

Unresolved grief in parents of children with CF: A pilot randomised controlled trial on the use and delivery sequence of disease-related education and psychotherapeutic support

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1 **Unresolved grief in parents of children with CF:A pilot randomised**
2 **controlled trial on the use and delivery sequence of disease-related**
3 **education and psychotherapeutic support**

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28

29 **ABSTRACT**

30 Diagnosis of chronic disease in a child can result in unresolved grief (UG) in parents. This
31 study aimed to evaluate the efficacy of psychological insight-oriented therapy (IOT) as a
32 treatment for UG compared to disease related education in parents of children with cystic
33 fibrosis (CF). Sequence of delivery, first IOT then disease related education (or *vice versa*)
34 was also examined, to let all participants experience both interventions.

35 Parents were screened for UG. Parents with UG were randomised to either five one-hour
36 sessions of IOT or five one-hour sessions of education. Measures were assessed pre-
37 intervention, after the first intervention period (primary efficacy assessment), and after the
38 second intervention period (swapping intervention).

39 Forty-seven parents were screened of which 46.8% (22/47) had UG. Median duration of UG
40 was 5 years (range: 6 months to 14 years). Anxiety (50% vs. 20%, $p = 0.03$) and stress (59%
41 vs. 28%, $p = 0.03$) were significantly more prevalent in parents with UG. There was no
42 difference between arms in the odds of UG resolving either following the first intervention
43 period (OR 0.88; 95%CI 0.5, 1.5) or the second intervention period (OR 0.91; 95%CI 0.5,
44 1.6). While not statistically significant, adjusted mean values of all nine mental health
45 measures were lower in the IOT (first) arm compared to the ED (first) arm, following the first
46 intervention period.

47 *Conclusions:* UG is a significant burden for families affected by CF. Provision of disease
48 related education and psychological support, regardless of sequence, can result in resolution
49 of grief.

50 *Trial registration number:* ACTRN12621000796886, *date of registration* 24 June 2021,
51 *retrospectively registered.*

52 **KEYWORDS**

53 Cystic fibrosis, diagnosis, grief, parental response, education, psychotherapy.

54

55 **INTRODUCTION**

56 Cystic Fibrosis (CF) is an autosomal genetic and life limiting disorder that affects
57 approximately 70,000 people [1; 2]. In many countries, CF is commonly diagnosed through
58 newborn screening [3]. The news of a CF diagnosis usually is a shock for families and
59 coincides with an important period of laying down the early foundations for parent-child
60 attachment[4], thereby potentially compromising these important relational bonds.

61

62 Parents report the period directly following diagnosis to be a time of shock, disbelief, grief
63 [5; 6], heightened emotional experiences and difficult thoughts [7]. This is also typically a
64 time of intense CF related education and engagement with a multidisciplinary team. For some
65 parents the initial disease related education has been found to be an overwhelming experience
66 in which coping capacities can vary greatly [7; 8].

67

68 Parents may dissociate or push away the information about diagnosis during this particular
69 period of crisis and struggle to reach a stage of resolution or acceptance about the diagnosis
70 [9]. The emotional process of resolution requires an acceptance of the emotional pain and
71 other feelings that arise with these news, as well as the acceptance of the long-term aspect of
72 parenting a child with these challenges [9]. As the features of being unresolved are closely
73 associated and parallel with many features of grieving, for the purposes of this study, we will
74 refer to a lack of resolution as unresolved grief (UG)

75

76 Prolonged UG can have long-lasting effects on the quality of the parent-child relationship in
77 terms of the parent-child attachment, and consequently on the child's developing sense of self
78 [10; 11]. Specific and targeted interventions to address parental unresolved grief following a
79 diagnosis of CF have not been studied to-date.

80

81 Insight oriented psychotherapy (IOT) aims to help patients develop new insights about their
82 suffering, and to bring about positive change in a person's internal world and state of mind.
83 In the context of potential UG in parents of children with diagnosed CF, IOT on the parental
84 level could be a valuable intervention for affected families.

85

86 We aimed to:

- 87 1. Identify (and quantify) a lack of resolution of UG in parents, following their child's
88 diagnosis with CF
- 89 2. Evaluate the effectiveness of IOT compared to disease related education in treating UG
- 90 3. Examine the sequence of delivery of IOT followed by disease related education (or *vice*
91 *versa*) in treating UG
- 92 4. Examine the impact of IOT and ED on anxiety, depression and stress.

93

94 **METHODS**

95 *Study design*

96 In this pilot study, participants were screened for UG. Those with UG were then randomised
97 to a pre-post design to assess two interventions.

98 *Setting, participants and screening*

99 Parents of children with CF aged 6 months to 18 years and living within, or close to, the
100 Perth (Australia) metropolitan area were identified through the only paediatric CF service in
101 Western Australia and approached with or mailed information about the study. In total 105
102 families were approached and 7 opted out of being contacted further by the study team.
103 Parents who gave written informed-consent to participate in the study were screened for grief
104 in relation to their child's diagnosis with CF with the Reaction to Diagnosis Interview (RDI)
105 [12] administered during a one-hour interview with an RDI-trained psychologist. The
106 interview was video recorded and later analysed by a separate investigator who was trained in
107 the analysis of the RDI interview. Parents with UG continued to randomisation, with one
108 parent enrolled per affected family.

109 *Randomisation*

110 Following enrolment, participants were randomised to receive either IOT (IOT arm) or
111 education (education arm). Randomisation was carried out by a third party (hospital
112 pharmacy) using randomised blocks of four.

113

114 Following the first period of intervention, participants had a one-to-five week break,
115 depending on participant availability, before moving on to the alternate intervention. The
116 decision to give each participant the opportunity to experience both interventions was based
117 on recommendations from clinicians in the hospital's CF service who felt it best all
118 participants were able to experience the IOT component of the research as parents of patients
119 had no other means of direct access to such services through the paediatric hospital setting.
120 Participants were assessed again for UG and emotional wellbeing following each period of
121 intervention. Therefore, participants were assessed for UG and emotional wellbeing at three

122 time-points pre (screening), post (after intervention one), and follow-up (after intervention
123 two).

124

125 *Interventions*

126 **Intervention One (insight-oriented therapy; IOT):** Comprised five one-hour sessions of
127 IOT spread out within a minimum of five weeks. The aim of short term IOT was to assist
128 parents to gain a deeper understanding of the impact of their UG surrounding their child's
129 diagnosis on their day-to-day coping, their interpersonal relationships, and their distress
130 surrounding the future of their child, with the aim of bringing about psychological change
131 which would lead to a sense of resolution in relation to their child's CF diagnosis.

132 **Intervention Two (disease related education; hereafter 'education'):** Comprised five one-
133 hour sessions of CF related education spread out over a period of at least five weeks.
134 Education was provided by experienced members of a CF care team including a paediatric
135 respiratory physician, CF nurse, physiotherapist, gastroenterologist, and dietician, providing
136 one-on-one information sessions about their areas of expertise in CF. The basic framework
137 for each session is provided in online resource 1. Sessions allowed for questions from
138 parents, and the information provided was tailored to parents' needs. The education sessions
139 aimed to address basic CF related information that parents may not have been receptive to at
140 initial meetings with the clinical CF care team shortly after having been informed about the
141 diagnosis, and that may not have been re-visited with subsequent routine clinical care visits.
142 The overall setting of education sessions could be described as optimistic and caring.

143

144 *Outcome measures*

145 Resolution of UG formed the primary outcome measure. Other assessed measures of
146 emotional wellbeing included anxiety, depression, and stress, measured using the Depression,
147 Anxiety, Stress Scale (DASS [13]), and the Parent Stress Index (PSI [14]).

148

149 *Data analysis*

150 Descriptive statistics were calculated for the full sample and per arm. Basic between-group
151 comparisons (at post and follow-up time-points) were made using a Chi-squared test or a
152 Student's t-test, as appropriate.

153

154 The primary analysis of whether UG become resolved, was assessed using a logistic
155 regression model, with an odds ratio (OR) and 95% confidence interval (95% CI) reported for
156 the odds of grief resolving in the IOT arm relative to the ED arm. For the other outcomes
157 (emotional wellbeing measures), data from the end of the first period (post) were analysed
158 using a linear regression ANCOVA framework, where the post-time-point measurement was
159 the dependent variable and the pre-time-point measurement was included as an independent
160 variable in the model. All regression models were adjusted for gender of the parent, age of
161 the parent, duration of grief, and, where appropriate the initial measurement of the dependent
162 variable for the period of analysis.

163

164 The secondary analysis of the follow-up time-point data were carried out via two different
165 approaches; firstly, via the same framework as for the primary outcome using only data from
166 the post- and follow-up time-points in place of the pre- and post-time-points (respectively)

167 and, secondly, using data from the pre- and follow-up time-points using a ‘sequence’ variable
168 to quantify overall change based on sequence of interventions.

169 Because of the exploratory nature of this pilot study, the analysis does not focus on the
170 statistical significance of the between arm (intervention) differences and instead focuses on
171 the magnitude of effects and variability; *p*-values are presented for some comparisons as a
172 further quantification of the observations against the underlying hypothesis of no difference
173 between arms. All analyses were carried out in using a reproducible research framework
174 implemented within R [15].

175 *Ethics*

176 The study as evaluated and approved by the West Australian Child and Adolescent Health
177 Service Research Ethics Committee RGS2520.

178

179 **RESULTS**

180 The CONSORT diagram is presented in Figure 1. Briefly, 47 parents (42 female) were
181 screened for eligibility. Anxiety was present in 34% (16/47), depression in 28% (15/47), and
182 stress in 40% (21/47). Of the 47, 22 (48%) had UG and were randomised. For these 22, the
183 median duration of UG was 5 years (range: 6 months to 14 years). Compared to the parents
184 without UG, parents with UG had higher levels of anxiety (50% vs. 20%, *p* = 0.03) and stress
185 (59% vs. 28%, *p* = 0.03). No difference was observed for depression. The remaining results
186 relate to the 22 participants that were randomised, of which twelve were randomised to IOT
187 and ten were randomised to education. Cohort descriptive statistics, by arm, are presented in
188 Table 1; the mean age of parents was 37.4 years, and the parents were predominantly female
189 (19, 86.3%).

190

191 **Primary outcome – Unresolved grief**

192 In the IOT arm, UG became resolved in four (of the 12) parents and remained unresolved in
193 four parents; four parents dropped out of the study before completion of this period of the
194 study. In the education arm, UG resolved in seven (of the 10) parents and remained
195 unresolved in two parents; one parent dropped out of the study before completion of this
196 period of the study. In the primary outcome analysis, those in the IOT arm had a non-
197 significant, reduced adjusted odds (OR 0.88; 95% CI 0.5, 1.5), of their grief resolving by the
198 end of the first intervention period.

199

200 Of the eight remaining parents in the IOT arm, UG resolved in an additional two parents and
201 returned in one, following the second period where they accessed the education intervention.
202 Of the nine remaining parents in the education arm, UG resolved in an additional one parent
203 and returned in one, following the second period where they accessed the IOT intervention.
204 Those in the IOT arm had a non-significant, reduced adjusted odds (OR 0.91; 95% CI 0.5,
205 1.6), of their grief resolving by the end of both intervention periods.

206

207 **Mental health outcomes (*post initial-intervention*)**

208 Mean values for all eight mental health outcomes (scales and totals), were all lower post-
209 invention compared to pre, in both arms (Table 2, Figure 2). This decrease was only
210 statistically significant for DASS-Stress in the IOT arm (-2.12; 95% CI -3.57, -0.68).

211 At the end of the first intervention period, the IOT arm had lower (though the confidence
212 intervals included the value of no difference) mean values for seven of the eight mental

213 health outcomes (DASS-Depression being the exception, mean difference estimate 0.0), see
214 Table 3, Figure 3. The difference estimates ranged in magnitude from -0.6 (95% CI; -3.3, 2.1)
215 for DASS-Stress to -4.2 for both PSI- Parent-Child-Dysfunctional Interaction and PSI-Total.

216

217 **Secondary analysis (*follow-up; post alternate-intervention*)**

218 Mean values for most mental health outcomes (scales and totals), were lower at the follow-up
219 time-point compared to the post time-point, in both arms (Table 2, Figure 2); exceptions
220 being DASS-Anxiety and PSI-Parent-Child-Dysfunctional Interaction in the IOT arm, noting
221 that this was following this cohort's education intervention.

222 At the end of the second intervention period, the IOT arm (who had just received education)
223 had higher (though the confidence intervals included the value of no difference) mean values
224 for six of the eight mental health outcomes (PSI-Defensive Response and PSI-Parent-Child-
225 Dysfunctional Interaction being the exceptions), see Table 3. The analysis of both
226 intervention periods (with follow-up time-point as the dependent variable, and overall
227 sequence (IOT then education) being the variable of interest) yielded no significant
228 differences for the eight mental health outcomes; five of the eight outcomes had a mean
229 between-sequence difference of <1.0 unit in magnitude.

230

231 **DISCUSSION**

232 Parental unresolved grief (UG) related to their child being diagnosed with CF was highly
233 prevalent amongst parents, and for some this had been present over a decade post diagnosis.
234 Anxiety and stress were more frequently present in parents with UG. IOT and CF education
235 (in either delivery sequence) appeared to mitigate parental grief and resulted in resolution of

236 grief. Whilst increased rates of depression and anxiety and higher levels of stress have been
237 well described in patients with CF and their caregivers, and UG related to other chronic and
238 permanent conditions has been described [16-20], to our knowledge this is the first study to
239 investigate UG related to a diagnosis of CF and also the first clinical trial to investigate
240 methods to facilitate resolution of such grief.

241

242 In our study population grief remained unresolved for up to 14 years; this extended duration
243 of grief is similar to grief related to loss of a close family member [21; 22], which is also
244 associated with adverse mental health outcomes, insecure attachment [23], and has been
245 demonstrated to associate with adverse effects on the child's developing sense of self [10;
246 11].

247

248 The high percentage of resolution of grief seen following the relatively brief interventions
249 applied in our study is encouraging, and may have implications for other conditions. For
250 example, substantial levels of UG has been reported in parents of children with other
251 permanent conditions such as Down syndrome, autism spectrum disorders, cerebral palsy,
252 epilepsy, and phenylketonuria [16-20].

253

254 The signal for resolution of grief was supported by strong signals for improvement of
255 comorbidities. All parents with both unresolved grief and depressive symptoms at screening
256 reported a decrease in depressive symptoms once both interventions were completed. Also,
257 ninety one percent of parents with both unresolved grief and anxiety at screening experienced
258 a reduction in anxiety symptoms once both interventions were completed. Increased rates of
259 depression and anxiety and higher levels of stress have been well described in caregivers of

260 children with CF [16-20]. Therefore, improvements in both anxiety and stress levels
261 following resolution of grief was encouraging.

262

263 The study had a number of limitations. Firstly, the sample size was limited. However, results
264 clearly showed that grief that had been present for years resolved for multiple participants
265 following the relatively brief study period. For some participants the limited intervention may
266 not have been enough to fully explore the depth of their feelings regarding their child's
267 diagnosis and result in resolution of grief, hence the resolution of grief seen with the limited
268 intervention was encouraging. Secondly, study participants were mostly mothers. The
269 implications of our findings to fathers therefore require further study. Thirdly, only one
270 certified RDI coder was used to analyse the data to make the UG diagnosis. Despite this, the
271 reduction in 'coder assessed UG' coincided with a substantial reduction in mental wellbeing
272 indicators, supporting the study's ability to objectively assess change following intervention.

273

274 The present study also demonstrated that revisiting CF related education proved to assist
275 parents to resolve UG. Clearly, offering CF related education to parents shortly after
276 diagnosis (i.e. during a time of high stress, shock and disbelief) may not be conducive
277 towards the information being taken on board and processed, and might result in distortion of
278 information. Providing parents with an opportunity to revisit CF education after a period of
279 adjustment may enable parents to obtain clarity and reprocess information more accurately.

280 The measurement of UG using the RDI is both time consuming and costly, it requires
281 clinicians to be trained in the administration and scoring of this complex measure. A recent
282 study by Sher-Censor *et al* [24] examines a less costly and more time-effective self-reported

283 measure of parental resolution, the Reaction to Diagnosis Questionnaire (RDQ), which
284 assesses parental resolution in relation to the child's diagnosis and looks to show promise.

285

286 In summary, UG is highly prevalent in parents of children with CF, can last over many years
287 and is associated with mental health comorbidities; however, it can potentially be resolved
288 with education and/or psychotherapy. Results suggest the need to investigate how to improve
289 practices around CF diagnosis and incorporate grief identification and appropriately targeted
290 intervention for parents. These results have potential application for other chronic conditions
291 diagnosed in infancy.

292

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374 **TABLES**

375 **Tab. 1 – Cohort descriptive statistics**

	IOT^a Arm *	Education Arm *	Full Sample
n	12	10	22
Age of parents in years median (range)	36.3 (6.8)	38.5 (7.9)	37.4 (7.2)
Gender (female : male)	10 : 2	9 : 1	19:3
Duration of grief	5.2 (4.4)	5.3 (4.8)	5.3 (4.5)

376 ^aIOT - insight oriented psychotherapy

377 * mean (sd), or count

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391 **Tab. 2 – Descriptive statistics for the mental health outcome measures**

Assessment	Subscale	IOT ^a Pre *	ED ^b Pre *	IOT ^a Post *	ED ^b Post *	IOT Follow-up (after ED ^b) *	ED ^b Follow-up (after IOT) *
n		12	10	8	9	8	7
DASS	<i>Stress</i>	8.3 (4.3)	10.2 (5.6)	6.0 (4.5) **	8.8 (5.3)	5.1 (4.2)	5.6 (3.9)
	<i>Depression</i>	4.1 (3.0)	5.2 (5.5)	3.0 (2.7)	4.4 (4.4)	1.5 (1.2)	2.0 (2.6)
	<i>Anxiety</i>	3.8 (2.9)	4.3 (4.7)	1.1 (1.2)	3.2 (4.3)	2.4 (2.6)	2.9 (4.1)
PSI	<i>Defensive Response</i>	21.7 (5.3)	21.4 (4.3)	18.1 (3.3)	20.7 (6.2)	17.0 (4.8)	18.1 (4.2)
	<i>Parental Distress</i>	34.3 (8.2)	33.0 (8.6)	29.2 (7.6)	34.1 (15.1)	27.6 (5.2)	28.9 (4.5)
	<i>Parent-Child-Dysfunctional Interaction</i>	19.1 (6.5)	23.4 (7.9)	16.4 (4.9)	22.6 (7.5)	17.2 (3.8)	21.1 (4.7)
	<i>Difficult Child Domain</i>	31.1 (12.0)	31.6 (8.3)	25.0 (6.5)	30.3 (8.2)	24.8 (7.9)	27.7 (5.9)
	<i>Total</i>	84.5 (22.7)	101.1 (44.6)	70.6 (14.5)	87.0 (27.0)	69.6 (13.1)	77.7 (10.1)

392 ^aIOT - insight oriented psychotherapy; ED^b - education

393 * mean (sd), or count

394 ** $p < 0.05$ for paired t-test with pre score

395 **Tab. 3 – Adjusted mean difference between arms, following the first and second study**
 396 **period and overall**

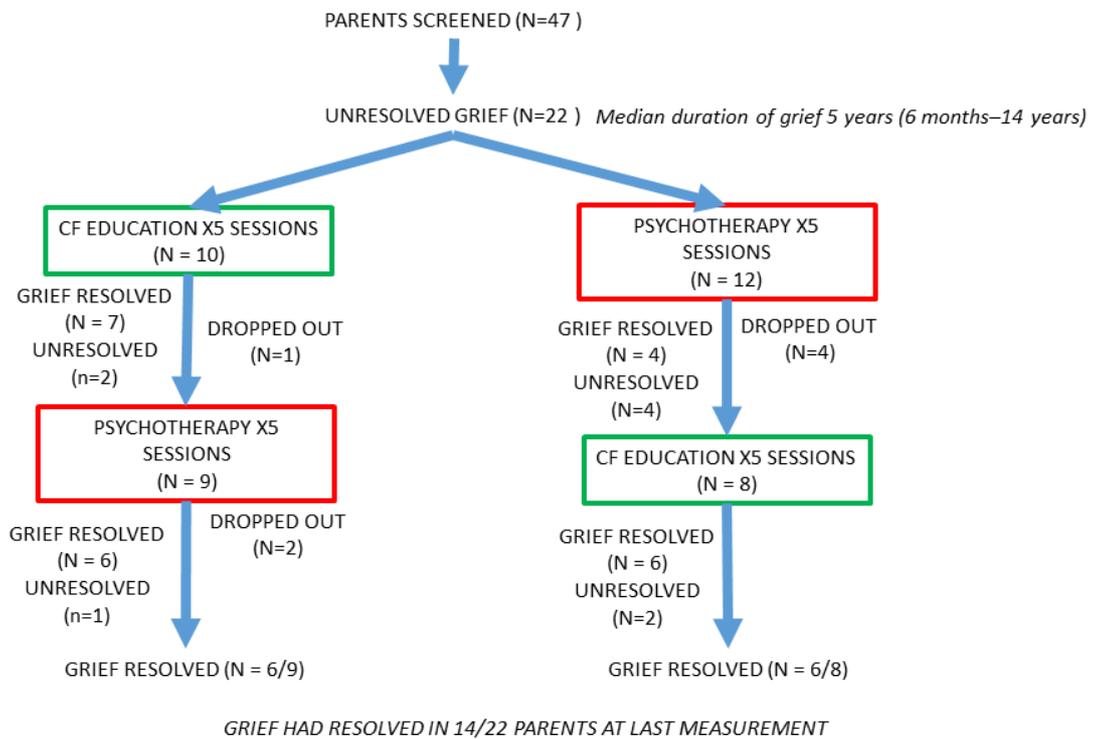
Assessment	Subscale	Adjusted IOT ^a effect (period 1) *	Adjusted IOT effect (period 2) *	Adjusted sequence effect (IOT then ED ^b) (periods 1 and 2) *
n		17	15	15
DASS	<i>Stress</i>	-0.6 (-3.3,2.1)	1.2 (-2.0,4.5)	0.5 (-3.9,5.0)
	<i>Depression</i>	0.0 (-2.2,2.3)	0.3 (-2.8,3.4)	0.2 (-3.2,3.5)
	<i>Anxiety</i>	-1.2 (-4.4,2.1)	0.8 (-4.8,6.4)	1.9 (-3.4,7.2)
PSI	<i>Defensive Response</i>	-1.3 (-6.8,4.3)	-0.7 (-5.2,3.9)	0.3 (-2.7,3.4)
	<i>Parental Distress</i>	-3.8 (-17.5,10.0)	0.0 (-6.9,6.9)	0.2 (-4.0,4.3)
	<i>Parent-Child-Dysfunctional Interaction</i>	-4.2 (-11.9,3.5)	-0.3 (-8.3,7.6)	-0.2 (-3.9,3.6)
	<i>Difficult Child Domain</i>	-3.0 (-11.5,5.5)	1.4 (-9.0,11.8)	1.6 (-2.9,6.0)
	<i>Total</i>	-4.2 (-21.3,13.0)	0.9 (-13.9,15.7)	2.9 (-3.4,9.3)

397 ^aIOT - insight oriented psychotherapy; ^bED - education

398 * Beta coefficient for the between group difference (IOT arm relative to education arm) from
 399 a linear regression model, using an ANCOVA framework, adjusted for parents age, duration
 400 of grief, gender, and measure at the start of the period being analysed

401 FIGURES

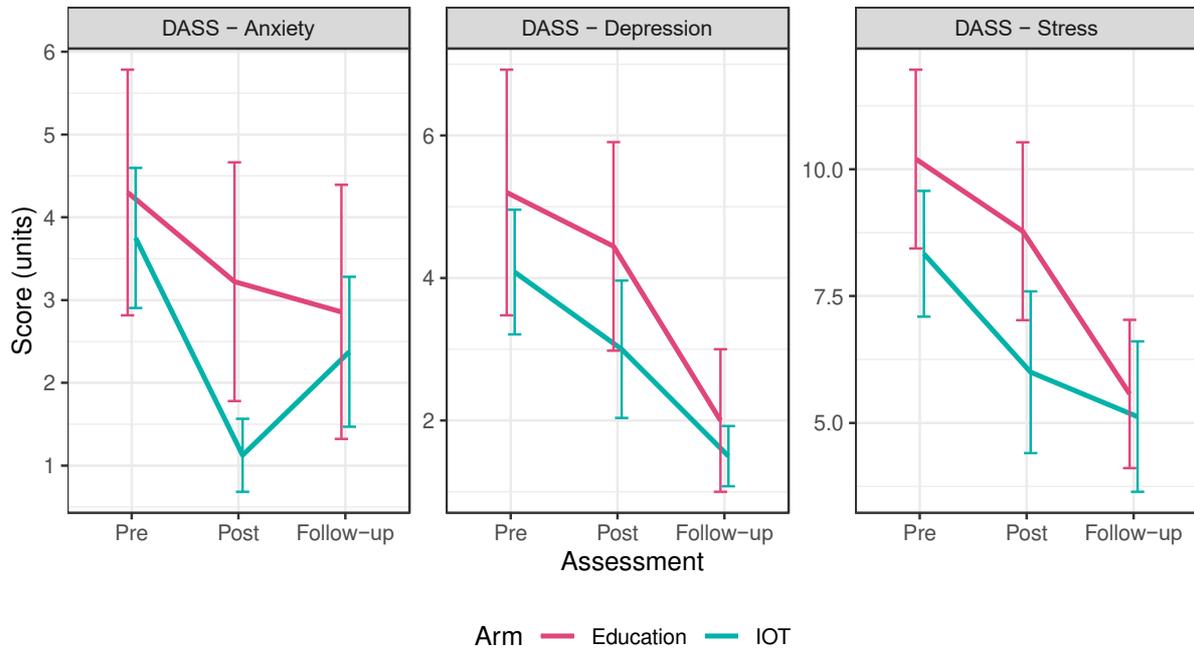
402 Fig. 1 – CONSORT diagram



403
404

405 **Fig. 2 – Mean DASS scores across three timepoints, by arm**

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408 *Plotted line is the raw mean of the observed DASS scores (by subscale) from each group;*

409 *error bars represent one standard error of the mean*

410

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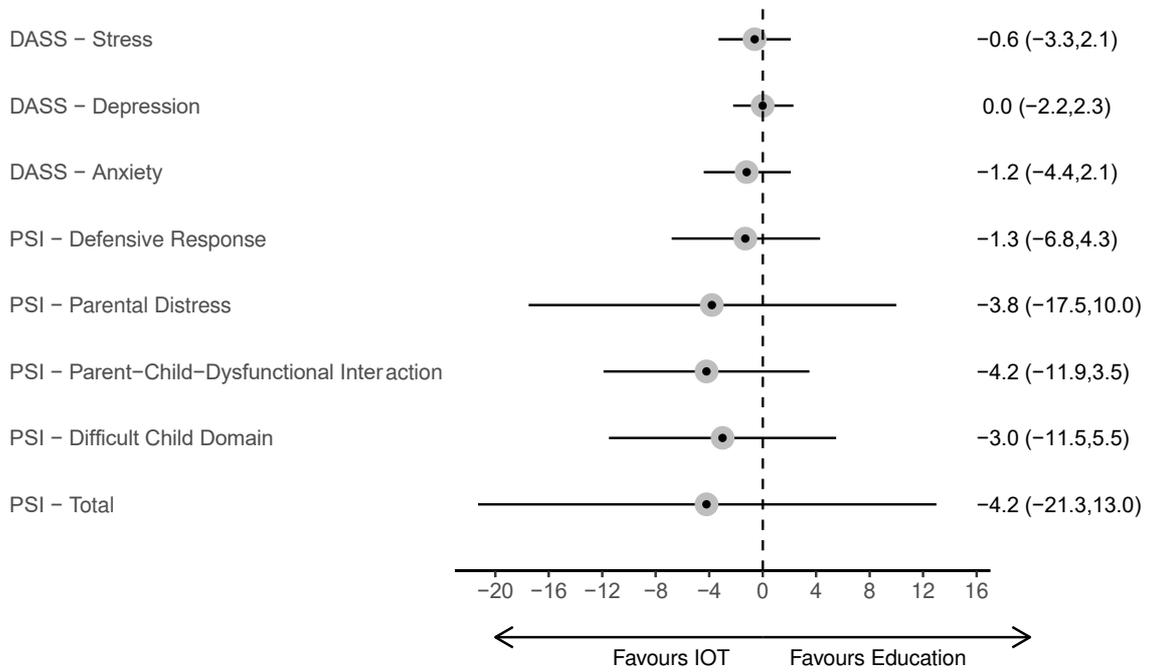
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416 **Fig. 3 - Adjusted mean difference in measures of emotional wellbeing following**
 417 **intervention**



418

419 *Plotted point is the adjusted between group difference (IOT arm relative to ED arm) from a*
 420 *linear regression model, using an ANCOVA framework, adjusted for parents age, duration of*
 421 *grief, gender, and measure at the start of the period being analysed; line width represents the*
 422 *95% confidence interval for the adjusted between group difference estimates*

Supplementary Files

This is a list of supplementary files associated with this preprint. Click to download.

- [OnlineResourceEJP.pdf](#)