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Evaluation of a newly introduced parenting support programme for families of children with 22q11.2 Deletion Syndrome

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Abstract

Aims

To evaluate the implementation of a parenting programme for parents of children with 22q11.2DS at a specialised 22q clinic.

Methods

Both parents of 8 children, age 6-10 years, with 22q11.2DS and reported behavioural difficulties were invited to take part in an 8-week parenting programme. Questionnaires were administered at three time points: prior to, immediately after, and at six-month follow-up after the programme to measure behavioural and emotional difficulties, parental stress and satisfaction. Parents set goals for themselves and for their children for the duration of the programme and provided written feedback.

Results

The course was completed by 5 families. No significant difference was found over time with regards to quantitative measures. However, parents indicated significant move towards achieving their individual parenting and child-related goals. Parents reported that participation in the group provided peer support and shared experiences. The group provided sense of understanding and belonging.

Discussion

The favourable feedback supports future integration of a parenting programme for 22q11.2DS families as part of an integrated care pathway for rare disorders.

Introduction

22q11.2 deletion syndrome (22q11.2DS) is the commonest microdeletion syndrome affecting 1:2000-1:3000 individuals[1]. The clinical manifestation varies, including a spectrum of congenital abnormalities and developmental delay. Autism spectrum disorder, attention deficit hyperactivity disorder (ADHD) and other neurodevelopmental conditions are diagnosed in 30-40% of the children with 22q11.2DS. From early childhood to adulthood affected individuals are at higher risk of presenting with behavioural difficulties, anxiety and mood disorders. About 30% of the 22q11.2 population experience symptoms of psychosis throughout their life span[2]. The complex clinical presentation of 22q11.2DS requires specialist care across the lifespan and support for caregivers. Research among parents shows high level of perceived burden, with behavioural difficulties and psychiatric illness presenting as most challenging[3].

A specialised 22q paediatric clinic is currently run in one of the paediatric hospitals in Ireland, supported by many disciplines including psychiatry. Previous research carried among the parents whose children attend the specialised 22q clinic explored specific needs and guided parenting support initiatives over time. The theme of managing behavioural difficulties frequently emerged, including the need of a parenting programme as part of the care for families provided by the clinic[4].

Skills training and emotional support (e.g. parenting programme) are integral part of the support interventions for family members and carers dealing with challenging behaviour and are included in clinical guidelines[5]. Parenting programmes are widely used by services providing autism care[6,7]. Parents Plus Children's Programme (PPCP, 6-12 years) is an evidence-based parenting programme developed in Ireland, aiming to promote positive parenting, reduce behavioural problems and improve the parent-child relationship by using structured theoretical modules and sets of practical modelling videos[8]. PPCP is used in various community settings to support parents of children with behavioural difficulties and suitable to be delivered online. Our aim was to introduce PPCP to our parent group and explore potential benefits in the context of the 22q11.2DS cohort.

Methods

Parents of 8 children with 22q11.2DS, age 6 to 10 years, with previously reported behavioural challenges and no history of intellectual disability or autism, accepted the offer to participate. The programme consisted of 8 two-hour sessions, including an introductory session held in person and 7 online sessions which followed the PPCP manual. Self-report questionnaires were used as measures at three different time points: prior to (T1), immediately after (T2), and at six-month follow-up (T3). Strengths and Difficulties Questionnaire (SDQ) contains 25 items identifying positive and negative behaviours in children calculating total difficulty score (TDS, score of above 17 is clinically significant). Parental Stress Scale (PSS, range 18-90) – measuring perceived level of stress (higher scores indicating higher level of stress), and Kansas Parental Satisfaction Scale (KPSS, range 3-21) – measuring satisfaction with self and the child (higher scores meaning higher level of satisfaction) were used. Parents were asked to identify individual and child-related goals and rate how close they were to reach goals at the start and in the end of the programme. Free text feedback was also obtained after each session. To support adherence parents were given weekly tasks to work on with their children between sessions. SPSS vs27 was used for statistical analysis.

Results

Five families completed the course. Sessions were attended predominantly by the mothers who took part in the evaluation. Mean baseline SDQ score was 18.2 (SD 5.07), PSS – 38.2 (SD 7.36), KPSS – 13.8 (SD 3.27). No significant difference was found at T2 and T3 for SDQ, PSS and KPSS. However, parents indicated significant move towards achieving their individual parenting and child-related goals (Table 1).

	T 1 (Baseline measures)	**T 2 (Post completion)**	**F_1**	**T 3 (6-month follow-up)**	**F_2**
SDQ Total Difficulties	18.2 (5.07)	19.0 (2.92)	0.083	18.4 (6.66)	0.591
Emotional Symptoms	3.8 (2.59)	3.8 (2.28)	0.218	3.8 (1.79)	1.000
Conduct Problems	2.4 (0.55)	2.4 (0.55)	0.789	2.8 (1.64)	0.541
Hyperactivity	8.2 (0.45)	8.2 (1.64)	0.272	8.6 (1.67)	0.621
Peer Problems	3.8 (2.59)	4.6 (1.67)	0.216	3.2 (2.17)	0.426
Prosocial	5.6 (1.82)	5.0 (3.32)	0.222	4.6 (2.88)	0.413
PSS	38.2 (7.36)	43.2 (10.71)	0.054	42.2 (9.20)	0.157
KPSS	13.8 (3.27)	14.4 (4.04)	0.798	15.3 (4.50)	0.613

Parenting Goals	2.50 (0.85)	5.90 (2.09)	0.018*
Child-related Goals	3.50 (2.36)	6.10 (1.96)	0.016*

Table 1. Means, Standard Deviation (in parentheses) and paired sample t-test for time effects
(F_1, F_2 and p-value). *$p < 0.05$

Free text written feedback from the participants demonstrated that the topics were relevant and helped parents reflect on their parenting approach: “I have better understanding of small actions I can take to help my child”; “I learned how to tune in with my child and connect through play”; “I learned how to react to inappropriate behaviour”. Parents found value in sharing their experiences with other parents of children with 22q11.2DS and being part of a support group: “This course was different as the facilitators had working knowledge of 22q”; “I learned from other 22q parents, … it was more relatable.”

Discussion

Parenting programmes are tailored to address the needs of parents in various contexts. This study aimed to evaluate the introduction of PPCP for parents of children with 22Q11.2 DS, commonly characterised by behavioural and emotional difficulties. Although there was no statistically significant change in the objective outcomes (SDQ, PSS and KPSS scores), parents reported finding personal value in participating in a support group of parents facing similar circumstances and learning from each other. There was a considerable positive shift in achieving set goals. Within the six months after the programme four of the children received a formal ADHD diagnosis and one child – autism diagnosis, which could account for the absence of statistically significant changes in the measured outcomes and the requirement for more specialised interventions and treatment.

Limitation of this pilot is the small sample size. The initial trial was offered to a smaller than usual group of parents to allow for better engagement with the group online and breaking into small groups for certain tasks. The online delivery was preferable to give equal access to parents despite geographical differences. Three families dropped out, the most common reason being child care demands, work commitments and needing to address other personal priorities.

The favourable feedback suggests that there is an opportunity for further exploration of the benefits of parenting group work for 22q11.2DS families and expanding towards other patient groups as part of an integrated care pathway for rare disorders.

Declarations of Conflicts of Interest:
None declared.

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