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Co-creation of a Novel Childhood Onset Rare Disease Self-Care Support Tool

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BACKGROUND

Children with chronic health problems are disproportionately affected by later life mental health disorders.[1–3] Treatment regimens, medical monitoring, and life under the 'shadow of threat' all contribute to poor mental health outcomes.[1,2] The negative impact on well-being is further compounded by the isolation that comes with a diagnosis of a rare disorder.[4] Key ameliorators of this negative impact are the engagement and empowerment of children in their own care.[3]

INNOVATION AND CONTEXT

Immune mediated inflammatory disorders (IMID) are characterised by regular hospital appointments, the use of immunosuppressive agents, and multi-site and multidisciplinary care. Families and carers of affected children initially handle these complex care processes, but as children approach the age of self-care they are expected to understand and manage their own care needs. In 2019, patient and family focus groups within Great Ormond Street Hospital, a national (United Kingdom) centre of care excellence for childhood IMIDs, identified the need for support around managing adolescence and expectations of self-care.[5]

A co-creation approach was undertaken for the development of a self-care tool. Using multiple existing 'patient passports' which provided useful learning, but which did not address the needs of this population,[6–10] content and format were developed through face to face structured interviews with 12 affected children and their families. The patients were aged 7yrs - 15yrs with disease duration ranging from 6 months – 10yrs, and had inflammatory eye, bowel disorders and / or rheumatological disorders. The interviews informed adaptation of

existing content, and development of novel content for the passport. The key stakeholder (families, children and young people affected by rare childhood onset inflammatory disorders) requested that the tool (1) could be personalised, (2) allowed logging of timelines of treatments and procedures and self-completed narratives on the experience of these events, (3) provided access to trusted information sources for their rare disorder, and (4) enabled documentation and descriptions of the clinical teams involved in care. The resulting tool was named 'CORDSS', the childhood onset rare disease self-care support passport. The descriptor of 'passport' was retained to reflect the "journey" described by many, ie the changing nature, over the disease course, of their lived experience as affected families.[5]

A prototype tool was piloted (n=6 stakeholder families, using the passport for 1 – 4 months), and then underwent two cycles of refinement, firstly to add additional links (embedded quick response, QR codes) to trusted information on generic health domains such as diet, health and mental well-being. The second addition allowed documentation of sources of support for families experiencing urgent care needs when in locations remote from their managing care teams. CORDSS (figure 1) is an editable electronic master document which is disseminated in paper form due to concerns around digital inequity and expressed preference for a material product. The tool is completed with input from clinical teams as considered necessary or appropriate. Setting specific adaptation is possible (for example, setting specific QR codes for disease or country specific patient groups and digital resources), but CORDSS has been designed for children with disorders characterised by rarity, chronicity, complexity, and multi-disciplinary care. Two versions of the cover page exist: one with a child-drawn image of a rainbow zebra, an icon for rare disorders, and a simpler front cover for older children, as per expressed preference. The tool has been in use since May 2022. We now present data from an initial survey of the family experience of the passport.

RESULTS

A convenience sample of n=8 families (ensuring representation of: female and male children, children and young people from the <8yrs, 8-12, and 12+ age groups); and white and non-white, specifically Black and South Asian, families) were given the tool at a clinic consultation. Children were affected by a range of disorders, comprising: sarcoidosis (n=1); inflammatory bowel disease (n=1); Blau syndrome (n=1), tubulointerstitial nephritis (n=1); and ocular inflammatory disease (n=4). A clinical nurse specialist (CT) spent between four and 20 minutes with them introducing the tool, and supporting completion of pages as directed by the families. At the following clinic consultation they were asked whether or not they would continue using the passport ("yes" / "no" / "unsure") , and asked for any comments they had about the tool. All families wished to continue using the passport (8/8, "yes" responses), and reported that completing the tool was a positive experience (sample quotes: *"I wish we'd had this at the beginning, it's been hard keeping track of everything"; "...filling it in themselves, and actually asking us....about {their} diagnosis...like {they} weren't trying to avoid talking about it for once"*). Families had used the tool to communicate with relatives, school and other healthcare professionals. Additional amendments were suggested, including future creation of an adapted tool that was parents and carer centred, and the addition of pages to which siblings could contribute if the directly affected child allowed. There were also requests for a digitised mobile version of the tool.

DISCUSSION

The co-created content and format within the passport has resulted in a tool which appears to support children and young people, and their families, in taking control of rare, chronic, complex conditions. This resource has been designed for use across different disorders, and

in different settings, with the tool introduced for use at any stage of the patient journey. The next steps will be more formal assessment of the impact of this tool as a complex intervention using outcome metrics which explore the impact on self-care behaviours and capture the well-being and global health outcomes of the child and their family. The editable version of the tool can be updated in response to changes in care delivery or disease care updates, and can be changed to address needs for site or disease specific adaptations. The Childhood Onset Rare Disease Self-Care Support (CORDSS) Tool is freely available for non-commercial use on application to the team.

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Figure 1. Images from the CORDSS tool

The authors all declare that they have no conflicts to disclose.

Declaration of Competing Interest

none