Reducing posttraumatic stress in parents of patients with a rare inherited metabolic disorder using eye movement desensitization and reprocessing therapy: a case study

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Abstract

Background

Parents of children with severe inborn errors of metabolism (IEMs) frequently face stressful events related to the disease of their child and are consequently at high risk for developing parental posttraumatic stress disorder (PTSD). Assessment and treatment of posttraumatic stress disorder (PTSD) in these parents is however not yet common in clinical practice. PTSD can be effectively treated by Eye Movement Desensitization and Reprocessing (EMDR), mostly offered in multiple weekly sessions which may preclude participation as parents are generally overburdened by the ongoing and intensive care for their child. We offered time-limited EMDR therapy with a maximum of four sessions over two subsequent days to two parents (from different families) of mucopolysaccharidosis type III (MPS III) patients to explore the potential effect of this approach in reducing post-traumatic stress symptoms and comorbid psychological distress.

Methods

Both qualitative and quantitative outcomes were used. The case conceptualisation, EMDR sessions and the effects reported by parents are described. The change in the severity of post-traumatic stress symptoms and comorbid psychological distress were evaluated with the Reliable Change Index (RCI).

Results

All traumatic memories and catastrophic fears of the future reported by parents were successfully processed and neutralized. Parents felt more competent to face future difficulties related to the disease of their child, and no adverse effects were reported. Quantitative outcomes showed a clinically significant decrease in post-traumatic stress symptoms and comorbid psychological distress from pre- to post treatment, and these beneficial effects were maintained at follow-up.

Conclusion

Time-limited EMDR might be a successful treatment for traumatized parents of children with mucopolysaccharidosis type III, and we suggest that this approach may have a wider application including parents of children with other severe IEMs. More awareness in clinical practice of the need for assessment and treatment of PTSD in parents of children with IEMs is essential to improve the psychosocial wellbeing of both parent and child.

Background

Inborn errors of metabolism (IEMs) constitute the largest group of disorders in childhood causing progressive intellectual and neurologic deterioration, often in the absence of a disease modifying treatment (1). Examples of such disorders are several of the lysosomal storage disorders (e.g. some of the mucopolysaccharidoses, Nieman-Pick disease type A, Gaucher's disease type 3, Sandhoff disease and Krabbe's disease), peroxisomal disorders (e.g. Zellweger spectrum disorders) and disorders of the mitochondrial respiratory chain (e.g. Leigh syndrome) (1, 2). Although these disorders are individually rare, their combined prevalence is substantial (3). Earlier studies showed that IEMs have a highly negative impact on the psychosocial functioning of parents (4–7), and that parents of children with IEMs report a lower health related quality of life compared to parents of other chronically ill children including paediatric cancer (8). This may be explained by the fact that parents of children with IEMs often have to deal with a continuous process of loss due to the progressive nature of the disease, facing an uncertain future with a realistic fear to lose their child often after a long diagnostic odyssey (15, 16). Moreover, parents frequently face potential traumatic medical events (*e.g.* during the diagnostic phase, but also related to palliative treatment and procedures related to clinical trials) followed by short- or long term stress responses (9).

Consequently, these parents are pre-eminently at risk for developing parental post-traumatic stress disorder (PTSD) (10–12). According to the 5th edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5), facing a life-threatening disease in one's child indeed qualifies as an event that may lead to PTSD (13). Parental PTSD is diagnosed when parents fulfil PTSD criteria related to their child's illness and experience symptoms such as intrusions (e.g. re-experiencing the trauma), avoidance (e.g. avoiding certain people or places that remind you of the trauma), negative alterations in mood and cognitions (e.g. distorted feelings of guilt), and hyper arousal (e.g. irritability) (10, 13).

Although the assessment of parental PTSD related to severe paediatric diseases such as cancer has gained significant interest (11, 14, 15), studies focusing on PTSD in parents of children with rarer diseases such as IEMs are lacking. Evaluating the presence of PTSD and, if needed, offering treatment may less often be considered when parents are continuously exposed to stressful events, in contrast to parents of paediatric patients who have experienced more delineated traumatic (*e.g.* a restricted period of intense treatment or an acute hospital admission). We have recently assessed post-traumatic stress symptoms in parents of mucopolysaccharidosis type III (MPS III, Sanfilippo syndrome) patients (16), a rare lysosomal storage disorder (birth prevalence of approximately 1:40.000) characterized by a developmental delay from the age of 2–4 years often followed by severe behavioural problems and progressive mental deterioration ultimately leading to severe dementia and premature death (17). Results indicated a remarkably high prevalence of PTSD (22%, compared to 3.8% in the general Dutch population (18)) in parents of MPS III patients, necessitating effective trauma treatment (16).

Until now, more than 30 randomized controlled trials (RCT) have demonstrated the efficacy of Eye Movement and Desensitization and Reprocessing (EMDR) in reducing symptoms of post-traumatic stress (19) and EMDR is recommend as treatment by the International Society for Traumatic Stress Studies [ISTSS] (20), the World Health Organization (21) and the National Institute for Health Care and Care

Excellence [NICE] guidelines in order to reduce posttraumatic stress symptoms (22). The clinical utility of EMDR for parental PTSD has, however, not yet been demonstrated. It seems likely that most parents of children with rare IEMs are not routinely assessed for the presence of PTSD in clinical practice as health care providers may not realize that caring for a progressively ill child may lead to PTSD. In addition, EMDR is generally provided in weekly sessions which may preclude participation of these parents as they are often overburdened by the complexities of parenting their child (4, 23). In order to make treatment more accessible for these overburdened parents, we offered four sessions of EMDR (1.5 hrs each) scheduled on two subsequent days, to two parents of MPS III patients (from different families). We report the preliminary effects of time-limited EMDR on an individual subject level, which is useful for critical deduction and provides important suggestions for future research (24, 25).

Methods

Participants and procedure

The two participating parents (a mother and a father from different families) were recruited by the Dutch expertise centre for MPS III (Amsterdam University Medical Centers). The age of the children was approximately 10 years. The children received the diagnosis MPS III when they were three and four years old. Parents were referred to the psychology unit in 2018 by the metabolic paediatrician of their child for treatment of presumed post-traumatic stress symptoms and had not previously received trauma treatment or other formal psychological therapy. The parents visited the hospital on three separate occasions; one visit for the intake session (1.5 hrs) and two visits in which they were offered EMDR (2 times 1.5 hrs per day). EMDR was provided by two licensed clinical psychologists (LH and CR), who have completed accredited training in EMDR (LH: level II, CR: licensed EMDR Europe Child and Adolescent trainer). Written informed consent from parents to describe their cases in the literature has been obtained. Minor details in this article have been amended to ensure patient confidentially.

EMDR therapy

EMDR therapy was delivered following the standard eight-phase protocol described by De Jongh and Ten Broeke (26). During the intake session, history taking consisted of a standardized case conceptualization assessing a hierarchy of stressful memories or flash forwards (a mental representation of a feared catastrophe) related to the disease of their child.

During EMDR, parents were asked to focus on the most distressing image related to the selected memory or flash forward (target image), eliciting the dysfunctional negative cognition (NC) related to the image, as well as accompanying emotions and somatic sensations. In the desensitization phase, parents focused on the target memory, while simultaneously focusing on an external distracting visual or tactile stimulus, namely following a light-bar with their eyes and/or holding buzzers in their hands. At regular intervals the parents had to rate the target memory with the subjective units of disturbance (SUD) score, with 0 = ‘no disturbance’ and 10 = ‘worst disturbance possible’, until the target memory was no longer disturbing (SUD = 0) and a functional cognition was rated a 7 on the 7 point Validity of Cognition (VOC)

scale, with 1 = ‘totally unbelievable’ and 7 = ‘totally believable’. At the end of the session the therapist checked for and processed any residual disturbing body sensations, followed by a positive closure and evaluation (26).

Assessments

Parents completed reliable, validated questionnaires to measure posttraumatic stress symptoms (related to the disease of their child) and comorbid psychological distress prior the start of EMDR (T0), one-month post treatment (T1) and at follow-up (T2, mean duration of six months after treatment).

Severity of post-traumatic stress symptoms was measured by the Dutch Impact of Event Scale-Revised (IES-R; Kleber & De Jong, 1998) (27, 28). The IES-R consists of 22 items, rated on a four-point scale according to how often each post-traumatic stress symptom has occurred in the past 7 days (0 = not at all, 1 = rarely, 3 = sometimes, 5 = often). The total score ranges from 0-110, where a higher score indicates more post-traumatic stress symptoms.

Comorbid psychological distress was measured using the Brief Symptom Inventory (BSI)(29, 30). The BSI consists of 53 items that assesses different psychological symptoms (including the symptom dimensions Somatization, Obsession-Compulsion, Interpersonal Sensitivity, Depression, Cognitive issues, Anxiety, Hostility, Phobic anxiety, Paranoid ideation and Psychoticism) rated on a five-point Likert scale (0 = none, 1 = some, 2 = quite, 3 = quite a lot, 4 = extremely). The total score consists of the mean score on all items, where a higher score indicates more symptoms.

Statistical analysis

First, the qualitative data of the case conceptualisation, content of the EMDR sessions and effects reported by parents were described. Second, the reliable change index (RCI), which controls for coincidence or error, was calculated for pre- to post treatment change scores on the IES-R and BSI. RCIs > 1.96 indicates a significant change at $p < .05$, suggesting a reliable change. In the current study, a *clinically* significant change is considered when the post treatment score falls within the range of the mean score minus/plus two standard deviations of the normative population (31). To calculate the RCI for posttraumatic stress scores, the SD and the test-retest reliability (α) of the norm scores of Olde et al. (32) were used, with SD = 13.0 and α = 0.88. To calculate the RCI for comorbid psychological distress, the SD and the test-retest reliability (α) of the norm scores of De Beurs et al. (33) were used, with SD = 0.72 and α = 0.97.

Results

Qualitative results: Case conceptualisation and EMDR sessions

Case parent 1 (father)

The most important psychological symptoms reported by the father were irritability, unable to tolerate bright light and loud sounds, sleeping problems, fatigue, troubles concentrating and remembering, feelings of sadness, and feelings of guilt towards a healthy sibling. The disturbing memories and flash forwards reported by father are listed in Table 1. During the first day of EMDR therapy (2 sessions), three out of four traumatic memories and flash forwards were processed until a SUD score of 0 and VOC score of 7 per memory was achieved. The residual flash forward was processed at the first session of the second treatment day. The fourth session was superfluous, due to the fact that all memories were neutralized (SUD = 0). The total duration of the EMDR therapy was 4,5 hours. The father reported feeling very surprised by the positive effect of the treatment in such a short amount of time. In the first couple of days after the EMDR session, he thought more about a number of the traumatic memories, both during day and night-time. However, afterwards he felt less easily irritated, tolerated bright light and loud sounds better and was more able to concentrate at work. Although the father still had the feeling that he had to divide his attention between his child with MPS III and the healthy sibling, he felt better equipped to balance the care for his children. He divided the attention for the ill child and healthy sibling more equally with his partner, which made him feel less guilty. Fatigue was still present after EMDR, but he did not experience feelings of sadness anymore. Father expressed to feel resilient and competent to face future difficulties related to the disease of his child. No adverse events occurred.

Case parent 2 (mother)

At baseline, this mother reported the following psychological symptoms; irritability, troubles concentrating and completing tasks, binge eating, feelings of sadness, worrying, and not being able to enjoy the interaction with her children. The disturbing memories and flash forwards that were reported during the intake session are presented in Table 1. Four out of six memories with the highest SUD score were already fully processed (SUD = 0 / VOC = 7 per memory) during the first treatment day. The other two flash forwards were processed during the third and fourth session on the second treatment day. She reported an immediate effect after the first treatment day. She felt more cheerful, was less easily irritated, was able to stop binge eating and managed to finish tasks. Moreover, she had more attention for the healthy sibling and was better able to enjoy the interaction with her children again. She still suffered from fatigue, but noticed that she did not have to sleep during the day anymore. Finally, she was better able to ignore worrying thoughts and felt more competent to handle future stressful events related to the illness of her child. No adverse events occurred.

Table 1
Most stressful memories and flash forwards related to the IEM of their child.

Stressful memories	
Case parent 1 (father)	*Case parent 2 (mother)*
Comforted the child in the hospital, saying that everything would be okay after a minor ENT operation (grommets). Now the diagnosis MPS III is known, it became clear that 'everything would not be okay at all' (failure as parent, SUD 7)	The paediatrician communicated the diagnosis MPS III to the parents (SUD 9)
The paediatrician communicated the diagnosis MPS III to the parents (SUD 6)	Termination of a subsequent pregnancy because the foetus was diagnosed with MPS III (SUD 8)
	Announcement from the hospital that the clinical trial (enzyme replacement therapy(34)), in which the child participated, was prematurely terminated (loss of hope, SUD 7)
	Attending the funeral of another MPS III patient (SUD 6)
Flash forwards	
Funeral of their child with MPS III (SUD 9)	Her child in a vegetative state with palliative care by deep sedation and withholding of fluids (SUD 10)
Image of the child in a special disability-inclusive transport necessitating a lot of medical equipment (tubes) and making repetitive movements and screaming sounds (SUD 8)	Her child in a wheelchair, no longer able to communicate by laughing, eye contact or movements (SUD 8)
	Sudden death of the child (SUD 7)
Note. SUD = Subjective units of disturbance score.	

Quantitative results

Case parent 1 (father)

Measurement post treatment (T1) showed a clinically significant decrease in post-traumatic stress symptoms and comorbid psychological distress compared to T0 (Table 2). Improvement was maintained at three months follow up (T2).

Case parent 2 (mother)

Measurement post treatment (T1) showed a clinically significant decrease in post-traumatic stress symptoms and comorbid psychological distress compared to T0 (Table 2). Improvement was maintained at nine months follow up (T2).

Table 2
Reliable change index (RCI) for posttraumatic stress and comorbid psychological distress.

	T0	T1	T2	RCI		
IES-R total score (0-110)				T0-T1	T0-T2	T1-T2
Parent 1	67	24	12	6.75*	8,64*	1.88
Parent 2	62	12	3	10.36*	10.52*	0.157
BSI total mean score (0–4)						
Parent 1	1,40	0,30	0,40	6,24*	5,67*	0,567
Parent 2	1,53	0,28	0,26	7,09*	7,02*	0,113
**Significant decrease between measurements at p < 0.001.*						

Discussion

This study reports the effects of a time-limited EMDR therapy in two different parents of MPS III patients. A maximum of four sessions of EMDR scheduled over two subsequent days resulted in a significant decrease of post-traumatic stress symptoms and comorbid psychological distress in these parents. Moreover, no adverse effects were reported.

These positive outcomes are comparable to the effect of EMDR in adults with PTSD related to personal traumatic events (35). The size and persistence of the effects at follow up in our study are remarkable, especially in the context of the progressive and grim course of the disease in patients with the common phenotype of MPS III, generally causing ongoing daily stress for the whole family (4, 16). Most people who are treated with EMDR are trying to cope with traumatic symptoms resulting from past events, delineated in time, in which there is a post trauma safety situation whereas parents of progressively ill children will experience multiple traumas during the course of the disease, and may even more suffer from threats of expected future medical crises and an early death. Although the cases described in this study only relate to parents of MPS III patients, we expect that a short course of EMDR may also be beneficial for parents of children with post-traumatic stress symptoms related to other rare, severe and progressive diseases for which there is still no disease modifying treatment. Future randomized controlled trials with larger sample sizes are necessary to validate the potential effect of EMDR in this specific population.

Early and effective treatment of parental PTSD is essential, both for the health of the parents as well as for the child. The presence of even a few post-traumatic stress symptoms can have a significant influence on various parenting domains, such as the ability to respond in a sensitive manner to the needs of the child or parenting satisfaction, which may all negatively impact the psychosocial functioning of the child (36–38). It has been shown in other paediatric populations, such as paediatric cancer, that

parental post-traumatic stress symptoms were associated with psychosocial problems (e.g. behaviour problems) in the child (39). We were not able to measure the psychosocial functioning of the children in our study by using the Child Behavior Checklist (CBCL, 1,5–5 years) because parents struggled to complete this questionnaire as most of the items were not applicable given the severity of the cognitive, social-emotional and physical impairment of their child. Considering the phenotypic heterogeneity of many rare IEMs, individualized specific measures instead of generic questionnaires could be considered as more appropriate outcomes in future research.

The scarcity of reports on parental PTSD in parents of children with rare progressive disorders is probably caused by underdiagnoses. Parents of severely ill children with multiple clinical problems may be less inclined to discuss their own health concerns as they prioritize the wellbeing and the clinical symptoms of their child. Our results, showing promising effects of time-limited EMDR therapy, underline the importance of structural (and continuous) screening of posttraumatic stress symptoms in this population of parents. Structural screening by patient reported outcomes measures (PROMs) may help to detect parents at risk for parental PTSD. For instance, parents can be asked to regularly complete the short screening instrument 'PTSD Checklist for DSM 5 (PCL-5)' (40) at home prior to a scheduled visit. Moreover, the traumatic memories and flash forwards outlined in Table 1 of this study may provide valuable information for medical and psychological professionals about potential traumatic events for parents of children with rare progressive disorders. Since both parents indicated that receiving the diagnosis of their child was one of the most stressful memories, we are convinced that an early referral to a health psychologist at the time of diagnosis should be standard care, which is common practice in less rare childhood disorders, such as paediatric cancer and cystic fibrosis (41, 42).

One limitation of this study was that the timing of the follow-up measurements was not equal for both parents. One participant (the mother) completed the third measurement at nine months instead of three months post-treatment, following several reminders by the researcher. This illustrates that even the completion of questionnaires may take a lot of effort of parents who are often overburdened by the intensive and complex care for their ill child. Therefore, the use of a brief screening instruments at standard times in future research is recommended.

Conclusion

This is the first study that shows the potential effects of EMDR as a time-limited treatment for parents experiencing ongoing post-traumatic stress reactions related to the disease of their child. More awareness in clinical practice for assessment and treatment of PTSD in parents of children with progressive rare disorders is essential to be able to improve the psychosocial wellbeing of both parents and the child.

Abbreviations

BSI

brief symptom inventory
EMDR
eye movement and desensitization reprocessing
IEM
inborn error of metabolism
IES-R
impact of events scale revised
MPS III
mucopolysaccharidosis type III
PTSD
posttraumatic stress disorder
RCI
reliable change index
RCT
randomized controlled trial
SUD
subjective units of disturbance
VOC
validity of cognition

Declarations

Ethics approval and consent to participate

Review of the Medical Ethical Committee was not applicable as treatment was part of standard care in clinical practice.

Consent for publication

Informed consent for publication was obtained from all participants of the study using institutional consent forms.

Availability of data and materials

Data that support the findings of this study are available from the corresponding author on reasonable request due to privacy restrictions.

Competing interests

TC, LH, FW and CR have no competing interest to declare.

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Not applicable.

Authors' contributions

CR and LH provided EMDR to parents and carefully recorded all information during the sessions. TC made substantial contributions to the acquisition and analysis of the data, interpretation of the data and wrote the first draft of the manuscript.

LH, FW and CR made substantial contributions to interpretation of the data and critically reviewed and revised the manuscript for important intellectual content. All authors read and approved the final manuscript.

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