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A meta-analysis of intervention effects on challenging behaviour among persons with intellectual disabilities

Keywords: meta-analysis, intellectual disability, mental retardation, challenging behaviour

Abstract

Background: Persons with intellectual disabilities (ID) often show challenging behaviour. We review distinct interventions that are applied to treat these challenging behaviours, and analyse intervention effects and moderating variables.

Method: A literature search was conducted using the databases *ERIC*, *PsycINFO*, *Web of Science* and *Medline*. A random-effects meta-analysis was carried out, supplemented with sensitivity, subgroup, meta-regression, and publication bias analyses.

Results: Eighty potential articles were identified, from which 30 contained sufficient data to enable statistical meta-analysis. From these 30 studies, 18 described a biological, 13 a psychotherapeutic and 9 a contextual intervention, either applied alone or combined. The overall standardised mean difference was 0.671 (SD = 0.051). As shown by sensitivity analysis, this summary effect size is robust. Assessed through subgroup and meta-regression analysis, all tested moderators showed no statistically significant association with the treatment effects. After applying a funnel plot-, a fail-safe N-, and Duval's and Tweedie's trim and fill-analysis, we conclude that our meta-analysis does not suffer much from publication bias effects.

Conclusions: Several biological, psychotherapeutic and contextual interventions effectively reduce challenging behaviours among persons with intellectual disabilities.

Introduction

Over the last decades, literature on the prevalence, impact, and treatment of challenging behaviour presented by individuals with intellectual disabilities (ID) has accumulated (Antonacci et al. 2008, Didden et al. 2006, Harvey et al. 2009, Kahng et al. 2002, McClintock et al. 2003, Moss et al. 1997, Prout & Nowak-Drabik 2003). Examples of challenging behaviour include verbal and physical aggression, property damage and destructiveness, disruptive and antisocial behaviour, overactivity, temper tantrums, screaming, stereotyped and repetitive behaviour, general delinquency, and self-injurious behaviour, like head punching, self-biting, skin picking, and hitting against hard objects or other body-parts (Allen & Davies 2007, Benson & Brooks 2008, Deb et al. 2001, Didden et al. 2007, Grey & Hastings 2005).

Challenging behaviours are especially prevalent in individuals with ID, and the more severe the disability, the higher the likelihood of the presence of these behaviours (Borthwick-Duffy 1994,

Cormack et al. 2000, Janssen et al. 2002, McIntyre et al. 2002). Most studies report prevalence rates of challenging behaviour among persons with ID between 10 and 20% (Emerson et al. 2001, Kahng et al. 2002, Lowe et al. 1998), while some authors reporting substantially higher rates (Allen 2000, Cooper et al. 2009a, Crocker et al. 2006, 2007, Deb et al. 2001, Dekker et al. 2002, Gavidia-Payne & Hudson 2002, Moss et al. 1997). The development of challenging behaviour is influenced by person- and environment-oriented factors, which often interact with each other. Examples of these factors that are frequently mentioned, are not only the age, gender and level of ID of persons, but also poor adaptive and social skills, psychological stress, inadequate problem-solving skills, impaired language, socioeconomic deprivation, negative life events, secondary disabilities and psychiatric disorders (Allen & Davies 2007, Aunos et al. 2008, Blacher & McIntyre 2006, Bradley et al. 2004, Chadwick et al. 2008, Collacott et al. 1998, Cooper et al. 2007, Crocker et al. 2007, de Ruiter et al. 2008, Dekker et al. 2002, Didden et al. 2007, Embregts et al. 2009, Hastings & Mount 2001, Hemmings et al. 2006, Holden & Gitlesen 2009, Janssen et al. 2002, Matson & Rivet 2008, McClintock et al. 2003).

Challenging behaviours have a negative impact on persons with ID and their environment, *inter alia* because they increase risks on a reduced quality of life, stressful events, obstacles to social integration, and the need for costly residential care (Benson & Brooks 2008, Blacher & McIntyre 2006, Cooper et al. 2009a, Didden et al. 2006, Harvey et al. 2009, Hassiotis et al. 2008, Knapp et al. 2005, Matson & Boisjoli 2009, McIntyre et al. 2002). Moreover, due to their recurrent character, challenging behaviours tend to become a lifelong challenge for the individuals with ID, their family, and the involved services (Cooper et al. 2009a, 2009b, Danquah et al. 2009, Murphy et al. 2005, Tenneij & Koot 2008, Totsika & Hastings 2009).

Because of the high prevalence and negative impact of challenging behaviours among persons with ID, considerably much research has been conducted on interventions in this area (Antonacci et al. 2008, Beail 2003, Deb et al. 2007, Grey & Hastings 2005, Kahng et al. 2002, McGillivray & McCabe 2006, Moss et al. 1997, Prout & Nowak-Drabik 2003, Sohanpal et al. 2007, Sturmey 2004, Taylor 2002, 2005, Willner 2005). Treatment strategies that are frequently employed are biological and psychotherapeutic interventions, sometimes supplemented with contextual strategies (Ager & O'May 2001, Benson & Brooks 2008, Bouras 1999, Dösen & Day 2001, Gavidia-Payne & Hudson 2002, Grey & Hastings 2005, Kahng et al. 2002, Mildon et al. 2008, Taylor 2002). Recently, some reviews and meta-analytic articles have studied these interventions for challenging behaviour among persons with ID (Brylewski & Duggan 1999, Didden et al. 2006, Lotan & Gold 2009, Matson & Neal 2009, Prout & Nowak-Drabik 2003, Shogren et al. 2004, Thomson et al. 2009a, 2009b). None of these articles included both the biological and the psychotherapeutic and contextual interventions, although it is interesting to compare effects of these three intervention types, either applied alone or combined, with each other. Further advantages of this meta-analysis that are often missing in previous reviews and meta-analytic articles, are the quality assessment of all included articles, the extensive moderator testing, and additional sensitivity and publication bias analyses.

The main purpose of the present study was to review distinct biological, psychotherapeutic, and contextual interventions that are applied to treat challenging behaviour among persons with ID, and to analyse intervention effects and moderating variables. We did this by performing a meta-analysis, in order to boost the total sample size and effect precision by combining ‘sufficiently’ homogeneous results across studies. This was done by first describing the selected articles and the interventions they report. Second, we performed a meta-analysis in order to broadly explore intervention effects and possible moderating effects of study and participant variables. Additionally, we report on the quality assessment we carried out for each study included, and examine the moderating role of study quality for reported intervention effectiveness. The meta-analysis, supplemented with sensitivity, subgroup, meta-regression, and publication bias analyses that were carried out, is described in this article.

Methods

Literature search and selection of studies

The literature search was carried out using four electronic databases: *ERIC*, *PsycINFO*, *Web of Science* and *Medline*. We searched for articles reporting on biological, psychotherapeutic, or contextual interventions for challenging behaviour among persons with ID by combining key terms describing the target group and outcome variables. Applied key terms referring to the target group were *intellectual disability*, *mental retardation*, *learning difficulty* and *complex/high support needs*. Used key terms describing outcome variables were *challenging behaviour*, *problem behaviour*, *behavioural problems*, *aggression*, *self-injury* and *self-injurious behaviour*. We searched for these terms in singular and plural, in US- and UK-terminology. We only included English-language articles that described empirical studies and that were published in peer-reviewed journals between January 2000 and November 2008. The main focus of the included articles had to be the implementation of an intervention targeting challenging behaviour among persons with ID. We excluded articles that only reported on diagnostic instruments or only presented review material. Because we wanted to perform a meta-analysis, the selected articles had to contain data or sufficient statistical information to make a quantitative analysis feasible.

Characteristics recorded for each study

Some features were recorded for all studies included in the meta-analysis: intervention characteristics, intervention effects, methodological features, study characteristics, and participant features. We selected these features because of their relevance to the domain, as described in prominent literature (Altman et al. 2001, Jadad et al. 1996, Khan et al. 2001, Oxman & Guyatt 1988, Shadish et al. 2002, van Tulder et al. 2003, Verhagen et al. 1998). First, we recorded for each study which treatment was implemented, thereby making a distinction between biological, psychotherapeutic, and contextual interventions, and possible combinations of two or three of these intervention types applied together in

a single study (multimodal interventions). Second, intervention effects were determined for each study. Examples of challenging behaviour that are often the target in intervention studies are self-injurious and stereotypic behaviour, and aggressive and destructive behaviour (Allen & Davies 2007, Campbell 2003, Didden et al. 2006, Grey & Hastings 2005, Totsika & Hastings 2009). Third, some methodological features were recorded. We looked at each study design and recorded whether the authors implemented an experiment, a quasi-experiment, a natural experiment, or a non-experimental design. Furthermore, we described the data collection procedure: did the authors use tests or physiological measures, questionnaires, observations, interviews, and/or other data collection methods? In addition, we looked whether indices of reliability of applied data collection instruments were reported. Fourth, we described two study characteristics: the length of the research period (the average time between the first and last measurement for each studied group of participants) and the continent where the research took place. Fifth, four participant features were recorded for each article: the number of participants, and their gender, age, and level of ID. In addition, we graded the quality of all articles included in the meta-analysis. This was done by applying a quality assessment instrument, containing criteria described by Walsh and Downe (2006) and Campbell and colleagues (2003). This instrument referred to seven stages (*Scope and purpose, Design, Sampling strategy, Analysis, Interpretation, Ethical dimensions, Relevance and transferability*) incorporating nine ‘head items’ or ‘essential criteria’ (*Clear statement of and rationale for research question(s) / aim(s) / purpose(s), Study thoroughly contextualised by existing literature, Method / design apparent and consistent with research intent, Data collection strategy apparent and appropriate, Sample and sampling method appropriate, Analytic approach appropriate, Data used to support interpretation, Demonstration of sensitivity to ethical concerns, Relevance and transferability evident*) that each included several specific items. We scored each specific item, essential criterion and stage by ‘A’, ‘B’ or ‘C’, respectively indicating that an item was properly, partially, or not properly addressed or stated. Articles with ‘good quality’ were coded by maximum one ‘C’ or two ‘B’ head items; articles with ‘moderate quality’ by minimum two ‘C’ or three ‘B’ head items, or a combination of both; and articles with ‘poor quality’ by minimum three ‘C’ head items.

Analysis procedure

In order to review different biological, psychotherapeutic, and contextual interventions that are applied to reduce challenging behaviour among persons with ID, and to analyse intervention effects and moderating variables, we performed a meta-analysis¹. A priori, we preferred a random-effects to a fixed-effect meta-analysis, because random-effects models take a between-study variance component as a descriptive index of variation into account; study weights are more balanced under the random-

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¹ However, combining studies should not be done lightly, considering assumptions behind the use of meta-analysis. The fixed-effect model assumes that the true effect size is the same in all studies, while for random-effects meta-analysis the true effects in the studies are assumed to have been sampled from a distribution of true effects (Borenstein et al. 2009).

effects model assigning less relative weight to large studies; random-effects models provide inferential results referring to a universe of more diverse studies than fixed-effect analyses; and because the width of the confidence intervals calculated by the fixed-effect model tends to imply a greater degree of precision than is actually the case, while analysis based on a random-effects model will generally be more ‘conservative’ and ‘realistic’ (Borenstein et al. 2009, Fletcher & Fletcher 2005, Petitti 1994, Wachter & Straf 1990, Wang & Bushman 1999). In our random-effects meta-analysis we allowed for variation of the *true effect size* between studies, and assumed that the studies in our analysis only represent a random sample of effect sizes that could have been observed (Adèr & Mellenbergh 1999, Borenstein et al. 2009, Cooper & Hedges 1994, Kline 2005). Therefore, the summary effect is our estimate of the mean of these effects. In order to conduct the meta-analysis, effect sizes (standardised mean difference d)² and variances were computed for all included studies’ intervention effects (Borenstein et al. 2009, Rosenthal 1991). We generated a summary effect (effect size and variance) with a 95% confidence interval, and measures of heterogeneity (Q-value, Tau-squared, I-squared) (Borenstein et al. 2009, Higgins & Thompson 2002, Thompson 1994, Thompson & Sharp 1999). The studies were weighted by study precision in order to minimize both the within-study error and the variation in the true effects across studies: studies that yield more precise estimates of the effect size are assigned more weight (Borenstein et al. 2009, Hedges & Vevea 1998). Next to that, we assessed the impact of moderating variables through subgroup and meta-regression analysis. The former implied analyses of variance to compare treatment effects across groups concerning categorical variables, while the latter explored the impact of continuous moderators (Borenstein et al. 2009, Egger et al. 2007, Thompson & Higgins 2002). Furthermore, a sensitivity analysis was performed by running the analysis 30 times, each time removing one study, in order to show each study’s impact on the combined effect (Borenstein et al. 2009, Egger et al. 1997a). In addition, we analysed the possible impact of publication bias by a funnel plot-, a fail-safe N-, and Duval’s and Tweedie’s trim and fill-analysis (Borenstein et al. 2009, Duval & Tweedie 2000, Egger et al. 1997b, Kulinskaya et al. 2008, Rothstein 2005, Soeken & Sripusanapan 2003, Sutton et al. 2000b, Thornton & Lee 2000). The classic fail-safe N asks whether we need to be concerned that the entire observed effect may be an artifact of bias, while trim and fill offers a more nuanced perspective by asking how the effect size would shift if the apparent bias were to be removed (Borenstein et al. 2009, Cooper 1998, Higgins & Green 2008, Hunter & Schmidt 2004, Lewis-Beck et al. 2004, Lipsey & Wilson 2001, Riegelman 2005, Rothstein 2005, Salovey & Rothman 2003, Vaughn & Howard 2009, Wolf 1986). Statistical analyses were conducted using Comprehensive Meta-Analysis Version 2.0 (Borenstein et al. 2009, <http://www.meta-analysis.com>).

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² The calculation of the standardised mean difference d is based on means and standard deviations, which are not reliable indicators of location and spread for skewed distributions (Larson 2006). Since the included studies did not report measures of skewness, we can not conclude whether more distribution-robust statistics such as medians and interquartile ranges should be used instead of the standardised mean difference (Everitt & Howell 2005, Larson 2006).

Results

Description of primary studies and participants

Guided by the described inclusion and exclusion criteria, 80 articles were considered for inclusion in our study. However, only 30 articles contained sufficient data to enable meta-analysis, and could be entered in the programme Comprehensive Meta-Analysis Version 2.0 (see <http://www.meta-analysis.com> for a list of all supported formats). Table 1 presents an overview of the 30 selected articles, representing 1444 participants. For each article, challenging behaviours shown by participants, interventions used to target these challenging behaviours, assessment of the articles' quality, and gender, level of ID and number of participants are reported. The references to these articles are drawn up in Appendix 1.

From those 30 articles, 18 described a biological, 13 a psychotherapeutic, and 9 a contextual intervention, with sometimes more than one implemented treatment discussed in a single article. More specific, 14 studies reported a unimodal biological, 5 a unimodal psychotherapeutic, and 2 a unimodal contextual intervention, next to 5 psychotherapeutic-contextual and 2 biological-psychotherapeutic treatments, 1 biological-contextual, and 1 biological-psychotherapeutic-contextual intervention. For the biological treatments, especially atypical antipsychotic medications (risperdone and olanzapine) were studied (9), next to typical antipsychotics (2) and other biological interventions (5). Two articles described sensory interventions. Regarding psychotherapeutic treatments, four behavioural, four systemic, three cognitive-behavioural and two other psychotherapeutic interventions were investigated. Concerning the explored contextual treatments, there were especially interventions situated within a multidisciplinary approach (6) and interventions focusing on changing the environment (2).

Describing methodological features, we looked at designs, data collection instruments, and their reported reliability. The most often implemented study design was an experiment (14), followed by a quasi-experiment (10), and a natural experiment (6). Regarding the data collection procedure, 22 authors applied questionnaires, 11 interviews, 9 tests or experimental measures, 9 observations, and 8 other data collection methods. In 16 articles indices of reliability were reported for data collection instruments. Turning to study characteristics, the length of the research period (the average time between the first and last measurement for each studied group of participants) was less than a year in 13 studies, more than one year in 10 studies, and not mentioned in 7 articles. The continent where most research took place was America (14), followed by Europe (11), Australia (4) and Asia (1). Finally, we recorded four participant features: the number of participants, and their age, gender, and degree of ID. Seven articles described 20 or fewer participants, 12 articles reported on interventions implemented for 21 to 50 participants, and 11 articles reported on intervention effects for more than 50 participants. In 21 studies there were more male than female participants, while for 6 articles the opposite was true. There were slightly more (16) studies describing a mean age of participants above

18, compared with 11 studies describing a mean age of participants under 18, while 3 articles did not mention participants' ages. Furthermore, 16 studies involved persons with mild ID, 14 moderate ID, 15 severe ID and 8 profound ID. Six articles did not mention the degree of ID of their participants. In grading the quality of the articles, we found that 22 articles were of 'good' and 8 of 'moderate' quality. Especially *Scope & Purpose*-, *Analysis*- and *Interpretation*-items scored very well. Regarding the categories *Design*, *Sampling* and *Ethics*, there was often information missing.

Meta-analysis

Combined effect sizes and their standard errors computed for all 30 articles are presented in Table 1. The applied interventions made an improvement in each study: all calculated effect sizes are positive³. Applying random-effects weights, the standardised mean difference is 0.671 with a 95% confidence interval of 0.570 to 0.771. According to Cohen (1988) this is a medium (effect size around 0.5) to large (effect size around 0.8) effect. The null hypothesis that the mean of these effects is zero, can be rejected, $Z(N = 30) = 13.070$, $p < 0.001$ ⁴.

In addition, we computed three measures quantifying heterogeneity. First, the p -value for the weighted sum of squares on a standardised scale, $Q = 30.277$ with $df = 29$, is 0.400. Applying 0.05 as criterion for statistical significance, we cannot reject the null hypothesis that all studies share a common effect size. Second, tau-squared, the variance of the true effects calculated on the same scale (squared) as the effects themselves, is 0.003, with a standard error of 0.021. Third, I-squared, the proportion of observed dispersion that is real, is 4.219%. This means that 4.219% of the observed variance comes from real differences between studies, and, as such, can potentially be explained by study-level covariates (Borenstein et al. 2009, Cooper & Hedges 1994).

A sensitivity analysis was performed to determine the robustness of our results by examining whether our conclusions might differ substantially if a study was dropped (Borenstein et al. 2009, Cooper & Hedges 1994, Petitti 1994, Sutton et al. 2000a). We calculated for each study the overall standardized mean difference when that study was hypothetically removed from the meta-analysis. Since the 30 overall effect sizes formed by omitting each study separately varied between 0.649 and 0.689, our results look very robust.

Furthermore, the impact of moderating variables was assessed through subgroup and meta-regression analysis. We applied analyses of variance to compare treatment effects across groups for the following

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³ Incidentally, if the null hypothesis of no treatment effect would be true, then there would be a 50% chance of observing positive effect sizes. Because the studies can be considered as independent events, this setup constitutes a binomial situation with $p = .50$. The probability to observe 30 successes out of 30 trials is $.50^{30}$, or in other words $p < .0001$.

⁴ Applying the fixed-effect model, the standardized mean difference is 0.670 with a 95% confidence interval of 0.572 to 0.768, and the null hypothesis that the mean of these effects is zero, can be rejected, $Z(N = 30) = 13.452$, $p < 0.001$. So, the results for the fixed- and random-effects analysis are analogous.

categorical variables: intervention type (biological, psychotherapeutic and contextual), intervention combination (unimodal, multimodal), quality assessment, study design (experiment, quasi-experiment, natural experiment), data collection (test, questionnaire, observation, interview, other), reliability, length of the research period, continent, and the gender, age, and degree of ID (mild, moderate, severe, profound) of participants. We present the descriptive statistics of these moderators in Appendix 2. After assessing the relationships between subgroup membership and effect size, we conclude for *all* these variables that differences between groups are *not* significantly related to the effect size.

Table 1: Characteristics of studies included in the meta-analysis

First author	Year published	Number of participants	Level of ID *	Male-female participants	Category of target challenging behaviour **	Intervention type ***	Quality of the article ****	Std diff in means *****	Standard error
Aman	2002	118	1,2	97-21	1	B	A	0.502	0.191
Buitelaar	2001	38	1	33-5	3	B	A	0.394	0.338
Capone	2008	23	3	20-3	1,4	B	A	0.723	0.236
Chan	2005	89	1,2,3	36-53	3	B	A	0.457	0.218
Dowling	2006	56	1,2,3	.	1,2	PC	A	0.662	0.279
Duker	2000	16	.	10-6	4	P	B	1.207	0.544
Gates	2001	77	.	.	1	PC	A	0.187	0.256
Holden	2000	28	1	17-11	1	BPC	B	0.814	0.285
Huang	2007	40	.	14-26	1	BP	B	0.730	0.179
Hudson	2003	115	1,2,3	81-34	1	PC	A	0.482	0.257
Janowsky	2003	22	3,4	8-14	1,3,4	B	B	0.567	0.305
Janowsky	2005	34	3,4	23-11	3,4	B	A	0.845	0.350
Janowsky	2003	20	1,2,3,4	9-11	1,3,4	B	A	1.380	0.570
Mace	2001	15	3,4	10-5	4	BP	A	0.269	0.488
McDonough	2000	7	3,4	5-2	4	B	B	0.769	0.434
Packman	2003	24	.	18-6	1	P	A	0.548	0.416
Pearson	2003	24	1,2	18-6	1,3	B	B	0.417	0.293
Plant	2007	74	1,2,3	55-19	1	PC	A	0.752	0.172
Read	2007	24	2,3,4	19-5	3,4	B	B	0.708	0.234
Roberts	2006	47	1	37-10	1	PC	A	0.855	0.308
Rose	2000	44	.	39-5	3	P	A	1.411	0.340
Rose	2005	86	.	71-15	3	P	A	1.109	0.234
Shapira	2004	8	1,2	3-5	4	B	A	1.158	0.460
Singh	2004	45	3,4	31-14	3,4	BC	B	1.075	0.336
Snyder	2002	110	1,2	83-27	1,3	B	A	0.659	0.197
Taylor	2002	19	1	19-0	3	P	A	1.306	0.509
Turgay	2002	77	1,2	57-20	1	B	A	0.223	0.308
Xenitidis	2004	84	1,2,3	36-48	1,2	C	A	0.703	0.195
Young	2006	60	2,3	38-22	1	C	A	0.182	0.259
Zarcone	2001	20	1,2,3,4	10-10	1	B	A	0.867	0.468

Missing information is depicted as ‘.’

*: 1=mild; 2=moderate; 3=severe; 4=profound

**: 1=aberrant/challenging/disruptive/maladaptive behaviour; 2=psychiatric disorders, psychotic symptoms; 3=aggression, destructive behaviour, anger; 4=self-injurious behaviour; 5=sexual offending; 6=stereotypical, repetitive behaviour

***: B=Biological intervention; P=Psychotherapeutic intervention; C=Contextual intervention; BP=Biological-Psychotherapeutic intervention; PC=Psychotherapeutic-Contextual intervention; BC=Biological-Contextual intervention; BPC=Biological-Psychotherapeutic-Contextual intervention

****: A=article with good quality; B=article with moderate quality

*****: Standardised difference in means

In addition, we explored the impact of one continuous moderator: the year of publication. For this moderator too, we cannot reject the null hypothesis of no effect. Applying the regression model (unrestricted maximum likelihood), we see that the total dispersion of studies about the grand mean

(Q -total) is 30.27744 with $p = 0.40025$ ($df=29$), meaning that the amount of total variance is less than we would expect based on the within-study error. Furthermore, the dispersion explained by the covariates (Q -model) is 0.26403 with $p = 0.60737$ ($df=1$), which means that the relationship between publication year and treatment effect is even weaker than we would expect by chance.

Finally, we address possible publication bias effects by a funnel plot-, a fail-safe N -, and Duval's and Tweedie's trim and fill-analysis. We refer to publication bias when studies included in an analysis differ systematically from all studies that could be included: particularly, studies with larger effects are more likely to be published, and this can lead to an upward bias in the summary effect (Borenstein et al. 2009, Cooper & Hedges 1994, Higgins & Green 2008, Hunter & Schmidt 2004, Lipsey & Wilson 2001). First, we explore possible publication bias effects by a funnel plot. In Figure 1 a measure of study size (precision, the inverse of standard error) is plotted on the vertical axis as a function of effect size on the horizontal axis. In the absence of publication bias, we would expect the included studies (white circles in Figure 1) to be distributed symmetrically around the combined effect size (Adèr & Mellenbergh 1999, Borenstein et al. 2009, Bowling & Ebrahim 2006, Petitti 1994, Rothstein 2005), and this seems to be the case for our meta-analysis.

In the following, we assess some statistical procedures to quantify this possible publication bias effect. The fail-safe N is 1336, meaning that we would need to locate and include 1336 'null' studies in order for the combined two-tailed p -value to exceed 0.050. Because this number is large, we can be relatively confident that the treatment effect, while possibly inflated by the exclusion of some studies, is real. Duval and Tweedie's trim and fill analysis addressing the left side of the mean effect suggests that four studies are missing. These four imputed studies are plotted in Figure 1. Under the random-effects model the standardised mean difference and 95% confidence interval for the combined studies is 0.67070 (0.57012, 0.77128). Using trim and fill, the imputed standardised mean difference is 0.63436 (0.52493, 0.74378), which is a little bit lower than our standardised mean difference. When addressing Duval and Tweedie's trim and fill to the right side of the mean effect, the method suggests that no studies are missing.

Figure 1: Funnel plot of precision by standardised difference in means: plot with observed (white circles) and imputed (black circles) studies

So, after applying a funnel plot-, a fail-safe N-, and Duval's and Tweedie's trim and fill-analysis, we conclude that our meta-analysis does not seem to suffer much from publication bias effects.

Discussion

In this meta-analysis of articles describing rigorous quantitative empirical studies of intervention effects on challenging behaviour among persons with ID we found effect sizes (standardised mean differences) from 0.223 to 1.411. So, the effect sizes vary between a small and a very large effect (Cohen 1988), all indicating positive treatment effects. The combined effect size over all studies is 0.671, with a 95% confidence interval of 0.570 to 0.771, which is a medium to large effect according to Cohen (1988). The implemented sensitivity analysis revealed that this effect is robust. Analyses of variance showed no significant different treatment effects for biological, psychotherapeutic, and contextual interventions. Differences between unimodal and multimodal treatments turned out to be not significant as well. The reported means and standard errors for the moderators *intervention type* and *intervention combination* show that there are only small differences between the mean effects for biological, psychotherapeutic, and contextual interventions, and for unimodal and multimodal treatments (Appendix 2).

In the general literature and in articles presenting qualitative and descriptive material, some authors claim that there is evidence for the effectiveness of pharmacological treatments (Deb et al. 2007, Grey & Hastings 2005), although many authors assert that they lack empirical effectiveness (Antonacci et

al. 2008, Brylewski & Duggan 1999, La Malfa et al. 2006, Matson & Neal 2009, Sohanpal et al. 2007, Taylor 2002, Thomson et al. 2009a, 2009b). Besides, there are many concerns regarding their adverse effects (Deb et al. 2007, McGillivray & McCabe 2006, Sohanpal et al. 2007). Concerning psychotherapeutic and contextual interventions too, there are authors advocating their effectiveness (Ager & O'May 2001, Beail 2003, Campbell 2003, Didden et al. 2006, Gavidia-Payne & Hudson 2002, Grey & Hastings 2005, Harvey et al. 2009, Mildon et al. 2008, Moss et al. 1997, Prout & Nowak-Drabik 2003, Shogren et al. 2004), while some say that the evidence is rather limited (Antonacci et al. 2008, Gustafsson et al. 2009, Sturmey 2004, Taylor 2005, Willner 2005). Our meta-analysis shows that there is evidence for the effectiveness of pharmacological, psychotherapeutic and contextual interventions, used alone or in combination. Furthermore, we found no indications for the superiority of one of the treatment approaches or combination types.

Treatment effects also did not vary much across groups for the categorical moderators included in our meta-analysis: *quality assessment, study design, data collection, reliability, length of the research period, continent, and gender, age, and level of ID of participants*. We also did not find any effect of the continuous moderator *publication year* on the treatment effect. Some authors have suggested that the nonsignificance of moderator effects is not uncommon, due to low statistical power for detecting interaction between a moderator variable and the independent variable (Aguinis & Beaty 2005, Aguinis & Stone-Romero 1997, Borenstein et al. 2009, Hedges & Pigott 2001, 2004, Shieh 2009). For our meta-analysis, we see in Appendix 2 that there exist only small differences between the mean effects for the tested categorical moderating variables, so even if we would increase the power, we would most likely not detect clinically important differences. As a particular strength of our study we want to mention that a funnel plot-, a fail-safe N-, and Duval's and Tweedie's trim and fill-analysis demonstrate that our meta-analysis does not seem to suffer much from publication bias effects.

In summary, interventions for challenging behaviour among persons with ID described in the 30 included articles were effective, with only small differences between the mean effects for biological, psychotherapeutic, and contextual treatments, and for unimodal and multimodal interventions. In contrast to claims in the literature that the evidence for one or another intervention is still rather limited, the effects in our meta-analysis were robust and convincing.

Further research should now focus on the working mechanisms of these successful interventions. We showed that challenging behaviour among individuals with ID can be successfully treated by diverse biological, psychotherapeutic, and contextual interventions, but it is still not very clear how and why each of these interventions works, either when applied alone or combined. Are there common working mechanisms behind these interventions, or do biological, psychotherapeutic, and contextual interventions function differently? And, how do they work combined? To this end, it would be interesting to have more longitudinal research focusing on differential effects of biological, psychotherapeutic, and contextual interventions, taking into account theoretically relevant moderators and mediators.

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Appendix 1: Articles included in the meta-analysis

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Appendix 2: Descriptive statistics of categorical moderators

Groups of moderators				Number of studies	Standardised mean difference	Standard error
Intervention characteristics	Intervention type	Biological	Yes	18	0.646	0.068
			No	12	0.707	0.081
		Psychotherapeutic	Yes	13	0.752	0.076
			No	17	0.607	0.067
	Intervention combination	Contextual	Yes	9	0.624	0.086
			No	21	0.699	0.066
		Unimodal		21	0.674	0.065
				9	0.677	0.089
Methodological features	Quality assessment	Good quality		22	0.650	0.060
		Moderate quality		8	0.732	0.103
	Study design	Experiment	Used	14	0.627	0.075
			Not used	16	0.711	0.072
		Quasi-experiment	Used	10	0.787	0.010
			Not used	20	0.631	0.058
	Data collection	Natural experiment	Used	6	0.637	0.102
			Not used	24	0.684	0.061
		Test	Used	9	0.726	0.090
			Not used	21	0.643	0.064
		Questionnaire	Used	22	0.647	0.060
			Not used	8	0.746	0.105
		Observation	Used	9	0.723	0.103
			Not used	21	0.653	0.061
		Interview	Used	11	0.716	0.076
			Not used	19	0.632	0.070
		Other	Used	8	0.530	0.088
			Not used	22	0.735	0.060
	Reliability	Reported		16	0.657	0.074
		Not reported		14	0.686	0.075
Study characteristics	Research period	Less than 1 year		13	0.624	0.085
		More than 1 year		10	0.699	0.096
		No information		7	0.705	0.096
	Continent	America		14	0.676	0.077
		Europe		11	0.745	0.093
		Other		5	0.563	0.107
Participant features	Gender	More female participants		6	0.680	0.105
		More male participant		21	0.693	0.064
		Equal / no information		3	0.472	0.181
	Age	Mean < 18 years		11	0.559	0.074
		Mean > 18 years		16	0.770	0.077
		No information		3	0.736	0.139
	Degree of ID	Mild ID	Yes	16	0.637	0.068
			No	8	0.646	0.113
			No information	6	0.795	0.115
		Moderate ID	Yes	14	0.592	0.065
			No	10	0.766	0.108
			No information	6	0.792	0.110
		Severe ID	Yes	15	0.653	0.073
			No	9	0.616	0.097
			No information	6	0.794	0.115
		Profound ID	Yes	8	0.774	0.126
			No	16	0.605	0.063
			No information	6	0.792	0.111