

Brief Communication article

“When I was younger, my story belonged to everyone else”:

Co-production of resources for adults living with craniosynostosis

Nicola M Stock, Bruna Costa, William Bannister, Charlotte Ashby, Nammie Matthews, Louise Hebden, Laura Melles, Zoe Hilton-Webb, Sally Smith, Kristian Kane, Lewis Carter, Anna Kearney, Katie Piggott, Charlotte Russell, Karen Wilkinson-Bell

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Abstract

Objective: Despite growing recognition that congenital craniofacial conditions have lifelong implications, psychological support for adults is currently lacking. The aim of this project was to produce a series of short films about living with craniosynostosis in adulthood, alongside a psychoeducational booklet.

Design: The resources were developed using multiple focus groups and meetings attended by researchers, patient representatives, a leading charitable organisation, an award-winning film production company, clinicians, and other experts in the field.

Results: An online mixed-methods survey was developed based on prior work to request feedback on the acceptability and utility of the resources from the craniosynostosis community. While data collection to evaluate the resources is ongoing, preliminary results ($n=22$) highlight an acceptability rating of 100%.

Conclusions: The resources developed represent a step forward in addressing the unmet information and support needs of adults with craniosynostosis and highlight the benefits of co-production in research.

Background

Craniosynostosis is a relatively rare and complex condition, in which the clinical presentation can vary considerably. It occurs when two or more of the cranial plates fuse prematurely and affects approximately one in every 2,000 live births¹. Approximately 25% of cases are syndromic¹. Varying according to the severity of the condition and associated medical needs, surgical intervention and multidisciplinary treatment throughout childhood is recommended.

Alongside medical care, community-based support for those affected by craniosynostosis is offered by Headlines Craniofacial Support (Headlines), a UK-wide organisation that became a registered charity in 1996. Following a strategic review and research priority-setting exercise that highlighted the need to investigate the experiences and support needs of adults with craniosynostosis, Headlines partnered with the Centre for Appearance Research (CAR) at the University of the West of England (UWE) to conduct an online mixed-methods survey across the UK. In line with the limited available research within the adult craniofacial population²⁻⁷, survey respondents reported more symptoms of anxiety, more appearance concerns, less satisfaction with social relationships, and lower levels of resilience compared to the general population⁸. Qualitative analysis of open-ended responses also identified a range of ongoing physical health problems, difficulties accessing treatment, employment bias, and concerns around starting a family due to the increased genetic risk⁸. Further, respondents were unaware of how to obtain information and support in adulthood and viewed Headlines exclusively as a children's charity⁸. These findings are also supported by related research in adults with other craniofacial conditions, such as cleft lip and/or palate (CL/P)⁹⁻¹².

It is estimated there are up to 10,000 adults living in the UK with a form of craniosynostosis that has lifelong impacts¹³. As it stands, UK craniofacial teams are not commissioned to offer routine treatment or support to those over the age of 18 years. Yet, as with other congenital craniofacial conditions such as CL/P, recognition for craniosynostosis as a lifelong condition is growing, as is an understanding that current care provision is not meeting adults' needs. Headlines and CAR subsequently acquired further funding to develop online resources which would help to address the needs identified and increase the scope of Headlines' existing services to encompass long-term outcomes. This project was known as 'ACCORD' (A D u l t s w i t h Cr an io s yn o s t o s i s : C r e a t i n g O n l i n e R e s o u r c e s t o r e d u c e D i s t r e s s). The word 'accord'

also means to give someone power and recognition, especially if they have not had their voice heard before.

Producing the Resources

At the start of the project, Headlines and CAR established a group of 10 adult representatives aged between 21-58 years and affected by different forms of craniosynostosis, ranging from single-suture to rare syndromic conditions. Between October 2021 and February 2022, CAR hosted a series of online group discussions with the adult representatives to stimulate the sharing of experiences and ideas for the development of the resources. Also drawing on current literature and guidance from specialist clinical psychologists, CAR then developed a proposal for the content and format of the resources, which was shared with adult representatives at an in-person workshop in London in March 2022. Adult representatives provided feedback on this proposal during the workshop, which culminated in group consensus that the resources would include a series of five films (Table 1), alongside an accompanying psychoeducational booklet. The booklet would provide detailed information and referrals to resources, while the films would bring the information to life, increase visibility and reach, and directly addressing the isolating experience reported by many adults of never having met anyone with the same condition.

CAR enlisted the expertise of Fine Rolling Media, an award-winning UK film production company with an established track record of working with charitable and community-based organisations. The whole team (CAR, Headlines, adult representatives) met with the film crew regularly to further develop the key concepts for each film and agree upon the questions that would be asked during the on-camera interviews. The film crew also spent 1-1 time with each of the 7 adult representatives who volunteered to feature in the films, to better understand the stories these adults wanted to share and to build relationships prior to filming. Filming took place in diverse locations across England chosen by adult representatives throughout Summer 2022 and were edited by the team over subsequent months.

In March 2023, the team organised a film premiere at an independent cinema in Oxford. In attendance were members of the craniosynostosis community, including adult representatives, friends, family members, Headlines Trustees, and specialist health professionals. In addition to screening the films, the day included a series of

talks from various team members and a 'Question and Answer' session with the adult representatives. The event was captured through film and photography.

Complementary to the films, the booklet expanded on the same key themes. Included in the booklet were quotations from the original survey, photographs taken as part of the ACCORD project, information about craniosynostosis and treatment in adulthood, strategies for coping with psychological challenges, and referrals to additional sources of information and support. Case studies were also included to address areas not able to be covered in the films, such as intersectionality. In addition to the existing team, expertise in the areas of equality law, clinical genetics, and nursing were sought.

Evaluating the Resources

The team developed an online mixed-methods survey based on prior work¹⁴ to request feedback on the acceptability and utility of the ACCORD resources from the craniosynostosis community. Ethical approval for the evaluation was granted by the Faculty Research Ethics Committee at UWE. Respondents are being asked a series of 7 questions with multiple choice answers about who they think the resources will benefit, what they particularly like about the resources and what could be improved, what the impact(s) of the resources might be, whether they would recommend the resources to others, and whether they would support widespread distribution of the resources via Headlines and/or the National Health Service. Three additional open-ended questions ask what respondents will take away from the resources, why they would/would not recommended the resources, and any additional comments they would like to share. While data collection in the UK and internationally is ongoing, current results show acceptability (as measured by the number of respondents who would recommend the resources to others) to be 100 percent.

To date, the survey has been completed by 22 members of the UK craniosynostosis community (including 8 adults with craniosynostosis, 7 specialist health professionals, and 7 parents/family members). Respondents believed that as a result of the new resources, adults with craniosynostosis will feel less isolated, be better equipped to deal with any challenges, and have better access to peer support networks, information, and emotional support. In addition to promoting wellbeing in adults with craniosynostosis, respondents felt that parents/family members, young people with craniosynostosis, non-specialist health professionals (such as GPs), friends, schools,

and members of the general public would benefit from the resources. Examples of qualitative responses collected via open-ended text-boxes are collated in Table 2.

In addition to the formal evaluation, adult representatives from the ACCORD team reflected on their experiences of co-production.

“The ACCORD project motivated me to share my experiences of living with craniosynostosis, probably for the first time. Getting to know other people with the condition and hearing their stories, even though our experiences have been different, has been so inspiring”.

“I wanted to share my story and get out of my comfort zone. I feel that sharing my experiences spreads that awareness and educates members of the public. Seeing it all on the big screen was just brilliant”.

“I got a chance to be part of a great team, and we’ve all become great friends. I got to help other people who are facing some of the challenges that I’ve faced in my life”.

“[The team] all invested so much in our stories. It was clear that they’d taken the time to really think about our stories and what we’d been through, and how to convey them accurately”.

“We can’t let such a valuable and empowering project end here. We must carry this forward to give voice and effect change”.

Project Outcomes

All five films and the accompanying booklet are accessible free of charge via the Headlines website. The film crew and one adult representative also produced blogs about the project, and the project featured strongly in the Headlines 2023 newsletters. The project was the subject of a feature-length episode of the international Appearance Matters podcast in Spring 2023 and has been presented at conferences and events across the UK, Europe and the USA between 2023 and 2024. One of the adult representatives has since begun a PhD at the UWE to continue CAR’s research in the area of craniosynostosis. A summary of all resources related to this work is provided in Table 3.

Conclusion

The ACCORD project has contributed valuable resources that hold promise in addressing the unmet information and support needs of adults with craniosynostosis. Yet, the project also identified a number of areas for further investigation, such as the need for commissioned multidisciplinary care in adulthood, the development of

national registries, a better understanding of physical symptoms related to craniosynostosis, the development of resources to increase awareness among non-specialist health professionals, additional work with older adults, and the need for further research with underrepresented groups, particularly in relation to ethnicity and culture. The team also believe the films could be utilised to make impacts beyond the scope of the original project, for example, positively influencing general population attitudes toward “difference”. We encourage all members of the craniofacial community to access the resources and to share their views by completing the online evaluation survey.

References

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Table 1: Key themes and related content

Themes	Content	Exemplar Quotations from Group Discussions
Being ‘Different’	Managing other people’s reactions; overcoming barriers; finding an identity; engaging with life’s opportunities	<p><i>“I’ve always just dealt with it, really. I’ve never let [craniosynostosis] stop me... I think the key is to embrace the difference and enjoy life”.</i></p> <p><i>“I was more than an Asian, Muslim female. I was also someone who society saw as having abnormalities... As an adult I realised my condition was a strength... This allowed me to</i></p>

		<p><i>work on my identity and personality outside of [craniosynostosis]</i>".</p> <p><i>"I've only got three digits on my hand, and I was worried I wouldn't be able to drive at all, including my eyesight as well...but I've got an automatic car now and I enjoy driving"</i>.</p>
Social Experiences	<p>Navigating friendships and social anxiety; romantic relationships; developing a network of peers</p>	<p><i>"[Social situations] are generally OK, but I do get nervous when I meet new people... What are they going to think? Are they going to ask questions?... I second-guess myself and replay the moment in my head afterwards"</i>.</p> <p><i>"Dating and relationships as someone with [craniosynostosis] is something I found really difficult and navigating that has been very confusing"</i>.</p> <p><i>"I found Headlines in 2003 and it was very family- and child-oriented. It was lovely to see how everyone rallied around the families and how things had changed since I was a child, but as an adult I still didn't fit in"</i>.</p>
Navigating Treatment	<p>Making decisions about treatment; taking responsibility for care in adulthood</p>	<p><i>"[The children's hospital] was like another home...and then I moved to [the adult hospital] and that was new and big, and that was scary"</i>.</p> <p><i>"Most [young adults] would be thinking about the transition to [university], or your career, or your life plans... I was spending more time worrying about whether I should have surgery"</i>.</p> <p><i>"You have to learn to advocate for yourself... Managing [care as an adult] is demanding. Emotionally, physically, and psychologically"</i>.</p>
Challenges with Employment	<p>Applying for jobs; addressing bias in the workplace</p>	<p><i>"The negative part is getting rejected all the time and having to keep applying... I have the job interviews, but then nothing happens beyond that. They say 'you don't have enough experience', but if that was true, why was I offered an interview in the first place? So, something is happening... I do feel discriminated against"</i>.</p> <p><i>"I worked on the front desk in a busy [environment]. A lot of people would come in and out and it was getting that mixed reaction from people... It was really uncomfortable at times"</i>.</p> <p><i>"I developed my own business so my skills and capabilities could speak for themselves. Work can now also be flexible...if my physical health isn't good that day"</i>.</p>
Growing Older	<p>Medical traumatic stress; physical health;</p>	<p><i>"I gathered my medical records and started building my own story. It took some doing...to build up a timeline... That was how I started</i></p>

	<p>continued access to care; starting a family</p>	<p><i>piecing together the snapshots of memories I had and making sense of what had happened”.</i></p> <p><i>“I have physical symptoms now that I connect to my [craniosynostosis], but getting anyone to understand that is, well, I didn’t know where to start”.</i></p> <p><i>“Immediately after the birth they rushed [my daughter] to the neonatal team... The doctor was fumbling around and I said ‘you’re trying to tell me she’s got [craniosynostosis] aren’t you?’ He said ‘yes’ and the whole of my world fell apart... I was going to have to go through that journey again”.</i></p>
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Table 2: Examples of comments received about the ACCORD resources

Quotations	Respondents
<p><i>“Outstanding work, long overdue. It’s clear the participant’s voice has been the driver for creating the content. It really feels like it belongs to them”.</i></p>	<p>Health professional</p>
<p><i>“An excellent resource. The films are very well-made and provide a very positive message, without glossing over the challenges”.</i></p>	<p>Health professional</p>
<p><i>“Compelling stories that are really authentic and inspiring”.</i></p>	<p>Parent of a child with craniosynostosis</p>
<p><i>“[The resource] is really accessible. The videos are short which prevents overwhelm. You can watch the ones that are pertinent to you and revisit the rest later”.</i></p>	<p>Adult with craniosynostosis</p>
<p><i>“(I feel) a great sense of community, and like I’m not alone in my experiences. It’s a really powerful resource”.</i></p>	<p>Adult with craniosynostosis</p>

Table 3: A list of resources related to the ACCORD project

Resource	Link
<p>All five films and the accompanying downloadable booklet</p>	<p>www.headlines.org.uk/for-adults</p>
<p>All five films in a single film roll</p>	<p>https://vimeo.com/804350602/b0f7a06ba2</p>
<p>A teaser film</p>	<p>https://vimeo.com/804270982/feaed82e0b</p>
<p>A promotional film recorded at the</p>	<p>https://vimeo.com/808037873/b1f5381e45</p>

premiere event (March 2023)	
A blog written by all members of the film crew	www.finerollingmedia.co.uk/telling-craniofacial-stories-perspectives-from-the-fine-rolling-team
A reflective blog by an adult representative	www.hannahsfund.co.uk/post/sally-and-the-accord-project
The Appearance Matters podcast (episode 79)	https://soundcloud.com/appearance-matters/adults-with-craniosynostosis-creating-online-resources-to-reduce-distress
Online evaluation survey	https://uwe.eu.qualtrics.com/jfe/form/SV_b1P5ljToXUs3zIY