

Developing a Booklet to Promote Psychosocial Health in New Families Affected by Craniosynostosis

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Summary

Recent research indicates that parents of children with craniosynostosis may be at risk of emotional distress. Yet, parents may not be accessing the support they need to cope with common challenges. The aim of this project was to develop a research-informed booklet to promote psychological health in new parents, and to assess acceptability of the booklet within the craniosynostosis community. The first draft was designed in close collaboration with leading UK charity (*Charity*), five parent representatives and three specialist clinical psychologists via online focus groups. This draft, attached to an online acceptability survey, was distributed to a broader group of parents and multidisciplinary specialists working in craniofacial teams in England for feedback. A total of 44 complete responses to the online acceptability survey were received. Acceptability (measured by the number of respondents who would recommend the booklet) was 100 percent. All respondents reported they 'agreed' or 'strongly agreed' with the UK-wide distribution of the booklet. Evidence for psychological intervention in the craniofacial field remains scarce, and specialist teams are currently under-resourced to effectively screen and support parents. It is hoped this booklet will begin to address the gap in psychological support for new parents of children with craniosynostosis.

Key words: craniosynostosis, parent, psychosocial wellbeing, intervention

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Introduction

A congenital craniofacial condition and its long-term treatment is expected to bring about a range of psychosocial challenges for those affected and their families (Feragen & Stock, 2017). For parents, the diagnosis can evoke a number of conflicting and fluctuating emotions, as they come to terms with the implications of their child's condition (Nelson, Glenny et al., 2012). Relative to research carried out with those affected by the most common craniofacial condition, cleft lip and/or palate, understanding of the psychosocial impact of rare conditions, such as craniosynostosis, is poor (Feragen & Stock, 2017). However, recent research has indicated that parents of children with craniosynostosis may be at risk of elevated anxiety and stress levels compared with the general population (Rosenberg et al., 2011), exacerbated by a lack of reliable information and support (Zerpe et al., 2020), and perceived social stigma (Zeytinoğlu et al., 2021).

A recent online survey distributed throughout the United Kingdom (UK) to parents of children with craniosynostosis (*manuscript under review*) offered a number of additional insights into the parenting experience. In comparison to norms, parents ($n = 111$) reported lower levels of optimism and resilience, less relationship satisfaction, higher levels of stress, and more symptoms of anxiety and depression. In addition, just over 92 percent reported emotional distress at the birth of their child, and almost 100 percent were concerned about the future. Participating parents also provided qualitative commentary describing their experiences in more detail. Common reports included a traumatic birth experience, a missed or delayed diagnosis, having their concerns dismissed by non-specialist health professionals, and difficulties accessing appropriate care. Although all five specialist craniofacial teams in the UK (four in England, one in Scotland) now include clinical psychologists, only 27 percent of survey respondents had accessed any form of psychological support to help them cope with these challenges (*manuscript under review*). Investigations in the wider health field have demonstrated the importance of early psychological intervention for families, not only to promote parental wellbeing itself, but to encourage family functioning as a whole and to prevent longer-term difficulties in the child (Sanger et al., 2015). In 2017, Pidgeon and colleagues found that the introduction of a parent information leaflet helped to reduce parental anxiety ahead of attending a craniofacial outpatient appointment, demonstrating that even small-scale interventions can be effective.

In response to recent findings advocating a need for increased psychological support for parents of children with craniosynostosis, the current paper outlines a short-term project designed to begin to address this gap. The aim of the project was to develop a research-informed booklet focused on promoting psychological health in new parents, and to assess acceptability of the booklet within the craniosynostosis community.

Materials and Methods

Design

Funding was received to develop and review the booklet over a 3-month period. A collaborative approach was taken, led by a senior researcher from (*University*), alongside

(*Charity*), NHS specialist clinical psychologists, and a group of parent representatives. Online focus groups were utilised to develop and refine the first draft of the booklet. A brief online survey was used to assess acceptability.

Procedure

The work involved three key phases (Figure 1). First, initial focus groups were held online with a) a group of five parent representatives and b) a group of three specialist clinical psychologists. Parents were recruited directly through (*Charity*). All specialist clinical psychologists currently working in one of the five NHS craniofacial teams were invited to participate. Focus groups were facilitated by the first and last authors, and were audio-recorded for notes purposes only with participants' permission. Parent focus groups lasted 3 hours on average. Clinical psychology focus groups lasted 1.5 hours on average.

The aim of the initial focus groups was to develop a framework for early parent/caregiver experiences in the context of craniosynostosis, and to discuss the ideal content and format of the booklet. To develop a framework, focus group participants were asked to identify key milestones within the parent journey. Six key milestones relevant to the early years were identified (Figure 2). So as not to overwhelm new parents, it was agreed that the initial booklet should focus solely on the first three milestones: Pregnancy and Birth, Diagnosis, and Accessing Treatment. Focus group summaries were sent to all participants and any further comments were invited.

Based on focus group discussions, existing craniofacial literature, and existing broader psychological resources (such as Acceptance and Commitment Therapy), the first and last authors developed an initial draft of the booklet content. This included: brief information about the condition and its treatment, common psychological concerns, available support, and how to manage challenges at home (Table 1). Photographs, diagrams, and quotations derived from previous research were also included.

Second, follow-up focus groups were conducted. The aim of these focus groups was to review the initial draft booklet and make any recommended amendments. Minor edits were made, summaries of the changes were sent to focus group participants, and all participants approved the final draft. The booklet was then sent to a graphic designer for editing.

Finally, a brief online survey was designed by the first and last authors for the purpose of assessing the acceptability of the booklet to the broader craniofacial community (Table 2). This survey was advertised to all parents who are members and/or followers of (*Charity*), in addition to the four NHS craniofacial teams in England. A downloadable copy of the draft booklet was included in the survey. The survey remained open for two weeks. Survey data were cleaned and only complete responses were included. Survey data were then summarised descriptively by the first author.

Participants

The clinical psychology focus group consisted of three specialist clinical psychologists representing two of the five NHS craniofacial teams in the UK. The parent focus group included four mothers and one father from four families living in the UK. Participating parents' children were aged between six and 28 years ($M = 14$ years), with half having received a syndromic diagnosis.

Online survey participants included 27 parents, three other family members (2 grandparents, 1 uncle), and 14 health professionals representing the four craniofacial teams in England. Parents/family members included mothers ($n = 18$) and fathers ($n = 9$) of children aged between eight weeks to 14 years, diagnosed with non-syndromic ($n = 17$) and syndromic ($n = 13$) forms of craniosynostosis. Health professionals included speech and language therapists ($n = 4$), clinical nurse specialists ($n = 3$), clinical psychologists ($n = 3$), neurosurgeons ($n = 2$), clinical geneticists ($n = 1$), and orthoptists ($n = 1$).

Results

A total of 44 complete responses to the online acceptability survey were received. Respondents believed the booklet would not only be useful for new parents ($n = 43$), but also for other family members (such as grandparents; $n = 41$), non-specialist health professionals (such as midwives, health visitors, and General Practitioners; $n = 35$), and parents' friends ($n = 33$). Three respondents also felt the booklet could be useful in early educational settings. Respondents reported that they liked the content ($n = 39$), quotations ($n = 35$), links to information and support services ($n = 34$), layout ($n = 32$), photographs ($n = 28$), and diagrams ($n = 24$). Improvements were also suggested, as follows: provide more detailed medical information ($n = 10$), include additional quotes from parents ($n = 3$), include more photographs of children with syndromic diagnoses ($n = 2$), and reduce the overall amount of text ($n = 1$).

As a result of the booklet, respondents felt families would feel reassured they are not alone on their journey ($n = 31$), be better equipped to cope with common challenges ($n = 19$), and would have better access to information about craniosynostosis ($n = 28$), peer support groups and networks ($n = 24$), and emotional support ($n = 23$). Respondents also believed that family and friends ($n = 25$) and non-specialist health professionals ($n = 21$) would be better informed about craniosynostosis as a result of the booklet. Acceptability, as measured by the number of respondents who would recommend the booklet, was 100 percent. All respondents reported they 'agreed' ($n = 13$) or 'strongly agreed' ($n = 31$) with the UK-wide distribution of the booklet via (*Charity*) and the NHS craniofacial teams.

Respondents provided a range of additional qualitative responses, outlining the benefits they believed the booklet would have (Table 3). Within these qualitative responses, parents and health professionals indicated they would like more booklets like this to be developed in future. In particular, and mapping onto the conceptual framework, this included information to help parents make decisions about and prepare for the child's primary surgery ($n = 9$), and information about what happens post-surgery (including nursery/school entry, and talking to the child about their condition; $n = 5$). Three participants felt a separate booklet for parents of children with syndromic diagnoses might be needed, due to differences in treatment pathways and outcomes.

Discussion

Recent literature in the craniofacial field has indicated a need for improved access to psychological support for parents of children with craniosynostosis. The aim of the current project was to develop a research-informed booklet focused on promoting psychological health in new parents, and to assess acceptability of the booklet within the craniosynostosis community.

Collaborative Working

A collaborative approach was taken for this work, involving an academic researcher, a leading UK charity, multidisciplinary members of specialist craniofacial teams, and parent representatives. While involving service users in research is no longer uncommon, the co-production of a resource, whereby users and professionals from a wide variety of backgrounds contribute their varying expertise simultaneously, offers a particularly strong model of collaborative working (Heaton et al., 2015). Research has demonstrated that diverse teams work most effectively when the project is end-user driven and focused on a coherent and shared objective, when team members are receptive to different forms of knowledge, and when all perspectives are equally valued (Boyle & Harris, 2009; Heaton et al., 2015). In the craniofacial field, especially when compared to other areas of health, such approaches remain in their infancy (Stock et al., 2018). Efforts to build long-term cohesive collaborations between key stakeholders to maximise reach and impact are therefore strongly encouraged.

Acceptability

Perhaps due in part to the collaborative approach of this work, the feedback received from parents and health professionals in response to the booklet was overwhelmingly positive. Survey respondents felt the booklet would improve knowledge and psychological health for a range of beneficiaries, in addition to fulfilling a gap in care and positively impacting clinical practice. All recommended the booklet for UK-wide distribution. Some suggestions for improvement were made, and most were actioned in the final version of the booklet. However, the request for a separate booklet for parents of children with syndromic diagnoses was carefully considered and ultimately discarded, as the existing literature (Stock & Feragen, 2019) and clinical experience suggests that similar psychological concerns tend to apply across the different types of condition. Similarly, the request for more detailed medical information was felt not to be appropriate, since the aim of this booklet was to focus on psychological health, and since other booklets containing medical information are already in circulation. In addition to parents who are actively seeking psychological support, the booklet could also be helpful for those who feel more reluctant to access mental health services, by normalising emotional reactions to the diagnosis and reducing the stigma associated with this form of care (Stuart, 2016).

Future Work

As was indicated by the conceptual framework and requested in the acceptability survey, additional booklets may be of benefit to the community. In particular, preparation for surgery, nursery/school entry, and talking about craniosynostosis within the family were identified as important foci. According to current craniofacial literature, the primary surgery is reported as being an especially emotive and challenging time, and treatment decisions may weigh heavily on parents (Nelson, Caress et al., 2012). Nursery/school entry is likely the first time children will have been regularly exposed to their peers, and may come into contact with questions, comments, and teasing (Rumsey & Harcourt, 2012). Knowing how to talk to their children about their condition, particularly if the child's appearance is affected, is another known concern for parents (Feragen et al., *in press*). Early interventions to address these common concerns are therefore warranted. More broadly, a better understanding of the psychosocial impact and support needs of the craniosynostosis population is essential. Currently, little evidence for psychological intervention exists in the craniofacial field (Norman et al., 2015), despite being the top priority in a number of recent relevant research priority-setting exercises (e.g. Petit-Zeman

& Cowan, 2013). Dedicated exploration of parents' and individuals' experiences in the context of craniosynostosis would help to build the evidence base and support future intervention development and evaluation.

Methodological Considerations

Despite the importance of the collaborative approach taken, the groups of representatives utilised in this project were relatively small. Similarly, the number of survey respondents was fairly limited, which can often be the case when the condition of interest is relatively rare. Not all disciplines were represented, and the new specialist unit in Scotland could not be contacted, which was likely compounded by the short timescale. Nonetheless, the amount achieved within the time available and the feedback that was collected is encouraging. It is hoped this project demonstrates a first step toward improving psychosocial support for parents of children with craniosynostosis, and toward building a long-lasting beneficial partnership between key stakeholders.

Conclusions

This short-term project has produced a booklet which was supported by parents/family members and health professionals, and which has demonstrated relevance and potential impact for clinical practice. Evidence for psychological intervention in the craniofacial field remains scarce, and specialist teams are currently under-resourced to effectively screen and support parents. It is hoped this booklet will begin to address the gap in psychological support for new parents of children with craniosynostosis.

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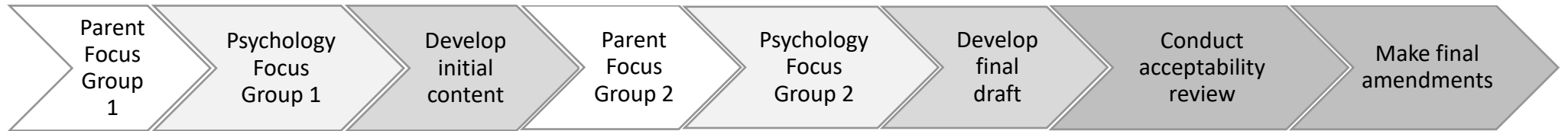


Figure 1: Activity Timeline

Key Milestones During the Early Years

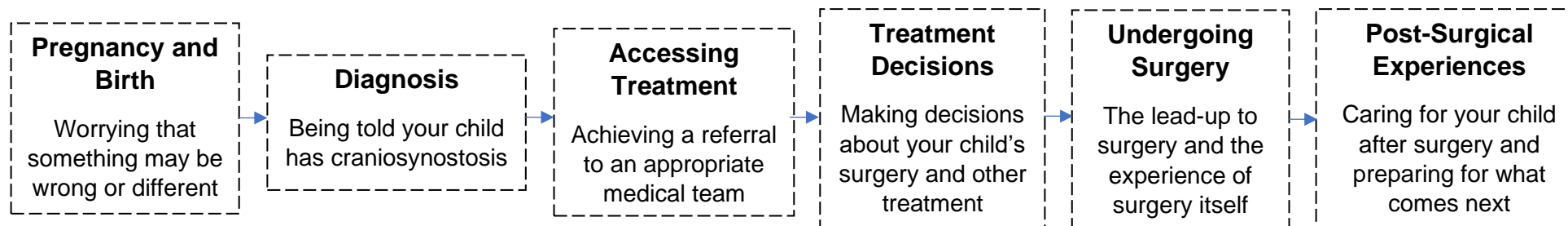


Figure 2: Conceptual Framework Illustrating Parents' Early Experiences

Table 1: An Overview of Booklet Content

Section Headings	Key Content
What is Craniosynostosis?	A brief explanation of how craniosynostosis occurs (including diagram of the skull)
	A brief explanation of non-syndromic and syndromic forms of the condition
	Estimated prevalence
How is Craniosynostosis Treated?	An overview of how treatment is offered in the United Kingdom
	Locations of the five designated craniofacial units in the United Kingdom (including map)
Common Concerns	An overview of the common and fluctuating emotions that parents experience, with an emphasis on normalising these emotions (including diagram of the 'Grief Cycle')
	Information about the first three key milestones and how to seek support (including diagram of conceptual framework)
Support for You and Your Family	Signposting to key sources of support for families, including Clinical Nurse Specialists, Clinical Psychologists, national charities, and Health Visitors
Managing Challenges at Home	Support for infant-parent bonding
	Relationship support (including partners, family, friends, child's siblings)
	Having a baby with a different appearance
	Supporting your own mental health (including breathing exercises and grounding statements)
	Advice from other parents
Your Child's Future	A reminder that support is available throughout the journey

Table 2: An Overview of the Online Acceptability Survey

Question	Response Options
Who do you think this booklet could be helpful for?	Tick all that apply Other (open-ended)
What do you like about the booklet?	Tick all that apply Other (open-ended)
What about the booklet could be improved?	Tick all that apply Other (open-ended)
What did you personally take away from reading the booklet?	Open-ended
What do you think could change as a result of this booklet being available?	Tick all that apply Other (open-ended)
Would you recommend this booklet?	Yes / I'm not sure / No
Please tell us why you would/would not recommend the booklet?	Open-ended
I would support the distribution of this booklet by (<i>Charity</i>)	0 = Strongly Disagree, 4 = Strongly Agree
I would support the distribution of this booklet by the specialist craniofacial teams	0 = Strongly Disagree, 4 = Strongly Agree
I have other suggestions for the distribution of this booklet	Open-ended
If you do not support the distribution of the booklet, please tell us why?	Open-ended
Do you have any other comments about the booklet that you would like to share?	Open-ended
Are there any additional booklets that you think could be helpful?	Open-ended

Table 3: Summary of Qualitative Comments Alongside Exemplar Quotes

Respondent	Topic	Exemplar Quotes
Parent/Family Member	Feeling Reassured	“Reassurance, a sense of not being alone, feeling we are supported... There are people who understand”
		“So reassuring to know others go through a similar journey and experience similar thoughts and emotions”
	Fulfilling a Gap in Care	“This is the information I really needed to read when my son was born. It would have helped me immensely. I wish this booklet had been around then”
	Raising Awareness	“I’m so glad someone is trying to spread the word and educate parents who desperately need it”
		“Although my child is older now, this booklet is still useful for sharing information about my child’s condition, and giving family and friends an insight into what we’ve been through”
		“Any health professional involved in the early stages should read this”
	Impact on Psychological Health	“It gives great practical tips on how to cope and provides validation of parents’ feelings”
		“It will put new parents’ minds at ease and covers many aspects, views, and concerns that most parents will have thought about at some point”
		“This booklet gives so many answers that as a parent you are searching for... It will also prompt further self-research”
	Writing Style	“The information is concise but detailed... Really clear, well written, very relatable”
		“Easy explanations without medical jargon... Clear and direct information which has been hard to come by in our experience”
General Praise	“It’s a brilliant resource that will be a huge support to so many people”	
Health Professional	Increased Empathy	“Compassion and strong empathy for the parents of a newly diagnosed child”
	Gaining of Psychological Knowledge	“I understand much more about the different reactions parents can experience after a diagnosis”
		“The booklet has helped me to recognise the emotional toll of the diagnosis on parents”
	Impact on Clinical Practice	“The booklet raises important issues which are often difficult to broach in clinic”
		“The booklet is less medical than others I’ve seen and more family-directed which is nice and much needed”

		"This booklet will impact parental wellbeing, which is so fundamental to their life, their attachment with their baby, and all aspects of their child's development"
	General Praise	"This booklet is just what is needed"
		"The booklet hits the mark on many levels"