Summary

Background:

Craniosynostosis is a relatively rare and complex craniofacial condition. Craniosynostosis and its treatment is expected to bring many challenges, and the physical and psychological wellbeing of those born with craniosynostosis and their families can be impacted. Access to appropriate information and support throughout this journey is therefore crucial for families, children, young people, and adults. Headlines Craniofacial Support is a UK-based charity dedicated to improving the lives of people affected by all types of craniosynostosis. Unfortunately, little research about the physical and psychological effects of craniosynostosis exists. This has meant that knowledge about craniosynostosis and how to best support those affected is limited.

Aims of the Project:

In 2017, Headlines published a list of the top ten priority questions for future research. In 2019, Headlines partnered with the Centre for Appearance Research, based at the University of the West of England, to apply for funding for Headlines' first major research project. A small grant was awarded to Headlines by the VTCT Foundation to address two of their priority research questions:

- **What are the long-term physical and psychological effects for individuals with syndromic and non-syndromic craniosynostosis?**
- **Are individuals with craniosynostosis likely to suffer from mental health difficulties, or are they more resilient?**

Due to the known importance of the health of the family as a whole, this project also aimed to gain a better understanding of the psychological **wellbeing of parents of children born with craniosynostosis.**

Study Design:

An online survey was designed. The survey consisted of standardised parent- and self-reported outcome measures and free-text boxes. Topics for parents of children born with craniosynostosis included: Experiences of their child's treatment, their child's wellbeing and development, their own emotional wellbeing and relationships, and experiences of support services. Topics for adults with craniosynostosis included: Emotional wellbeing, relationships, starting a family, education and employment, satisfaction with appearance, physical health, experiences of treatment, and experiences of support services. The survey was based on previous research (where available) and was reviewed by members of the specialist NHS craniofacial teams and by families and adults affected by craniosynostosis at a Stakeholder Workshop in September 2019. The survey was launched in October 2019 and remained open until April 2020.

Findings:

Parents

Parents' psychological wellbeing and relationships were considerably impacted by their child's diagnosis and ongoing treatment. Parents reported higher levels of stress and anxiety than the general population and described symptoms of medical traumatic stress. Parents reported lower levels of optimism and resilience than the general population, although many also described positive outcomes of their child's condition.

Children and Young People

Parents reported their children to have more behavioural difficulties, more emotional problems, more difficulties with peers, and more difficulties overall in comparison to the general population. However, these scores were not considered to be of immediate clinical concern. Parents also reported their children to have many physical complaints, such as hearing, vision, speech, and movement difficulties.

Adults

Adults with craniosynostosis reported higher levels of anxiety and appearance concerns than the general population and were less comfortable in adult relationships. Others lacked experience of dating altogether and felt that having craniosynostosis posed a challenge to forming meaningful social connections. Starting a family was a cause of concern for many, due to the increased risk of future generations being affected. Physical health, care coordination, making treatment decisions, and gaining employment were also highlighted as ongoing concerns in adulthood. Adults' levels of optimism and resilience were lower than the general population, yet adults also described positive aspects to their condition and their self-esteem did not appear to be affected overall.

Syndromic and Non-Syndromic Conditions

Few differences were found between individuals with syndromic and non-syndromic craniosynostosis on standardised measures, but a greater impact of having a syndrome was suggested in parents' and adults' free-text responses.

Psychological Support

Only a minority of parents and adults had received psychological support for challenges associated with craniosynostosis or had directly engaged with Headlines' existing services.

Recommendations:

There is a clear need for routine psychological screening and support for individuals and families throughout childhood and into adulthood. Routine physical assessments are also needed to identify troubling symptoms and prevent them from worsening. While the provision of support early on may increase resilience and prevent long-term distress for those currently engaged in services, ways of supporting 'older' adults requires further consideration. Education for non-specialist health professionals (such as midwives and General Practitioners) is of high importance to prevent delayed diagnosis and upsetting interactions, and to improve access to care. Going forward, Headlines could consider an evaluation of their current services against the support needs described in this report to highlight any gaps. Future research with young people, other family members (such as grandparents and siblings), and minority groups is suggested. Above all, continued collaboration between Headlines, its members, researchers, the specialist craniofacial teams, and other charities is needed wherever possible to carry out further high-quality research and to continue to improve support and long-term outcomes for individuals and families.

Acknowledgments:

It has been a pleasure and a privilege to work with Headlines Craniofacial Support on this project and we are highly motivated to continue our collaboration. We are grateful to the VTCT Foundation for funding this project and to everyone who contributed to the development of the study, including those who attended the stakeholder workshop, the Clinical Psychologists from the four highly specialist craniofacial teams, and in particular Dr Natasha Rooney from Great Ormond Street Hospital. We are especially grateful to all study participants for candidly sharing their experiences with us.

PARENTS OF CHILDREN BORN WITH CRANIOSYNOSTOSIS

111 parents took part in the study

41 years old was the average age of the parent

91% were mothers

Most parents were white, married, and educated to at least a pre-graduate level

In comparison to the general population, parents reported:

- Lower levels of resilience
- Lower levels of optimism
- Less satisfaction with their relationships
- Higher levels of stress
- More symptoms of anxiety
- More symptoms of depression

When their child was born:

- 93% of parents felt emotionally distressed
- 74% of parents were shocked by the diagnosis
- 99% of parents were worried about the future

ONLY **27%** of parents had received psychological support.

Many parents also reported:

- A traumatic birth experience
- A lack of awareness among non-specialist health professionals, such as midwives and GPs
- A delay in achieving a diagnosis for their child
- An emotional impact of craniosynostosis on siblings and grandparents
- Extreme stress at the time of their child's surgery
- Difficulties making decisions about treatment
- An ongoing burden of care
- Positive consequences of having faced these challenges

Parents provided advice to others:

- Talk things through
- Ask for help and information
- Trust your instincts
- Join support groups
- Take one day at a time
- Focus on your child's strengths
- Take time to enjoy your child
- Accept the journey will be difficult but there will be many good times too

CHILDREN AND YOUNG PEOPLE WITH CRANIOSYNOSTOSIS

111 parents reported on their child's health and wellbeing

10 years old was the average age of the children

62% The most common diagnosis was single suture craniosynostosis

Most children were white

Compared to the general population, children with Craniosynostosis had:

More **emotional** problems

More **behavioural** difficulties

More **social** difficulties

Few differences were found between children with **syndromic and non-syndromic** forms of craniosynostosis

Ongoing physical difficulties included:

Speech

Hearing

Vision

Movement

Eating and drinking

Other challenges included:

Teasing or **bullying**

Exclusion from activities

Learning difficulties

Gaining appropriate support in school

Time out of school for hospital appointments

Parents

Believed their **child was happy overall**

Didn't think craniosynostosis had held their child back in life

Felt craniosynostosis **had helped** their child develop a strong character and positive outlook

Were proud of their children for coping with challenges

ADULTS WITH CRANIOSYNOSTOSIS

36 adults took part in the study

31 years old was the average age of the adults

Most adults were white, single, and educated to at least a pre-graduate level

69% were female

Diagnoses included Crouzon syndrome, single suture craniosynostosis, Apert syndrome and Saethre-Chotzen syndrome

In comparison to the general population, adults with craniosynostosis reported:

- More fear of being judged by others
- Poorer body image
- More pressure to conform to societal appearance ‘ideals’
- Lower levels of optimism
- Lower levels of resilience
- Less satisfaction with relationships
- Less comfort in adult relationships
- More symptoms of anxiety
- Similar levels of overall self-esteem
- Few differences were found between adults with syndromic and non-syndromic forms of craniosynostosis

Compared to the general population, adults with craniosynostosis reported higher rates of:

- Eating disorders
- Self-harm
- Suicidal ideation

Ongoing physical difficulties included:

- Hearing
- Fatigue
- Vision
- Teeth
- Migraines

Adults also reported:

- An ongoing burden of self-management of physical and psychological symptoms
- Ongoing and significant difficulties accessing appropriate treatment
- Difficulties making decisions about treatment
- Difficulties gaining employment
- Concerns that future generations of their family would be affected by the condition

ONLY 3% of adults had received psychological support.

Advice to others included:

- Not being defined by your condition
- Asking for help and information
- Self-acceptance
- Self-compassion
- Findings others who are accepting of your condition
- Being determined
- Identifying the gifts that come with being different