statements of perceived implications. Although we had some great discussion around implications, these were not always clearly worded, specific, stated implications in the way that I would have liked. We maybe could have tried using online technology, but often we were struggling with technology as it was, and these difficulties were a barrier to introducing any further tools*".

Report of Activity 4b

ACTIVITY: Activity 4: Reach consensus over clinical implications and guide knowledge translation activities. **Activity 4b: Guide knowledge translation activities**	
1. Aim	
Task aim	To guide knowledge translation activities for the SUCCESS project.
2. Methods	
Who was involved?	All members of the stakeholder group had opportunities to contribute to discussions around the dissemination plan. People attending meetings at which the dissemination plan was specifically discussed were: 06-07-2020 Attendees: Doreen McClurg, Lorna Booth, Pauline Campbell, Alex Pollock, Andy Elders, Claire Torrens, Deb Smith, Clare Millington, Margaret Ogden, June Rogers, Davina Richardson 09-09-2020 Attendees: Doreen McClurg, Lorna Booth, Pauline Campbell, Alex Pollock, Andy Elders, Julie Cowie, Deb Smith, Clare Millington, Jonathan Sutcliffe, June Rogers, Davina Richardson, Gemma Kierczuk 28-04-2021 Attendees: Doreen McClurg, Lorna Booth, Pauline Campbell, Margaret Ogden, Brenda Cheer, Davina Richardson, Gemma Kierczuk, Clare Millington, Jonathan Sutcliffe 29-06-2021 Attendees: Doreen McClurg, Pauline Campbell, Margaret Ogden, Davina Richardson, Suzanne Hagen, Andy Elders, Karen

	Jankulak 13-09-2021 Attendees: Doreen McClurg, Pauline Campbell, Lorna Booth, Margaret Ogden, Debs Smith, Clare Millington, Karen Jankulak, Brenda Cheer 25-01-2022 - Attendees: Doreen McClurg, Lorna Booth, Pauline Campbell, Alex Todhunter-Brown, Margaret Ogden, Davina Richardson, Deb Smith, Jonathan Sutcliffe, Clare Millington, Karen Jankulak 15-03-2022 - Attendees: Alex Todhunter-Brown, Margaret Ogden, Deb Smith, Jonathan Sutcliffe, Karen Jankulak, Brenda Cheer, Lorna Booth, Pauline Campbell 20-04-2022 - Attendees: Alex Todhunter-Brown, Margaret Ogden, Deb Smith, Karen Jankulak, Jonathan Sutcliffe, Clair Torrens, Brenda Cheer
When was the involvement?	An initial (draft) dissemination strategy was included in the funding application. This was added to over a series of meetings, with relevant notes taken. There were three updated versions drafted over the course of the project. Specific meeting and relevant items discussed at meetings were: 06-07-2020 Dissemination plan 09-09-2020 Dissemination plan 28-04-2021 Dissemination plan 29-06-2021 Abstracts for ICS

	13-09-2021 Interactive maps & report writing 25-01-2022 Report writing 15-03-2022 Dissemination plan 20-04-2022 Dissemination ‘products’
What happened?	06-07-2020, 09-09-2020, 28-04-21 – at each of these meetings the research team recapped on some of the dissemination ideas that had been proposed at the last SG meeting and continued to develop the dissemination plans 29-06-2021 During the meeting, the stakeholders were informed that Two abstracts were submitted to the ICS and have been accepted in the Open Discussion Section and are now published on the web-site Nos 411 and 412. These relate to the Complementary and Economic reviews. 13-09-2021 Following a presentation of some draft evidence maps, stakeholders provided feedback about the layout, content and dissemination of these. 25-01-2022 A draft outline for the NIHR report was presented and discussed, and agreements made on the content of and contributions to some of the chapters (see Results). 15-03-2022 and 20-04-22 Version 2 of the dissemination plan was discussed and key priorities for dissemination agreed. After the meeting, this was integrated into Version 3 of the dissemination plan, which was shared by email.

Level of involvement?	Stakeholders led key decisions about the products that should be produced and where these should be disseminated; therefore the level of involvement was categorised as "controlling".
3. Results	
Outcomes—Report the results of SG in the study, including both positive and negative outcomes	**<u>Dissemination Plan</u>** Version 3 of the Dissemination Plan is below. Stakeholders agreed that the aim should be to make the evidence available for every aspect of the public to use, and that we should have the following key outputs: - Published protocols - NIHR final report - Interactive evidence map(s) - High quality journal publications - Lay report - Leaflets / infographics / visual abstracts. The following were noted as key points relating to dissemination: - **Personal stories**, and the use of professional and public newsletters were also recommended by SG members. - **Pyramid model -** It was highlighted that the Pyramid model may make sense in professional documents but will be less accessible to the general public. - **Journal publications.** *M*ain findings should be published in several relevant peer reviewed journals such as the 'BMJ', 'Child: care, health and development' and the Nursing Times 'Continence Supplement'. Also consider journal read by GPs. - **Social media / online forums** – cascading key findings via key stakeholders and social media platforms is important. We should include the charities involved in the project and on-line forums used by parents and patients as identified by our SG. - **Stakeholder support** – important that we work with our third sector partners, e.g. ERIC – the Children's Bowel and Bladder Charity, Bowel and Bladder, UK and our SG, to develop the findings into clear, clinically relevant evidence-based treatment pathways, as well as an accessible guide for parents. - **Symposiums / Conferences / meetings** – relevant conferences include those held by: *International Continence Society, Association*

for Continence Advice, ERIC Paediatric Continence Care Conference, Physiotherapy networks (Annual conference of the professional network, Pelvic, Obstetric and Gynaecological Physiotherapists). Also consider including surgical meetings and relevant meetings of Royal college of paediatrics.
    - A presentation was made at the Bladder & Bowel UK National Continence symposium for health care professionals in March 2022.

- **Parent information –** Bladder & Bowel UK have parent and professional information and online resources whilst ERIC hold parent and carer training days and have multiple on-line resources to which we could add our findings.
- **Commissioners -** barriers to delivery care are not just about not being precise about treatment, but that it is the infrastructure that it is delivered within. Papers are rarely written about how to develop an infrastructure for a service like CFC, which is multidisciplinary, cross-sector. This means that there will be a deficit in our review because there are not going to be a lot of papers. Suggested that we draft something that would influence commissioners to highlight where things could be improved within the NHS. Consider dissemination to NHS England (continence board) and NICE implementation team. Consider publication in Health Service Journal.
- **Education** - Suggestion that dissemination needs to take place within education and child-care sectors – e.g. primary teachers and people doing NVQ’s in child-care. The following suggestions were made; There is an e-learning module on bowel care in The Royal College of Paediatrics. There is new module that is about to be launched in the RGCP in Scotland. There has been guidance written for nursery schools and colleges on bladder and bowel issues in children (joint Bladder and Bowel UK, and ERIC document been widely disseminated). There has been a lack of training health care professionals, but profile of bladder and bowel care has been raised among nursing and midwifery council.
- **Special needs community** – dissemination could be effective via online forums. Possibly also something in SEN magazine. School governors may be appropriate to provide information to.
- **YouTube videos –** proposed as an effective / efficient / accessible mode of dissemination for a variety of audiences

<u>NIHR final report</u>

The following was agreed in relation to the stakeholder involvement in

	the writing of the NIHR final report: - Co-production of chapter 3. It was agreed that chapter on stakeholder involvement should be co-produced. It was agreed that the researchers should do a first draft and then stakeholders should work collaboratively, in an iterative fashion, to contribute to the writing of this chapter. - Reflections sections. It was also proposed and agreed that all chapters of the report should have a ‘reflections’ section, for individual members of the stakeholder group to add their personal thoughts. - GRIPP2 reporting guidelines. Stakeholders discussed the GRIPP2 reporting guidelines and agreed that the GRIPP2-LF should be used, checking the relevant sections at the end and writing in the page numbers. - Plain English: It was agreed that the report will be written in plain English wherever possible. Consider including a plain English summary of findings in each chapter. **Evidence maps** It was agreed that the evidence maps would be a fantastic output from this project, providing a legacy database which will be made available in an interactive map that everyone can access. This will also make reference lists available to other researchers and interested clinicians which will be valuable to this community. General feedback about the presentation of evidence maps included: - Important to have the interactive maps explained e.g. a video - Important to make sure accessible to all sections of society e.g. on phones/paper - Like to keep as simple as possible - Look at colours
4. Discussion & conclusions	
Outcomes—Comment on the extent to which SG influenced the study overall. Describe	The stakeholders led key decisions about the products that should be produced and where these should be disseminated. The stakeholders had many ideas and proposals relating to dissemination and products that could be produced. The ability of the research team to address all of these was limited by time and resources. However,

positive and negative effects	stakeholders were able to highlight priorities. The key priority was to have different 'layers' of information, so that different audiences could access information to different levels of detail: *"we need pick and mix short presentations"*.
5. Reflections / critical perspective	
Comment critically on the study, reflecting on the things that went well and those that did not, so others can learn from this experience	**Stakeholder reflections** *"produce the information in the format that others can pick it up and use it"* On co-production of the Stakeholder involvement chapter: "*Maybe I should have been more enthusiastic about the PPI paper – after all it's what I've been wanting all along. But it's a fantastic opportunity. Am just a bit apprehensive about finding time to dedicate to it…….. suggestion about working with manageable chunks should help us not feel too overwhelmed*". "[our organisation] *will happily share on all of its social media platforms, and if you just send anything then I will make sure it gets to the right place……we have a very efficient media person who would do that*". "*To me, telling people about the project is a way of promoting the concept…..it lets me tell people 'be aware of constipation'*" Evidence maps will be valued by clinicians as "*you can get to the source data and you feel respected…..this will be really good*". "*if we can't do professional films…..we can produce something that is good enough to influence the outcomes*" **Research team reflections** *"A key lesson to me has been that in future funding applications I need to make sure that we have a budget for 'creative' input…..the stakeholders had fantastic ideas about products which could be shared on social media, but I wasn't sure that we had the skills – or time – in the research*

	team to do these justice" *"'Co-production' of written work is challenging. There still needs to be a lead writer so that things get brought together into one whole piece. I feel this chapter has had really substantial stakeholder input….but does this class as 'co-produced' writing…?"* *"If we can't produce all the fancy animated things we want, we can produce the information to go into these….. a bit dry but ready to be wrapped up in a nicer format…."*

SUCCESS Dissemination plan (02/05/2022)

<u>Social-media based dissemination plan:</u>

Audience	**Products (need to clarify the *objective* for each product)**	**Details / notes**	**Responsible for producing**	**WHERE is this being disseminated?**	**Responsible for disseminating**	**Deadline / completion date**
Parents, carers and children with CFC	Single A4 page, plain language summary of findings		Research team to draft, PPI stakeholders to edit	To be hosted on GCU SUCCESS webpage – links to this from tweets / FB	PPI, ERIC and Bladder & Bowel UK?.	
	Plain language tweet		Research team to draft, PPI stakeholders to edit	@ERIC_UK, @NMAHPRu Personal accounts @BladderBowelUK	All	
	Flier / Facebook 'announcement'	suitable for sharing on FB which says ""this has been done and this is where you can find more information" …. promoting the project and the concept…….be aware of constipation " Lay information with links to more information	Research team to draft, PPI stakeholders to edit	Personal accounts of stakeholders	All	

	3-minute videos covering key messages from project	“I think having different people speaking would be good”	Research team + stakeholder volunteers	Hosted on GCU website. Shared by Twitter & FB	All	
Clinicians	Flier	Overview of project – links to more info	Research team + stakeholder feedback	Twitter, FB, websites. @VivJBennett @WeSchoolNurses @ACAContinence @iHealthVisiting Tweets targeting: dieticians, psychology, GP, Community paeds, general paeds, paeds gastro, adult gen surgeons and paed surgeons, clinical pharmacists, social workers, education sector, child-care workers, professionals working with vulnerable children	Stakeholders to circulate. Send to NIHR, NICE, NHS (4 nations). Professional interest groups.	
	Visual abstracts	One for each of reviews / one page infographic pointing people to maps / further information	Research team (to consult with GCU brand & marketing)			
	2-page summary	To be linked to from other products	Research team + stakeholder feedback	Hosted on GCU website.		
	Recorded powerpoints (max 3.5	“pick and mix short presentation	Research team	Hosted on GCU website.		

	minutes)	s” - Covering all aspects of project		Linked to from other products		
	3-minute film, targeting continence teams	Key messages for continence teams	? Brenda			
	3-minute film, targeting consultant teams	Key messages for consultant teams	? Jonathan			
	Evidence maps	Hosted on GCU SUCCESS website	Research team	Linked to from visual abstract, 2-page summary, recorded PPT		
	Summary of all above resources	Share with BIG (Bowel Interest Group)(facilitated by E4H and Coloplast) and other relevant organisations	Doreen	BIG webinars, workshops, online resources	Doreen	
Commissioners	Targeted information for NICE implementation website	Approach to ask what they would like for website	Research team			
	Targeted information for NHS England (Continence board)	Approach Liz Wrigley for advice				
	3-minute film, targeting commissioners	Key messages for commissioners	? Jonathan / ? Liz Wrigley	Suggested: HSJ, Academy of Fabulous Stuff, NHS Networks, Integrated		

				Care Systems (ICS) around 30 of thesis in England.		
Third sector	Information suitable for third sector organisation to use to create / inform content for educational products etc	"produce the information in the format that others can pick it up and use it"	Research team to discuss with ERIC (Alina)			
	Summary of all above resources			Send to Brenda to be shared by ERIC and Davina to be shared by Bladder & Bowel UK		

Conference / meeting dissemination plan

Audience	**Products (need to clarify the *objective* for each product)**	**Details**	**Responsible for producing**	**WHERE is this being disseminated?**	**Responsible for disseminating**	**Conference date**
Clinicians	Bladder & Bowel UK conference		Doreen		Doreen	March 2022
	UK Colorectal group	Attend clinician meetings	Jonathan			
	International continence society (ICS) conference	2 presentations: Complementary review Economic review	Doreen	ICS conference	Doreen	Sept 2021

	ERIC conference	Overview of project & findings PPI view	Brenda	ERIC conference	Brenda to coordinate - ? Pauline & Karen to present	10th Oct
	Surgical / Gastro / Consultant teams meeting		Jonathan?			
Third sector	Link with National Bowel & Bladder UK Health Project (led by NHS England)		Brenda to discuss at next National Bladder & Bowel UK meeting			

<u>Publications plan:</u>

- PROSPERO protocol publication
- NIHR Final report
- Journal publications for:
  - Scoping review + effectiveness review
  - Implementation review
  - Economic evaluation
  - Logic model
  - Stakeholder involvement

<u>NB: to consider -</u>

Clinicians to include: school nurses, health visitors, physiotherapists, continence nurses, GPs, community paediatrics, general paediatrics, paeds gastro, adult general surgeons, paed surgeons, dieticians, psychologists. **Parents and Carers** to include special need community. **Third sector** to ERIC, Bladder & Bowel UK, Bowel Interest Group. **Researchers and International Collaborators** to include: NIHR, International Continence Society, and wider research community (list of authors). **Education establishments** to include: nursery, primary and secondary school staff, colleges offering child-care modules. Commissioners to include: NHS England (continence board), equivalent for other nations, NICE Implementation team

ACTIVE framework of involvement in a systematic review

Framework Constructs	***Categories***		***Key / Icon***
Who is involved?	Patients, carers and / or their families		🧍
	Patients, carers and / or their families + other stakeholders		🌐🧍🧍
	Other stakeholders only		🌐🧍
How are people recruited?	Open 🚪	Fixed	
		Flexible	🚪 Flexible
	Closed 🔒	Invitation	🔒 Invite
		Existing group	🔒 Group
		Purposive sampling	🔒 Sample
	Other / Unclear		?
What happened? *Approach?*	One-time		x1
	Continuous		🔄
	Combined (i.e. both one-time and continuous)		⧉
What happened? *Methods?*	Direct interaction		💬
	No direct interaction		🖥

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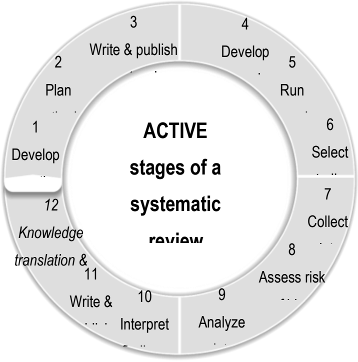
<tr><td rowspan="6">Stage & Level?</td><td rowspan="5">

(see Figure 2)</td><td>Leading</td><td>Lead</td></tr>
<tr><td>Controlling</td><td>Control</td></tr>
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Report of conversations with children and parents

During the project the core group of stakeholders raised the need to strengthen the “voice” of children and parents. We therefore decided to conduct some interviews with children and their parents and we were also provided access to a video, which had been recorded within the Leeds paediatric colorectal unit, in which a boy (aged 8) talked about living with CFC.

Ethical approval was provided by Glasgow Caledonian University to conduct the interviews with separate Information and consent forms for children 6-12, 13-15 and their parents/guardian. We have permission to use anonymised direct quotes. Leeds University Hospital Trust and the participants of the video provided permission for the core team to view the video.

We advertised for participants through the Movicol Mummies Facebook and received interest from 5 parents/children. Three interviews, using Zoom and lasting approximately 45 minutes were conducted during July 2020. All three children were female and were aged 7-10.

CFC began in one child after struggling to become ‘dry’ as a toddler and being prescribed Oxybutynin at 5 years and 3 months. Constipation with soiling began shortly after this and was prescribed Movicol but with no follow-up. The Oxybutynin was discontinued following a referral to a urologist, however several episodes of impaction has occurred since and was presently going through disimpaction (week 3) and currently on 12 sachets of Movicol per day. She has also tried enemas and glycerol suppositories. Due to lockdown feels a lack of support but access to sites such as ERIC and Movicol MUMS helpful. Has a good diet and is active. There had been one meeting with the School Nurse just before lockdown. Generally, she is a happy positive child but is worried about being teased if she has to continue to wear pull ups.

The second interview was with a 7-year old girl and her mum. The child had one severe stomach upset when aged 4 which had been due to an infection and she has been suffering from CFC since then. She has been taking ½ Movicol per day and was referred to a Continence Nurse for night time wetting. Had a recent referral to a Child Development paediatrician and was currently taking 12 sachets of Movicol, 20 mls pecosulphate and 5mc Oxybutynin and was to continue with this to allow the ‘rectum to shrink’. Biweekly support phone calls with a Specialist Nurse was helpful and important for support. Had tried dairy free, but had made no difference. Has a good general diet and is fairly active. Has had no issues at school. Uses social media for support and information.

The third girl aged 8 had issues with potty training and had refused to use it. Also issues with bed wetting. School nurse and GP had tried to help and then referred to a Paediatrician who was helpful. Now taking Movicol, picolax and oxybutynin. Has been going through the process of disimpaction lately with some success, being at home due to lock down was making this easier, quite a dark time but seemed to be coming out the other end. Diet and exercise seemed good. Uses social media for support and information, and uses mindfulness.

Main issues from all interviews and video

1. Getting the right help at the right time is difficult

2. Knowing the best social media to access is not always apparent

3. CFC impacts the child's social and school life but also has social implications for the rest of the family

4. Taking large amounts of Movicol seems to be the main treatment but is not easy to take.