Do parents of children with metabolic diseases benefit from the Triple P - Positive Parenting Program? A pilot study

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Abstract

Background: Parents of children with metabolic diseases report more parenting stress, anxiety, depression and dysfunctional parenting styles than parents of children without metabolic diseases. In addition, their children have more behavioral problems. Beside the fact that meta- bolic diseases are rare, they form a relatively large proportion in the morbidity and mortality of chronically ill children.

Methods: In this pilot study 14 parents of children with metabolic diseases, aged between 2.5 and 13 years, participated in a quasi-experimental pre-post-follow-up study.

Results: After participating in the Level 4 Group Triple P-program there were small effects in decreasing child behavioral problems and large effects in decreasing dys- functional parenting styles. There was a moderate to large reduction of parental stress and a large reduction of parental anxiety. Only the effects on the behavioral problems and the parenting style 'laxness' were no longer significant at 6 months follow-up.

Conclusions: In summary it can be said that the existing Triple P-program has good effects, with a great degree of satisfaction, for parents of children with metabolic dis- eases in reducing dysfunctional parenting styles, parent- ing stress and behavioral problems of their children. One should not wait for a specialized program to reach these parents, but further research is necessary as a greater effect can be expected when this program is adapted to these parents.

Keywords: behavioral problems; children with metabolic diseases; parental anxiety; parental stress; parenting styles; Triple P-program.

Introduction

Childhood chronic illnesses threaten the health and development of children and their families [1]. These children develop more emotional, behavioral and social problems [1–10]. Moreover these emotional and behavioral problems place children at a higher risk of child abuse and neglect [1]. The parenting style plays an important role in prevent- ing and dealing with these problems [11]. But families at risk, such as families of chronically ill children, use more dysfunctional parenting styles [8].

In addition to regular parenting issues, parents of chronically ill children are also facing the

medical care of their child [12]. This results in more parenting stress [4, 9, 12–15], depression and anxiety [9, 12, 16–19]. All these factors result in a decline of the quality of life of the family [1–7, 12–18] and in a decline of parents' self-efficacy for managing their child's disease [1, 9]. The impact on chil- dren and families depends on the severity and the nature of the chronic disease [1]. Parents of children with meta- bolic diseases report an even lower quality of life, due to the uncertain course and limited life expectancy of many metabolic diseases [6]. In addition behavioral, emotional and psychological problems are more prevalent in chil- dren with metabolic diseases [20–22]. The reasons why this population is more vulnerable is not yet elucidated and must be based in part on the complexity of the under- lying disease with, for instance, stressful dietary interven- tions and a degree of neurobiological impairment [20]. But despite their rare occurrence metabolic disorders form a relatively large proportion of the morbidity and mortal- ity of chronically ill children [23].

It is important that this group of families is properly assisted. However, there is a paucity of evidence-based parenting approaches for them [1]. The Positive Parenting Program (Triple P) is an existing, multi-level, evidence- based parenting program for parents of children up to 16 years of age [24–26]. It focuses on five principles of pos- itive parenting. As Triple P has proved to be effective in many family contexts in reducing dysfunctional parenting styles and behavioral problems of children [26–29] and in improving parental emotional well-being (less stress and depression) [26, 29] and given the vulnerability of parents of children with metabolic diseases the following study hypotheses raised: after attending the Triple P- program there is a decline of behavioral problems of children with metabolic diseases, there is a decline of dysfunctional par- enting styles, parenting stress, depression and anxiety.

Patients and methods

This pilot study used a quasi-experimental pre-post-follow-up design.

Patients

Nine parents of six children with metabolic diseases were included in 2008 and five parents of five children with metabolic diseases were included in 2012 (Table 1). The focus of this study was on preadolescent children, aged between 2.5 and 13 years, as different developmental issues arise as children transit into adolescence and self-management of their condition becomes increasingly important [1]. Recruitment took place in the Centre for Inherited Metabolic Disorders in Antwerp (CEMA). The included metabolic disorders were: sucrase-isomaltase deficiency, succinyl CoA:3-oxoacid CoA transferase deficiency, mito- chondrial respiratory chain defect, galactosemia, pyruvate dehydrogenase complex deficiency, methylmalonic academia, urea cycle defect and congenital lactic academia. Exclusion criteria were: chil- dren without a metabolic disease, children who did not go to school, children in special needs schools (except type 8) and parents who had insufficiently mastered the Dutch language.

Expected parameters, such as effect size, standard deviation (SD) or difference in values were unknown for primary outcomes, as this was the first study of its kind. The sample size, calculated to detect an effect size of 0.7 at 80% power, was 19 participants.

Methods

All the parents of the children who were followed in the CEMA were contacted by phone for participation. There was a participation rate of 12.2%.

Results

The parents participating in the Triple P-program reported that their children had significantly fewer emotional

The parents who participated in 2012, each followed at least 60% of all sessions. Parents who missed a session, usually through acute illness of their child, were asked to go through the missed sub- jects at home. The data from the parents of 2008 are missing.

The number of missing data were: 2.31% without drop-outs in the 'pre-test' questionnaires, 3.06% without drop-outs in the 'post- test' questionnaires and 2.86% with four drop-outs in the 'follow-up' questionnaires. The study protocol was approved by the Committee of Ethics of the University of Antwerp.

Measures

The 'pre-test', 'post-test' and 'follow-up' questionnaires, each existed of four standardized questionnaires. The 'post-test' questionnaire also contained a satisfaction questionnaire.

Family background questionnaire: This questionnaire, developed by Turner et al. in 2002 [30], collected the family demographics.

Strengths and Difficulties Questionnaire (SDQ): This questionnaire screens for children at risk of psychosocial problems. The parent version for children from 4 to 16 years was used. The questionnaire consists of 25 statements in five subscales with acceptable internal consistency [31].

Parenting scale: This scale measures dysfunctional parenting styles in 30 items and a seven-point Likert-scale. It consists of a total score and three factor scores [32]. The original version of the scoring key was used as it has higher values of internal consistency, predic- tive validity and test-retest reliability [33].

Depression -Anxiety-Stress (DAS) scale: To assess the emotional wellbeing of parents the DAS scale was used. It consists of three sub- scales with adequate internal consistency. Parents scored 42 items on a seven-point Likert-scale [34].

Satisfaction questionnaire: This questionnaire, which is an adap- tation of the Client Satisfaction Questionnaire [35], consists of 12 questions around the received information and help, how well the program met the needs of the parent and child and whether they would recommend the program.

Data analysis: Analyses were undertaken using SPSS 20.0. To test the hypotheses, paired t-tests and Cohen's d were used. We chose to use the Cohen's d instead of the Bonferroni correction because weak effects may also have biological importance in behavioral science but will not pass the Bonferroni correction, which leads to publication bias [36, 37]. Multiple linear regressions were used to assess whether the socio-demographic variables affected the outcome values. A mul- tiple imputation was used to correct for the missing data. All tests were carried out with a level of significance of $p < 0.05$.

Firstly, parents filled in ‘pre-test’ questionnaires, afterwards they followed the 8-week Level 4 group Triple P-program for parents of children up to 12 years. The program ended with completing the ‘post-test’ questionnaires. Six months later ‘follow-up’ question- naires were sent by (e-)mail.

size at post-intervention and a large effect size at 6 months follow-up (Table 2). At the population level, it is important to determine whether the Triple P-program helped people from the clinical range to achieve a normal score.

The ‘clinical behavioral problems’ (reduction of 7.2% at post-intervention, 28.6% at follow-up), ‘clinical emo- tional problems’ (reduction of 21.4% at post-intervention, 54.3% at follow-up), ‘clinical problems with peers’ (reduc- tion of 35.7% at post-intervention, 47.1% at follow-up) problems (large effect size at 6 months follow-up), conduct problems (small effect size at post-intervention), prob- lems of ‘hyperactivity and attention deficit’ (small effect size at post-intervention, medium effect size at 6 months follow-up) and problems with peers (large effect size at 6 months follow-up). There was no significant reduction of ‘problems with prosocial behavior’. The total problem score of the Strengths and Difficulties Questionnaire (SDQ)-scale improved significantly, with a medium effect and ‘clinical problems with pro-social behavior’ (reduc- tion of 7.2% at post-intervention, 14.3% at follow-up) all decreased. There was a decrease of 21.4% from a clini- cal score to a borderline score of ‘hyperactivity’ at post- intervention and a decrease of 30% to normal scores at follow-up.

The second hypothesis could be confirmed. The dys- functional parenting styles – laxness, overreactivity and verbosity were all improved statistically significantly at post-intervention and at 6 months follow-up with large effect sizes. Only ‘laxness’ was no longer significant at follow-up (Table 2). There were also decreases in the clini- cal dysfunctional parenting skills at post-intervention and at follow-up: a decrease of 25.7% and 23.4% for ‘laxness’, a decrease of 30.8% and 27.4% for ‘overreactivity’ and a decrease of 40.6% and 41.4% for ‘verbosity’, respectively.

The third hypothesis was partially supported. The subscale ‘stress’ improved significantly at post-inter- vention and at 6 months follow-up with, respectively, a medium and large effect size. ‘Anxiety’ also improved with a large effect size at follow-up. In addition, the total score improved initially with a small effect to a large effect size at follow-up (Table 2). The results reveal a clinical decline of 7.2% in the number of parents who are stressed at post-intervention and a decline of 14.3% at follow-up.

As possible confounders the following were studied: gender of the parent, relationship to the child, current marital status, having a paid job, seeking help from a psychologist, psychiatrist, social worker or other, age and gender of the child. Only the gender of the child had influence on the emotional issues of this child at 6 months follow-up ($t = 3.42$ $p = 0.011$), this is because the emotional problems between boys and girls differed already before the start of the program ($t = 3.69$, $p = 0.003$).

In the experimental group 75% of parents, who partic- ipated in 2012, was satisfied with the received information and help. However, only 25% of parents felt strong enough to cope with future educational problems. A quarter of parents would like to see the program more focused on the metabolic condition of their child. The data regarding the satisfaction of the parents,

who participated in 2008, are missing.

Discussion

After completing the Triple P-program parents of chil- dren with metabolic diseases report a small reduction of behavioral problems of their children. This is consistent with findings in children without metabolic diseases

[26–29], but the effects in our study are smaller compared to a meta-analysis [27]. However, behavioral, emotional and psychological problems are more prevalent in chil- dren with metabolic diseases [20–22]. Especially children with metabolic diseases of the 'intoxication type', like phenylketonuria or respiratory chain defects, experi- ence more disruptive behavior disorders and externaliz- ing problems [21]. Normally Triple P has good effects in non-metabolic sick children with disruptive behavior and attention/hyperactive difficulties [38]. In our study, 55% of children had a respiratory chain defect or metabolic disease of the 'intoxication type', but none of them had phenylketonuria which is mainly associated with attention deficit hyperactivity disorder [21]. Forty-five percent of children had an energy metabolism disorder, and these experienced fewer externalizing problems than the other groups [21]. Thus, it is possible that the current Triple P-program insufficiently meets the specific behavioral problems of children with metabolic diseases. Interest- ingly Triple P has proven to be effective in reducing inter- nalizing or externalizing behavioral problems in children with type 1 diabetes [39]. The fact that the 'problems with prosocial behavior' did not improve could be attributed to insufficient focus of the Triple P-program on this item. Nevertheless the reduction in behavioral problems can be explained by a reduction of dysfunctional parenting styles, which maintain or worsen behavioral problems [32], or by imposing less problematic behavior or more positive behavior by the child [29]. It is unclear which factor has the greatest influence as behavioral problems are complex and determined by characteristics of the child [32].

The large reductions in dysfunctional parenting styles is in keeping with past research in parents of children without metabolic diseases [26–29]. These studies show that the total score of the parenting scale has a greater reduction than the subscore 'overreactivity', which in turn decreases more than 'laxness' and 'verbosity'. However, in this study, 'laxness' has in the first instance a greater reduction than 'verbosity', but this decline is not sus- tained at follow-up. One possible explanation is that parents of children with metabolic diseases used, before the program, the parenting style 'laxness' more, because they attributed the misconduct of their child to their illness [7]. It is striking that the Triple P-program in this study has greater effects on the dysfunctional parenting styles than a meta-analysis [28]. One possible explana- tion is that parents of children with metabolic diseases, through their great access to the health care, already received some parenting support and could implement the information more quickly.

The medium to large reduction of stress among parents of children with metabolic diseases is not surpris- ing as literature shows that the Triple P-program mainly responds to the stress level [28]. As a metabolic disease is a life-long condition, even small reductions in stress across time can be fruitful, and may have indirect health benefits for parents. The large

reduction of the anxiety at 6 months' follow-up is surprising as parents of children with metabolic diseases face other fears than parents of children without metabolic diseases, because of the sometimes limited life expectancy and uncertain course of some metabolic disorders [6].

Some subscales of the SDQ-scale (i.e. 'hyperactivity and attention deficit' and 'total problem score'), the sub- scale 'stress' and the 'total score' of the DAS scale have a further increase of the effects at follow-up. The literature is contradictory here. Some studies claim that the effects sustain rather than improve, while others indicate that there is an improvement [29]. The increase of the effects in this study could be explained by the fact that parents of children with metabolic diseases were more motivated to implement the strategies because they want their child to experience a good development, despite their disease. The continuation or improvement of the standard care or better coping with the disease, may also contribute to an increase of effects.

Limitations

Firstly, because of the rarity of metabolic disorders (incidence 40/100 000 [40]), there was a modest sample size whereby the statistical power of this study is limited. Because of the lack of epidemiological data regarding the prevalence of behavioral problems of children with meta- bolic diseases, dysfunctional parenting styles and paren- tal emotional wellbeing the impact on a population level is unclear.

A second limitation is that participation in this study was voluntary, so there is possibly a non- responder bias. The participants may not have been a representative sample of the population as regards to the type of metabolic disease (e.g. there is no patient with phenylketonuria), ethnicity and social-economic status. Parents of children with metabolic diseases are more often consanguineous due to the autosomal reces- sive inheritance of the diseases and more often both of the parents do not speak the Dutch language, in which the Triple P-program was given. We could try to contact interpreters if this is the case. A third limitation is that the inclusion of the participants took place at different times, which can lead to bias due to changing financial and living conditions. In particular, limited financial resources may be a reason why parents could not participate. On the other hand, participation was free of charge, which made the program more accessible but may have led to the participation of less motivated parents.

The low percentage of parents, who participated in all sessions, is a fourth limitation, as less guidance leads to diminished effects. After a missed session parents were asked to read the missed advice and so received all the contents of the program. To increase the participation, one could opt to choose individual counselling whereby parents can reschedule appointments. A disadvantage is that the peer interaction is lost. We can also try to provide care for the children during the sessions. Another option to further enhance the recruitment of parents could be to offer the program as usual care and raise awareness among parents about the importance of good quality of life for the whole family.

A next limitation is that the results were obtained by self-reporting. This gives greater effects than obser- vational instruments [29] and could have led to overes- timation. Another cause of

overestimation, were it is not corrected for, are the effects of regular care and previous parenting support. In addition, because of the absence of a valid control group, it is unclear whether the effects were obtained by the program itself or by other aspects such as peer contacts. Furthermore, no account is taken of the type of metabolic disease and associated behavioral problems, the severity, duration and life expectancy of the condition and whether or not the offered advice and exer- cises were applied.

A final limitation is that the Triple P-program is a 'do- program' for which parents need energy. This can be prob- lematic for parents of children with metabolic diseases as they are taxed more heavily in daily life.

Suggestions for further research

Some people propose that parenting programs should focus on generalized illness aspects [1]. Nevertheless we found that only a minority of the parents in our study felt strong enough to solve future educational problems and that the Triple P-program is not responsive to their specific needs. Specific requirements described in lit- erature are: learning to cope with being the carrier of the responsible gene [41] and dilemmas about future pregnancies [23, 41], the need of more information about food-related problems [6, 41], learning to deal with the uncertain course of the disease [6], the long-term effects [42] and the limited life expectancy of some metabolic disorders [6]. It could be interesting to subsequently organize further meetings after finishing the level 4 group Triple P-program, so parents can discuss with their peers how they implement the lessons learned in daily life with regard to the metabolic disease of their child. Parents who can give tips to other parents are also increased in their sense of competence, which increases their resilience. To improve the sustainability of the positive effects after the program it is possible that the parents may benefit from a more intensive parenting support program (such as level 5), additional support such as home visits and individual counselling or a telehealth refreshment course at 3–6 months after the program. Because 40% of the children with a metabolic disease has a below average intelligence [21], it is prom- ising to investigate the effect of the Stepping Stones Triple P program for families of a child with a disability [43, 44] in this group. Also the Self-Directed Teen Triple P-program shows promising results on disease-related conflicts in children with type 1 diabetes [45]. Further- more it is interesting to examine whether the effects will still be present in adolescence. It is useful to extend this study to other age groups and parents of children with other chronic diseases, like type 1 diabetes, as the program probably will work for them to.

Applying the Triple P-program in these groups of parents could lead to a reduction in health costs and social problems. However, more cost-effectiveness studies are needed.

References

1. Morawska A, Calam R, Fraser J. Parenting interventions for child- hood chronic illness: a review and recommendations for inter- vention design and delivery. J Child Health Care 2015;19:5–17.

2. Zashikhina A, Hagglof B. Mental health in adolescents with chronic physical illness versus controls in Northern Russia. Acta Paediatr 2007;96:890–6.

3. Hysing M, Elgen I, Gillberg C, Lie SA, Lundervold AJ. Chronic physical illness and mental health in

children. Results from a large-scale population study. J Child Psychol Psychiatry 2007;48:785–92.

4. Adams CD, Streisand RM, Zawacki T, Joseph KE. Living with a chronic illness: a measure of social functioning for children and adolescents. J Pediatr Psychol 2002;27:593–605.

5. Ray LD. Parenting and childhood chronicity: making visible the invisible work. J Pediatr Nurs 2002;17:424–38.

6. ten Hoedt AE, Maurice-Stam H, Boelen CC, Rubio-Gozalbo ME, van Spronsen FJ, et al. Parenting a child with phenylketonuria or galactosemia: implications for health-related quality of life. J Inherit Metab Dis 2011;34:391–8.

7. Antshel KM, Brewster S, Waisbren SE. Child and parent attribu- tions in chronic pediatric conditions: phenylketonuria (PKU) as an exemplar. J Child Psychol Psychiatry 2004;45:622–30.

8. Morawska A, Gregory C, Burgess S. Parental beliefs about behaviour problems of their asthmatic children and interven- tions to support parenting. J Child Health Care 2012;16:75–90.

9. Mitchell AE, Fraser JA, Ramsbotham J, Morawska A, Yates P. Childhood atopic dermatitis: a cross-sectional study of relationships between child and parent factors, atopic dermatitis management, and disease severity. Int J Nurs Stud 2015;52:216–28.

10. Agrawal J, Kumar R, Malhi P, Dayal D. Prevalence of psycho- social morbidity in children with type 1 diabetes mellitus: a survey from Northern India. J Pediatr Endocrinol Metab 2016;29:893–9.

11. Botello-Harbaum M, Nansel T, Haynie DL, Iannotti RJ, Simons- Morton B. Responsive parenting is associated with improved type 1 diabetes-related quality of life. Child Care Health Dev 2008;34:675–81.

12. Major DA. Utilizing role theory to help employed parents cope with children's chronic illness. Health Educ Res 2003;18:45–57.

13. Cappelleri JC, Gerber RA, Quattrin T, Deutschmann R, Luo X, et al. Development and validation of the WEll-being and Satis- faction of CAREgivers of children with diabetes questionnaire (WE-CARE). Health Qual Life Outcomes 2008;18:6:3.

14. Kakleas K, Kandyla B, Karayianni C, Karavanaki K. Psychosocial problems in adolescents with type 1 diabetes mellitus. Diabetes Metab 2009;35:339–50.

15. Hauenstein EJ. The experience of distress in parents of chroni- cally ill children: potential or likely outcome? J Clin Child Psychol 1990;19:356–64.

16. Rodrigue JR, Geffken GR, Clark JE, Hunt F, Fishel P. Parenting satisfaction and efficacy among caregivers of children with diabetes. Child Health Care 1994;23:181–91.

17. Mullins LL, Wolfe-Christensen C, Pai AL, Carpentier MY, Gillaspy S, et al. The relationship of parental overprotection, perceived child vulnerability, and parenting stress to uncertainty in youth with chronic illness. J Pediatr Psychol 2007;32:973–82.

18. Lindstrom C, Aman J, Norberg AL. Increased prevalence of burn- out symptoms in parents of chronically ill children. Acta Paediatr 2010;99:427–32.

19. de Silva KS, de Zoysa P, Dilanka WM, Dissanayake BS. Psycho- logical impact on parents of children with congenital adrenal hyperplasia: a study from Sri Lanka. J Pediatr Endocrinol Metab

2014;27:475–8.

20. Smith I, Knowles J. Behaviour in early treated phenylketonuria: a systematic review. Eur J Pediatr 2000;159:S89–93.

21. Simons A, Eyskens F, De Groof A, Van Diest E, Deboutte D, et al. Cognitive functioning and psychiatric disorders in children with a metabolic disease. Eur Child Adolesc Psychiatry 2006;15:207–13.

22. Trifiletti RR, Packard AM. Metabolic disorders presenting with behavioral symptoms in the school-aged child. Child Adolesc Psychiatr Clin N Am 1999;8:791–806.

23. Waisbren SE, Rones M, Read CY, Marsden D, Levy HL. Brief report: predictors of parenting stress among parents of children with biochemical genetic disorders. J Pediatr Psychol 2004;29:565–70.

24. Kleefman M, Jansen DE, Reijneveld SA. The effectiveness of Stepping Stones Triple P: the design of a randomised controlled trial on a parenting programme regarding children with mild intellectual disability and psychosocial problems versus care as usual. BMC Public Health 2011;11:676.

25. Sanders MR. Triple P-Positive Parenting Program: towards an empirically validated multilevel parenting and family support strategy for the prevention of behavior and emotional problems in children. Clin Child Fam Psychol Rev 1999;2:71–90.

26. Sanders MR, Kirby JN, Tellegen CL, Day JJ. The Triple P-Positive Parenting Program: a systematic review and meta-analysis of a multi-level system of parenting support. Clin Psychol Rev 2014;34:337–57.

27. de Graaf I, Speetjens P, Smit F, de Wolff M, Tavecchio L. Effectiveness of the Triple P Positive Parenting Program on behavioral problems in children: a meta-analysis. Behav Modif 2008;32:714–35.

28. de Graaf I, Speetjens P, Smit F, de Wolff M, Tavecchio L. Effective- ness of the Triple P positive parenting program on parenting: a meta-analysis. Fam Relat 2008;57:533–66.

29. Nowak C, Heinrichs N. A comprehensive meta-analysis of Triple P-Positive Parenting Program using hierarchical linear mod- eling: effectiveness and moderating variables. Clin Child Fam Psychol Rev 2008;11:114–44.

30. Turner KM, Markie-Dadds C, Sanders MR. Facilitator's manual for group Triple P (Edition II revised). Milton, QLD: Triple P Inter- national, Pty.Ltd., 2002.

31. Goodman R. Psychometric properties of the strengths and difficulties questionnaire. J Am Acad Child Adolesc Psychiatry 2001;40:1337–45.

32. Arnold DS, O'Leary SG, Wolff LS, Acker MM. The parenting scale: a measure of dysfunctional parenting in discipline situations. Psychol Assess 1993;5:137–44.

33. Salari R, Terreros C, Sarkadi A. Parenting scale: which version should we use? J Psychopathol Behav Assess 2012;34:268–81.

34. Lovibond SH, Lovibond PF. Manual for the depression anxiety stress scales, 2nd Ed. Sydney, NSW: Psychology Foundation of Australia, 1995.

35. Sanders MR, Markie-Dadds C, Tully LA, Bor W. The Triple P-Positive Parenting Program: a comparison of enhanced, stand- ard, and self-directed behavioral family intervention for parents and children with early onset conduct problems. J Consult Clin Psychol 2000;68:624–40.

36. Garamszegi LZ. Comparing effect sizes across variables: gener- alization without the need for Bonferroni correction. Behavioral Ecology 2006;17:682–87.

37. Nakagawa S. A farewell to Bonferroni: the problems of low statistical power and publication bias. Behavioral Ecology 2004;15:1044–45.

38. Bor W, Sanders MR, Markie-Dadds C. The effects of the Triple P-Positive Parenting Program on preschool children with co-occurring disruptive behavior and attentional/hyperactive difficulties. J Abnorm Child Psychol 2002;30:571–87.

39. Westrupp EM, Northam E, Lee KJ, Scratch SE, Cameron F. Reduc- ing and preventing internalizing and externalizing behavior problems in children with type 1 diabetes: a randomized con- trolled trial of the Triple P-Positive Parenting Program. Pediatr Diabetes 2015;16:554–63.

40. Applegarth DA, Toone JR, Lowry RB. Incidence of inborn errors of metabolism in British Columbia, 1969–1996. Pediatrics 2000;105:e10.

41. Read CY. Using the impact of event scale to evaluate psychologi- cal response to being a phenylketonuria gene carrier. J Genet Couns 2004;13:207–19.

42. Lambert C, Boneh A. The impact of galactosaemia on quality of life – a pilot study. J Inherit Metab Dis 2004;27: 601–8.

43. Roux G, Sofronoff K, Sanders M. A randomized controlled trial of group Stepping Stones Triple P: a mixed-disability trial. Fam Process 2013;52:411–24.

44. Shapiro CJ, Kilburn J, Hardin JW. Prevention of behavior prob- lems in a selected population: Stepping Stones Triple P for parents of young children with disabilities. Res Dev Disabil 2014;35:2958–75.

45. Doherty FM, Calam R, Sanders MR. Positive Parenting Program (Triple P) for families of adolescents with type 1 diabetes: a ran- domized controlled trial of Self-Directed Teen Triple P. J Pediatr Psychol 2013;38:846–58.

Table 1: Socio-demographic, medical and psychological characteristics of the study population

	Study population
Parents: n	14
Gender: n (%)	
Man	3 (21.4%)
Woman	11 (78.6%)
No answer	0 (0%)
Relationship to the child: n (%)	

Mother/Father	13 (92.2%)
Stepmother/Stepfather	1 (7.1%)
Foster mother/Foster father	0 (0%)
Other	0 (0%)
No answer	0 (0%)
Civil status: n (%)	
Married	5 (35.7%)
Cohabiting	7 (50%)
Divorced	2 (14.3%)
Separated	0 (0%)
Never married/cohabiting	0 (0%)
Single	0 (0%)
Widow/widower	0 (0%)
No answer	0 (0%)
Paid job: n (%)	
Yes	13 (92.9%)
No	1 (7.1%)
No answer	0 (0%)
Help from a psychologist, psychiatrist, social worker or other in the last six months : n (%)	
Yes	3 (25.0%)
No	9 (60.0%)
No answer	2 (13.3%)
Children: n	**11**
Gender child: n (%)	
Boy	5 (45.5%)
Girl	6 (54.5%)
Metabolic disease of the child: n (%)	
Congenital lactic acidemia	1 (9.1%)
Sucrase-isomaltase deficiency	1 (9.1%)
Succinyl CoA: 3-oxoacid CoA transferase deficiency	1 (9.1%)
Mitochondrial respiratory chain defect	2 (18.2%)
Methylmalonic acidemia	1 (9.1%)
Urea cycle defect	1 (9.1%)
Galactosemia	2 (18.2%)
Pyruvate dehydrogenase complex deficiency	2 (18.2%)
Age of the child in years:	
Mean M *(SD)*	5.72 *(3.03)*

Table 2: Effect of the Triple P-program at post- and follow-up intervention in the experimental group

	Pre (n=14) M *(SD)*	Post (n=14) M *(SD)*	FU (n=10) M *(SD)*	Effect Pre – Post t	p	Effect score Cohen's d	Effect Pre – FU t	p	Effect score Cohen's d
Strengths and Difficulties Questionnaire (SDQ)									
Emotional problems	5.00 *(3.55)*	4.21 *(2.35)*	2.09 *(0.84)*	0.97	0.330	0.26	3.02	0.003*	1.13
Conduct-problems	1.93 *(1.68)*	1.21 *(1.33)*	1.32 *(0.77)*	2.69	0.007*	0.48	1.26	0.210	0.47
Hyperactivity & attention deficit	5.86 *(3.13)*	4.64 *(2.73)*	4.13 *(1.91)*	2.80	0.005*	0.42	1.97	0.050*	0.67
Problems with peers	2.64 *(2.00)*	2.07 *(2.13)*	1.00 *(0.96)*	1.26	0.208	0.28	3.68	0.000*	1.05
Problems with prosocial behavior	7.71 *(1.68)*	8.21 *(1.53)*	8.30 *(0.90)*	-1.71	0.087	-0.31	-1.33	0.183	-0.50
Total problem score	15.43 *(7.26)*	12.14 *(5.48)*	8.64 *(2.43)*	2.20	0.028*	0.51	3.84	0.000*	1.25
Impact score	2.14 *(2.15)*	1.55 *(1.56)*	0.41 *(0.68)*	1.91	0.057	0.31	2.90	0.004*	1.08

Parenting scale									
Laxness	27.01 *(5.40)*	22.13 *(5.11)*	24.50 *(4.13)*	2.85	0.004*	0.93	1.59	0.112	0.52
Overreactivity	27.67 *(6.81)*	21.88 *(6.49)*	20.05 *(6.36)*	3.89	0.000*	0.87	5.12	0.000*	1.16
Verbosity	25.29 *(4.65)*	21.14 *(5.02)*	20.90 *(3.26)*	2.30	0.022*	0.86	2.57	0.010*	1.09
Total score	86.65 *(10.44)*	71.90 *(14.82)*	72.59 *(13.07)*	3.94	0.000*	1.15	3.64	0.000*	1.19
Depression-Anxiety-Stress scale (DASS)									
Depression	3.71 *(3.98)*	2.27 *(4.14)*	1.78 *(3.22)*	1.73	0.083	0.35	1.85	0.064	0.53
Anxiety	2.21 *(1.83)*	1.57 *(2.24)*	0.83 *(1.20)*	1.21	0.226	0.31	2.04	0.042*	0.89
Stress	7.64 *(5.66)*	4.71 *(5.78)*	3.70 *(3.64)*	2.47	0.013*	0.51	2.72	0.007*	0.83
Total score	13.57 *(10.22)*	8.92 *(10.69)*	6.50 *(6.66)*	2.51	0.012*	0.44	2.46	0.014*	0.82

* $p < 0.05$