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Research Article

An Exploratory Study Examining the Long-Term Mental Health Impact of Stevens-Johnson Syndrome and Toxic Epidermal Necrolysis on Paediatric Patients and their Parents

Christopher Phillips*

Department of Dermatology, Churchill Hospital, Oxford, United Kingdom

ABSTRACT

Background: Steven Johnsons Syndrome (SJS) and Toxic Epidermal Necrolysis (TEN) are rare but devastating skin conditions. In adults, the long-term psychological sequelae are known, however, there is no such research in the paediatric population. Our study's aim was to explore the experiences of Children and Young People (CYP) and their parents during and after their illness and the subsequent physical and psychological sequelae.

Method: A retrospective qualitative study with a grounded theory approach was undertaken using detailed semi-structured interviews. 13 CYP and families who had been admitted to a tertiary referral centre were interviewed. Interview transcripts were analysed by three different researchers and themes emerging were identified.

Results: Age range of those interviewed was 10-24 years. Following discharge, patients frequently had ongoing physical issues and psychological morbidity. Common themes included inadequate information on discharge and a desire for more psychological aftercare for both CYP and parents. SJS/TEN and its causes did not appear to have been clearly explained. While there was marked heterogeneity in the experiences and impact on CYP, all parents interviewed cited symptoms of health anxiety, and some believed they had developed subsequent mental health issues.

Conclusion: Subsequent physical disability, health anxiety, psychological morbidity and little understanding of SJS/TEN were themes that were prevalent from our interviews. Families desire a more coordinated and holistic aftercare plan which may contribute to a better understanding of SJS/TEN as well as give opportunity to address the long-term physical and psychological morbidity of SJS/TEN.

Keywords: Steven Johnsons Syndrome (SJS); Toxic Epidermal Necrolysis (TEN); Health anxiety; Physical disability

INTRODUCTION

Stevens-Johnson Syndrome (SJS) and Toxic Epidermal Necrolysis (TEN) are rare but devastating skin conditions that lead to significant morbidity and mortality. The disease process in Children and Young People (CYP) has demonstrated lower mortality and higher morbidity rates when compared to the adult population [1,2], making long term management of particular importance. The underlying cause amongst the adult population is more commonly due to a medication, however, in CYP infectious causes can also contribute [3], and as such recurrences are more commonly seen [4]. Guidelines were produced for the management of SJS/TEN1 in CYP which also highlighted these important differences between adults and CYP. Whilst previous research has demonstrated psychological sequelae in surviving adults [5], these guidelines suggested a need for further research into these long-term psychological outcomes in CYP.

This qualitative study investigates the psychological impact that SJS/TEN has on CYP and their families. All CYP under the age of 18 who had been treated in Oxford over a 12-year period (2007-2019) were identified (23). Semi-structured interviews were performed by the same interviewer and where possible parents

Correspondence to: Christopher Phillips, Department of Dermatology, Churchill Hospital, Oxford, United Kingdom, E-mail: Chris.Phillips@ouh.nhs.uk

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were invited to be included in the interviews. Interviews were transcribed and then coded using *NVivo* 12 software where thematic analysis occurred. 3 were involved in the coding of the interviews. 13 (10 male, 3 female) consented to semi-structured interviews. Mean age at time of diagnosis was 6.6 years (range 3-12 years). Mean time between diagnosis and interview was 8.3 years (range 4-16 years). The cause was identified as an infection in seven cases, medication in one case and inconclusive in five cases. Duration of hospitals stay ranged between 7 and 42 days. 5 cases demonstrated subsequent recurrent episodes.

METHODOLOGY

Interview questions explored experiences of the event, care during and after discharge and long-term sequelae. 9 interviews were able to include the parents and in these cases interviews were conducted individually and with both child and parent present. This report focuses on thematic analysis of these interviews with representative quotes outlined in Table 1.

CYPs' physical experiences of their acute illness centered around significant pain and disruption of basic functions such as eating, talking and passing urine. Long-term, most still have ongoing physical complaints although many have improved with time. The most common ongoing physical complaint was ophthalmic with many requiring ongoing regular eye drops for dry eyes. Skin scarring or pigment change is frequently a problem, particularly one patient with skin Type VI Fitzpatrick classification, and 3 boys required subsequent circumcision. CYP and parents' experience of hospital and healthcare was frequently positive, expressing gratitude and positivity towards the healthcare professionals who cared for them. However, some of the most traumatic memories for both parents and children whilst in

Theme	Participant quotation
CYP's experience of psychological trauma in hospital	'About 10 of them have come in and pinned me down and literally put it (naso-gastric tube) up my nose and I was screaming and when they'd gone I just looked at my mum and I was crying. I think if I was an eight year old now in hospital I would want a doctor to tell me exactly what the prognosis is, i.e., "if you don't pick this option, this is going to happen" and let them patient sit with that for a little bit. Parents are there to deliver that type of message, transparency I think is so key.' (24-year-old female, age 9 at diagnosis)
Long-term psychological impact-CYP	'Yeah, so it (flashbacks) was mainly night-time, but it was the thought of him dying, and the thought of me dying, and his dad dying, him ending up in hospital again, yeah, he would wake up shaking, screaming, yeah.' (mother of son, age 8 at diagnosis) 'I think mostly, yeah, like missing out on school it was like missing that, making friends, learning to handwrite, socialising skills I guess, and I think that's (episode of TEN) affected me down the line.' (21-year-old-male, age 5 at diagnosis)
	'Oh, he was depressed for the whole time, yeah, totally. I mean from a child that does rugby, and football, he basically became disabled, can't walk, can't throw a ball, and definitely can't run. So he just wanted to be physically fit again basically, so he was very depressed.' (Mother of son, age 6 at diagnosis)
Long-term psychological impact-Parent	'Yeah, I don't think I'm the same person I was before it happened, for sure, and I'm just an anxious person and I definitely think I've had some post-traumatic stress from it, for sure, yeah, much more than he (child) has.' (mother of son, age 5 at diagnosis) Mother: 'I don't give him any medicine-ever, not a drop, not a cough sweet-nothing. And he hasn't had any for six years.' (mother of son, age 5 at diagnosis)
Experience of lack of longer-term psychological support	'I think there's a very strong suggestion that this may not impact you right now, but this is going to affect you, and I can't see this happening and it not affect you.' (father of daughter, age 3 at diagnosis) 'Yeah, and the aftermath of the hospitalisation was handled really poorly: We were discharged without any follow-up plans.' (mother of son, age 8 at diagnosis)
	It just felt very abrupt, that it went from very intense focused, lots of attention, to absolutely nothing.' (mother of daughter, age 6 at diagnosis)

Table 1: Exerts from qualitative interviews with CYP and their parents who had experienced SJS/TEN.

hospital were iatrogenic in nature. This was most commonly in the form of venesection or cannulation but included other interventional procedures also. One female CYP described a naso-gastric tube being inserted whilst being held down by doctors and nurses. On reflection, she felt that there was inadequate explanation given to her despite feeling she would have been old enough to understand at the time. Parents also recalled traumatic invasive interventions surrounding eye care, describing episodes of "eye-rodding" which they felt occurred without sufficient explanation surrounding the event. Negative experiences of the hospitals and healthcare such as this and others centred around poor communications. Some described, it wasn't so much the interventional procedure itself, but rather the lack of communication of how and why it was being done that caused the distress. The theme of poor communications was further demonstrated in the delivery of information. 2 parents described they were told briefly and flippantly that their child may go blind just before they underwent an invasive procedure on the eyes. This was also reflected in parents and CYP who felt poorly informed regarding what was happening whilst in hospital and in some cases they were mis-informed. One mother recalled being told by a doctor that their child was never going to have an episode of SJS again; however, 6 months later they were back in hospital with a recurrence. The mother felt that this misinformation had led to her being slower to re-present to hospital on the second occasion. General understanding of SJS/TEN was limited among interviewees and they reported that resources for this age group were lacking. There was frequently a lack of clarity over the exact cause of SJS/TEN in their case.

RESULTS AND DISCUSSION

CYP's subsequent psychological morbidity commonly manifested as night terrors and symptoms of Post-Traumatic Stress Disorder (PTSD), as well as symptoms of depression and anxiety for many years afterwards. A longer hospital stay was also more likely to correlate with psychological morbidity. Only 3 went onto get professional psychological support, with a further six retrospectively identifying that they likely had one or more of these psychological diagnoses and expressed a wish that there had been appropriate psychological support available.

Parents were able to recall the events with much more clarity than the CYPs themselves. All parents interviewed exhibited health anxiety towards their child subsequent to the event which took various forms. Some parents described being quick to go into "panic mode" when their child became subsequently sick and would have a lower threshold for presenting to emergency services. Others described themselves as being overprotective, described as "wrapping their child in cotton-wool" acknowledging this had a negative effect on their child. Parents and CYPs were aware that medications and viruses were generally implicated but most felt that uncertainty around the specific cause and likelihood of recurrence was an additional source of anxiety. Notably, even in cases where infections were reported to be the likely aetiology, most CYPs and parents showed a fear of medications with one claiming to be "terrified" of medication and some avoiding medications entirely.

In some cases, parents suffered more significant psychological morbidity with symptoms such as generalised anxiety, Post-Traumatic Stress Disorder (PTSD) and depression for many years after the event. One father interviewed described it taking him 4 years to talk about the event afterwards and only 6 years afterwards did he feel that he was now coming out of a depression. Another mother felt that she had had post-traumatic stress much more than her child had. Families perceived there was a lack of aftercare and described a sharp contrast from the intense care in hospital to discharge back home where they felt alone and unsupported with lack of follow-up care. Some reported they would have benefitted from more long-term support for their child, both in terms of improving their understanding of the condition but also their ongoing physical and emotional needs. Parents frequently described that they themselves would have benefitted from ongoing psychological support also with some finding that difficult to navigate and others describing that the need for this came much later after the event.

The study's limitations are the relatively small sample size due to the rare nature of this condition. However, response rate and engagement was good and saturation of themes was achieved after 10 interviews. The long lag-time between the acute event and interview could be also seen as a limitation. Conversely, this may also allow for more balance and reflection in the responses and allows us to identify longer-term impacts. This rich dataset of patients' voices highlights the significant long-term psychological trauma experienced by both child and parent after an episode of SJS/TEN. From these interviews it is possible to identify factors which may be contributory. SJS/TEN is a life-threatening illness sometimes requiring prolonged Intensive Care Unit (ICU) stay and subsequent PTSD in this instance has been described [6]. There are specific features of this illness, however, that may also contribute to subsequent psychological morbidity. The illness itself is mutilating and visually traumatising for patients and parents. There is often a lack of clarity surrounding the exact cause and as such parents are unclear how to avoid it happening again which can lead to over-protective behaviours. Even in cases where there is thought to be an infectious etiology, fear of medication can still exist. Having recurrent episodes or at least being aware that the condition can be recurrent can also perpetuate anxiety and fear surrounding their child's health.

How we can impact on improving psychological outcomes for these children and their parents can be divided into care given whilst they are inpatients and care given as outpatients. Whilst in hospital, providing the best clinical care through using up to date clinical guidelines for management of SJS/TEN is likely to improve psychological morbidity by reducing severity or illness and length of hospital stay. Having an awareness, however, that interventions can be psychologically traumatic is important and as such only be performed if there is a clinical need. If they are required, then time must be allowed for explanation to the parents and child in an age-appropriate way. There must also be awareness that in the case of obtaining consent for procedures in outlining risks and benefits, this may in some cases be akin to breaking bad news. As such these conversations with patients and children should be done so with appropriate time and sensitivity. Providing accurate information on the condition and individualising it to their case is important to prevent misinformation and accurate understanding of what, if any, medications should be avoided in the future. Resources have since been developed for this age-group including a template discharge letter and bad information leaflet which should be used to help address these issues [7].

Following discharge, follow up care may be difficult to co-ordinate, especially if these children and families had been transferred from another area. It is, therefore, important that an appropriate follow-up is arranged as this was a clear need expressed by families and CYP. At an outpatient appointment, there is an opportunity to go through the facts again, debrief and re-iterate the likely cause in their case. If there is uncertainty this risk can be discussed and conversations on how to manage this risk appropriately can be had. Follow up appointments are also a chance to screen for any subsequent physical health complaints. Enquiring about the patient's psychological wellbeing is also important and offering psychological support if needed. Enquiring about the parent's psychological well-being is also required as in some cases their needs in this area may be greater than the child's.

CONCLUSION

In summary, this research has revealed that the psychological burden that CYP have following SJS/TEN is significant, not just amongst CYP but their parents also. Having an awareness of this will be important for clinicians who care for these patients as they have the power to impact upon these both whilst they are inpatients but also following discharge to ensure they have access to longer-term care.

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