Institution: University of the West of England, Bristol

Unit of Assessment: 3

Title of case study: Improving the global provision of psychological support for individuals and families affected by craniofacial conditions

Period when the underpinning research was undertaken: 2012 – 2019

Details of staff conducting the underpinning research from the submitting unit:

Name(s):	**Role(s) (e.g. job title):**	**Period(s) employed by submitting HEI:**
Nicola Stock	Senior Research Fellow	2010 – present
Nichola Rumsey	Professor	1990 – 2017

Period when the claimed impact occurred: 2014 – 2020

Is this case study continued from a case study submitted in 2014? No

1. Summary of the impact

One in approximately 600 babies is born with Cleft Lip and/or Palate globally. Stock and Rumsey developed an intervention framework to address the resulting psychological distress suffered by those living with this condition. This framework led to the following impacts: (i) all 16 Cleft Teams in the UK adopted the framework, resulting in improved patient care and service delivery; (ii) the UK's only national support charity for cleft developed new services, managed by a new full time post, to support adults with Cleft Lip and/or Palate; (iii) global adoption of the framework across more than twenty-five countries resulted in more effective identification and treatment of psychological issues associated with Cleft Lip and/or Palate.

2. **Underpinning research**

Research by Stock and Rumsey highlighted that patients with cleft lip and/or palate (CL/P) are at risk of a range of poor psychological outcomes such as withdrawn-depressed behaviour, social anxiety and appearance concerns (**R1**, **G1**). They proposed individually tailored face-to-face interventions using a stepped approach that involved delivering psychological care across the lifespan to optimise outcomes. Alongside other findings, the research identified that patients with CL/P require continuous support into adulthood and that there is an urgent need to further understand the influence of race, culture and social grouping on psychological adjustment to cleft. Furthermore, the research also highlighted the additional needs of children and adults from resource-poor countries with CL/P (**R1**, **G1**, **G2**).

Previous evidence on the benefits of psychological interventions for patients with CL/P was inconclusive. To address this issue, the world's largest CL/P research program, '*The Cleft Collective*', was launched in 2012, funded by the Healing Foundation and Universities and Hospital Trusts based in Manchester and Bristol. As of May 2020, the '*The Cleft Collective*' had recruited 9,032 participants from 3,223 families into their programme. Stock, in her role as a Healing Foundation Research Fellow, and Rumsey, were key members of this collaborative programme, which has provided a comprehensive resource of information about living with CL/P (**R2**, **G1**). '*The Cleft Collective*' provided the platform for clinicians and researchers to agree a method of measuring psychological adjustment to CL/P in the early years of life (**R3**, **G1**). Led by Rumsey and Stock, in collaboration with clinical psychologists, a comprehensive conceptual framework was established. This included age-appropriate measures such as parent and infant interaction, social anxiety, appearance and psychological well-being (**R3**).

Stock led the further development of the framework to understand (i) the psychological support being delivered in clinical practice, and (ii) whether the services provided were adequate from the perspectives of health professionals representing all 16 UK CL/P surgical sites (**R4**, **G1**, **G3**). The research identified common psychological concerns experienced by individuals and families affected by CL/P, alongside associated risk and protective factors for psychological distress. Additionally, the research proposed a comprehensive understanding of current clinical practice in the UK, and a framework for the standardized assessment of holistic individual and familial wellbeing (**R4**). Stock and Rumsey also achieved a UK-wide consensus in the academic and clinical measurements of psychological adjustment in CL/P for patients aged 8 years onward (**R5**, **G1**, **G3**).

Stock and Rumsey further explored the continuing psychological adjustment and support needs of adults born with CL/P, who would have typically been discharged from the cleft service at the age of 18. They identified a range of new issues related to the workplace, relationships and the heritability of cleft, indicating a need for psychological support in adulthood and continued access to treatment throughout the lifespan (**R6**, **G3**).

Further research has applied the intervention framework to other craniofacial conditions. This includes the first UK-wide study to investigate long-term outcomes in Craniosynostosis (**G4**), and a multi-million-dollar, international and multidisciplinary project to address the healthcare burden and psychological adjustment in individuals and families affected by Craniofacial Microsomia (**G5**, **G6**).

3. References to the research

R1 Rumsey, N. and Stock, N. M. (2013) Living with a cleft: psychological challenges, support and intervention. In: Berkowitz, S., ed., (2013) *Cleft Lip and Palate: Diagnosis and Management.* 3rd ed. Berlin: Springer-Verlag, pp. 907-915. https://www.springer.com/gb/book/9783642307690

R2 Stock, N. M., Humphries, K., St Pourcain, B., Bailey, M., Persson, M., Ho, K.M., Ring, S., Marsh, C., Albery, L., Rumsey, N. and Sandy, J. (2016) Opportunities and challenges in establishing a cohort ctudy: an example from cleft lip/palate research in the United Kingdom. *The Cleft Palate–Craniofacial Journal.* 53(3), pp. 317-325. https://doi.org/10.1597/14-306

R3 Stock, N. M., Hammond, V., Owen, T., Kiff, J., Shanly, A. and Rumsey, N. (2016) Achieving consensus in the measurement of psychological adjustment to cleft lip and/or palate. *The Cleft Palate–Craniofacial Journal.* 53(4), pp. 421-426. https://doi.org/10.1597/15-071

R4 Stock, N. M., Zucchelli, F., Hudson, N., Kiff, J. and Hammond, V. (2020) Promoting psychosocial adjustment in individuals born with cleft lip and/or palate and their families: current clinical practice in the United Kingdom. *Cleft Palate-Craniofacial Journal*, 57(2), pp. 186-197. www.doi.org/10.1177/1055665619868331

R5 Stock, N. M., Hammond, V., Hearst, D., Owen, T., Edwards, Z., Ridley, M. and Rumsey, N. (2020) Achieving consensus in the measurement of psychological adjustment to cleft lip and/or palate at age 8+ years. *The Cleft Palate-Craniofacial Journal.* 57 (6), pp. 746-752. https://doi.org/10.1177/1055665619898596

R6 Stock, N. M., Feragen, K.B. and Rumsey, N. (2015) “It doesn’t all just stop at 18”: psychological adjustment and support needs of adults born with cleft lip and/or palate. *The Cleft Palate-Craniofacial Journal.* 52(5), pp. 543-554. www.doi.org/10.1597/14-178

Evidence of the quality of the supporting research

G1 Rumsey, N. *Generation Cleft*, The Healing Foundation, 2012 – 2017, £2,397,123.

G2 Rumsey, N. *Appearance Matters: Tackling the physical and psychosocial consequences of dissatisfaction with appearance*, European Cooperation in Science and Technology (COST Action), 2013 – 2017, £542,635.

G3 Rumsey, N. *Funding to support the work of researchers in The Centre for Appearance Research (CAR)*, The VTCT Foundation, 2017 – 2020, £1,000,000.

G4 Stock, N. *Psychological adjustment in individuals born with craniosynostosis and their families*, VTCT Foundation UK, 2019 – 2020, £20,644.

G5 Stock, N. (Co-I) *Psychosocial Needs in Craniofacial Microsomia and Microtia,* Center for Clinical and Translational Research, Seattle Children's Research Institute, 2018-2019, £10,812.

G6 Stock, N. (Co-PI) *Craniofacial microsomia: Accelerating Research and Education (CARE)*, National Institutes for Health (USA), 2020 – 2025, £2,127,236.

4. Details of the impact

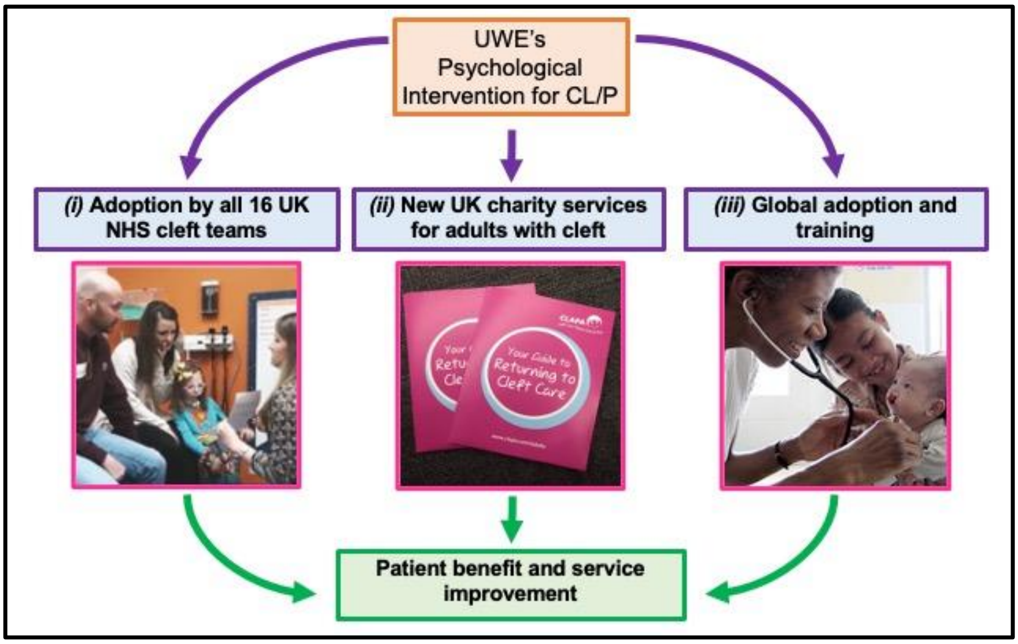

Figure 1. Summary of the impact. Image under (i) is a family from Seattle Children's Hospital craniofacial routine clinic, (ii) shows the CLAPA Leaver's Pack, (iii) is from ReSurge International (https://www.flickr.com/photos/interplast/119428083).

(i) Adoption of the framework by all 16 cleft teams in the UK to improve patient care and service delivery

The core set of measures for psychological adjustment developed by Stock in conjunction with key stakeholders (**R1**, **R3**, **R4**, **G1**, **G2**, **S1, S2**) have been implemented by all 16 UK Cleft Teams to assess all 5-year-olds. The Cleft Teams use these data, alongside the National Minimum Audit protocol, to provide key psychological evidence for intervention (**S1**). Furthermore, Stock's research has led to the formal assessment of the psychological wellbeing of parents across all NHS teams for the first time (**R4**, **S1**).

Clinics at Alderhey and the Royal Manchester Children's Hospital, for example, have adopted the framework and measures, replacing an existing screening protocol that lacked in relevant areas of psychosocial concerns. Stock and Rumsey's framework:

> '*is now used at the 5-year clinic alongside the nationally agreed minimum audit protocol…to more fully screen their psychosocial needs and to identify those at risk.*' (**S2**).

The Chair of the Cleft Psychology Clinical Excellence Network (2013-2019) and Consultant Clinical Psychologist highlighted that Stock's research also provided the foundation for service change for adult patients. The Chair reported, '*when an adult returns to the cleft service, rather than being referred straight to a surgeon or a dentist, many cleft teams will [now] schedule a psychology consultation*' (**S3**).

(ii) New UK charity services for adults with cleft

The UK charity for CL/P (CLAPA) originally focused on services aimed at children, families and young people, with minimal support provided for adults. Stock's research (**R3**, **R4**, **R6**) highlighted the psychological distress and on-going support needs of adults with CL/P. Additionally, her research provided a strong evidence base that led CLAPA to secure funding to provide services directed to previously unmet need. As a result, CLAPA established a full-time Adult Services Manager post in 2017 to develop a programme of adult services and put in place one-to-one peer support (accessed by 73 people, as of March 2019) and expand online services (**S4**, **S5**). The Adult Services Manager reported that they had developed multi-modal resources including videos and podcasts (959 views and 1456 plays, respectively, as of October 2020) (**S5**). Furthermore, young adults transitioning from routine care are now supported with a UK-wide 'Leaver's Pack' (distributed since January 2020) to address their common queries and concerns and provide information on how to access services going forward. Adults using the services reported that they felt more confident visiting the GP or dentist and understood more about cleft treatment options (**S5**). A service user with CL/P stated that the new support CLAPA provides, '*helped us to build a community and get adults the help they need*' (**S6**).

(iii) Global adaptation of the framework for patient benefit

Rumsey and Stock adapted their intervention framework for use worldwide by applying a three-tiered approach. Tier 1 facilitates teams in countries with minimal resources to explore patient-centred outcomes, while Tier 3 allows teams from the best resourced countries to comprehensively assess patient-centred outcomes, audit practice and share data worldwide. Furthermore, in 2017 Rumsey and Stock trained 468 health professionals from 15 disciplines in 26 countries in how to identify and address psychological issues in their patients using the tiered measures and intervention framework (**S7**).

The former Chief Co-ordinator of the Cleft and Craniofacial Centre, Sri Ramachandra University, India, which treats over 1000 young people each year, said that '*the tiered measures … are helping to promote patient-centred care in developing countries especially'* (**S7**). She further stated that the training provided by Rumsey and Stock in 2017 '*has made a positive difference to the way [attendees] work, focusing more on [the] patients' perspective in making treatment decisions*' (**S7**).

Stock's framework and measures have also influenced both prenatal services and adult-led services in The Netherlands (**S8**). The Amsterdam University Cleft Team now includes counselling in prenatal consultations to help communicate the diagnosis appropriately and provide support to parents (**S8**). A Professor of the Amsterdam University Cleft Team, who is the current President of the European Cleft Palate Craniofacial Association, reported that Stock's research '*has improved the way parents received information about the cleft once it was diagnosed during the 20-week ultrasound*' (**G2**, **S8**).

Stock’s framework has also been applied in the care of patients with other craniofacial conditions. The Associate Professor of Paediatrics at the Seattle Children’s Hospital and University of Washington School of Medicine drew on Stock’s work from 2014, which provided an evidence base for practice (**S9**). Expected psychological outcomes are now discussed for the first time with new families with young infants diagnosed with Craniofacial Microsomia (**G5**-**G6**, **S9**).

5. Sources to corroborate the impact

S1 Testimonial from Professor Emeritus of Orthodontics, Inaugural Dean of Health Services, Former Principal Investigator of The Cleft Collective, University of Bristol

S2 Testimonial from Clinical Psychologist, Alder Hey Children’s NHS Trust

S3 Testimonial from Consultant Clinical Psychologist at South Wales Cleft Team, Chair of Psychology Clinical Excellence Network of the Craniofacial Society of Great Britain and Ireland (CEN) 2013-2019

S4 Testimonial from the CEO of Cleft Lip and Palate Association (CLAPA)

S5 Testimonial from Adult Services Co-ordinator at Cleft Lip and Palate Association (CLAPA)

S6 Testimonial from adult with cleft and volunteer with Cleft Lip and Palate Association (CLAPA)

S7 Testimonial from the Chief Co-ordinator, Cleft and Craniofacial Centre, Sri Ramachandra University, Chennai, India

S8 Testimonial from Professor, Cleft Palate Team Amsterdam, University of Amsterdam

S9 Testimonial from Associate Professor of Paediatrics, Seattle Children’s Hospital