

The Official Academic Journal of the European Association of Clinical Psychology and Psychological Treatment



CLINICAL PSYCHOLOGY IN EUROPE

The Official Academic Journal of the European Association of Clinical Psychology and Psychological Treatment



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Editorial



The (Neuro)-Science Behind Resilience: A Focus on Stress and Reward

Chantal Martin-Soelch 1 (D)



[1] Reach Lab, Unit of Clinical and Health Psychology, Department of Psychology, University Fribourg, Fribourg, Switzerland.

Clinical Psychology in Europe, 2023, Vol. 5(1), Article e11567, https://doi.org/10.32872/cpe.11567

Published (VoR): 2023-03-31

Corresponding Author: Chantal Martin-Soelch, University of Fribourg, Department of Psychology, Rue P-A Faucigny 2, CH-1700 Fribourg. Tel: +41 26 300 76 87. E-mail: chantal.martinsoelch@unifr.ch

Mental disorders represent one of the major causes of disability worldwide, with depressive disorders being the leading causes of burden among mental disorders in all age categories above 14 years old, followed by anxiety disorders (GBD 2019 Mental Disorders Collaborators, 2022). Although knowledge concerning their etiology has improved, it is still unclear why one person will develop a mental disorder while another will not when facing adversities. In this context, the identification of resilience mechanisms is crucial. Adopting an approach based on mechanisms allows us to have a transdiagnostic and transtheoretical approach and to target specific processes for the development of psychological interventions.

Indeed, better knowledge of resilience mechanisms allows the development of targeted interventions in at-risk populations. Some risk factors for the development of mental disorders have been well identified, such as childhood abuse or in general early adverse childhood experiences (ELS) (Kessler et al., 2010; Mandelli et al., 2015). ELS have received increased attention from research, which led recently to the development of a consortium in the framework of the global traumatic stress collaboration dedicated to the investigation of socio-emotional consequences of ELS (Pfaltz et al., 2022). A further well-investigated risk factor is being the offspring of one or more parents suffering a mental health condition, particularly depression, bipolar disorder or schizophrenia (Rasic et al., 2014).

Resilience can be defined as the capacity of an individual to adapt successfully to highly adverse events and keep a healthy functioning by harnessing resources (Southwick et al., 2014). It is most often measured by questionnaires, although these may be limited by issues of internal validity in particular because little is known about the



different elements that make up resilience. These questionnaires therefore often focus on one or a set of measures related to well-known protective factors or resources, such as feelings of self-efficacy, self-esteem, sense of mastery, optimism, positive affect, good emotion regulation skills or sense of coherence (Southwick et al., 2014). Neuroscience has provided new insights in this area and indicates that neurocognitive and neuroaffective factors, such as cognitive flexibility or reactivity to stress or reward may play a role in resilience. These two processes- reward and stress- are linked to well-defined brain systems that are considered to be crucial for human motivation and adaptation (Godoy et al., 2018; Schultz, 2000). Blunted neural responses to reward have been consistently observed in depressive disorders, and have been hypothesized to underly the symptoms of anhedonia, apathy and loss of interest observed in these conditions (Pizzagalli et al., 2009). And a large body of empirical evidence shows the importance of stress in the development of several psychopathological conditions, among others depression (Liu & Alloy, 2010). Recently, it has been postulated that not only the responses to reward or the effect of stress, but rather an interaction between both is involved in the etiology of mental disorders. Thus, a high reactivity of the brain to stress and a reduced brain reactivity to reward, also conceptualized as an imbalance between the neural responses to stress and to reward, has been hypothesized to be a vulnerability factor for the development of mental disorders, in particular depressive disorders (Admon et al., 2013). This model has been completed with research works showing that not only the neural responses during the presentation of stressful stimuli or rewarding information is important, but also the neural recovery after these events, in particular longer recovery after stress and shorter recovery after reward, might play a role. This has been conceptualized as emotional inertia and brought in relationship with difficulties in emotion regulation (Koval et al., 2015).

Our laboratory, the IReach lab at the Department of Psychology of the University of Fribourg (Switzerland), has been particularly interested in the stress-reward interactions and their role in understanding the development of disorders in a transdiagnostic approach based on clinical neuroscience research results. Preliminary studies from our group suggest that in children of parents suffering from depression, reactions to rewards are impacted differently than in a control group under acute stress conditions (Gaillard et al., 2020; Martin-Soelch et al., 2020). These results are interesting because our participants had no clinical symptoms, but they showed different neural activation to reward stimuli and to the effect of stress on their processing. This may suggest a form of latent vulnerability that is not observable at the behavioral level. These results are in line with differences observed in response to rewarding information (without stress) in offspring of depressed parents (McCabe et al., 2012).

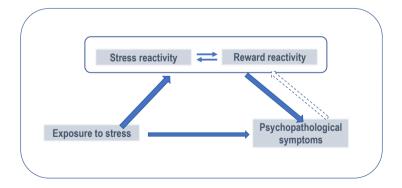
Understanding and integrating the interactions between the reward and stress systems in a model (see Figure 1) can serve as basis for developing and testing psychological prevention and/or treatment interventions that target these mechanisms. On this basis,



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we developed for instance a multi-modal stress management program that has shown effects in activating resources in general and increasing the feeling of reward in daily life in particular (Recabarren et al., 2019). Other therapeutic programs have also shown significant effects on reward processing. For instance, a study by Dichter et al. (Dichter et al., 2009) suggests that behavioral activation restores the brain's reactivity to reward in association with improvement of depressive symptoms in individuals diagnosed with major depressive disorder. Furthermore, a recently developed and validated intervention, the Mindfulness-Oriented Recovery Enhancement (MORE) program, which was originally developed for the management of substance use and addiction problems, in particular opioid use in connection with chronic pain management (Garland et al., 2022), also seems to show beneficial effects on the brain's responses to reward and to increase positive affect and emotion regulation (Garland et al., 2017). This group intervention program combines cognitive-behavioral techniques, mindfulness and meditation methods with savoring training. This 8-weeks training has shown significant beneficial effects on opioid use and improvement in chronic pain symptoms in large clinical trials in the USA (Garland et al., 2022). A current study of our group is interested in investigating whether this program can improve pain symptoms as well as affective symptoms of women suffering from fibromyalgia at a clinical level and to investigate as well the neural responses to reward changes and in the functioning of dopamine, a neurotransmitter that has been linked to reward, before and after the intervention (Ledermann et al., 2021).

Figure 1Simplified Schematic Model of the Stress-Reward Interaction as Mediator of the Relationship Between Stress Exposure and the Development of Psychopathological Symptoms



Note. A higher neural reactivity to stress and a lower reactivity to reward are hypothesized to be a vulnerability factor for psychopathology. Interventions targeting one or both mechanisms can be used in a preventive manner or as treatment.



What Does This Mean for Psychologists?

Integrating results and approaches from other disciplines such as neuroscience to understand and identify neural mechanisms that are important for the development of disorders and that are directly associated with psychological mechanisms allows the development of targeted psychological interventions that can be used preventively in groups of individuals at risk of developing psychological disorders, for example in offspring of depressed parents. These interventions can also be used in addition to or in complement to usual psychotherapeutic treatment for individuals currently diagnosed with a mental disorder in order to offer targeted treatment. These mechanism-based interventions enrich the clinical psychologist's range of interventions and allows for a transdiagnostic approach. Finally, as their neural correlates are known, it is possible to perform neuroimaging measures of these mechanisms before and after the intervention in randomized controlled trials to show the effect of the psychological interventions not only at a clinical level, but also at a neural level. This approach is therefore a promising avenue for the development of new clinical psychological interventions either for the prevention or the treatment of mental disorders.

Funding: The author has no funding to report.

Acknowledgments: The author has no additional (i.e., non-financial) support to report.

Competing Interests: The author has declared that no competing interests exist.

Twitter Accounts: @Psychologiecli1

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Clinical Psychology in Europe (CPE) is the official journal of the European Association of Clinical Psychology and Psychological Treatment (EACLIPT).



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Research Articles



Did a Nocebo Effect Contribute to the Rise in Special Education Enrollment Following the Flint, Michigan Water Crisis?

Siddhartha Roy^{1,2} , Keith J. Petrie³ , Greg Gamble⁴ , Marc A. Edwards¹

[1] Department of Civil and Environmental Engineering, Virginia Tech, Blacksburg, VA, USA. [2] UNC Water Institute, Gillings School of Global Public Health, University of North Carolina, Chapel Hill, NC, USA. [3] Department of Psychological Medicine, University of Auckland, New Zealand. [4] Department of Medicine, University of Auckland, Auckland, Auckland, New Zealand.

Clinical Psychology in Europe, 2023, Vol. 5(1), Article e9577, https://doi.org/10.32872/cpe.9577

Received: 2022-05-28 • Accepted: 2023-02-21 • Published (VoR): 2023-03-31

Handling Editor: Winfried Rief, Philipps-University of Marburg, Marburg, Germany

Corresponding Author: Siddhartha Roy, The Water Institute, University of North Carolina, 4114 McGavran-Greenberg Hall, Chapel Hill, NC 27516, USA. E-mail: sidroy@vt.edu

Supplementary Materials: Materials [see Index of Supplementary Materials]



Abstract

Background: Exposure to waterborne lead during the Flint Water Crisis during April 2014-October 2015 is believed to have caused increased special education enrollment in Flint children.

Method: This retrospective population-based cohort study utilized de-identified data for children under six years of age who had their blood lead tested during 2011 to 2019, and special education outcomes data for children enrolled in public schools for corresponding academic years (2011-12 to 2019-20) in Flint, Detroit (control city) and the State of Michigan. Trends in the following crisis-related covariates were also evaluated: waterborne contaminants, poverty, nutrition, city governance, school district policies, negative community expectations, media coverage and social media interactions.

Results: Between 2011 and 2019, including the 2014-15 crisis period, the incidence of elevated blood lead in Flint children ($\geq 5\mu g/dL$) was always at least 47% lower than in the control city of Detroit (p < .0001) and was also never significantly higher than that for all children tested in Michigan (p = 0.33). Nonetheless, special education enrollment in Flint spiked relative to Detroit and Michigan (p < .0001). There is actually an inverse relationship between childhood blood lead and special education enrollment in Flint.



Conclusion: This study failed to confirm any positive association between actual childhood blood lead levels and special education enrollment in Flint. Negative psychological effects associated with media predictions of brain damage could have created a self-fulfilling prophecy via a nocebo effect. The findings demonstrate a need for improved media coverage of complex events like the Flint Water Crisis.

Keywords

blood lead, lead exposure, Flint Water Crisis, nocebo effect, special education

Highlights

- Waterborne lead exposure during the Flint crisis did not correlate with special education enrollment.
- Flint children were repeatedly labeled as lead poisoned and brain damaged in the aftermath.
- · A nocebo effect could have contributed to negative educational outcomes in Flint.
- Erroneous, negative media labels can be internalized and lead to psychological harm in children.

In April 2014, the city of Flint, Michigan stopped purchasing treated Lake Huron water from Detroit and switched to corrosive Flint River water as a cost savings measure. The city also interrupted the addition of corrosion control chemicals to the treated water, which were required under federal regulations to reduce the leaching of the neurotoxin lead from lead pipes and home plumbing. This increased lead levels in tap water and children's blood mainly in the months of June-August 2014 (Roy et al., 2019). In response to residents' concerns, two of the authors assisted with sampling 269 Flint homes in 2015, proving the 90th percentile water lead level (27 μg/L) was almost twice the US Environmental Protection Agency (EPA) action level of 15 μg/L (Pieper et al., 2018). It was later revealed that the proportion of children < 6 years of age with elevated blood lead, i.e., ≥ 5 μg/dL US Centers for Disease Control and Prevention (CDC) reference level, increased following the water switch (Hanna-Attisha et al., 2016), primarily in June-August 2014 (Roy et al., 2019). Michigan officials later announced a Legionnaire's Disease outbreak that killed at least 13 people (Rhoads et al., 2017). These events became known in the media as the Flint Water Crisis (FWC). After the water problems were exposed, Flint reconnected to Detroit water in October 2015, a federal emergency was declared in January 2016, and over US\$1.2 billion in relief funds have been appropriated for residents including free bottled water (through April 2018), free lead faucet filters, health interventions, settlement money for lead-exposed children, special education services, and replacement of around 12,000 lead pipes to be completed in 2023 (Bosman, 2020; City of Flint, 2022; Roy & Edwards, 2019a; Roy & Edwards, 2020). Flint water has met all federal standards since late 2016 and many residents still consume only bottled water due



to lost trust (Flint Cares, 2018; Fonger et al., 2019; Reuben et al., 2022; Roy, 2017; Roy & Edwards, 2019b; Sobeck et al., 2020).

Recent media reports (see Supplementary Materials [SM] Table S1) attribute increasing rates of special education enrollment and diagnoses of learning disabilities in Flint children to lead exposure and "lead poisoning" from the FWC (Alfonsi, 2020; Green, 2019) but none of these conclusions are based on peer reviewed data. Blood lead levels have been steadily dropping in the United States and in Flint for the past 50 years following the banning of lead from gasoline, paint and pipes (Dignam et al., 2019; Gómez et al., 2018). The peak childhood blood lead levels during the FWC (2014-15) were well below those recorded in Flint during 2011 (Gómez et al., 2018; Roy et al., 2019).

In this study, we investigate the hypothesis that increased negative educational outcomes were caused by lead exposure from the FWC as has been stated by the media and experts (ACLU, 2016; Alfonsi, 2020; Green, 2019; Redlener, 2018; Riley, 2018; Strauss, 2019). Trends in blood lead levels of Flint children were compared to the control city of Detroit, which has comparable socioeconomic and racial make-up (Table 1) and also used the same drinking water for over 50 years except for the 18 months of the FWC. We also compare Flint to state-wide trends from Michigan, and evaluate relevant extraneous factors that may have affected educational outcomes in Flint children.

 Table 1

 Key Demographic Factors of Comparison for Flint and Detroit (Control City)

Measure	Flint	Detroit (Control)
Water source during:		
1950s - Apr 2014	Lake Huron	Lake Huron
Apr 2014 – Oct 2015	Flint River	Lake Huron
Oct 2015 – present	Lake Huron	Lake Huron
Approximate count of lead service line	Pre-2016: 12,000 (40%)	80,000 (40%)
connections (% of total water	Current: <1,400 (4.7%)	
connections)		
Drinking water source in public schools	Bottled water	Bottled water
	(Sep 2015-Feb 2022)	Aug 2018-Aug 2019)
	Filtered water	Filtered water
	(Feb 2022-present)	(Aug 2019-present)
Net change in population (2011 to 2019),	-8.4%	-8.6%
%* [#]	(105,391 to 96,559)	(738,223 to 674,841)
Population < 5 years old (range during 2011-19), %*	7.5-8.3%	7.0-7.3%
Persons per household, 2014-18	2.36	2.55
Net change in unemployment rate (2011	-52.1%	-58.1%
to 2019), % [†]	(19% to 9.1%)	(20.5% to 8.6%)



Measure	Flint	Detroit (Control)
Net change in median household income	+8.3%	+10.9%
(2011 to 2019), %*	(\$26,621 to \$28,834)	(\$27,862 to \$30,894)
Health outcomes (range during	77-82 of 83	81-83 of 83
2011-2019), overall rank in Michigan	(Genesee Co.)	(Wayne Co.)
Percent below poverty level (range during 2011-19), %*	38.8-41.9%	35-40.9%
Worst American city to live in, rank (based on 2015 data)	#1	#3
% decline in total students attending public schools in 10 years (2009-10 till 2018-19)	43.1%	68.4%
% of total resident students attending	45.6%	37.9%
charter schools, 2018-19 (national rank in charter school enrollment)	(#3)	(#2)

^{*}Data from American Community Survey 5-Year Estimates Data Profiles via US Census (US Census Bureau, 2022).

Other data references: City of Flint, 2022; David et al., 2017; Goetz, 2022; Mack, 2019; Sauter et al., 2017; University of Wisconsin Population Health Institute, 2022.

After demonstrating that covariates unlikely played a primary role (see Text S1, Supplementary Materials), we probe the possibility of a nocebo effect (Barsky et al., 2002; Petrie & Rief, 2019) or a self-fulfilling prophecy, associated with repeated predictions of brain damage to Flint children via the intense publicity associated with the FWC. Research has shown that parents' and teachers' negative expectations of children can have adverse effects on educational outcomes. Past studies also suggest that these effects are cumulative and have a greater impact on disadvantaged populations (Jussim et al., 2009; Madon et al., 1997; Madon et al., 2011; Rosenthal & Jacobson, 1968). To examine the interaction between media stories and community perceptions, we evaluated a) representative national and local media stories and associated social media interactions, b) public statements of government, medical and school leaders, and c) resident feedback in media's news stories, highlighting the purported effects of lead and "lead poisoning" during the FWC period on children and their educational outcomes in Flint and the control city of Detroit.



^{*}US Census language: Estimates are not comparable to other geographic levels of health estimates (due to methodology differences that may exist between different data sources).

[†]Data from Michigan Bureau of Labor Market (Michigan Department of Technology Management and Budget, 2022).

Materials and Method

Elevated Blood Lead

Childhood blood lead testing is required under Medicaid, where all children receive a screening blood lead test at ages 1 and 2 years, and up to 5 years (Cantor et al., 2019; US Preventive Services Task Force et al., 2019), but not all children receive such tests in practice. The State of Michigan sampling methodology and reporting guidelines have not changed markedly since 1998 (Michigan Department of Health and Human Services, 2020). The percentage of children under six years of age with blood lead above the 2012-21 CDC reference level of 5 μ g/dL, and the pre-2012 CDC "level of concern" of 10 μ g/dL, were calculated for Flint, Detroit, and Michigan for the years 2011-19 using a dataset with 1,445,808 blood lead levels of all Michigan children tested, obtained from the Michigan Department of Health and Human Services (MDHHS) through a Data User Agreement (#202103-144) following IRB approval (IRB #202103-04-NR). Separately, de-duplicated data were also provided to us after MDHHS epidemiologists extracted the highest blood lead values per child per year using the following standard criteria (in order of preference):

- The highest venous blood lead test result available during the calendar year
- If there is no venous test result available, the highest capillary blood lead test result available during the calendar year
- If there is no test result with blood type available, the highest test result available during the calendar year

Educational Outcomes

The data on all special education outcomes and general education 3rd grade reading proficiency for students enrolled in Flint Community Schools, Detroit Public Schools Community District, and all public schools in Michigan for the academic years 2011-12 until 2019-20 (or, latest available) were downloaded from the Michigan Department of Education's website www.mischooldata.org. The special education enrollment data during 2006-07 to 2010-11 was obtained through Freedom of Information Act requests to the Michigan Department of Education.

Poverty and Nutrition

The rates of poverty and households with children aged 0-18 years receiving food assistance (i.e., on Supplemental Nutrition Assistance Program) for Flint, Detroit and Michigan for 2011-19 were obtained from the US Census Bureau (US Census Bureau, 2022).



Media Coverage and Social Media Interactions

A representative list of national and local media stories on lead exposure and educational outcomes of Flint children and Detroit children during October 2015-January 2021 (Table S1) was gathered using Google searches with keywords "lead", "children", "education", "Flint" with and without the term "-Detroit" (i.e., removes all search results with "Detroit"), and "Detroit" with and without "-Flint". The CrowdTangle extension v3.0.29 in Google's Chrome Browser was utilized to gather total "interactions" (reactions, comments, and shares) of all Facebook users and total follower counts of public pages (e.g., celebrities, news organizations, and politicians) and public groups who shared the media stories on Facebook from publishing date until the time of conducting research (August 2020-September 2021). The representative negative expectations commentary of community leaders, teachers, parents and schoolchildren about lead exposure during the FWC period and educational difficulties for Flint and Detroit (Table S2) were gathered through manual screening of articles, posts and videos published during October 2015-January 2021, which were in turn obtained through open-ended Google searches using multiple keywords, including "Flint" (Flint only), "Detroit" (Detroit only), "Flint Water Crisis" (Flint only), "lead", "poisoning", "education", and "children". Separately, the total count and number of interactions data for all posts and web links shared on official Facebook pages of Michigan local media (Data S1) with the keywords "lead poisoned" during January 2016-November 2020 were downloaded from CrowdTangle (www.crowdtangle.org) and network maps were plotted in Gephi v0.9.2 (CrowdTangle, 2020).

Statistical Analyses

All analyses were conducted in Excel® 2016 (Microsoft), SAS® 9.4 (SAS Institute, Cary NC), or GraphPad PRISM 8.4.3 (GraphPad Software). General linear mixed-effects modeling was used to model changes in binary effects over time, between Flint and Detroit (both nested within Michigan). Data are presented as mean with 95% confidence intervals. Pairwise planned comparisons sliced through each year were made and p < .05 was considered significant after false discovery rate adjustment within each outcome. All tests were two tailed. No further adjustment for multiple comparisons was performed. Ordinary least squares regression lines were fitted between log of percentage students enrolled in special education and log %EBL in the same years and the slopes compared within GraphPad PRISM.

Results

This retrospective population-based cohort study utilized longitudinal datasets (de-identified aggregated yearly data) for Flint, the control city Detroit, and the entire State



of Michigan to examine the hypothesized link between lead exposure and educational outcomes.

Elevated Blood Lead

The proportion of children < 6 years with elevated blood lead (%EBL) at or above the 5 µg/dL CDC reference level decreased significantly from 2011 to 2019, $p_{\rm (time)}$ < .0001, in Flint, Detroit and Michigan overall. The %EBL in Flint steadily decreased by 65.8% between 2011-19 from 5.42% to 1.85%, Risk Ratio = 0.43, 95% CI [0.33, 0.56], p < .0001, notwithstanding the FWC increase that occurred in the months of June-August 2014 immediately following the water switch (1,17). The corresponding %EBL in Detroit and Michigan also saw large decrements of 41.3%, Risk Ratio = 0.69, 95% CI [0.65, 0.74], p < .0001) and 55.3%, Risk Ratio = 0.58, 95% CI [0.56, 0.61] from 2011-2019, respectively (Figure 1a).

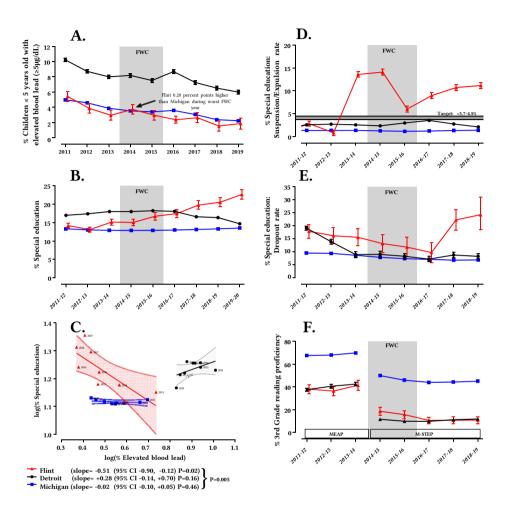
There were also substantial differences in %EBL between Flint, Detroit and all of Michigan (Figure 1a) ($p_{(\text{time*center})} < .0001$). Specifically, %EBL in Flint was 47-77% lower than for Detroit during 2011-19. Even in the worst FWC year of 2014, children in Detroit had more than double the %EBL of Flint. The %EBL in Flint (which comprised 2.2% to 2.4% of the State population) was also 13-35% lower than for the State of Michigan between 2012-19, with the exception of 2014 when Flint exceeded the %EBL in Michigan by 0.20 percentage points (i.e., 3.72% in Flint vs. 3.52% in Michigan). In other words, the net effect of the FWC, was to temporarily raise the blood lead of Flint children, up to the average for all data reported by the State of Michigan.

The relative trends between Flint, Detroit, and Michigan at the pre-2012 CDC 10 $\mu g/dL$ "level of concern" blood lead threshold (%EBL10) were somewhat similar (Figure S1) to those seen at the 5 $\mu g/dL$ level (%EBL). The %EBL10 for Flint was statistically indistinguishable from Michigan during 2011-19 even during the 2014 and 2015 FWC years. Finally, the %EBL10 for Flint was 65-77% lower than Detroit (p < .00001) for the entire 2011-19 time period.



Figure 1

Childhood Blood Lead and Educational Outcomes



Note. Trends in (A) percentage of children < 6 years of age with elevated blood lead $\geq 5 \,\mu\text{g}/\text{dL}$ (%EBL), (B) enrollment of public school students in special education programs, (D) special education suspension/expulsion rates, (E) special education dropout rates, and (F) general education 3rd grade reading proficiency*, for Flint, Detroit, and Michigan, 2011-19 (and corresponding school years of 2011-12 to 2019-20). Error bars are +/- 95% confidence intervals and maybe contained within symbols. (C) Scatter plot between %EBL vs. special education enrollment rate for Flint, Detroit, and Michigan by year. 95% confidence bands for the ordinary least squares fits are shown. p value shown is for comparison of slopes.

*The State of Michigan followed the Michigan Educational Assessment Program (MEAP) testing standards until 2013-14 and then switched to Michigan Student Test of Educational Progress (M-STEP) starting 2014-15.



Analysis at the individual child-level was conducted to consider isolated cases of anomalously high blood lead from acute exposure during the FWC that were possibly masked by yearly aggregated trends (i.e., Figure 1a). Plotting blood lead measurements of every tested child in Flint and Detroit with blood lead $\geq 5~\mu g/dL$ (Figure 2a-2b) and comparing the count and percentage of children in Flint, Detroit, and Michigan with blood lead ≥ 5 , 10, 20, 25 and 40 $\mu g/dL$ (Figure 3) during the FWC period of 539 days (April 25 2014-October 16 2015) revealed:

- a. The mean blood lead of all children with blood lead $\geq 5 \mu g/dL$ in Detroit was 12.4% higher than in Flint (unpaired two-tailed *t*-test; p < .05).
- b. The count of Detroit children was higher than Flint children in every blood lead category (\geq 5-40 µg/dL).
- c. Detroit children had statistically higher blood lead than Flint children at and above the 5 and $10 \mu g/dL$ blood lead thresholds.
- d. Data for Flint children were statistically indistinguishable from that reported for all State of Michigan children in every blood lead category ($\geq 5-40 \,\mu\text{g/dL}$).
- e. There were 28 children who tested at or above 40 μ g/dL in Michigan during the FWC period, of which half (14) were in Detroit and none (0) in Flint.
- f. There were four Flint children with blood lead at or above 25 $\mu g/dL$, both during the FWC and in the same time duration pre-FWC (November 1 2012 April 24 2014).

Overall Educational Outcomes

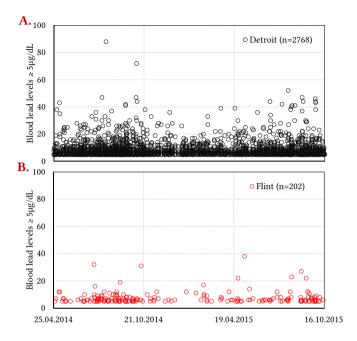
Of 23 special education outcomes monitored each academic year, nine worsened, nine improved and five did not change (<±1% change) in Flint after the crisis vis-à-vis before the water crisis (see Text S2, Supplementary Materials). In a simple comparison relative to Detroit, only three outcomes worsened and another three improved in Flint.

Despite these overall neutral trends, four worsening outcomes in Flint were nonetheless emphasized and attributed to lead exposure from the FWC by the national media and experts (ACLU, 2016; Alfonsi, 2020; Green, 2019; Redlener, 2018; Riley, 2018; Strauss, 2019) including: a) special education enrollment; b) suspension or expulsion for children in special education; c) dropout for children in special education, and d) worsening reading proficiency of 3rd grade students in general education.



Figure 2

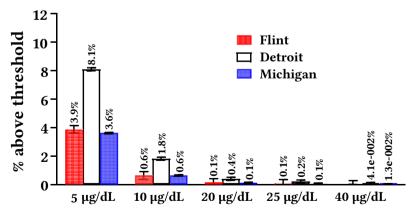
Individual Child-Level Blood Lead Measurements ≥ 5 μg/dL During the FWC Period (April 25 2014-October 16 2015)



Note. (A) Detroit and (B) Flint. The data is de-duplicated; i.e., only highest blood lead value per child is shown.

Figure 3

Percentage of Children < 6 Years of Age With Blood Lead $\geq 5-40~\mu g/dL$ in Flint, Detroit, and Michigan During April 25, 2014 – October 16, 2015



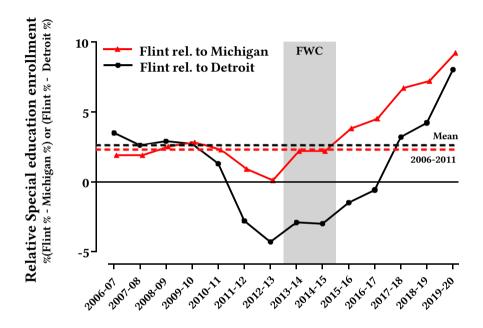
Note. The CDC threshold for elevated blood lead was 40 $\mu g/dL$ between 1973-75, 25 $\mu g/dL$ between 1985-90, 10 $\mu g/dL$ until 2012 and 5 $\mu g/dL$ until 2021.



Each of these attributions is examined in greater detail for Flint Community Schools, Detroit Public Schools Community District, and all public schools in Michigan using data for the academic years 2011-12 until 2019-20. Special education enrollment was also examined from 2006-07 onwards to identify a baseline (Figure 4), and 3rd grade reading proficiency was not analyzed in 2019-2020 since tests were cancelled due to the COVID-19 pandemic.

Figure 4

Special Education Enrollment in Flint Relative to Detroit and Michigan, 2006-20



Special Education Enrollment

Special education enrollment trends are routinely gathered under federal and state laws (Individuals with Disabilities Act, 2004; Michigan Department of Education, 2020; Weiss & Mettrick, 2010), and the data are significantly different for Michigan, Detroit and Flint (p < .0001, Figure 1b). Overall, in the State of Michigan, the proportion of children in special education remained stable between 2011-12 and 2019-20 at 12.9-13.5%. The special education enrollment rate in Detroit slightly increased from 2011-12 to 2016-17 followed by a downtrend during 2017-18 to 2019-20 (Figure 1b). The special education enrollment rate in Flint started lower than Detroit (p < .0001) in 2011-12 as would be expected due to lower blood lead levels alone, but began rising in 2015-16 (relative to the previous three academic years) after the federal emergency and aggressive national and



international reporting on the FWC (Green, 2019; Jackson, 2017; Pew Research Center, 2017). Flint special education enrollment even surpassed that in Detroit in 2017-18 (p < .0001). Notably, the spike in Flint special education enrollment rate only occurred in the 6-21 year age group, whereas the age group that would be considered most vulnerable to water lead exposure (i.e. those in the womb or up to age 1 during the FWC) saw no significant increase (Figure S2).

The special education enrollment rate in Flint relative to Michigan during 2013-15, including the first FWC year, was comparable to the 2006-11 baseline, but began to spike in the second FWC year (2015-16), when media coverage on the crisis increased markedly (see Figure 4, "Flint relative to Michigan"). This was associated with a strong diverging trend between the special education rates for Flint and Detroit starting in 2016-17 (Figure 1b). Similarly, Flint special education enrollment was much lower relative to Detroit between 2011-16 (see Figure 4, "Flint relative to Detroit"), became comparable in 2016-17, and increased dramatically from 2017-20. There is actually a strong inverse relationship (Figure 1c) between %EBL and special education enrollment rate in Flint, r = -0.79, 95% CI [-.96, -0.18], p = .021, but there is no such relationship for the same time period in Detroit, r = 0.20, 95% CI [-0.59, 0.79], p = 0.63, or Michigan, r = 0.09, 95% CI [0.66, 0.75], p = 0.83).

Special Education Suspension/Expulsion Rates

The special education suspension/expulsion rates in Flint increased 7.4 times in 2013-14 (13.6%) before the FWC began (Figure 1d) compared to the previous two school years, and peaked in the first FWC year 2014-15 (14.1%), before dropping more than half in the second FWC year 2015-16 (6%). Rates progressively rose during 2016-19 (9% to 11.2%) after the FWC came to light.

Special Education Dropout Rates

The special education dropout rates in Flint roughly doubled in the 2017-20 school years (22.1%) versus 2014-17 (11.5%), after a steady decline during 2011-17 analogous to that occurring in Detroit and Michigan (Figure 1e).

General Education Reading Proficiency

After the State of Michigan adopted the stricter Michigan Student Test of Educational Progress (M-STEP) standard in the 2014-15 school year, both Flint (22.3 percentage points) and Michigan (19.9 percentage points) witnessed identical drops (around 20 percentage points) in 3rd grade reading proficiency between 2013-14 and 2014-15, but Detroit fell even more precipitously (31 percentage points) (Figure 1f). During the FWC (2014-16 school year) and until the 2018-2019 school year, 3rd grade reading proficiency stayed roughly the same in Detroit, however Flint continued to decrease until reaching the same level as Detroit.



Potential FWC Covariates That Could Explain Rising Special Education Enrollment

Analyses of trends in covariates including waterborne contaminants besides lead, poverty, poor nutrition, City of Flint's administration and emergency management decisions, and Flint Community Schools' policies and funding do not appear to be primarily associated with the post-FWC rise in special education enrollment in Flint (see Text S1, Supplementary Materials).

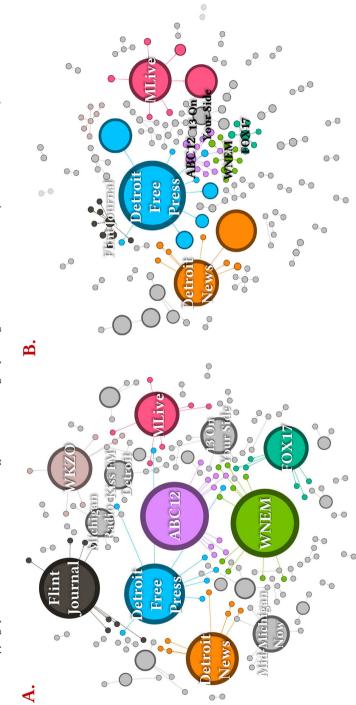
Negative Community Expectations and Media Coverage

The aggregated data from Facebook, the dominant social media platform in the United States (Perrin & Anderson, 2019), obtained using the CrowdTangle (www.crowdtangle.com) public insights tool owned and operated by Facebook, indicate news around lead "poisoning" of Flint children and worsening educational outcomes was interacted with hundreds of thousands of times and potentially reached tens of millions of users on Facebook alone between 2015-21 (Table S1). In contrast, there were just two articles about Detroit children that saw just over 22,000 interactions. On average, 12.2% web traffic to news websites originate from social media, and, therefore, these reported values are gross underestimates of the total "reach" of news, which would include all newspaper and magazine hard copies read, news channel broadcasts watched, and radio programs and podcasts heard (Alexa, 2020). To illustrate, the 60 Minutes Flint special education episode (Alfonsi, 2020) alone was interacted with over 27,000 times and potentially reached ~10 million users on Facebook, and its television broadcast was also watched by over 10 million viewers on the CBS channel (Table S1). Negative pronouncements about lead exposure during the FWC period and educational difficulties in the media disproportionately originated from Flint community leaders describing Flint children, but similar claims were not made publicly by Detroit community leaders (Table S2) despite the much higher blood lead for Detroit children (Figure 1a, Figure 2a, 2b, Figure 3 and Figure S1).

A search for all posts and weblinks shared on official Facebook pages of Michigan local media (Data S1) with the keywords "lead poisoned" and no mention of "Flint" or "Detroit" during January 2016-November 2020 time period using CrowdTangle revealed over 80% articles (Data S2) discussed the FWC. Network mapping of these posts (Figure 5) revealed the media outlets who shared the most articles and links to be from Flint (WNEM TV5, ABC12, and The Flint Journal/MLive; collective n = 32) followed by prominent state-level newspapers Detroit Free Press and The Detroit News. Moreover, the postings from Michigan's Top three dailies by circulation (i.e., Detroit Free Press, The Flint Journal/MLive, and Detroit News) (Agility PR, 2020) saw the most interactions (~35,000) in the form of reactions, shares, and comments.



Network Mapping of All Posts and Weblinks Shared on Official Facebook Pages of Michigan Local Media With the Keywords "Lead Poisoned," Jan 2016-Nov 2020 Figure 5



Note. (A) Media outlets arranged by total number of posts/links shared. (B) Media outlets arranged by total interactions (reactions, shares, and comments) on posts/ inks shared. Raw values for all media bubbles in the maps are provided in SI (Table S4).

(i) The size of the media bubbles is relative; i.e., higher the metric of interest, larger the bubble. The numerous links emerging from each bubble indicate resharing of he posts/links to other Facebook pages and the representative bubbles are also relatively sized according to the metric of interest. (ii) The Flint Journal belongs to he parent media company MLive and both have separate Facebook pages. Therefore, while the pages appear separately in the left network map as they do in Facebook, their interaction metrics are aggregated in text.



Discussion

This investigation confirms that the proportion of young children with elevated blood lead in Flint, Detroit and Michigan as a whole has been declining over the past 10 years. The %EBL trend in Flint is very similar to that observed across Michigan and has always been much lower than Detroit. In fact, the %EBL in Flint has now dropped below data for the United States (US Centers for Disease Control and Prevention, 2020).

For additional perspective, the geometric mean blood lead even during the worst FWC year as reported in previous research (Gómez et al., 2018), was lower than that reported in the European nations of France and Poland (Table S3). Paradoxically, since the FWC was revealed in 2015 and residents were further protected from exposure to waterborne lead, Flint saw a dramatic spike in special education enrollment, while such enrollment remained steady across Michigan and even declined after 2015-16 in the control city of Detroit.

While lead is a neurotoxin with known potential for worsening educational outcomes (Jusko et al., 2008; Mendelsohn et al., 1998; Surkan et al., 2007; Watt et al., 1996), analysis of the data in Flint relative to Detroit is inconsistent with the attribution of rising special education enrollment in Flint to lead exposure. The worst lead exposure from the FWC was of relatively short duration (about one-sixth of the entire time on Flint River water; Roy et al., 2019), and is set against a historic decline in blood lead in Flint as well as Detroit, Michigan and nationally (Dignam et al., 2019; Gómez et al., 2018). The elevation in Flint childhood blood lead was above the relatively new 5 μ g/dL CDC reference threshold but not the 10 μ g/dL threshold "level of concern" exceeded in Washington DC children during its 2001-04 lead in drinking water crisis (Roy et al., 2019). The number of individual children testing \geq 25 μ g/dL, a threshold above which it is reported that 20% of children require an average of nine years of special education (Swinburn, 2016), was 18 times higher in Detroit (0.21% of all children tested; n = 73) than in Flint (0.08% of all children tested; n = 4) during the FWC.

As early as January 2016, it was acknowledged that the worst-case incidence of elevated blood lead during the FWC was always less than half of the incidence in other Michigan cities of Detroit, Grand Rapids, and Muskegon, and 3,800 other communities across the United States (Frazier, 2018; Lanphear, 2017a; Mack, 2016; Pell & Schneyer, 2016; Wilkinson, 2016). Moreover, since %EBL in Detroit was always at least twice that in Flint before, during and after the FWC, worse outcomes, whether concurrent or lagged, would always be expected for Detroit children, but such an impact is not observed in the time period of interest. Instead, there is an incongruous inverse relationship between childhood blood lead and special education enrollment in Flint, while no such relationship exists for Detroit and Michigan.

Despite an equal number of overall special education outcomes worsening and improving (see Text S1, Supplementary Materials), only those that superficially appeared to be worsening were publicized in the media. Our detailed analysis shows these outcomes



are insignificant or inconsistent with the actual lead exposure that occurred. Specifically, the seven-fold jump in suspension/expulsion rates of special education students had occurred in 2013-14 before the onset of the crisis, and the comparison with Detroit further discounts an association with lead exposure. Indiscriminate enforcement of suspension/expulsion policies before the FWC (D.R. v. Michigan Department of Education, 2016) may have contributed to this spike. The special education dropout rates in Flint only started to rise in 2017-18 post-FWC after expectations of such an outcome was widely publicized in the media starting late 2016. Finally, the reduction in Flint general education 3rd grade reading proficiency after adoption of a new academic standard in 2014-15 was also observed in the Detroit control group, and could be attributed to the changed tests.

The rise in special education enrollment in Flint following the FWC was not associated with confounders of waterborne contaminants besides lead during the FWC, poverty, poor nutrition, and emergency management. The Flint schools' failure to properly enforce special education policies and a severe budget deficit since the early 2010s may have contributed to less Flint students being enrolled in special education programs pre-FWC, but the enrollment rate had returned to historical norms during the FWC.

A nocebo effect is consistent with the trend of rising special education enrollment after the FWC was exposed (Colloca & Barsky, 2020; Petrie & Rief, 2019). As a top news story of 2016, the crisis engendered negative psychological effects described by residents as "Flint fatigue," and the surrounding international media coverage has continued for over five years with negative headlines (Adams, 2016; Associated Press, 2020; Cuthbertson et al., 2016; Goodnough & Atkinson, 2016; Heard-Garris et al., 2017; May, 2016). The news reports and their popularity on social media (Table S1, Figure 5) and negative perceptions of Flint community leaders and parents (Table S2) could have heightened negative expectations about the effects on children, who readily accept and act on information from those they trust (Harris & Corriveau, 2011; Jaswal et al., 2010; Landrum et al., 2013). Contaminated water creates high public anxiety compared to other environmental concerns (Petrie et al., 2001). For example, the psychological impact of the FWC caused increased tap water avoidance amongst US children nationwide after the FWC came to light (Rosinger & Young, 2020). The early speculation and worst case predictions of impacts on Flint children were also made in a vacuum of trust, uncertainties in the timing and magnitude of the water lead exposure due to manipulation of official test results, and an acknowledged "failure of government at all levels" that caused the FWC (Roy & Edwards, 2019a).

From 2016-18 arguments over the possible negative consequences of labeling Flint children "poisoned" versus "exposed" played out in the media (Clark & Filardo, 2018; Drum, 2017; Gómez & Dietrich, 2018; Mays, 2018; Schneider et al., 2016; Shell, 2016). The worst negative expectations for special education enrollments in years following the FWC appear to have been realized, even though comprehensive blood and water



lead analyses eventually published in 2018-20 (Gómez et al., 2018; Gómez et al., 2019; Roy et al., 2019; Roy & Edwards, 2020) contradict the popular belief that Flint children experienced an unprecedented environmental lead exposure (Figure 1a). Moreover, in many cases, the national media – e.g., *The New York Times* (Green, 2019) and *CBS 60 Minutes* (Alfonsi, 2020) – have provided even worse prognoses, labeling Flint children as brain damaged or lead poisoned (Table S1). No comparable media labeling was applied to children in Detroit (Table S1, Figure 5) or the other Michigan cities with much higher %EBL incidence. A significant percentage of Flint households experience water crisis-related stress and other negative psychological effects, are meeting criteria for psychological trauma, report behavioral problems in their children, and believe that "the crisis would never be fixed" (Bosman & Greeson, 2020; Brooks & Patel, 2022; Ezell & Chase, 2021; Jones et al., 2022; Reuben et al., 2022; Sneed et al., 2020; Trejo et al., 2022).

A perception that Flint's water is still unsafe and a source of ongoing community concern is supported by continued high rates of bottled water use five years after the switchback to Detroit water. Bottled water use has persisted despite distribution of free lead filters, replacement of over 90% of lead pipes, and independent tests showing current Flint water lead levels to be lower than observed in other Michigan cities with old pipes (Alfonsi, 2020; City of Flint, 2022; Flint Cares, 2018; Reuben et al., 2022; Roy & Edwards, 2019a, 2020). In fact, it is reported that some of Flint's youngest children have only bathed in and consumed bottled water their entire lives (Alfonsi, 2020; Fonger et al., 2019; Herndon, 2018).

Exposure to feared contaminants such as lead is known to create nocebo responses (Blettner et al., 2009; Crichton et al., 2014; Gruber et al., 2018; Petrie et al., 2005; Small & Borus, 1987; Witthöft & Rubin, 2013). Other suspected water contamination incidents have caused health complaints that were difficult to explain by the level of toxicological exposure (David & Wessely, 1995; Page et al., 2006). In the Camelford water contamination incident in Cornwall, England, health complaints were intensified by media interest, concerns about a conspiracy and litigation (David & Wessely, 1995). However, in contrast to the FWC, those studies did not have direct data from continuous monitoring of the contaminant of concern in the blood of the affected population, or suitable control groups for comparison as in the research results presented herein.

It has also been argued that the actual (and small) magnitude of elevation in children's blood lead from the FWC does not matter in terms of the resulting health harm (e.g., Hanna-Attisha et al., 2018; Kuehn, 2016; Oleske et al., 2016; Schmidt, 2018; Schneider et al., 2016; Stateside Staff, 2018). However, the epidemiological study by Lanphear and colleagues noted an inverse, supralinear dose-response relationship: a net decrease of 6.9 IQ points, 95% CI [4.2, 9.4] for blood lead increment of 2.4 to 30 μ g/dL, with the steepest drop of 3.9 IQ points, 95% CI [2.4, 5.3] occurring for the lowest blood lead range of 2.4 to 10 μ g/dL (Lanphear et al., 2005). While the underlying (biological)



mechanism has not been elucidated (Lanphear, 2017b), the supralinear curve confirms the scientific principle that "the dose makes the poison" for lead.

These data suggest that the rising enrollment in special education attributed to the FWC, may be associated with widespread negative expectations and not an elevation in blood lead. This possible nocebo effect in Flint represents an unfortunate natural large-scale experiment, in which a population has been repeatedly informed by trusted national and international media sources that an unprecedented lead exposure event had occurred with severe long-term adverse repercussions to children, even when the data indicate that the actual lead exposure was normal for the State and less than nearby communities.

Two of this paper's authors (MAE/SR) personally witnessed such expectations during a science outreach program for over 1,000 K-12 Flint students in March 2017 (Edwards, 2017; Jacques, 2018), where several teachers openly expressed their belief that Flint children had been brain damaged, were incapable of learning, and that there was little point in trying to teach them (Bouffard, 2018; Edwards, 2017; Jacques, 2018; Roy & Edwards, 2019c). Trust of teachers in students and parents is a significant predictor of student achievement (Goddard et al., 2001). These and similar expectations have been broadcast in the media for over five years (e.g., Tables S1 and S2, Figure 5) and can strongly influence children's school performance and behavior, such as those previously documented in younger and stigmatized children from African-American and lower socioeconomic backgrounds (Jussim et al., 1996).

Students who require and receive special education services do benefit from them (Ballis & Heath, 2021) and higher special education enrollments are not necessarily indicative of permanent brain damage or health harm from the water lead exposure. In fact, part of the rise might be viewed as part of a proactive effort to compensate for the failures of government at all levels that caused the FWC (Wagner & Kennedy, 2016). In any event, the media have never publicized this possible positive interpretation.

Importantly, the media messaging has not changed, in spite of ample evidence that the actual lead exposures in Flint were not abnormally high relative to all of Michigan and were much lower than neighboring Detroit. It is possible that the harm from such messaging is continuing. For instance, the special education enrollment rate in Flint for 2019-20 (22.7%) is now over 1.5-1.7 times the rates in Detroit (14.7%), and is higher than Michigan (13.5%) and the United States overall (14.1%) (US Department of Education, 2020), despite the fact that Detroit children have always had more than double the incidence of %EBL than Flint (Figure 1a). This trend may even be accelerating due to universal special neuropsychological screening now being conducted for Flint children, which has recently indicated an 80% diagnosis rate for "language, learning or intellectual disorders" that are attributed to lead exposure from the FWC (Alfonsi, 2020; Chambers, 2019).



Our study has limitations. This study is limited by reliance on existing blood lead datasets collected under standard screening practices, covering about 23% of young children in Michigan, 40% in Detroit and 39% of Genesee Co. in 2016 (Michigan Department of Health and Human Services, 2018). Since this is a population-based study, we did not have educational outcomes data at the individual level to adjust for potential confounders or to identify if multiple adverse outcomes were occurring for the same children. Our study is also limited by the lack of charter school data. And finally, our central analyses were correlational, and should be interpreted with caution. In contrast, the strength of this population-based study is the utilization of over 1.44 million individual childhood blood lead measurements and annual monitoring of outcomes in general and special education occurring under uniform Michigan educational policies in two cities with comparable demographics, using the same source of treated drinking water from Lake Huron except for the 18 months Flint was served by the Flint River and suffered the manmade public health crisis. The educational outcomes data are representative as they are weighted by city-level population instead of individual schools. The novel contribution of this study is uncovering of possible nocebo effect in the aftermath of a public health emergency involving a known neurotoxin, via an unfortunate natural experiment that could never have been studied intentionally.



Funding: US Environmental Protection Agency (#8399375); Spring Point Partners, LLC.

Acknowledgments: We are grateful to Michigan Department of Health and Human Services' Childhood Lead Poisoning Prevention Program and especially their Departmental Specialist Mr. Daniel Albright (access to the complete 2011-19 Michigan blood lead dataset and review of this manuscript under the Data User Agreement), Facebook CrowdTangle and their former Academics and Research Lead Ms. Naomi Shiffman (onboarding and access to Facebook CrowdTangle), and the Described and Captioned Media Program (access to the 2017 "Voices of Flint" documentary).

Competing Interests: MAE and SR worked with Flint residents to expose the Flint Water Crisis, and their data, testimony and emails have been subpoenaed in several lawsuits. They are not party to any of these lawsuits. MAE has been subpoenaed as a fact witness in many of the lawsuits, but he has refused all financial compensation for time spent on those activities. SR is serving as a scientific consultant in a Flint lawsuit for VNA starting December 21 2022 on biosolids research, a topic unrelated to this manuscript, and is expected to be financially compensated for that work. All other authors declare they have no competing interests.

Author Contributions: S.R., K.J.P, and M.A.E. designed research; S.R., M.A.E. and G.D.G. performed research; S.R. and G.D.G. analyzed data; and S.R., K.J.P, G.D.G., and M.A.E. wrote the paper.

Twitter Accounts: @siddharthaxroy, @keithpetrie

Data Availability: All education data are publicly available on Michigan Department of Education's website www.mischooldata.org. Blood lead data were obtained from Michigan Department of Health and Human Services under a Data User Agreement (DUA #202103-144) following IRB approval (IRB #202103-04-NR). The data can be made available from MDHHS upon completion of a data use agreement with the agency. The authors assume full responsibility for the analysis and interpretation of the data. All poverty and food assistance data are publicly available from the US Census. All media coverage and Facebook interactions data downloaded from CrowdTangle are available in the Supplementary Materials.

Supplementary Materials

The Supplementary Materials contain the following items (for access see Index of Supplementary Materials below):

- · Figures S1 to S2
- Tables S1 to S4
- Texts S1 (including Figure S3) to S2 (including Figures S4 to S22 and Table S5)
- References
- · Data S1 to S2



Index of Supplementary Materials

Roy, S., Petrie, K. J., Gamble, G., & Edwards, M. A. (2023). Supplementary materials to "Did a nocebo effect contribute to the rise in special education enrollment following the Flint, Michigan Water Crisis?" [Additional information]. PsychOpen GOLD.

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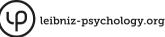
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EACLIPT

Clinical Psychology in Europe (CPE) is the official journal of the European Association of Clinical Psychology and Psychological Treatment (EACLIPT).



PsychOpen GOLD is a publishing service by Leibniz Institute for Psychology (ZPID), Germany.







Research Articles



Clinical Psychology and the COVID-19 Pandemic: A Mixed Methods Survey Among Members of the European Association of Clinical Psychology and Psychological Treatment (EACLIPT)

Julia Asbrand 1 0, Samantha Gerdes 2 0, Josefien Breedvelt 3,4 0, Jenny Guidi 0, Colette Hirsch 6,7,8 0, Andreas Maercker 9,10 0, Céline Douilliez 11 0, Gerhard Andersson 12,13 0, Martin Debbané 14,15 0, Roman Cieslak 16 0, Winfried Rief 17 0, Claudi Bockting 4,18 0

[1] Department of Psychology, Humboldt-Universität zu Berlin, Berlin, Germany. [2] NHS Veterans' Mental Health and Wellbeing Service, Camden and Islington NHS Trust, London, United Kingdom. [3] NatCen Social Research, London, United Kingdom. [4] Centre for Urban Mental Health, University of Amsterdam, Amsterdam, The Netherlands. [5] Department of Psychology "Renzo Canestrari", University of Bologna, Bologna, Italy. [6] Institute of Psychiatry, Psychology and Neuroscience, King's College London, Denmark Hill, Camberwell, London, United Kingdom. [7] National Institute for Health Research (NIHR) Biomedical Research Centre, South London and Maudsley Hospital, London, United Kingdom. [8] South London and Maudsley NHS Foundation Trust, Denmark Hill, Camberwell, London, United Kingdom. [9] Department of Psychology, Division of Psychopathology and Clinical Intervention, University of Zurich, Zurich, Switzerland. [10] Wissenschaftskolleg Berlin-Institute of Advanced Study, Berlin, Germany. [11] Université catholique de Louvain, Psychological Sciences Research Institute, Louvain-La-Neuve, Belgium. [12] Department of Behavioral Sciences and Learning, Department of Biomedical and Clinical Sciences, Linköping University, Linköping, Sweden. [13] Department of Clinical Neuroscience, Division of Psychiatry, Karolinska Institute, Stockholm, Sweden. [14] Psychoanalysis Unit, Research Department of Clinical, Educational and Health Psychology, University College London, London, United Kingdom. [15] Developmental Clinical Psychology Unit, Faculty of Psychology and Educational Sciences, University of Geneva, Geneva, Switzerland. [16] Department of Psychology, SWPS University of Social Sciences and Humanities, Warsaw, Poland. [17] Department of Clinical Psychology and Psychotherapy, University of Marburg, Marburg, Germany. [18] Department of Psychiatry, Amsterdam University Medical Centers (location AMC), Amsterdam, The Netherlands.

Clinical Psychology in Europe, 2023, Vol. 5(1), Article e8109, https://doi.org/10.32872/cpe.8109

Received: 2022-01-10 • Accepted: 2022-11-21 • Published (VoR): 2023-03-31

Handling Editor: Cornelia Weise, Philipps-University of Marburg, Marburg, Germany



[§]These authors contributed equally to this work.

Corresponding Author: Josefien Breedvelt, Josefien Breedvelt, NatCen Social Research, London, UK, 35 Northampton Square, London EC1V 0AX, United Kingdom. E-mail: josefienbreedvelt@gmail.com

Supplementary Materials: Materials [see Index of Supplementary Materials]



Abstract

Background: The COVID-19 pandemic has affected people globally both physically and psychologically. The increased demands for mental health interventions provided by clinical psychologists, psychotherapists and mental health care professionals, as well as the rapid change in work setting (e.g., from face-to-face to video therapy) has proven challenging. The current study investigates European clinical psychologists and psychotherapists' views on the changes and impact on mental health care that occurred due to the COVID-19 pandemic. It further aims to explore individual and organizational processes that assist clinical psychologists' and psychotherapists' in their new working conditions, and understand their needs and priorities.

Method: Members of the European Association of Clinical Psychology and Psychological Treatment (EACLIPT) were invited (N = 698) to participate in a survey with closed and open questions covering their experiences during the first wave of the pandemic from June to September 2020. Participants (n = 92) from 19 European countries, mostly employed in universities or hospitals, completed the online survey.

Results: Results of qualitative and quantitative analyses showed that clinical psychologists and psychotherapists throughout the first wave of the COVID-19 pandemic managed to continue to provide treatments for patients who were experiencing emotional distress. The challenges (e.g., maintaining a working relationship through video treatment) and opportunities (e.g., more flexible working hours) of working through this time were identified.

Conclusions: Recommendations for mental health policies and professional organizations are identified, such as clear guidelines regarding data security and workshops on conducting video therapy.

Keywords

psychotherapy, video therapy, online therapy, blended therapy, clinical psychology, COVID-19

Highlights

- Rapid change in psychotherapy delivery occurred due to the COVID-19 pandemic.
- Clinical psychologists and psychotherapists report challenges (e.g., reluctance among patients) and opportunities, resulting from changes to the work environment.
- Data security is crucial as well as access to treatment via video therapy.
- National policy and organizational guidance is crucial to support clinical psychologists and psychotherapists in their work.



Health care services globally have faced unprecedented challenges due to the COVID-19 pandemic. Alongside the physical health consequences of the COVID-19 virus, mental health problems are also increasing, with reported increases for anxiety, depression, psychological distress and sleeping problems (Bohlken et al., 2020; Liu, Heinzel, Haucke, & Heinz, 2021; Rajkumar, 2020; Salari et al., 2020; Vindegaard & Benros, 2020; Xiong et al., 2020). Furthermore, there has been an estimated additional 53.2 million cases of major depressive disorder and an estimated additional 76.2 million cases of anxiety disorders globally (Santomauro et al., 2021). As a consequence, mental healthcare needs to be prioritized and clinical psychologists and psychotherapists¹ play an important role in the prevention and treatment of these adverse consequences of the COVID-19 pandemic. However, as yet little is known about how well clinicians and services have adapted to the increased demand and additional challenges presented by the COVID-19 pandemic, and what might be done to improve mental health care for those who have suffered psychologically as a consequence of the COVID-19 pandemic.

Clinical psychologists and psychotherapists had to find rapid alternatives to face-to-face treatment such as telephone-based or video therapy (Békés & Aafjes-van Doorn, 2020; Humer, Stippl, et al., 2020), or in-person sessions whilst adhering to their COV-ID-19 national containment measures from the start of the pandemic. Prior studies have shown that the implementation of changes to service delivery can take an average of sixteen years to implement in a health care system (Rogers et al., 2017). In contrast, during the pandemic, change in service delivery was rapid and unexpected, and there was little supervision or guidance available for clinicians (e.g., Boldrini et al., 2020; Probst, Stippl, & Pieh, 2020). Moreover, the pandemic itself led to significantly higher stress levels in clinical psychologists and psychotherapists, especially in younger and less experienced professionals (Aafjes-van Doorn et al., 2020; Probst, Humer, Stippl, & Pieh, 2020). Additionally, fear of infection and other issues related to the pandemic itself were also reported by clinical psychologists and psychotherapists (Humer, Pieh, et al., 2020).

In the midst of such rapid and unforeseen changes to practice, several reassuring and thought-provoking phenomena have been observed. For instance, preliminary evidence showed video therapy to be more effective than previously expected (Humer, Stippl, et al., 2020). Interestingly, the ability to adapt to conducting therapy via video is related to the individual clinical psychologists' and psychotherapists' attitudes and is influenced by their past experiences with video therapy (Békés & Aafjes-van Doorn, 2020). Further, challenges have been reported by mental health professionals regarding the lack of interpersonal interactions, feelings of isolation and other technical issues whilst conducting therapy online (McBeath et al., 2020). The aforementioned studies provide an interest-

¹⁾ We use the term "clinical psychologists and psychotherapists" throughout, however in order to accommodate for different definitions between countries, the term includes clinical psychologists as well as psychotherapists, scientist practitioners, and all other mental health professionals who provide psychological therapy.



ing, yet heterogeneous, picture of the impact of the COVID-19 pandemic on mental health professionals. However, most of the studies used closed questions and quantitative methods (e.g., Békés & Aafjes-van Doorn, 2020; Boldrini et al., 2020), thus limiting the possibility for participants to provide their own insight into offering psychotherapy during a global pandemic.

Professional organizations and other commissions have taken the initiative to provide the public and mental health care professionals with information regarding COVID-19 (e.g., UK², Germany³, Austria⁴, Belgium⁵). However, it is also important for mental health care professionals who work 'on the ground' to share their experiences, in order for organizations to find ways to best support their clinicians. The current survey aimed to gather information 'from the field' to gain an understanding of the experiences of clinical psychologists and psychotherapists working during the COVID-19 pandemic, across different European countries. Members of the European Association of Clinical Psychology and Psychological Treatment (EACLIPT) were consulted; EACLIPT is an association that aims to foster research, education and dissemination of scientifically evaluated findings on clinical psychology and psychotherapy. The current study seeks to provide a first European wide insight into the perceived changes to clinical practice and research of clinical psychologists and psychotherapists, as well as the barriers and opportunities, in order to improve support to people as part of the response to the COVID-19 pandemic. The study also aims to gather information to highlight helpful ways for clinical psychologists and psychotherapists to approach, prioritize and manage their work in the context of the pandemic. Finally, it aims to provide information on how organizations and organizational bodies (such as EACLIPT) can best adapt to pandemic related changes.

Method

Participants

The survey (see Appendix A, Supplementary Materials) was targeted at clinical psychologists and psychotherapists across Europe who are members of EACLIPT. Potential participants were recruited via the EACLIPT members database (n = 698) from 25th May 2020 to 1st September 2020, when COVID-19 restrictions were still in place in most countries. However, it should be noted that restrictions at this time were often not as strict as they were during the first wave of the pandemic, and there were also



²⁾ https://www.bps.org.uk/coronavirus-resources/professional

³⁾ https://psychologische-coronahilfe.de/

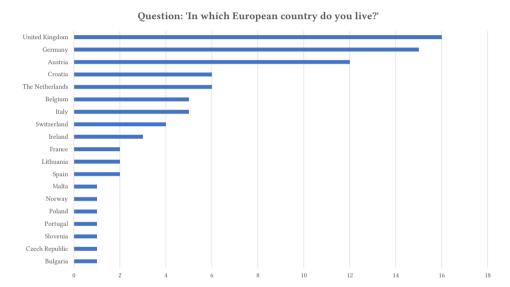
⁴⁾ https://www.boep.or.at/psychologische-behandlung/informationen-zum-coronavirus-covid-19

⁵⁾ https://www.compsy.be/fr/coronavirus

substantial differences between countries. Overall, n = 92 participants (13% of EACLIPT members) voluntarily agreed to participate in the survey. Most of the participants were from the United Kingdom (17.6%), Germany (16.5%), and Austria (13.2%; see Figure 1). Further, most participants worked in a university or other academic institute (34.4%), hospital (14%), public community clinic (15%) or private clinic (18%). The other 30.7% responses included: academic hospital (6.5%), university clinic (7.5%), retired (1%), Courts (1%), prison (1%), and not for profit (1%). Finally, most participants self-identified as working in an urban area (79.1%) compared to rural areas (15.4%), suburban areas (3.4%) and national coverage (1.1%).

Figure 1

Country of Origin of Participants



Note. Please note that n = 6 participants chose to not comment on their country of origin.

Procedure and Measures

Socio-demographic information was collected using nine closed questions (e.g., country of origin, place of work and most commonly presenting patient need during the pandemic). Five open questions were used to gain information on perceived changes in the work place, challenges and opportunities during the crisis, the effect of COVID-19 safety measures on their practice, and other implications. The survey was open for completion between 25th May 2020 and 1st September 2020. The last response that was included was submitted on 19th August 2020.



Quantitative and Qualitative Analysis

The six phases of thematic analysis (Nowell, Norris, White, & Moules, 2017) were followed by the first two authors (J.A. and S.G), including familiarization with the data (Phase 1), generating initial codes (Phase 2), searching for themes (Phase 3), reviewing themes (Phase 4), defining and naming themes (Phase 5) and producing the report (Phase 6). The third author (J.B.) supervised their work and checked the data during Phase 4, in order to review the themes that had been generated. This enabled research bias to be evaluated and the interpretation of the data to be confirmed. The first two authors screened the answers independently in Phases 1, 2, and 3 and formed their own categories, which were then compared and agreed on and a list of themes per question was finalized. Themes were then listed in terms of frequency for each question. The authors were each based in different countries, therefore all meetings took place over remote platforms.

Regarding the overall process, reflexivity is considered as a key aspect of the thematic analysis process (Nowell et al., 2017). Therefore, the first two authors kept their own reflexive journal to document the logistics and methodological considerations as well as their own personal reflections. The precise analysis was then conducted in line of the six step technique by Braun and Clarke (2006).

Results

The results have been analyzed according to the six-phase method by Braun and Clarke (2006) i.e., familiarization with the data, generation of initial codes, searching for themes, reviewing themes, defining and naming themes, and producing the report. Data are organized and summarized in the results section and the interpretation regarding significance and implications follows in the discussion.

Quantitative Results

Changes in Patients Seeking Help

Based on the question that asked if participants were seeing more or less patients, the number of patient contacts (i.e., number of patients seen by a clinician) seemed to remain relatively stable during the first wave of the COVID-19 pandemic, as reported by 42% of the participants who indicated no change in the number of patient contacts. Nonetheless, almost 40% reported to see less patients, while 17% reported to see more patients.

Further, 78.8% reported that patients displayed similar psychological problems as they did prior to the COVID-19 pandemic. However, 48.3% also reported that their patients seemed to be more distressed compared to one year before, whereas 28.7% reported no change in their patients' distress. The most frequently reported patients' clinical issues

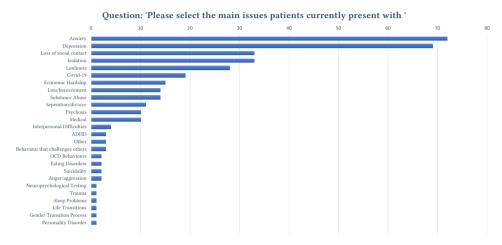


encompassed anxiety (86%), depression (82%), loss of social contacts and isolation (each 39%) (see Figure 2).

In terms of clinicians' working practices, most responders reported that COVID-19 had changed their work routines (73.6%), mostly in ways that they perceived to be undesirable.

Figure 2

Main Mental Health Issues Reported by Patients



Note. Please note that not all patients were seen in standard psychotherapeutic environments which is why alternative topics are listed as the presenting problem. This refers to other medical conditions, neuropsychological testing and other non-identified topics.

Qualitative Results

The overarching themes that were identified in the data were: changes to clinical practice; changes to other work activities and contexts; the challenges and opportunities; the effect of COVID-19 measures on clinical practice and further reflections. Within these themes, the following categories were found including: Changes to working practices such as online working; psychotherapists reflections on the changes and an exploration of what could be improved and, implications for clinical practice and organizations.

Perceived Changes in Clinical Practice

Perceived changes in clinical practice were mostly in regards to working online, e.g., conducting video therapy, and working from home. Further, several participants reported changes in treatment frequencies (more/less patients, more sessions per patient), hygiene measures (such as wearing face masks, social distancing in assessments), challenges in



providing treatment while wearing personal protective equipment (such as face masks), redeployment and logistical difficulties if patients were not able to use online platforms.

Citation regarding hygiene measures: "every patient has to wash first his hands, more disinfection, mouth-nose-protection, plexiglass for breath protection, safety distance, and more time and space are needed between the appointments for disinfection"

Citation regarding personal protective equipment: "Wearing masks, me and patient, which is very disturbing while there is no emotional expression."

Some participants also reflected on patients' concerns regarding treatment, such as more anxiety and individual differences in motivation to access online treatment. Further, therapists' concerns were also mentioned (e.g., if their hygiene procedure is correct).

Citation regarding therapists' concerns: "the first thought in every step is 'how correct is my procedure?'"

Overall, changes in patient contact (i.e., less appointments, fewer face-to-face contact, more support for patients) were named.

Perceived Changes to Other Work Activities and Contexts

Not all participants were necessarily working in clinical practice, and changes in research and teaching were also reported. Participants noted that procedures in the work environment were modified according to COVID-19 safety measures, often leading to a lack of contact between colleagues.

In an additional question, general aspects of the working environment were covered. Here, once again digitalization was mentioned as a central change, as not only video therapy but also remote meetings with colleagues that had been introduced.

Citation regarding digitalization: "No face to face clinics, therefore replying on phone and video contact. Working in isolation more and away from my team to do working from home."

Some participants reported that there was an increasing lack of contact between colleagues due to the increasing division in teams as a result of remote working.

In answers to this question, participants also highlight the adherence to hygiene measures in the work environment such as social distancing, wearing masks and more cleaning. Several participants mentioned that they were mainly working from home and some were holding therapy sessions outside. Additionally, two participants were engaged in extra activities regarding COVID-19 (i.e., at a phone support line). Three participants said that there were significant changes to their research, such as delays in recruitment, or needing to stop research entirely.



Challenges and Opportunities in the COVID-19 Pandemic Era

The challenges and opportunities that participants reported were wide ranging, and there was not always a clear distinction between what constituted a challenge or an opportunity (e.g., only replying "tele-therapy"), and sometimes different participants reported the same issue as a challenge, whereas others saw it as an opportunity. There were several participants who listed an opportunity that arose as a result of the COV-ID-19 pandemic and also at the same time reported it as a challenge of working during the COVID-19 pandemic (e.g., no commute vs. constant working from home).

Change in Work Logistics — The change to a predominantly technology-based work practice appeared to be either a challenge or an opportunity for participants. While several participants reported problems regarding technical knowledge and support or internet connection issues, as well as lack of equipment (such as laptops), remote working was also perceived by some as an opportunity to improve their own technology skills.

Citation regarding technical factors: "videocalls are more tiring, but effective"

A similar pattern of both challenges and opportunities emerged for working logistics: additional childcare, the need to develop new work-related rituals and a higher strain of videocalls were mentioned, as well as no time between meetings. However, several positive aspects were also mentioned, such as less need to travel, more flexibility at work, and the opportunity to access patients who may not have had the possibility to receive treatment otherwise. Some also felt that video therapy works very well, and some had been able to further develop their self-care strategies.

Citation regarding working logistics: "The main challenge was managing childcare alongside working while the nurseries were closed."

Citation regarding working logistics: "working from home so less MDT [multidisciplinary teams] working, not being able to provide a service for those with sensory impairments primarily hearing loss, increased competing demands on my time, my own response to COVID and lockdown and depleted resources over time. New ways of working do include being able to offer video or remote access to appointments not requiring people to travel and being able to support people who are shielding"

Citation regarding working logistics: "I had to develop new rituals at the end of the work day, digital work exhausts me more than working from the office"



Clinical Issues — Interestingly, the therapy-related factors also included both challenges and opportunities. Interventions for some mental health problems appeared to be more challenging to deliver online (e.g., depression, trauma) compared to the pre-pandemic face-to-face settings (e.g., difficulty in finding new options to increase activity, more insecurity in trauma treatment due to a lack of stabilizing measures). Furthermore, participants reported that they were at a greater physical distance to patients during face-to-face interactions, whereas online sessions provided less opportunity for non-verbal feedback and therapeutic engagement. Participants reported having to spend more time preparing for sessions, and there were concerns about a lack of consent and choice of therapy modality for patients. Conversely, less cancellations were noticed. Additionally, participants were able to receive contextual information about their patients by seeing their environment.

Citation regarding therapy-related factors: "Less cancellations and non-attendance at sessions. Harder developing rapport and doing therapy without the same transference or cues."

Citation regarding therapy-related factors: "video sessions allow for less non-verbal feedback/assessment (negative for diagnosis and treatment recommendation); video sessions allow impression of the patient's home environment (important context information and opportunity for the patient to illustrate problems that occur at home = positive)"

Citation regarding therapy-related factors: "Reachability was better for some, but worse for others, especially mothers (closed schools) and women in abusive relationships (often had to talk in their car)"

Team and Organizational Factors — Several factors were portrayed as rather challenging, as participants reported difficulties regarding social factors at work, such as a worsening of team cohesion, staff absence, staff conflicts and social isolation. Although some found this to be a positive as they could decide who they spent time with.

Citation regarding social factors: "All disciplines of staff not offered same opportunities to work from home causing conflict/envy; opportunity to avoid toxic colleagues at work."

Citation regarding social factors: "prevent social isolation also in my staff without forcing collaborators back to work"

Furthermore, a number of organizational factors were mentioned. For example, the rapid change of regulations (e.g., weekly changes) and the lack of guidelines and unified standards were reported to make working processes even more difficult.



Citation regarding organizational factors: "Trying to keep up with the constant information changes, rapid decision making and trying to look after myself too"

Alongside organizational factors, data security issues were also mentioned. This often highlighted the problem of patient confidentiality and keeping data safely stored while working from home.

Finally, participants reported difficulty adapting to new ways of working initially, however this appeared to develop into a new and practiced working routine over time.

Effects of the COVID-19 Pandemic Measures on Clinical Practice

Participants mainly focused on the effects of COVID-19 emergency measures on their clinical practice. The general restrictions of contact, i.e., lockdown, social distancing, restricted entrance to buildings and building closures were mentioned by half of all participants. These were often brought into close relation to other themes such as increased psychopathology in patients.

Citation regarding general restrictions: "Restrictions concerning certain hours for meeting the patients."

Citation regarding general restrictions: "Lockdown and the unlocking of lock down introducing new anxieties and worry"

Citation regarding general restrictions: "Full Lock-down, both in effect on my work directly and how it seeps into clients' existing struggles"

Another topic mentioned was the effect of wearing masks.

Citation on wearing masks: "Mask....very hard to work without seeing emotional expression. Especially hard with kids"

Citation on wearing masks: "Mask wearing - conceals the faces of both client and counselor, lack of nonverbal cues"

Additionally, effects of hygiene measures on treatment were often mentioned, such as wearing protective clothing, opening windows during sessions, no face-to face contacts and short-notice cancellations due to patient concerns about showing COVID-19 symptoms.

Citation regarding hygiene measures: "Wearing protection clothing - it is necessary and important but it makes work harder"

Both general restrictions issued by the state and individual restrictions in the workplace had a significant effect on participants and their work, such as closure of nurseries,



quarantine, shielding, and loss of freedom as well as concerns around travelling on public transport.

Citation regarding individual restrictions: "Closure of nurseries - having to provide childcare between a working couple means a massive reduction in available working time."

Participants also reported on the effects on patients, such as changes in psychological symptoms and less motivation to seek help or engage in online sessions.

Citation regarding effects on patients: "People with mental health conditions hold their breath: that is do not seek help because they are afraid to get COVID-19 and because there is a pause in social life"

Citation regarding effects on patients: "COVID-19 measures, at least in Italy, did not help in containing the viruses, as people were terrified by the official information, so did not ask for help or did not dare to go to hospitals, and were hampered from going to parks."

Further Reflections

Participants shared a variety of interesting insights into prospective changes concerning both mental health professionals and government policies.

One major theme was the wish to collect, share and discuss their experiences of using video therapy. This included both concerns (e.g., regarding effectiveness and data security), and desire for specific training in psychotherapy delivered online. Participants also shared that they had a new understanding of the importance of being connected to their colleagues.

Citation regarding sharing with colleagues: "More practice in online therapy, share data concern effectiveness of online therapy versus on said therapy"

Citation regarding sharing with colleagues: "I think it would be a good idea to set up a section in Clinical Psychology in Europe [journal of the EACLIPT] and invite practicing clinical psychologists to describe their experience with new forms of work. I would be motivated by such an opportunity to contribute, and I would also learn from the experience of colleagues."

In terms of policy implications, it was argued that the importance of mental health should be further promoted at national levels, particularly given the collective impact on mental health. A greater flexibility (e.g., introducing video therapy into health insurance plans) and the possibility of choosing the most suitable treatment modality (e.g., face to face or video) were two major points raised by respondents. Furthermore, the effects



of the current pandemic on both research (e.g., regarding long-term effects on mental health), and the healthcare system was mentioned, including implications for research funding. Finally, government policy implications regarding the implementation of policy guidelines, such as closure of nurseries and schools, resulted in a dilemma for many parents who are having to work from home while caring for children.

Citation regarding political implications: "Research funding being so FAST and associated huge number of reviews; being on funding panels. Existing research in NHS stopped due to redeployment."

Citation regarding political implications: "Acknowledgement of the impact of COVID-19 on the mental health of the population should be acknowledged at a national/international level. Awareness needs to be raised in governments and there needs to be a way to address the increased level of distress that the population will undoubtedly experience."

Citation regarding political implications: "Productivity whilst working from home is understood although not overtly acknowledged to be more limited when children to be cared for at home too which creates unnecessary guilt when torn between roles. Some managers (not mine) did not stand up to look after staff by leading and issuing clear guidance."

Several participants pointed out that there will be long-term consequences of the COV-ID-19 pandemic on mental health.

Citation regarding long-term consequences: "To be prepared that COVID-19 has a long-lasting effect on young people especially adolescents and students"

Discussion

The current study demonstrates that the COVID-19 pandemic brought about unprecedented changes in clinical practice for clinical psychologists and psychotherapists across EACLIPT members in Europe. Changes to the clinical practice of psychologists and psychotherapists were sudden, for example the digitalization of therapy, which was at odds with previous attempts to implement digital mental health approaches in healthcare (Mohr, Riper, & Schueller, 2018). Some opinions and evidence have suggested that this has been a 'black swan' moment, where the COVID-19 pandemic has led to a rapid change in how mental health care is provided, including more opportunities for online working (Wind, Rijkeboer, Andersson, & Riper, 2020), also in low- and middle-income countries (Fu et al., 2020). Additionally, the current study showed that clinical psychol-



ogists and psychotherapists managed to provide treatment throughout the COVID-19 pandemic, despite the additional challenges of working in this context, to patients who were perceived to be experiencing a greater level of distress. Although challenges were clearly identified in the current study, participants also identified opportunities from working through the pandemic, such as reduction in commuting time, increased work flexibility and accessibility for patients.

Despite such a significant change in working context for clinical psychologists and psychotherapists, only one previous study looked at the impact of the pandemic on clinical psychologists and psychotherapists and also used a mixed-methods analysis of qualitative and quantitative data (McBeath et al., 2020). In that study, clinical psychologists and psychotherapists who were mostly based in the UK, were recruited via social media and, similar to our results, found that clinical psychologists and psychotherapists were able to cope with the rapidly changing work, and managed immediate problems with imagination and engagement. They also described a significant change of psychotherapeutic treatment, especially in relation to video therapy.

Digitalization

Even though clinical psychologists and psychotherapists have not yet reached a consensus regarding whether they plan to continue using video therapy in the long run (Aafjes-van Doorn et al., 2020), the opportunities conferred via video therapy are clearly shown, both in the current study and other research (e.g., Humer, Stippl, et al., 2020). More than ten years ago, Simpson (2009) pointed out the opportunities and challenges of video therapy, naming the lack of research regarding efficacy as one major research goal. Simpson (2009) also pointed out that efficacy might be strongly related to patient and therapist's personality and interpersonal style, as well as therapist skills and experience in the use of technology. A pilot project with university students (Simpson, Guerrini, & Rochford, 2015) and the analysis of the recent, COVID-19 induced changes (Simpson et al., 2021) clearly points out the potential of video therapy if used correctly. It seems likely that the pandemic has shaped therapists' attitudes towards technology and led to a more positive view of it now they are more experienced in conducting treatment remotely, even if prior to COVID-19 they would not have elected to do so (Aafjes-van Doorn et al., 2020). As the success is highly dependent on therapists' overall attitudes and self-confidence regarding technology and remote therapy (e.g., Aafjes-van Doorn et al., 2020), training courses and supervision in this regard is essential. As Simpson (2009) already pointed out, some barriers that prevent access to psychotherapy and counselling might be tackled with video therapy such as geographical distance between major cities and remote and rural communities, and a lack of adequate or affordable transport between them. Furthermore, video therapy can be used by patients who are immobile (Connolly, Miller, Lindsay, & Bauer, 2020). It might also encourage patients to engage who are indecisive about treatment and worry about stigma. Finally, most studies overall tend



to conclude that video therapy will not be the new standard medium for psychotherapy (e.g., Aafjes-van Doorn et al., 2020; Connolly et al., 2020), but a useful addition under certain circumstances and considering specific adaptations such as providing a rationale for video therapy, maintaining therapeutic boundaries and finding a new way of risk management (for an overview see Simpson et al., 2021; for an exemplary analysis of patients with Borderline Personality Disorder see Ventura Wurman, Lee, Bateman, Fonagy, & Nolte, 2021).

Conducting Therapy With Personal Protective Equipment (PPE)

While the changes to working as a result of video therapy clearly brought opportunities, conducting therapy in person while using protective equipment such as face masks presented significant challenges. Clinical psychologists and psychotherapists who conducted face-to-face treatment during the pandemic mostly wore face masks and thus covered more than half of their face. Thus, while the disadvantage of video therapy is erased (e.g., no technological difficulties), others might appear: it has been argued both in our study and previous opinion pieces (e.g., Hüfner, Hofer, & Sperner-Unterweger, 2020) that emotions are harder to read if someone is wearing a face mask, which can then cause difficulties in the patient-therapist relationship. Interestingly, initial evidence from basic research has shown mixed findings. Some found that emotions are harder to read when the conversational partner wears a face mask (Grundmann, Epstude, & Scheibe, 2021), while others found in a longitudinal design that participants change which cues they use to detect an emotion, suggesting they adjust to the presence of masks (Barrick, Thornton, & Tamir, 2021). One study of school-aged children who are currently constantly interacting while wearing masks concludes that masks pose a challenge but, in combination with other contextual cues, are unlikely to dramatically impair social interactions (Ruba & Pollak, 2020). Translating these findings to the clinical context, one can assume that psychotherapy with masks is somewhat more challenging than without masks, but could still lead to a good patient-therapist relationship and successful treatment outcomes. However, more research on the effects of masks on psychotherapy is necessary.

Restriction of Contact

It is important to acknowledge that clinical psychologists and psychotherapists have not only experienced a significant change in their working logistics, but also in their everyday life outside of work – similar to their patients and all other citizens. As some participants have highlighted, social support at work was less available and the team cohesion diminished, thus personal and work resources were limited. Furthermore, additional tasks added to stress (e.g., working from home without a proper place to work; child and other family care etc.). These factors indicate the importance of social support



among the clinical psychologists' and psychotherapists' community in difficult times, and the importance of leadership from professional and governmental organizations.

Reflections for EACLIPT

It has been an unprecedented working environment for clinical psychologists and psychotherapists during the COVID-19 pandemic. Clinical psychologists and psychotherapists were required to adapt their approach to work at very short notice during the first wave of the COVID-19 pandemic. However, survey respondents reported that they managed to convert working logistics efficiently and have been providing much needed care for patients ever since, even though the examination of the efficacy of treatment still needs more research. This was often done on an individual basis or by smaller groups of colleagues. An important next step is to collect, share and discuss experiences of, and develop guidelines for, video or phone therapy or intervention. This has been done locally (e.g., UK, Simpson, Richardson, Pietrabissa, Castelnuovo, & Reid, 2021. However, the EACLIPT as an organization has provided a position statement and a summary of national statements on what is needed for both patients and clinical psychologists and psychotherapists as well as future research endeavors regarding mental health⁶ by integrating perspectives from a wide range of clinical psychologists and psychotherapists across multiple countries.

Reflections for Government Policy and Other Institutions

Although the sample of the current survey was limited to EACLIPT members, arguably, this data could be useful to inform the policies of government and other institutions, as the views of clinical psychologists and psychological therapists are represented from across a broad range of occupationals settings, across multiple European countries. The current findings emphasize the importance of including mental health issues in current policy considerations on how to manage the pandemic in the longer term. The long-term effects on mental health as a result of the COVID-19 pandemic are still not clear (e.g., de Figueiredo et al., 2021). Based on previous research on epidemics, further symptom increases in the upcoming one to three years are expected in anxiety, anger, depression, post-traumatic stress symptoms, alcohol abuse, and behavioural changes such as avoiding crowded places and cautious hand washing (e.g., Kathirvel, 2020). This needs to be considered both in research (e.g., which factors could lead to mental health problems in the long run, de Figueiredo et al., 2021) and in health care (e.g., further flexible inclusion of video therapy into health insurance plans; enlarging mental health treatment provision; Kathirvel, 2020). In addition, the uptake of video therapy by clinical psychologists and psychotherapists during the COVID-19 pandemic offers the opportunity to take part



in treatment long distance (the therapist in one country, the patient in another) which calls for cross-border guidelines.

Limitations and Implications

The current study was implemented during the first wave of the COVID-19 pandemic, from May to September 2020. The pandemic is still ongoing and, thus, the situation is continually changing. To keep the questionnaire as short as possible to encourage participants to complete the survey, we did not include detailed information on the sociodemographic background and we did not ask for detailed numbers and facts, e.g. regarding the number of patient contacts before and during the pandemic. We rather opted to assess the personal estimation of change, which relies on the therapist's perception of the number of patient contacts and could include inaccuracies. Additionally, we are aware that only a small number of members completed the survey (i.e., 13% of EACLIPT members) and, thus, the results have to be considered in light of a rather limited and selective sample. However, our results provide a qualitative and quantitative picture of the first abrupt changes to the work of clinical psychologists and psychotherapists as a result of the COVID-19 pandemic.

Furthermore, in the current study, responses to open questions were often quite short, which at times limited the scope of interpretation. However, many answers pointed to similar conclusions as shown above.

The current study highlights the tremendous challenges that both patients and clinical psychologists and psychotherapists have experienced during the pandemic. Consequently, there are calls for specific training for therapists and clear guidelines regarding the use of technology, data security and solutions to the psychotherapeutic challenges of delivering therapy remotely. However, more research (such as the follow-up to this survey that is underway) is necessary to identify the long-term effects of the COVID-19 pandemic on both patients and clinical psychologists and psychotherapists, and to comprehensively influence policy and future healthcare considerations.



Funding: The authors have no funding to report.

Acknowledgments: The authors have no additional (i.e., non-financial) support to report.

Competing Interests: Winfried Rief is one of the Editors-in-Chief and Colette Hirsch is a Subject Editor of *Clinical Psychology in Europe*. Both authors played no editorial role in this particular article or intervened in any form in the peer review process. Andreas Maercker, Céline Douilliez, Gerhard Andersson, Martin Debbané, Roman Cieslak, and Claudi Bockting are Editorial Board Members of *Clinical Psychology in Europe* but did not intervene in any form in the peer review process.

Twitter Accounts: @julia_asbrand, @SamGerdes1, @JosefienUMH, @JennyGuidi, @DrColetteHirsch, @cdouilliez, @profGerhardA, @MartinDebb, @rocie, @WRief1, @clbockting

Supplementary Materials

The Supplementary Materials contain the questionnaire which was used in the study (for access see Index of Supplementary Materials below).

Index of Supplementary Materials

Asbrand, J., Gerdes, S., Breedvelt, J., Guidi, J., Hirsch, C., Maercker, A., Douilliez, C., Andersson, G., Debbané, M., Cieslak, R., Rief, W., & Bockting, C. (2023). Supplementary materials to "Clinical psychology and the COVID-19 pandemic: A mixed methods survey among members of the European Association of Clinical Psychology and Psychological Treatment (EACLIPT)"

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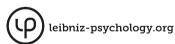
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EACLIPT

Clinical Psychology in Europe (CPE) is the official journal of the European Association of Clinical Psychology and Psychological Treatment (EACLIPT).



PsychOpen GOLD is a publishing service by Leibniz Institute for Psychology (ZPID), Germany.







Research Articles



Interoception and Premonitory Urges in Children and Adolescents With Tic Disorders

Christina Schütteler¹ , Katrin Woitecki², Manfred Döpfner^{2,3} ,

Alexander L. Gerlach 1 0

Department of Psychology, Clinical Psychology and Psychotherapy, University of Cologne, Cologne, Germany.
 School of Child and Adolescent Cognitive Behavior Therapy (AKiP), Faculty of Medicine and University Hospital Cologne, University of Cologne, Cologne, Germany.
 Department of Child and Adolescent Psychiatry, Psychosomatics and Psychotherapy, Faculty of Medicine and University Hospital Cologne, University of Cologne, Cologne, Germany.

Clinical Psychology in Europe, 2023, Vol. 5(1), Article e8185, https://doi.org/10.32872/cpe.8185

Received: 2022-01-20 • Accepted: 2022-10-25 • Published (VoR): 2023-03-31

Handling Editor: Winfried Rief, Philipps-University of Marburg, Marburg, Germany

Corresponding Author: Christina Schütteler, Department of Psychology, Clinical Psychology and Psychotherapy, University of Cologne, Pohligstr. 1, 50969 Cologne, Germany. Tel.: 0221 470 5381. E-mail: c.schuetteler@uni-koeln.de

Supplementary Materials: Data, Preregistration [see Index of Supplementary Materials]





Abstract

Background: Compared to healthy controls (HCs), adult Tic Disorder (TD) patients exhibit a lower interoceptive accuracy (IAcc) in heartbeat perception. Since the lower IAcc is not evident in children, the age at which tics develop, but in adults only (Pile et al., 2018, https://doi.org/10.1007/s10803-018-3608-8), lower IAcc may reflect a pathological mechanism relevant with regard to tics, premonitory urges (PUs) or the resulting impairment. Although tics are a motor phenomenon, up to date, IAcc has been assessed only with a heartbeat-counting task. This study aims at comparing cardiac and muscular IAcc using two different paradigms and investigates how IAcc is related to premonitory urges in youth.

Method: Interoceptive measures (heartbeat-counting task, muscle tension paradigm) of 28 youth with TD were compared to 23 control participants and related to self-rated premonitory urges and tic symptoms.

Results: TD patients did not differ from HCs in any IAcc measures. However, within TD patients, IAcc explained additional variance in PUs when controlling for tic severity. Muscular IAcc in TD patients is related to urges and tics, but the direction of this association is unclear. IAcc is lower in TD patients than in HCs, indicating imprecise sensory input which is more easily overcome by priors within the predictive coding framework.



Conclusions: Muscle tension feedback tasks could extend interoceptive trainings aimed at improving IAcc to improve accuracy of urge perception (more precise sensory input) to foster the ability to control tics via HRT. Longitudinal studies could provide further insights in causal relationships between IAcc, premonitory urges and tics.

Keywords

heartbeat, muscle, EMG, interoceptive accuracy, predictive coding

Highlights

- A muscle tension paradigm assessed interoceptive accuracy.
- Patients with tics did not differ from healthy controls in interoceptive accuracy.
- Muscular interoceptive accuracy in patients relates to premonitory urges and tics.
- · Muscular tension feedback tasks could improve treatment via habit reversal training.

Tics are sudden repetitive movements or vocalizations that occur in up to 21% of children (Cubo et al., 2011; Kurlan et al., 2001). In most cases tics disappear with increasing age and remain stable in only about 1% of people worldwide (Robertson & Cavanna, 2008). Tics regularly are preceded by an unpleasant premonitory urge or sensation (PU). PUs are often perceived as an urge to move, an impulse to move, inner tension or restlessness and mostly occur in the face, neck, shoulders, arms or hands (Kwak et al., 2003).

In psychotherapy, the perception of PUs is both necessary and problematic. On the one hand, in line with habit reversal training, a precise perception of PUs improves the ability to successfully suppress tics (McGuire et al., 2015). On the other hand, PUs illicit tics, negatively reinforce tics and correlate with tic severity (Li et al., 2019).

The capability to perceive bodily signals ('interoception') entails several different facets: IAcc is defined as the process of accurately detecting and tracking internal bodily sensations (Garfinkel et al., 2015). Interoceptive sensibility refers to the self-reported attention given to and detection of interoceptive information. Finally, interoceptive awareness refer to the metacognitive correspondence between objective IAcc and self-report of interoceptive information (Garfinkel et al., 2015). In the following, we will only focus on IAcc based on the notion that IAcc may be an underlying dimension in PUs, necessary to perceive interoceptive sensations.

Adult individuals suffering from a TD exhibit a lower IAcc in a heartbeat perception tasks whilst reporting a heightened perception of sensory stimuli (interoceptive sensibility) as compared to individuals without tics. However, this lower IAcc is not evident in children but in adults only (Pile et al., 2018). So far, IAcc in individuals with TDs has exclusively been assessed with a heartbeat-counting task (Schandry, 1981). Arguably, tic symptoms are muscle movements. Thus, assessing IAcc by looking at the ability of an individual to perceive heart activity may not be the best test of the possible involvement of IAcc in TD. Consequently, this study plans to investigate whether IAcc in a muscle



tension perception paradigm is associated with PUs in addition to or over the ability to perceive heart activity.

According to a predictive coding account of bodily symptom perception, bodily changes such as heart activity or muscle tension often are weak and imprecise signals. Against this background, a heightened interoceptive sensibility for tic related symptoms may be the result of overly precise interoceptive priors. Specifically, in TDs, an overactive putamen and insula may lead to overly precise predictions at hierarchically higher levels, overriding the actual weak and imprecise sensory inputs. The resulting prediction error may be reduced by performing an ,involuntary' tic and/or be the basis for the perception of an unpleasant PU (Rae et al., 2019).

On the behavioral level, PUs may represent a conditioned response to aversive external stimuli such as criticism, offenses or social marginalization as result of tic execution. In the course of a TD, tics may become associated with those negative emotional valences that subsequently constitute the PU. After a tic is executed, the unpleasant PU dissolves and ticking is negatively reinforced, promoting maintenance of tics (O'Connor, 2002). Following the O'Connor model, an attentional focus on PUs and tics may, over time, enhance the overly precise prior even further. As a result, unpleasant PUs are perceived even more, impairing patients' quality of life. Indeed, adult TD patients with a long history of the experience of tics exhibit lower IAcc (Ganos et al., 2015; Rae et al., 2019). Simultaneously, in adults, physical sensations are self-reported more often (interoceptive sensibility) compared to individuals without tics (Rae et al., 2019). Since a lower IAcc is not evident in children but in adults only (Pile et al., 2018), lower IAcc in adults may reflect a failure to develop, over time, a better IAcc if individuals suffering from TD, which is commonly found in healthy individuals (Murphy et al., 2019). In consequence, given this overly precise prior, the experience of PUs continues and may even be strengthened into adulthood. However, given that IAcc, so far, has been assessed only with a heartbeat-counting task, it is important to additionally assess whether IAcc in children with TDs may be increased with regard to the perception of muscular activity, since muscles are involved in the execution of tics. We opted to assess facial muscle tension given that most TD patients experience at least one tic in the face (McGuire et al., 2016).

Thus, in this study we wanted to test the hypotheses, that children and adolescents with pathological tics exhibit lower IAcc with regard to both heart activity as well as facial muscle tone as compared to children and adolescents without tics. We also test the hypotheses, that variance in PUs is explained by IAcc scores. Furthermore, we compare muscular to cardiac IAcc, using two different paradigms.



Method

Participants

A total of 51 children and youth between 10 and 19 years old were recruited at the University Hospital Cologne (28 patients and 23 control participants) and surrounding schools. One patient fulfilled the criteria of a chronic TD, 27 patients fulfilled the criteria of Tourette's Syndrome according to ICD-10. Inclusion criteria were a previously diagnosed tic disorder, age 10-21 years and fluency in German. Exclusion criteria were insufficient German language skills and the absence of any tic during the last week. Twenty of these 28 TD patients (71%) were male (13 of 23 HCs, 57%). 7 TD patients were diagnosed with a comorbid disorder via diagnostic checklists (5 ADHD, 1 OCD, 1 conduct disorder, 1 trichotillomania). Two TD patients received anti-tic medication (Aripiprazole, Tiapride), three received medication targeting ADHD (Methylphenidate). TD patients and HCs did not differ with respect to gender ($\chi^2 = 1.229$, p = .268) or age. CBCL total scores differed significantly between groups, but not YSR scores (Table 1).

Procedure

A two-group design compared TD patients with control participants not suffering from a TD (HC). TD patients and their parents additionally completed questionnaires regarding tic symptomatology and other psychopathology measures. Participation took between 70 to 90 minutes. The experimental paradigms measuring IAcc were presented via computer screen. Participants received an allowance of 8€ per hour. The current study was carried out according to the Declaration of Helsinki. The Ethics Commission of the University of Cologne's Faculty of Medicine approved the study (CSHF0044) and the study was pre-registered (see Supplementary Materials). All participants and their legal guardians gave informed consent. The data that support the findings of this study are openly available in figshare (see Supplementary Materials).

Questionnaires

The German version of the Child Behavior Checklist (CBCL; Döpfner et al., 2014) is a caregiver report and assesses a variety of psychopathological symptoms. In the current study, the internal consistency of the total score was excellent (α = 0.93). The German version of the Youth Self Report (YSR; Döpfner et al., 2014) aims at children and youth, is constructed equivalently to the CBCL, and assesses self-reports of a variety of psychopathological symptoms. The total score exhibited good internal consistency in the current study (α = 0.84). Both CBCL and YSR consists of the subscales Aggressive Behavior, Anxious/Depressive Symptoms, Attention Problems, Rule-Breaking Behavior, Somatic Complaints, Social Problems, Thought Problems, and Withdrawal/Depression.



The self-rated Symptom Checklist for Tic Disorders SCL-TIC-S (SCL-TIC-S) and SCL-TIC-P (parent-rated) are part of the DISYPS-III diagnostic system (Döpfner & Görtz-Dorten, 2017). They each assess the number of tics. On a 5-point Likert-type scale for each tic the respective intensity (very mild to severe, irritates others), frequency (a few times a week to constantly, every few minutes), and overall impairment (very low, hardly disturbs to extreme) is assessed. Additionally, the SCL-TIC-S assesses overall controllability (very low to very high). A tic symptom score (range: 0 – 16) is calculated by multiplying intensity with frequency for each tic, summing up the results and dividing the sum by the number of tics (Döpfner & Görtz-Dorten, 2017). Internal consistency of the SCL-TIC total score in the current study was excellent (α = 0.91 for SCL-TIC-S, α = 0.92 for SCL-TIC-P).

The Premonitory Urge To Tic Scale (PUTS) consists of 10 items and assesses PUs (Woods et al., 2005). We used the German translation (Rössner et al., 2010). The 10th item asks about tic controllability and is usually excluded or interpreted separately to sustain internal consistency (Woods et al., 2005). PUTS' internal consistency in the current study was acceptable (α = .75 for PUTS-9, α = .71 for PUTS-10).

Experimental Measures

The Mental Tracking paradigm (Schandry, 1981) was employed to assess IAcc based on cardiac sensibility. Participants were instructed to concentrate on their heartbeats for three randomly presented time intervals (à 25s, 35s and 45s) and silently count the perceived heartbeats. They were instructed to only count heartbeats that they felt (Koch & Pollatos, 2014). After a trial run, ECG-electrodes assessed the participant's ECG (sample rate: 512 Hz) and heart beats were assessed online using the software Uvariotest (Gerhard Mutz, Cologne, compare Meyerholz et al., 2019). A sound signal indicated begin and end of each time interval. Participants were not allowed to measure their pulse or time and were not informed about their average heart rate nor the length of each time interval. The Mental tracking paradigm is applicable for children and youth at least 10 years old (Koch & Pollatos, 2014). In the current study, internal consistency of the IAcc score for heart activity (HIAcc) based on the scores of the three time intervals was good (α = 0.94).

Facial EMG was assessed by skin electrodes placed on the masseter and corrugator supercilii on the left side of each participant's face. EMG placement followed the EMG guidelines by (Fridlund & Cacioppo, 1986). Muscle tension IAcc was assessed with a paradigm originally developed by Flor et al. (1992). Reported muscle tension and EMG measures were correlated to form an IAcc score for the masseter (MIAcc) and corrugator supercilii (CIAcc), respectively. We opted for two facial muscles that have previously been used in muscle discrimination tasks, because most TD patients display facial tics (Flor et al., 1992). During the task, participants looked at a screen that represented their muscle tension as measured by EMG. Muscle tension was visualized by a soccer ball



that moves along a line colored in red, green and yellow. Participants were instructed to keep the soccer ball in the green target zone for 2.5 seconds by tensing their facial muscles accordingly. A sound signal indicated the successful completion of the task. After regulating the respective muscle, participants reported the degree of muscle tension they believed the task required on a Likert-type scale ranging from 1 to 5. Since the axis of the colored line changed with the task (i.e., the green zone represented different intensities of muscle tension), participants needed to rely on interoceptive information only. Overall, participants went through 16 tasks as described above for each muscle. The required muscle tension levels varied in equal parts between 15%, 30%, 45% and 60% of the maximal achievable tension level for each participant. This maximal achievable tension level for each participant had been measured directly before the perception task by asking participants to tense the respective muscle as much as possible.

Data Analysis

The correlation between perceived and via EMG measured muscle tension represent masseter and corrugator IAcc scores, respectively. The heartbeat perception accuracy score (HIAcc) indicated the ability to perceive one's own heartbeat accurately and was calculated by employing the following formula with i = time intervals (25s, 35s, 45s) and no = measured heartbeats ns = counted heartbeats:

$$HIAcc = \frac{1}{3} \sum_{i=1}^{3} \left(1 - \frac{|no_i - ns_i|}{no_i} \right)$$

The resulting scores ranged between 0 to 1 with higher scores indicating higher cardiac accuracy. Independent samples two-tailed t-tests or chi²-tests were used to compare clinical between group measures and group differences in IAcc scores. Effect sizes are indicated by Cohen's d. All statistical tests are two-sided with p < .05. Pearson-product-moment-correlations and multiple regressions determined the relation between IAcc scores and tics or PUs, respectively. We used SPSS for these calculations.

Results

Interoception in Youth With Tic Disorders Does not Differ From Healthy Controls

When investigating IAcc scores, N=2 masseter datasets, N=5 corrugator datasets and N=1 HIAcc dataset needed to be excluded due to technical failure. On average M=13.18 corrugator trials (range from 2 to 16 trials) and M=14.92 masseter trials (range from 10 to 16 trials) were valid and could be included.



Table 1 displays descriptive statistics for each group and group comparisons between TD patients and HCs. TD patients' and HC's masseter and corrugator IAcc scores are shown in Table 1. Masseter and corrugator IAcc scores did not significantly differ between TD patients and HCs. TD patients and HCs did not significantly differ in any of the three IAcc measures, even when TD patients with comorbidities were excluded and the analysis was repeated (HIAcc: t = -0.414, df = 49, p = .68; MIAcc: t = -1.795, df = 38, p = .08; CIAcc: t = -1.309, df = 40, p = .20).

Table 1Group Differences

	TD	TD patients		hy Controls				
Variable	n	M (SD)	n M (SD)		t(df)	p	Cohen's d	
Age	28	12.65 (2.21)	23	12.86 (2.47)	t = -0.324 (df = 49)	.75	-0.09	
CBCL Total Score	27	32.19 (17.13)	21	11.24 (7.84)	t = 5.188 (df = 46)	< .001***	1.51	
YSR Total Score	26	49.96 (18.50)	23	45.26 (10.62)	t = 1.072 (df = 47)	.29	0.31	
Interoceptive Accura	cy Scores							
HIAcc	27	0.59 (0.28)	23	0.61 (0.29)	t = -0.244 (df = 48)	.81	-0.07	
MIAcc	26	0.42 (0.32)	23	0.53 (0.33)	t = -1.115 (df = 47)	.27	-0.32	
CIAcc	26	0.26 (0.36)	20	0.43 (0.34)	t = -1.614 (df = 44)	.11	-0.48	

Note. CBCL = Child Behavior Checklist; YSR = Youth Self Report; HIAcc = heartbeat perception accuracy score; MIAcc = masseter interoceptive accuracy score; CIAcc = corrugator interoceptive accuracy score.

***p < .001.

IAcc in a Proprioceptive Perception Task Explains Variance in Premonitory Urges in Youth

SCL-TIC-S score was M = 5.54 (SD = 2.98, N = 26), mean SCL-TIC-P score was M = 6.22 (SD = 3.33, N = 27). The mean PUTS total score for 9 items was M = 18.14 (SD = 5.02, N = 28). CIAcc and MIAcc correlated substantially with each other indicating internal validity, while not correlating significantly with HIAcc. Table 2 gives an overview over correlations between interoception scores and tic symptoms including PUs.

Table 2Pearson Correlations

Variable	1	2	3	4	5	6
1. PUTS-9						
r	-	.14	.47	06	.39	.24
p		.50	.01	.77	.05	.22
n		(27)	(26)	(26)	(26)	(27)



Variable	1	2	3	4	5	6
2. SCL-TIC-P						
r		-	.34	.06	.11	.00
p			.08	.77	.59	.99
n			(27)	(26)	(25)	(27)
3. SCL-TIC-S						
r			-	.12	.36	.07
p				.57	.08	.73
n				(26)	(25)	(27)
4. CIAcc						
r				-	.58**	30
p					.00	.14
n					(25)	(26)
5. MIAcc						
r					-	23
p						.27
n						(25)
6. HIAcc						_

Note. PUTS-9 = Premonitory Urge to Tic Scale (9 items); SCL-TIC-S = self-rated Symptom-Checklist for Tic Disorders; SCL-TIC-P = parent-rated Symptom-Checklist for Tic Disorders; CIAcc = corrugator interoceptive accuracy score; MIAcc = masseter interoceptive accuracy score; HIAcc = heartbeat perception accuracy score. $^*p < .05. ^{**}p < .01.$

 Table 3

 Linear Model of Predictors of PUTS-9 Total Scores With Confidence Intervals Reported in Parentheses

		95% CI					Zero-order	Partial
Predictors	b	LL	UL	SE B	β	p	correlation	correlation
Step 1								
Constant	13.45	9.39	17.50	1.96		< .001**		
SCL-TIC-S	0.78	0.13	1.42	0.31	.46	.02*	.462	.462
Step 2								
Constant	9.70	4.00	15.40	2.73		.002*		
SCL-TIC-S	0.50	-0.15	1.14	0.31	.30	.12	.462	.340
HIAcc	5.14	-1.51	11.79	3.19	.29	.12	.279	.339
CIAcc	-4.40	-10.62	1.82	2.98	31	.16	059	313
MIAcc	8.09	0.88	15.31	3.46	.52	.03*	.378	.464

Note. R^2 = .21 (p = .020) for Step 1, ΔR^2 = .22 (p = .08) and R = .43 (p = .02) for Step 2. PUTS-9 = Premonitory Urge to Tic Scale (9 items); SCL-TIC-S = self-rated Symptom-Checklist for Tic Disorders; SCL-TIC-P = parent-rated Symptom-Checklist for Tic Disorders; CIAcc = corrugator interoceptive accuracy score; MIAcc = masseter interoceptive accuracy score; HIAcc = heartbeat perception accuracy score. *p < .05. **p < .01.



In youth, tic symptoms vary substantially with age parallel to PUs, so tic severity should be accounted for when investigating PUs. Multiple regression analysis was used to investigate how IAcc relates to PUs. With PUTS total score as the dependent variable, self-reported tic total score was entered in Step 1 to control for tic severity. HIAcc and CIAcc and MIAcc scores were entered in Step 2. Adding the IAcc scores in Step 2 led only to a marginally significant change in \mathbb{R}^2 . Note, however, that the MIAcc score explained significant variance in PUs in addition to tic severity in Model 2 (compare Table 3).

Discussion

In the present sample of children and adolescents with and without tics, we found that neither HIAcc nor proprioceptive IAcc scores differed between these groups. These results are in line with recent studies that also did not find any difference in interoceptive accuracy (IAcc) in children with TD (Pile et al., 2018). Our study extends those findings to the perception of muscle tension. Although IAcc scores were numerically lower in participants with TD, these differences did not reach significance (d = -.07 - .43). Whereas lower HIAcc compared to HCs has been established in adult TD patients, we were not able to demonstrate such differences in our sample of children and adolescents. However, such differences in IA may evolve with increasing age of the TD, equal to increasing duration of a childhood-onset TD and may be compared to a model of altered interoception in children with chronic pain: Top-down processes, such as expectations of uncontrollably ticcing, and bottom-up processes, such as a stressed bodily state, may lead to altered interoception over time (Hechler, 2021). It would consequently be highly interesting to assess a sample of adult individuals with TD using our muscle tension paradigm.

Within TD patients, recent studies found IAcc to be positively correlated with PUs in adults (Ganos et al., 2015; Rae et al., 2019). Our findings corroborate this assumption. Adding IAcc scores (MIAcc, CIAcc and HIAcc) to tic severity scores when predicting PUs, a substantial (ΔR^2 = .22) additional amount of variance in PUs was explained. Note, that neither masseter nor corrugator IA scores were significantly correlated with HIAcc. Consequently, these different measures of interoceptive ability may cover different facets indicating that different body domains may matter when assessing IA. When taking a closer look at the results, the MIAcc (r = .39) correlated more strongly with the PUTS than the CIAcc (r = -.06), which even correlated negatively with the PUTS. In contrast to the corrugator supercilii, the masseter is more frequently contracted deliberately and thus might it be easier to control and it might be easier to estimate its tension. Arguably, this may result in a more reliable measure. However, the corrugator supercilii is linked with emotional expression (Tan et al., 2012) and, arguably, the location of the corrugator supercilii overlaps with locations most frequently affected by tics (McGuire et al., 2016).



We therefore recommend that future studies nonetheless continue to assess both facial muscles when looking at IA with regard to muscular tension.

Herbert et al. (2012) compared interoceptive accuracy in eating disorders across two bodily domains – cardiac and gastric –and showed them to be inversely correlated. Similarly, we found cardiac and muscular IAcc to be inversely correlated in TD ($r_{\rm CIAcc}$ = -.30, $r_{\rm MIAcc}$ = -.23), although not reaching significance. Following the interpretation by Herbert et al., trying to control muscles that feel uncontrollably at times, might increase activation in the sympathetic nervous system which in turn might increase cardiac IAcc. In line with Flack et al. (2017), activation in the sympathetic nervous system might as well be the result of heightened levels of fear when focusing on muscles associated with unpleasant tic execution.

HIAcc score did marginally significantly correlate with the PUTS in our sample of children and adolescents. Children have smaller hearts and a lower stroke volume, associated with a higher heart rate which influences heartbeat detection positively (Knapp-Kline & Kline, 2005). In light of this it may be concluded that increased HIAcc promotes the perception of PUs in children.

In summary, these results on the association between IA and PU provide an ambiguous picture. On the one hand, IA clearly is associated with PU, but the direction of this association remains unclear. Following our hypotheses, we would have assumed that IA should be negatively associated with PU, as was the case for corrugator perception. In contrast, we find a clear positive association between the perception of cardiac activity as well as tension of the masseter and PU. Obviously, more research is needed here.

Note that the findings linking interoception and PUs rely on the PUTS to represent PUs. However, the PUTS is a self-report measure that more likely represents interoceptive sensibility. Interoceptive sensibility is known to be altered in children and adults with TD (Owens et al., 2011; Rae et al., 2019) and the measure is challenged by the usual problems associated with self-reports. The relative relationship of interoceptive accuracy, interoceptive sensibility und interoceptive awareness constitute PUs' presence and cognitive, emotional and clinical consequences (Garfinkel et al., 2015).

We find it especially intriguing to interpret our findings within the predictive coding framework (Ainley et al., 2016; Friston, 2010; Khalsa et al., 2018; Rae et al., 2019). In this framework, a difference between sensory input and prior expectation results in a prediction error. Following Bayesian Inference, the prediction error may be resolved by updating the prior expectation or executing a movement to change sensory input. In TD patients, in hierarchical higher brain structures, an over-precise interoceptive prior might predict movements. If the weak bottom-up sensory input does not correspond with this prediction, the insula has to resolve the resulting interoceptive prediction error. The anterior insula is hypothesised to update predictions to reduce prediction errors (Seth, 2013; Seth et al., 2012) and is known to show functional abnormalities in TD patients. Due to the prior's over-precision and weight, imprecise sensory input may be



overcome by the prediction and the prediction error is being 'explained away' by the anterior insula as a premonitory sensation (urge to move). In adults, lower IAcc is found which, arguably, indeed indicates weaker priors. However, in line with previous research in adults (Ganos et al., 2015), we found that lower IA (heart activity and tension of the masseter) is correlated with lower PUs. Assuming that good IA, over time, leads to weaker priors, a smaller prediction error may result in less PU. In contrast, IA for the tension of the corrugator, was negatively correlated with PU. Following this finding, one could argue that the worse the perception of actual physical sensations in areas in which tics occur the more top-down predictions will overshadow interoceptive sensations and result in the perception of PUs (perceptual inference).

There are a number of limitations to the current study that offer opportunities for future research. Studies investigating the perception of muscle tension in adult TD patients are yet to be conducted to gain further insights on the development of PUs over the life span. Since the current cross-sectional study allows correlative interpretations only, the longitudinal comparison of chronological changes in HIAcc, MIAcc and CIAcc scores, PUTS and urge thermometers over the lifespan would provide further information on their etiological meanings. In addition, the exploratory findings on IA in child and adolescent TD patients need to be replicated, preferably in larger samples. Our study exclusively focused on IAcc in youth with TD. Future studies may extend our findings to interoceptive sensibility to disentangle the influence of interoceptive sensibility and IAcc on the self-reported perception on premonitory urges. Altogether, our sample size was relatively small, compromising statistical power to some degree.

TD patients usually differ from HCs not only with regard to tics but also with regard to comorbidities such as ADHD or OCD, depression or anxiety. When comparing TD patients to typically developing children, our study cannot account for the impact of comorbidities such as ADHD or OCD due to the relatively small sample size. Therefore, the relationship between interoception and tics and PUs in TD patients with multiple comorbid diagnoses is hard to disentangle. Panic and somatic ratings, for example, were found to correlate with higher HIAcc in adults in children (Eley et al., 2007). Comparing TD patients with a control group exhibiting matching comorbidities could help to disentangle the complex influences of comorbidities on IAcc and PUs. In line with Pile et al. (2018), additional instructions could be added to muscle tension perception tasks to reduce effects of inattention, a frequent comorbid symptom in TD patients, and task-misunderstanding.

The PUTS measures PUs as a whole but does not differentiate between context- and time-dependent urges as a state and the PU as a general trait. It is yet to be examined how context- and time-dependent urges, measured by urge thermometers, vary in relation with changes in PUs as a trait and changes in IAcc.

Similar to the PUTS that measures PUs as a general trait, the SCL-TIC-S and SCL-TIC-P measure self- and parent-reported mean tic severity over the course of a week. It



is not clear how accurate self-assessed tic frequency reflects actual tic expression. On the one hand, both child and adult patients underestimate their tic expression (Müller-Vahl et al., 2014; Pappert et al., 2003). On the other hand, parent-reports cannot be accurate either. At least adolescent patients are not observed by their parents most of the day and tic frequency highly depends on context. Another study compared children's self-reported account of tic frequency to objective video ratings of tic frequency in various experimental situations. Self-reported tic frequency related to objective measures depended on the situation. Interestingly, the higher children scored on the PUTS, the better their self-report predicted objective tic-frequency (Barnea et al., 2016). This implies that self-reported tic frequency depends on PUs and probably IAcc. Still, as our multiple regression analysis showed, IA explains variance in addition to self-reported tic frequency. The moderate correlation with parent-reported tic-frequency (r = .34) further validates self-reported tic-frequency.

The current study holds clinical implications. Interoceptive trainings specifically targeting interoceptive domains that are impaired in TD may be more beneficial than multisystem interventions (Khalsa et al., 2018), so research on the impact of interoceptive trainings aimed at improving heartbeat perception (Schaefer et al., 2014) could be expanded by adding muscle tension biofeedback tasks to improve accuracy of urge perception to foster the ability to control tics via HRT. Observations from intervention studies examining muscle tension biofeedback in TD would provide further insights in causal relationships between IAcc, PUs and tics.

Funding: The authors have no funding to report.

Acknowledgments: The authors have no additional (i.e., non-financial) support to report.

Competing Interests: Christina Schütteler reports no conflict of interest. Katrin Woitecki received royalties from treatment manuals published by Hogrefe. Manfred Döpfner received consulting income and research support from Lilly, Medice, Takeda, and eyelevel GmbH and research support from the German Research Foundation, German Ministry of Education and Research, German Ministry of Health, and Innovation Fund. He received income as head, supervisor, and lecturer of the School of Child and Adolescent Cognitive Behaviour Therapy at the University Hospital Cologne and as consultant for Child Behaviour Therapy at the National Association of Statutory Health Insurance Physicians (Kassenärztliche Bundesvereinigung). He also received royalties from treatment manuals, books and psychological tests published by Beltz, Elsevier, Enke, Guilford, Hogrefe, Huber, Kohlhammer, Schattauer, Springer, Wiley. Alexander L. Gerlach reports no conflict of interest.

Data Availability: For this article, a data set is freely available (Schütteler, Woitecki, Döpfner, & Gerlach, 2020).



Supplementary Materials

The Supplementary Materials contain the following items (for access see Index of Supplementary Materials below):

- · Pre-registration protocol
- · Research data

Index of Supplementary Materials

- Schütteler, C., Woitecki, K., Döpfner, M., & Gerlach, A. L. (2020). Supplementary materials to "Interoception and premonitory urges in children and adolescents with tic disorders" [Preregistration protocol]. OSF Registries, https://doi.org/10.17605/OSF.IO/V3ZKY
- Schütteler, C., Woitecki, K., Döpfner, M., & Gerlach, A. L. (2021). Supplementary materials to "Interoception and premonitory urges in children and adolescents with tic disorders" [Research data]. figshare. https://doi.org/10.6084/m9.figshare.17121632

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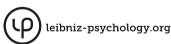
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Clinical Psychology in Europe (CPE) is the official journal of the European Association of Clinical Psychology and Psychological Treatment (EACLIPT).



PsychOpen GOLD is a publishing service by Leibniz Institute for Psychology (ZPID), Germany.







Research Articles



Skill Improvement Through Learning in Therapy (SKILT): A Study Protocol for a Randomized Trial Testing the Direct Effects of Cognitive Behavioral Therapy Skill Acquisition and Role of Learning Capacity in Depression

Sanne J. E. Bruijniks ^{1,2}, Ulrike Frank ¹, Brunna Tuschen-Caffier ¹, Jessica Werthmann ¹, Fritz Renner ¹

[1] Department of Psychology, Clinical Psychology and Psychotherapy, University of Freiburg, Freiburg, Germany.

[2] Department of Clinical Psychology, Utrecht University, Utrecht, the Netherlands.

Clinical Psychology in Europe, 2023, Vol. 5(1), Article e8475, https://doi.org/10.32872/cpe.8475

Received: 2022-03-02 • Accepted: 2023-01-06 • Published (VoR): 2023-03-31

Handling Editor: Cornelia Weise, Philipps-University of Marburg, Marburg, Germany

Corresponding Author: Sanne J. E. Bruijniks, Albert-Ludwigs University of Freiburg, Engelbergerstrasse 41, 79106, Freiburg, Germany. E-mail: s.j.e.bruijniks@uu.nl

Supplementary Materials: Materials [see Index of Supplementary Materials]



Abstract

Background: To improve psychological treatments for major depressive disorder (MDD), a better understanding on how symptoms ameliorate during treatment is essential. In cognitive behavioral therapy (CBT), it is unclear whether procedures focused on the acquisition of CBT skills play a causal role in the improvement of CBT skills. In this randomized trial, we isolate a single CBT Skill Acquisition Procedure (CBTSAP) and test its direct effects on CBT skills and related therapy processes (i.e., change in (idiosyncratic) dysfunctional thinking and reward processing). We hypothesize that the CBTSAP causes improvements in CBT skills and related therapy processes compared to an active control condition. In addition, we hypothesize that individual differences in attentional bias and memory functioning (defined as learning capacity) moderate the effects of CBTSAP on outcomes and that using mental imagery as a cognitive support strategy to strengthen the effects of the CBTSAP will be most beneficial for patients with low learning capacity.

Method: 150 patients with MDD will be randomized to one of three conditions: 1. an active control condition, 2. CBTSAP, 2. CBTSAP plus mental imagery, all consisting of three sessions. Primary outcomes will be change in CBT skills, changes in (idiosyncratic) dysfunctional thoughts



and behaviors, reward processing. Depressive symptoms are a secondary outcome. Measures of learning capacity will be conducted at baseline and tested as a potential moderator.

Discussion: Knowing whether and for whom the acquisition of CBT skills leads to change in therapy processes and a subsequent reduction of depressive symptoms will inform on how to personalize and optimize psychotherapy outcomes for depression.

Trial registration: The trial is registered at the German Clinical Trial Register (DKTR; registration number: DRKS00024116).

Keywords

 $major\ depressive\ disorder\ (MDD),\ cognitive-behavioral\ the rapy\ (CBT),\ cognitive\ behavioral\ the rapy\ skills,\ mental\ imagery,\ experiment$

Highlights

- Study protocol for a randomized trial to test direct effects of a procedure focused on CBT skill acquisition in MDD.
- Outcomes are CBT skills, dysfunctional thinking, reward processing and depressive symptoms.
- The role of learning capacity as a moderator will be investigated.
- Results will inform on the direct effects and individual differences in effects of CBT procedures.

Background

Current psychological treatments for depression are only effective for half of the patients (Cuijpers et al., 2021). Response to psychological treatments is limited and relapse rates are high (Steinert et al., 2014; Verduijn et al., 2017; Vittengl et al., 2007). To improve and innovate psychological treatments, a better understanding of how symptoms improve during treatment is essential.

Psychotherapies aim to reduce depressive symptoms by mobilizing therapy processes that seem central to the development and maintenance of depressive symptoms. Therapy processes can be defined as the mechanisms inside the mind of the patient that are activated by the therapeutic procedures delivered by the therapist with the intent of producing change (Bruijniks et al., 2018). In cognitive behavioral therapy (CBT; Beck et al., 1979), one of the most investigated treatments for depression, therapeutic procedures focus on three major therapy processes: dysfunctional thinking, behavioral activation and the acquisition of CBT skills (Barber & DeRubeis, 1989; Lorenzo-Luaces et al., 2016). First, cognitive change procedures aim to change the process of dysfunctional thinking (Garratt et al., 2007). Dysfunctional thoughts can be organized into different levels, some thoughts seem to occur more on a superficial level (negative automatic thoughts), while other thoughts are derived from more deeply integrated dysfunctional mental representations, that can include rules, expectations, or assumptions (attitudes) and sometimes



even originate from early experiences in the childhood (schemas) (Dozois & Beck, 2008). Second, CBT includes procedures aimed at behavioral activation in order to improve deficits in reward processing (Dimidjian et al., 2011), such as a reduced response to reward or an oversensitive response to negative feedback (Chiu & Deldin, 2007; Eshel & Roiser, 2010; Smoski et al., 2009). The third therapy process, the acquisition of CBT skills, is related to both dysfunctional thinking and behavioral activation. CBT skills are defined as the ability to re-evaluate the accuracy of one's own dysfunctional beliefs (CT skills) and in this way change patterns of dysfunctional thinking and the ability to engage proactively in pleasurable activities as a way to target reward experience (BT skills) (Strunk et al., 2007).

The acquisition of CBT skills is maybe one of the most promising therapy processes of CBT for depression. In contrast to the procedures focused on cognitive change and behavioral activation, CBT skill acquisition is a therapy process that emphasizes the patients' ability to use these cognitive change and behavioral activation procedures themselves, outside the therapy sessions. In addition, successful use of CBT skills may protect the patient from developing new future episodes after successful treatments (Strunk et al., 2007). Research shows that after successful treatment, impairments such as dysfunctional mental representations (Arntz, 2020; Sheppard & Teasdale, 2004), negative processing of information (Elgersma et al., 2019; Spinhoven et al., 2018; Woody et al., 2017) or blunted reward processing (Dichter et al., 2012; Pechtel et al., 2013) may remain, thereby possibly increasing the risk of new depressive episodes. The acquisition of CBT skills might be essential to transfer learned content from the therapy session to daily life and to cope with dysfunctional therapy processes or symptoms in future scenarios outside the therapeutic context.

However, although multiple studies have pointed out that the acquisition of CBT skills is associated with reduced depression (Adler et al., 2015; Forand et al., 2018; Strunk et al., 2014; Webb et al., 2019) and seems specific to CBT (Bruijniks et al., 2022), it is still unknown whether CBT skill acquisition directly causes a reduction of symptoms of depression. In order to test the causal effects of a certain therapeutic procedure, it is necessary to isolate the procedure and investigate its direct effects on the hypothesized changes in therapy processes and outcome (Bruijniks et al., 2018). Two preliminary experiments that focused on the acquisition of CT skills already evaluated how a procedure focused on the acquisition of CT skills could be isolated (Bruijniks et al., 2018) and showed that a short cognitive skill acquisition procedure in the form of a group masterclass led to better CT skill acquisition compared to an active control procedure in a sample of distressed students (Bruijniks, Los, & Huibers 2020). A next step towards the clinical application of this finding would be to evaluate how a procedure focused on CBT skill acquisition causally affects the acquisition of CBT skills and subsequent symptom reduction in a clinically depressed sample.



Nevertheless, skill acquisition seems to be a multifaceted process that requires different cognitive and neurobiological resources (Anderson et al., 2016, 2018; Basak et al., 2011; VanLehn, 1996), which may be impaired in depressed patients. Compared to healthy individuals, depressed individuals have biased attention towards negative rather than positive information (Fu et al., 2008; Liu et al., 2012; Marchetti et al., 2018; Roiser et al., 2012) and suffer from a variety of deficits related to executive functioning, such as inhibition, planning and working memory (Snyder, 2013). Recent studies supported the hypothesis that individual differences in cognitive or neurobiological impairments may interfere with the success of psychological treatments. In a systematic review, cognitive and neurobiological impairments showed to be associated with impairments in dysfunctional thinking and reward processing while depressed patients with better cognitive control, but more emotional bias, before start of CBT seemed to benefit more from CBT's procedures (Bruijniks, DeRubeis, et al., 2019). Possibly, patients who show more emotional bias are better able to tolerate and therefore target emotions as part of the CBT (Stange et al., 2017) while individuals with better cognitive capacities are more capable of integrating and implementing new information that was retrieved in the therapy session. CBT might help individuals with increased emotional bias, but limited cognitive capacity, to regain (emotional) control (Siegle et al., 2006). A recent experiment supports this suggestion, as results indicated that in healthy participants who received a stress induction, executive control under stress, but not under non-stressful circumstances, predicted the ability to reappraise negative material to become less negative (Quinn & Joormann, 2020). Investigating whether individual differences in cognitive or neurobiological impairments are associated with the success of a CBT skill acquisition procedure will provide insight in for whom CBT skill acquisition procedures will be more or less effective.

If individual differences in cognitive or neurobiological impairments are related to the success of CBT skill acquisition, this also means that the success of CBT skill acquisition might be improved by increasing the patients' capacity to learn from these procedures. One way to address and improve cognitive and neurobiological processes during treatment is by providing cognitive support. Examples are the use of memory strategies within sessions of CBT (Harvey et al., 2014, 2017) or providing short retrieval tests between the sessions (Bruijniks, Sijbrandij, et al., 2020). The major hypothesis is that by enhancing recall for the session content, the success of psychotherapy outcomes for depression can be improved and some studies provided preliminary evidence for this hypothesis (Dong, Lee, et al., 2017). However, to improve psychotherapy it might not only be important to improve recall of the session content but also improve and develop the therapy process. Yet, while current cognitive support strategies such as retrieval of newly learned information may improve recall of the session content, it might not be enough to improve CBT skill acquisition. According to theories on skill acquisition, the process of skill acquisition starts with learning new information (this can also be seen



as the 'declarative' part of skill acquisition), but then repeated practice is necessary to turn it into a more procedural form in which the newly learned skill becomes more and more automatized over time (Anderson et al., 2018; Tenison & Anderson, 2016; VanLehn, 1996). To increase skill acquisition, it might therefore be necessary to use a strategy that supports both the declarative and procedural parts of memory.

One strategy that seems promising in affecting both declarative and procedural memory is mental imagery. Mental imagery refers to perceptual experiences in the absence of sensory input and constitutes a non-verbal way of information processing, closely related to the experience of emotions (Holmes & Mathews, 2010). Mental imagery allows us to simulate past and future experiences and because of this allows us to "try-out" different courses of actions and their emotional consequences (Ji et al., 2016; Moulton & Kosslyn, 2009). Given these properties of mental imagery, when applied to CBT skills, imagery could be used to simulate skill application (Renner et al., 2021). Mental imagery has been linked to improved acquisition of skills in non-clinical settings, such as tennis performance or the development of surgical skills (Anton et al., 2017; Dana & Gozalzadeh, 2017; Gregg et al., 2011; Kim et al., 2017; Kraeutner et al., 2016), but also to increased BT skills in a clinically depressed population (Renner et al., 2017). Additionally, mental imagery has been related to the improvement of cognitive functioning, such as recall of memories (Dalgleish et al., 2013) and prospective memory (i.e., memorizing to execute a previously formed intention at some point in the future; McFarland & Glisky, 2012; McFarland & Vasterling, 2018). We suggest that simulating applying CBT skills using mental imagery might be a potential efficient way to increase skill acquisition during psychotherapy (Renner et al., 2021).

The aims of this randomized trial are two-fold. The first aim is to investigate and compare the direct effects of three procedures (active control, CBT skill acquisition (CBTSAP), CBTSAP + mental imagery) on changes in therapy processes (the acquisition of CBT skills, changes in idiosyncratic dysfunctional thoughts and behaviors, general dysfunctional thinking and reward processing) and depressive symptoms in a sample of patients with a diagnosis of major depressive disorder who do not currently receive other psychological treatment. We expect that compared to an active control procedure, the procedures focused on CBT skill acquisition (CBTSAP and CBTSAP + mental imagery) will lead to more improvement in the therapy processes and depressive symptoms. Second, we will investigate whether the effect of the therapeutic procedures is moderated by individual differences in learning capacity. Learning capacity will be defined as the presence of memory functioning and emotional bias (i.e., where more emotional bias and better memory functioning are defined as better learning capacity). Following earlier literature on the measurement of memory (Unsworth, 2010; Wilhelm et al., 2013), the measurement of memory functioning will be composed of both working memory and long-term memory tasks. We expect that, compared to the active control procedure, better learning capacity will be associated with larger improvements in the therapy



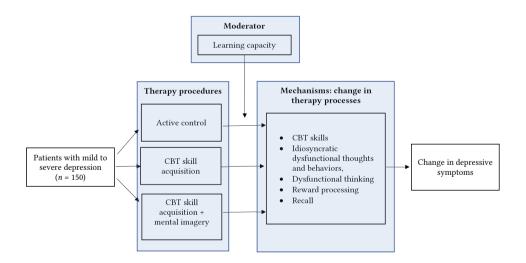
processes and depressive symptoms in both the CBTSAP's. In addition, we expect a difference between CBTSAP with versus without mental imagery: patients with low learning capacity will have most benefit from mental imagery and lower learning capacity will therefore be associated with more improvement in the therapy processes and depressive symptoms in the CBTSAP with mental imagery compared to the CBTSAP without mental imagery condition.

Besides the two main aims of the study, additional secondary analyses will be conducted. Because earlier studies suggested that cognitive support might improve the effects of therapy by increasing memory of the session content (Dong, Lee, et al., 2017), we additionally included a measure of session recall in the study and will test whether session recall will differ between the procedures. Hypotheses of these secondary analyses are in line with our hypotheses for the main study aims: we expect session recall to be larger in the CBTSAP with mental imagery compared to the CBTSAP without mental imagery. In further secondary analyses we will investigate whether the effect of the procedures on depressive symptoms is mediated through one of the therapy processes and/or session recall and whether these mediation effects are specific to the CBTSAP's (compared to the active control procedure).

A conceptual model for the proposed study can be found in Figure 1.

Figure 1

Conceptual Model for the Proposed Study



Method

Design

Between-subject experimental design with three parallel conditions, each having an equal length of 3 x 45-minute sessions: 1) Active control procedure (n = 50), 2) CBT skill acquisition procedure (CBTSAP) (n = 50), 3) CBTSAP with mental imagery (n = 50). The Ethic Committee of Freiburg University approved the study (registration number: 20-1022) and the trial is pre-registered at the German Clinical Trial Register (DKTR; registration number: DRKS00024116).

Participants

We aim to include 150 patients with a primary diagnosis of a major depressive disorder (MDD) (excluding non-dysthymic persistent major depressive disorder) as indicated by Structural Clinical Interview for DSM-5 Disorders (SCID-5-CV) and a score on Beck's Depression Inventory II (BDI-II) ≥ 14 to ensure sufficient symptom severity. Patients should be aged between 18-65 and have sufficient knowledge of the German language (because therapy sessions will be held in German). To prevent any potential interference with the therapeutic procedures and/or measurement of learning capacity, patients with the presence of a previously stated diagnosis of attention-deficit/hyperactivity disorder or attention-deficit disorder, current drug or alcohol use disorder according to the Structural Clinical Interview for DSM-5 (SCID-5-CV) or a cluster A or B personality disorder known by admission to the treatment center are excluded. To ensure the effects are attributable to the current therapeutic procedures, patients who receive currently (other) psychological treatment or have received CBT focusing on a major depressive disorder in the previous year are excluded. To reduce risk of adverse events, patient who show a high risk of suicide according to the intake staff or a score > 1 on BDI_II item 13 (Suicidal thought or wishes) will be excluded.

Sample Size

Based on a medium effect size, alpha = .05, power =.80, number of experimental conditions = 3, number of repeated measurements = 2 to 4 (G^* power (Faul et al., 2007)), a total sample size of 102 to 120 participants would be needed to detect a main effect, and 42 to 57 participants to detect an interaction in a repeated measures ANOVA. Simulation studies suggest 80 to 100 participants to detect an interaction effect between three groups (Shieh, 2019) while simulation studies on multilevel analyses suggest n = 80 participants to detect a medium effect size with power =.80 (Aarts et al., 2014). Taking into account 20% drop-out, we aim to include a total of 150 participants.



Recruitment

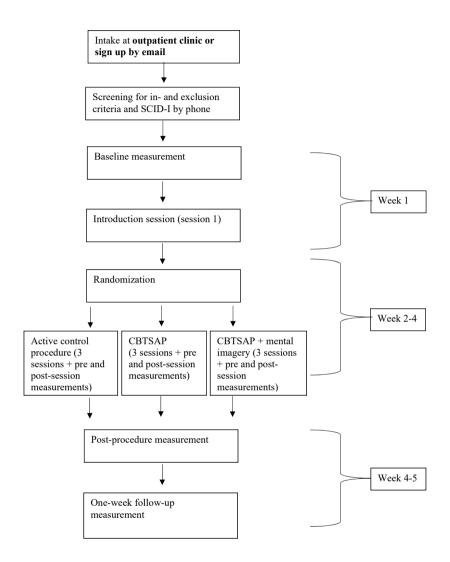
Patients will be recruited in two different ways. First, patients will be recruited from the academic outpatient treatment center at the Department of Psychology, Unit for Clinical Psychology and Psychotherapy at the Albert-Ludwigs University of Freiburg. Patients with various mental health problems and with a large variety of socio-demographic backgrounds seek treatment at the clinic. Patients can receive up to 80 sessions individual CBT at the clinic in accordance with the German national health insurance regulations. Treatment seeking individuals with severe mental disorders (e.g. schizophrenia) or acute suicidality are referred to other specialized services outside the clinic or in-patient treatment if indicated. During the intake patients will be checked on in- and exclusion criteria and receive the patient information letter if they are potentially eligible for study participation. After one week, patients will be called to check whether they are interested in participating in the study. The remaining in- and exclusion criteria will be checked, and the Structural Clinical Interview for DSM-5 Clinical Version (SCID-5-CV) interview will be conducted by phone. The procedures will take place while the patient is on a waiting list for regular treatment at the outpatient clinic. Second, individuals can sign up for the study independently of treatment in the academic outpatient treatment center. Information about the study will be put online and distributed in local health care centers. If interested, individuals will be send the patient information letter, called after one week to check remaining in- and exclusion criteria and a SCID-5-CV will be planned.

Randomization and Procedure

Patient who are eligible to participate in the study will complete a baseline measurement and an introduction session on different days. The baseline measurement will take place in the lab and includes a measurement of learning capacity. Informed consent will be signed before the baseline measurement. The order of the measurements during the baseline measurement will be randomized for each participant in order to control for potential fatigue effects. The introduction session is conducted by the therapist and focuses on introducing the principles of CBT and completing the Core Belief Interview (CBI; McBride et al., 2007). After the introduction session, patients will be randomized into one of the three conditions using a computer script performing block randomization (1:1:1, block size = 15). Block randomization will be done by a researcher who is not involved in the study measurements. Randomization will be pre-stratified on severity of depression (mild [Beck Depression Inventory-II (BDI-II) = 14-19] vs. moderate to severe [BDI-II ≥ 20]). Therapy sessions will be completed weekly and the total study procedure from baseline measurement to the post measurement will take a maximum of 5 weeks. The researchers who perform the study measurements are blind for the therapeutic procedures. The full study procedure is also presented in Figure 2. Participants do not receive financial incentives for participation in this study.



Figure 2
Recruitment and Study Procedure



Note. The exact time point of each measurement is given in Table 1.



Therapists

The therapeutic procedures will be conducted by 5 licensed therapists who are working at the outpatient treatment center of the University of Freiburg. All therapist involved in this study had completed a 3-year fulltime psychotherapist CBT training course required and strictly regulated in Germany to obtain a license as clinical psychological psychotherapist. Therapist have between 5 and 29 years therapy experience. Before start of the study, the therapists received 8 hour training consisting of advanced training in CBT skills by dr. Strunk (Ohio State University), advanced mental imagery training conducted by dr. Renner (University of Freiburg) and elaborate training on the different protocols for each therapeutic procedure in the study. All therapists will be involved in the delivery of all different procedures.

Introduction Session

The introduction session will focus on introducing the principles of CBT (central focus will be on the relation between thoughts, behaviors and mood) and completing the CBI. During the CBI, the therapist and patient try to gain insight in the current three most relevant dysfunctional beliefs and current three most relevant dysfunctional behaviors for the patient. These beliefs and behaviors will be used in the subsequent sessions to discuss in relation with depressive symptoms (active control procedure) or to practice CBT skills (CBTSAP and CBTSAP + mental imagery condition).

Therapeutic Procedures

All therapeutic procedures use techniques from the protocol for cognitive behavioral therapy (CBT; Beck et al., 1979) for depression and use agenda setting to structure the sessions. In addition, each procedure will focus on targeting the idiosyncratic beliefs and behaviors that were established during the CBI. However, the procedures differ in the number of active ingredients (see Figure 3 and data supplement 1). During the active control procedure, the therapist and patient will focus on discussion of dysfunctional thoughts and behaviors only. Therapists in this condition will be explicitly instructed to focus purely on exploring the relation between dysfunctional thinking and behavior and depressive symptoms, and not to engage in evaluating dysfunctional thinking or behavioral activation. During the CBT skill acquisition procedure (CBTSAP), the therapist and patient will choose one of the cognitive or behavioral skills from a predefined list of CBT skills (i.e., consisting of behavioral activation (behavioral therapy skill) and questions used for evaluating dysfunctional thoughts (cognitive therapy skill). Subsequently, the therapist and patient will discuss how this skill could be or have been applied in past or future situations in which idiosyncratic beliefs and behaviors may (have) lead to negative mood. During the CBTSAP plus mental imagery, the therapist and patient will not only discuss application of the skill but in addition, engage in a mental imagery exercise



of skill application. The mental imagery exercise is based on a guided mental imagery procedure and has been shown to increase motivation for goal directed behaviors (Heise et al., 2022; Renner et al., 2019). During the mental imagery procedure, participants are instructed to imagine as vividly as possible and focusing on the positive aspects of the image. The procedure consists of the following steps: 1) imagine the contextual cues (e.g., place, date) of a future or past situation with depressive symptom(s), 2) engaging in multi-sensory imagery of applying the CBT skill in this situation, 3) imaging and experiencing the positive aspects related to successfully applying the CBT skill. All sessions will be videotaped for treatment fidelity checks. Research intervision will take place regularly. The agenda for each therapeutic procedure and the list of CBT skills that can be chosen from and practiced in the CBTSAP procedures is given in data supplement 1 and 2.

Figure 3

Therapeutic Elements per Procedure







Note. Detailed information on the session content can be found in data supplement 1 and 2.

Instruments

An overview of all patient measurements is given in Table 1. An overview of all measurements completed by the therapists or third observers can be found in Table 2.



 Iable 1

 Overview of Patient Instruments per Time Point

		Introduction				One day after	
Measurement instruments	Baseline	session	Session 1	Session 2	Session 3	Session 3	Follow-up
		Primary or	Primary outcomes: therapy processes	processes			
CBT skills							
Ways of Responding (WOR)	×						×
Behavioral Activation for	×	×	×	×	×		×
Depression Scale – short form							
(BADS-SF)							
Cognitive Change Sustained	×	×	×	×	×		×
Change (CCSC)							
Idiosyncratic thoughts and behaviors	rs						
Core Belief Interview (CBI)		×	×	×	×	×	×
Dysfunctional thinking							
Cognition Checklist (CCL)	×					×	×
Reward processing							
Reward Probability Index (RPI)	×					×	×
Temporal Experience of Pleasure	×					×	×
(TEPS)							
		Seconda	Secondary outcomes: symptoms	ıptoms			
Depression							
Beck Depression Inventory II (BDI-II)	×	×	×	×	×		×
Symptoms other than depression							
Brief Symptom Inventory (BSI)	×						×



		Introduction				One day after	
Measurement instruments	Baseline	session	Session 1	Session 2	Session 3	Session 3	Follow-up
		Potential mo	Potential moderators: Learning capacity	g capacity			
Memory functioning							
Verbal working memory: n-back	×						
Visual working memory: Single probe detection task	×						
Long-term memory Paired- associates task	×						
Emotional bias							
Free viewing eye-tracking task	×	×				×	
			Other measures				
Recall							
Patient Recall Test (PRT)			×	×	×	×	×
Diagnostics							
SCID-5-CV	×						
Demographics	×						
Treatment evaluation							×
Manipulation check			×	×	×		
Procedure check							×

Note. CBTSAP = CBT Skill Acquisition Procedure; MI = Mental Imagery; Structural Clinical Interview for DSM-V Clinical Version (SCID-5-CV).

×



Expected success

 Table 2

 Overview of Therapist/Observer Instruments per Time Point

Measurement		Introduction				Post-	
instruments	Baseline	session	Session 1	Session 2	Session 3	procedure	Follow-up
Therapy processes							
Therapist-rated recall			X	X	X		
Manipulation check							
Therapy integrity							X
Protocol deviations			X	X	X		

Primary Outcome: Therapy Processes

CBT Skills — CBT skill acquisition will be measured in two different ways. First, before and one week after the therapeutic procedure patients will complete the Ways of Responding (WOR; Barber & DeRubeis, 1992). During the WOR, CBT skills of the participants are tested by asking them to think about themselves in various situations and to tell what they would think and do in such situations. The WOR will reflect the level of CBT skills demonstrated by the patient. Patients will receive three scenarios before treatment and three different scenarios after treatment. Answers to each scenario will be coded into 25 different categories (more categories per answer possible) and given a rating of the overall quality of the response (i.e., the raters' judgment on how well the response would be in improving mood or adjusting to the individual's needs, range = 1 (very negatively) to 7 (very positively)). The total score will be composed of the number of responses on positive categories (responses considered consistent with CBT) minus the number of responses on negative categories (depressotypic statements). Interrater reliability showed to be high (ranging from $\alpha = .91$ to $\alpha = .98$) and discriminant and convergent validity have been supported (for example: the WOR showed no correlation with a measure of self-control, but was correlated to self-report measure of CBT skills) (Barber & DeRubeis, 2001; Strunk et al., 2014). Second, change in CBT skills during the procedures will be measured using the Behavioral Activation for Depression Scale – short form (BADS-SF) (BA skills) and Cognitive Change Sustained Change (CCSC) (CT skills). The BADS-SF consists of nine items, each rated on a 7-point Likert Scale and internal consistency ($\alpha = .81$) and construct and predictive validity were supported (for example: the BADS-SF was positively related to measures of reward, negatively related to measures of avoidance and predicted time spent in high and low rewarding behavior; Manos et al., 2011). An example item from the BADS-SF is: 'There were certain things I needed to do that I didn't do'. The CCSC consists of 9 items rated on a 7-point Likert scale and internal consistency was supported ($\alpha = .93$) and the scale showed convergent and discriminant validity by showing a relation with a self-report scale of CBT skills and no relation with a measure of attributional styles (Schmidt et al., 2019). An example item from the CCSC is: 'I noticed myself thinking less negatively.'



Idiosyncratic Thoughts and Behaviors — Idiosyncratic thoughts and behaviors will be measured with the Core Belief Interview (CBI; McBride et al., 2007). This interview will be completed by the therapist during the introduction session. Together the therapist and patient will form an idiosyncratic top three of dysfunctional thoughts and top three of dysfunctional behaviors. Note that the behaviors can both exist of the presence of unhelpful behaviors or the absence of rewarding behaviors. Based on the identified beliefs and behaviors, six idiosyncratic visual analogue scales (VAS) (0-100) will be constructed for each patient (i.e., three dysfunctional beliefs, three dysfunctional behaviors). For the dysfunctional beliefs, credibility of the beliefs and strength of related emotions will be rated. Presence, reward and pleasure related to the behaviors will be measured. The CBI has been used successfully before to establish idiosyncratic dysfunctional thoughts (Bruijniks, Los, & Huibers, 2020; Renner et al., 2018). The exact items of the CBI are given in data supplement 3.

General Dysfunctional Thinking — General dysfunctional thinking will be measured using the Cognition Checklist (CCL; Taylor et al., 1997). The CCL consists of 26 items rated on a 5-point Likert Scale and can be divided into two subscale measuring dysfunctional thoughts related to depression versus anxiety. Internal consistency (ranging from α = .91 to α = .93) and validity was supported in an outpatient sample (i.e. the depression subscale showed a higher relation to other depression measures compared to the anxiety subscale, and the same was shown in the reverse direction; Steer et al., 1994). An example item of the CCL is: 'When I am with a friend I think: I'll never be as good as other people are.'

Reward Processing — Reward processing will be measured using the Reward Probability Index (RPI; Carvalho et al., 2011) and the Temporal Experience of Pleasure Scale (TEPS; Gard et al., 2006). The RPI is a 20-item self-report instrument that measures the presence of environmental reward, while the TEPS is an 18-item self-report instrument that measures the ability to experience pleasure. Reliability (RPI: α = .93, TEPS: α = .75) and discriminant and convergent validity have been supported for both instruments (for example: the RPI was related to another measures of reward, but not to measures of anxiety and support, and was related to experiencing rewarding behavior; the TEPS was relatable but also distinguishable from other measures of motivation and pleasure; Carvalho et al., 2011; Gard et al., 2006; Simon et al., 2018). An example item of the RPI is: 'I have many interests that bring me pleasure.' (RPI). An example item of the TEPS is: 'I enjoy taking a deep breath of fresh air when I walk outside'.

Secondary Outcome: Psychological Symptoms

Depression — Depression will be measured with the Beck Depression Inventory-II (BDI-II; Beck et al., 1996). The BDI-II is a 21-item self-report instrument assessing depressive



symptoms during the last two weeks. The items are rated from 0 to 3, higher scores representing more symptom severity. A score 0–13 indicates minimal depression, 14–19 mild depression, 20–28 moderate depression and 29–63 severe depression. Reliability and validity have been supported (i.e., test retest reliability between .73-.96, α = .85, convergent and discriminant validity; Beck et al., 1988; Wang & Gorenstein, 2013). For the purpose of this study, the BDI-II will be adjusted to assess depressive symptoms during the past week.

General Psychological Distress — Additional psychological symptoms will be measured using the Brief Symptom Inventory (BSI; (Derogatis & Melisaratos, 1983). The BSI consist of 53 items rated on a 0 (not at all) to 4 (extremely) scale and includes the following subscales: somatization, obsessives-compulsive, interpersonal sensitivity, depression, anxiety, hostility, phobic anxiety, paranoid ideation and psychoticism. Reliability and validity of the scale have been supported (i.e., Cronbach's alpha of .85, concurrent and divergent validity was supported; de Beurs & Zitman, 2006; Geisheim et al., 2002).

Potential Moderators: Learning Capacity

Memory Functioning: Verbal Working Memory — Verbal working memory will be measured using the N-back task (Braver et al., 1997). The n-back task measures verbal working memory. During the n-back task participants will be asked if a letter on the screen matches a letter previously (1-back, 2- back, 3-back) presented for 500 ms with an interval of 2000 ms. WM load increases as the task progresses from 1-back to 3-back. Accuracy of responses (total of correct hits (% correctly identified *n*-backs) and correct no hits (% correctly identified no presence of a *n*-back)) are measured and will be used as an outcome measure. The *n*-back task has been considered as a valid measure of working memory (Cronbach's alpha = .92; Schmiedek et al., 2014; Wilhelm et al., 2013).

Memory Functioning: Visual Working Memory — Visual working memory will be measured using the probe change detection task (PCDT; Dai et al., 2019). The PCDT consists of the following steps: 1. Participants are instructed by an arrow on their screen to focus on the left or right side of the screen (200 ms), 2. After a short break (300 ms) the screen is filled with colored squares on a gray background (100 ms). The squares are equally distributed between the left and right side of the screen. Participants are instructed to remember only the squares on the side that was instructed under Step 1, 3. After a second blank screen (900 ms), participants see again a field with colored squares (750 ms) and have to indicate whether the squares on the side of the screen are the same as under Step 2. Set sizes of the trial different between 8 to 12 colored squares in total. Participants will receive a total of 300 trials. Reliability and validity has been supported (i.e., test retest reliability between .52-.75; Dai et al., 2019).



Memory Functioning: Long-Term Memory — Long-term memory will be measured using the paired associates task (PAT; Unsworth et al., 2009). Long-term memory can also be considered as 'secondary memory', i.e., the part of memory where information is stored when the primary memory, where new information is temporary maintained is full. During the PAT, participants will be given three lists of 10 non-semantically related word pairs. All words are common nouns, and the word pairs will be presented vertically for 2 sec each. Participants will be told that the cue would always be the word on top and that the target would be on bottom. After the presentation of the last word (which takes 20 seconds), participants will see the cue word and "????" in place of the target word. Participants will be instructed to type in the target word from the current list that matches the cue. Cues will be randomly mixed so that the corresponding target words are not recalled in the same order as that in which they had been presented (i.e., this means that the time between encoding and recall varies and lies between the 2 and 70 seconds). Participants will have 5 sec to type in the corresponding word. A participant's score is the proportion of items recalled correctly. Words will be taken from the Toronto Word Pool (Friendly et al., 1982). The paired associates task has been considered a valid task of long-term memory (Unsworth et al., 2009; Wilhelm et al., 2013).

Emotional Bias: Sustained Selective Attention to Emotional Stimuli — Selective attention will be measured using a free-viewing eye-tracking task (Klawohn et al., 2020). Participants will view two blocks of neutral and happy and neutral and sad faces in counterbalanced order while their gaze patterns are concurrently recorded as index of selective spatial attention. Each block will take 30 trials that last 6 seconds. Each trial will show 16 different faces and participants will be asked to freely view the trials. Participants' gaze location and duration will be assessed using Eyelink eye-tracker software (https://www.sr-research.com/). The present study will use the exact same task as was recently used and validated by Klawohn and colleagues (2020). To maximize reliability of the task (MacLeod et al., 2019), it will be completed twice at baseline. To further investigate the predictive value of the task, an additional post procedure measurement (i.e., at one week follow-up) will be completed.

Other Measures

Recall — Patient recall will be measured using the Patient Recall Test (PRT; Lee & Harvey, 2015). The PRT measures recall of the previous session content. Following procedures of Lee & Harvey, the patient will be given 10 minutes to remember as much treatment points from the previous session as possible (past session recall). In addition, cumulative recall (i.e., what is remembered from all sessions) will be measured at the follow-up session. Treatment points will be defined as remembering insights, skills and strategies of the CBT model. Scores will be rated by two independent raters, inconsistencies in scoring will be resolved by discussion. Interrater reliability between raters will



be computed. The PRT showed good interrater reliability in previous studies (ICC = .92; Dong, Zhao, et al., 2017). In addition to recall of the patient, therapists are also asked to give a rating of recall on a 1-10 VAS scale (1 = patient has no memory of the previous session, 10 = patient remembers everything perfectly).

Manipulation Check — To check if patients in the CBT skill acquisition + mental imagery condition engaged in vivid mental imagery, they will be asked to note how vivid the imagery of the skills practiced in this session was on a scale from 1 (not vivid at all) to 10 (extremely vivid). To check and potentially control for self-efficacy, motivation and anticipated reward in the analyses, participants in all conditions will complete questions on a 0-10 scale and asked to rate based on today's session how capable they feel in coping with their dysfunctional beliefs and behaviors, their motivation to use the content of today's session to do something different in the upcoming week and their anticipated reward of doing something different in the upcoming week based on today's session. In addition, expected success of the SKILT study sessions in reducing depressive symptoms will be asked at baseline and after the first session. At the end of the study, patients will be asked to rate on a 0-10 scale to what degree the received sessions contributed to an improvement in depressive symptoms.

Adherence — *Protocol Deviations*. After each session, the therapist will complete a short questionnaire to check 1. how many skills and application of these skills were discussed, 2. How many mental imagery exercises were conducted, and 3. ask for the presence of deviations to the protocol in that session.

Procedure Integrity. To ensure the procedures differ in the presence of active components (i.e., CBT skill application and use of mental imagery) all sessions will be video-taped. A questionnaire will be developed that measures the presence and duration of the different components in the therapeutic procedures. This questionnaire will be completed by two independent raters.

Diagnosis — The SCID-5-CV (First et al., 2019) will be completed by phone during the recruitment phase.

Data Analyses

All statistical tests will be two-tailed (significance level alpha .05). Descriptives (means, standard deviations) for all measures will be provided for each condition. All analyses will be intention-to-treat and missing data will not be imputed.

Main Analyses

The main analyses will be conducted in Stata. First, to test the direct effects of the different procedures on change in therapy processes and symptoms, differences on the



primary and secondary outcomes between conditions (CBTSAP versus active control; CBTSAP versus CBTSAP + mental imagery; active control versus CBTSAP + mental imagery) will be tested using multilevel analysis with maximum likelihood estimation (measurements [Level 1] nested within patients [Level 2] nested within therapists [Level 3]). Because the WOR is only measured at two time points, differences between conditions on the WOR will be tested using a repeated measures ANOVA.

Second, to test whether the effect of the procedures is moderated by individual differences in learning capacity, learning capacity will be added as a moderator in the model. Moderation will be tested by adding learning capacity as a main factor and the interaction between learning capacity and condition to the multilevel regression model. For the WOR, the interaction will be added to the repeated measures ANOVA. Moderation of learning capacity will be tested separately for memory functioning and emotional bias. Memory functioning will be tested separately for each component of memory functioning (i.e., verbal working memory, visual working memory, long-term memory), while controlling for type I error (p < .016).

Mediation Analyses

The potential role of therapy processes and session recall as mechanisms of change will be tested by testing mediation within latent difference score (LDS) models. In separate LDS models (i.e., a different model for each mediator), we will test the relation of the procedure (CBTSAP's versus active control) on subsequent change in the mediator (i.e., therapy processes: CBT skills, idiosyncratic dysfunctional thinking and behaviors, general dysfunctional thinking, reward processing, and session recall) on subsequent change in the outcome (depressive symptoms). Note that we will merge the two CBTSAP's to test mediation of the active control versus the CBTSAP's. LDS models allow tests of mediation, include the temporality of the effects and are therefore capable of testing potential reverse causality (Grimm et al., 2017; McArdle, 2009).

Discussion

We presented a protocol for a randomized controlled study that isolates an often-used therapeutic procedure focused on the acquisition of cognitive behavioral therapy skills (CBTSAP) to test its causal effects on psychotherapy outcomes. The CBTSAP will be compared to an active control condition and CBTSAP with mental imagery. We hypothesized that, compared to an active control procedure, the CBTSAP's would lead to direct improvement in CBT skills, related therapy processes (dysfunctional thinking and reward processing) and subsequent reduction of depressive symptoms. In addition, we suggested that individual differences in cognitive and neurological impairments (referred to as learning capacity) in depressed patients may interfere with the successful acquisition of



CBT skills and that especially the patients with low learning capacity will benefit from added mental imagery to the CBTSAP.

One major strength of the present study is that it will be the first to investigate the direct effects of an isolated procedure focused on the acquisition of CBT skills in a depressed sample. Although the potential of investigating isolated procedures has been recognized (Craske, 2016; MacLeod & Grafton, 2016; Teasdale & Fennell, 1982), experimental studies that isolate therapeutic procedures in the field of depression have been scarce so far (Bruijniks et al., 2018). In addition, by performing an experiment that informs us about which therapeutic procedure works best for whom, the proposed study taps into the field of personalized medicine (i.e., optimizing the effects of treatment by matching the treatment to the patient (Cohen & DeRubeis, 2018)). By increasing insight in the direct effects of therapeutic procedures on how and for whom they reduce depression, the present study will not only contribute to the research field of personalized medicine, but has the potential to inform and improve clinical practice (i.e., informing on what technique might be helpful for whom).

This study is also the first that elaborately assesses learning capacity at baseline to investigate the moderating role of learning capacity on the effects of isolated CBT procedures. Earlier studies have indicated that depressed patients with more emotional bias and more memory functioning might show better improvement during CBT (Bruijniks, DeRubeis, et al., 2019), but these studies mostly investigated the role of learning capacity on the complete treatment package (i.e., a full CBT that includes multiple CBT procedures), primarily used neurobiological measures or where conducted in an elderly depressed population. The present study will be able to inform on the specific role of learning capacity in a key therapeutic procedure, the acquisition of CBT skills, in CBT for depression. In addition, a better understanding of the role of learning capacity and a CBT skill acquisition procedure on CBT outcomes might open up new avenues for future research on the role of skill acquisition and learning capacity in psychotherapy for depression in general. Another strength of the study is the repeated measurement of therapy processes, which will allow us to investigate how learning capacity affects the hypothesized mechanisms underlying the success of a CBT skill acquisition procedure and also how the CBTSAP might lead to reduction of depressive symptoms through these mechanisms. A final strength of the present study is that it includes a multimodal assessment, using not only self-report instruments but also a CBT skill test, idiosyncratic measures of therapy process change, behavioral tasks and eye-tracking. A limitation of the present study is that it is powered to find medium to large effects and will be underpowered to find small effects between the three treatment conditions.

In conclusion, while there are a number of effective evidenced based treatments for depression, many patients do not improve in treatment and progress in treatment innovation has been slow. One way forward is to isolate specific therapeutic procedures and test their direct effects on therapy processes and outcomes. Based on this experimen-



tal research framework we will conduct a randomized clinical study testing the direct effects of a key CBT procedure, CBT-skills, with or without a cognitive support strategy compared to an active control condition. The results of this study will contribute to a better understanding of individual differences in the effects of key CBT procedures.

Funding: This project is funded by a Rubicon research grant granted to S. Bruijniks by the Dutch Research Council Social Sciences and Humanities from NWO. FR and JW are supported by a Sofja Kovalevskaja Award from the Alexander von Humboldt Foundation and the German Federal Ministry for Education and Research.

Acknowledgments: The authors thank Anna Boehncke, Stephanie Heinrichs, Max Heise, Capucine John, Lisa Krause, Janina Reus, Svenja Schmedding, Adriana Soyoung-Steinborn and dr. Lena Zirn for their (ongoing) contribution to the study.

Competing Interests: The authors have declared that no competing interests exist.

Author Contributions: SB designed the study and wrote the manuscript. UF, BTC, FR and SB are involved in the coordination of the recruitment of patients and data collection. JW is involved as an eye-tracking expert. All authors read, contributed to, and approved the final manuscript.

Twitter Accounts: @SBruijniks

Data Availability: Data sharing is not applicable to this article as no datasets were generated or analyzed during the current study.

Supplementary Materials

The Supplementary Materials contain more exact information on the different therapeutic procedures and the outcomes of the core belief interview as these are used in the study (for access see Index of Supplementary Materials below).

Index of Supplementary Materials

Bruijniks, S. J. E., Frank, U., Tuschen-Caffier, B., Werthmann, J., & Renner, F. (2023). Supplementary materials to "Skill Improvement Through Learning in Therapy (SKILT): A study protocol for a randomized trial testing the direct effects of cognitive behavioral therapy skill acquisition and role of learning capacity in depression" [Additional information]. PsychOpen GOLD. https://doi.org/10.23668/psycharchives.12574



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Appendix: List of Abbreviations

BADS-SF - Behavioral Activation for Depression Scale - short form

BSI - Brief Symptom Inventory

CBI - Core Belief Interview

CBT - cognitive behavioral therapy

CBTSAP - cognitive behavioral therapy skill acquisition procedure

CCSC - Cognitive Change Sustained Change

CCL - Cognition Checklist

PAT - Paired associates task

PCDT - Probe change detection task

PRT - Patient Recall Test

RPI - Reward Probability Index

LDS - Latent difference scores

SCID-5-CV - Structural Clinical Interview for DSM-V

TEPS - Temporal Experience of Pleasure Scale

VAS - Visual analogue scale

WOR - Ways of Responding

EACLIPT

Clinical Psychology in Europe (CPE) is the official journal of the European Association of Clinical Psychology and Psychological Treatment (EACLIPT).



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Latest Developments



The Cultural Supplement: A New Method for Assessing Culturally Relevant Prolonged Grief Disorder Symptoms

Clare Killikelly 1,2 D, Andreas Maercker 1 D

[1] Department of Psychology, University of Zürich, Zurich, Switzerland. [2] Department of Psychiatry, University of British Columbia. Vancouver. Canada.

Clinical Psychology in Europe, 2023, Vol. 5(1), Article e7655, https://doi.org/10.32872/cpe.7655

Received: 2022-03-02 • Accepted: 2022-11-14 • Published (VoR): 2023-03-31

Handling Editor: Cornelia Weise, Philipps-University of Marburg, Marburg, Germany

Corresponding Author: Clare Killikelly, Department of Psychology, University of Zurich, Binzmuehlestrasse 14/17, CH-8050 Zurich, Switzerland. E-mail: c.killikelly@psychologie.uzh.ch

Abstract

Background: The new diagnosis of prolonged grief disorder (PGD) is both an opportunity and a challenge for researchers, clinicians, and bereaved individuals. The latest definition of PGD includes a refreshing and novel feature: the cultural caveat, i.e., clinicians must determine that the grief presentation is more severe and of longer duration than would be expected by an individual's culture and context. Currently, there are no guidelines on how to operationalize the cultural caveat in mental health care settings.

Method: To respond to this important demand we have developed, piloted, and tested the cultural supplement module of the International Prolonged Grief Disorder scale (IPGDS). The cultural supplement aims to provide clinicians with a catalogue of culturally relevant symptoms of grief that indicate probable PGD alongside a simple framework for cultural adaptation for use in specific clinical settings.

Results: In this short report we outline the rationale and aim of the cultural supplement and provide a summary of our latest validation studies of the IPGDS with bereaved German-speaking, Chinese and Swiss migrant individuals. We also provide a step-by-step framework for adaptation of the cultural supplement that clinicians and researchers may use when working with different cultural groups.

Conclusion: To date, this is the first PGD questionnaire based on the ICD-11, and the first to include a cultural supplement that can be adapted to different contexts and groups. This cultural supplement will provide clinicians and researchers an easy-to-use assessment tool with the aim to improve the global applicability of the ICD-11 PGD definition.



Keywords

prolonged grief disorder, ICD-11, International Prolonged Grief Disorder scale, cultural adaptation

Highlights

- We explore the role of culture and the new diagnosis of prolonged grief disorder.
- We provide the framework for a new method of cultural adaptation for a grief assessment measure.
- We summarize new research using this new method in different cultural groups around the world.
- We provide key recommendations for clinical practice.

Prolonged Grief Disorder

In 2022 the latest revision of the ICD-11 was implemented in clinical and research settings around the world. Prolonged grief disorder (PGD) is a new mental health disorder included in the ICD-11. The inclusion of grief as a mental disorder has been hailed as both an opportunity and a challenge for researchers, clinicians, and patients (Bryant, 2014; Killikelly & Maercker, 2017; Stelzer, Zhou, Merzhvynska, et al., 2020; Stroebe et al., 2008). In the latest iteration of the ICD-11 the WHO outlined a new remit for the structure and content for disorder definitions. A strong emphasis on clinical utility and global applicability was prioritized over further delineation of accessory symptoms and subtypes (Keeley et al., 2016). This led to the inclusion of refreshing new features in the diagnostic definition of PGD. The cultural caveat purports that for a diagnosis to be assigned, the symptoms of PGD must be more intense, more severe and of longer duration than would normally be expected for the individuals' cultural or religious context. This is an exciting and novel feature for a diagnostic definition. It holds the promise of a more inclusive, globally applicable classification system, that may improve diagnostic accuracy, therapeutic rapport and treatment outcomes (Aggarwal, 2013). However, the ICD-11 falls short of providing clear guidance on how to operationalize the cultural caveat. Questions remain about how to differentiate symptoms of 'normal' bereavement in different contexts (e.g., child loss, unnatural violent loss, ambiguous loss) alongside 'disordered' symptoms in different cultures around the world.

Historically the fields of culture, psychology, and psychiatry have only recently intersected to develop models and frameworks to explore the contribution of culture to psychopathology. Earlier in the history of psychiatry, it had been largely assumed that the symptoms of disorder expressed in North American and European populations were representative globally. Recent research has confirmed that the symptom content and structure, duration, chronicity and response to treatment can be highly dependent on culture (Kohrt et al., 2014; Nichter, 1981). This relativist view of disorder purports that the boundary between normal and abnormal is a social judgment or a social/cultural



norm and that the definition of abnormal will change depending on particular culture norms (Canino & Alegría, 2008). There are several examples of how disordered grief may manifest differently in different cultures. Unique culturally bound symptoms of PGD have been identified worldwide (Killikelly et al., 2018; Rosenblatt, 2008). For example, in traumatically bereaved Kurdish refugees one common expression of severe grief was to imitate the behaviours of the deceased (Hall et al., 2014), 52% of Cambodia refugees reported dreams of the deceased and this was associated with elevated PGD symptoms (Hinton et al., 2013), in Japan bereaved individuals will control their grief at funerals as they do not want to make others uncomfortable (Killikelly et al., 2022).

On the other hand, the universalist approach (or pan cultural approach) suggests that mental disorders have core symptoms of internal disorder however these symptoms may manifest differently in different contexts (Canino et al., 1997). A famous study on experiences of grief around the world examined the expression of emotion after bereavement in 78 cultures (Rosenblatt et al., 1976). They concluded that it is a basic human characteristic to react with emotions towards bereavement and for the majority of societies these emotions included crying, overt anger, and fear. In an early study comparing Dutch and Slovenian spouses who lost their partner due to unnatural causes it was found that there were more similarities than differences between cultures (Cleiren et al., 1996). Although in Slovenian people symptoms of depression were slightly higher the overall pattern was very similar. There is a gap in the research field, as currently there are no up to date studies that directly compare symptoms of PGD across cultures, particularly the latest ICD-11 definition of PGD. Additionally, there are no culturally adapted questionnaires or measures of prolonged grief disorder. It is therefore difficult to ascertain if PGD symptoms follow a relativist or universalist trend, especially without adequate assessment measures.

New research is consistently demonstrating the importance in cultural adaptation of mental health assessment interviews and questionnaires (Hall et al., 2016). However currently, there is discourse and debate in the field over the level of cultural adaptation that is required to successfully evaluate mental disorders in cultures outside of Europe and North America (Harper Shehadeh et al., 2016; Heim & Kohrt, 2019). There are currently two broad approaches to the development or adaptation of culturally sensitive mental health tools. The etic approach refers to a questionnaire or intervention developed outside of the culture whereas the emic approach refers to a questionnaire or intervention developed from within a culture (Triandis & Marin, 1983). Both of these approaches have been used in the cultural clinical psychology field to varying degrees (Heim et al., 2017; Killikelly et al., 2018; Rasmussen et al., 2014) and with conflicting results. Some clinicians and researchers argue that the etic approach is enough to provide a clear and valid understanding of disorder, while others argue that the evaluation must stem from within the culture in order to be valid (Aggarwal et al., 2014; Berry, 1969). Currently there is a dearth of both etic or emic approaches to assessment in the field of prolonged grief



diagnosis, assessment and treatment. Here we consider both the emic and etic approach in a new combined PGD assessment methodology.

IPGDS and Cultural Supplement

The International Prolonged Grief Disorder Scale (IPGDS) is a two-part assessment questionnaire. This questionnaire is unique as it includes both emic and etic methodology within one questionnaire. The first part is the 'standard scale'; a 14-item scale developed directly from the latest narrative definition of PGD from the ICD-11. This represents the etic approach. It contains two core items (longing or yearning for the deceased, preoccupation with the deceased), accessory items including examples of emotional pain, time and impairment criteria and the cultural caveat. The standard scale can be used to determine a preliminary diagnosis of PGD and is a clinical diagnostic tool. The second part of the IPGDS is the cultural supplement and uses an emic approach. The cultural supplement was developed from focus groups and key informant interviews from health care professionals and bereaved individuals from a range of cultural backgrounds. The aim was to collect a catalogue of possible PGD symptoms that may be culturally relevant above and beyond the standard ICD-11 items (Table 1). The cultural supplement is intended to provide a more in-depth assessment of possible PGD symptoms that may improve treatment decision making with clinical guidance. For example, recently a novel study of bereaved Balinese family members revealed a probable caseness of 0% for PGD, 1% for posttraumatic stress disorder and 2% for depression. These findings are striking as usually rates of PGD are expected to be at least around 1% and more commonly less than 10% of the population. The authors conclude that there are perhaps aspects of the Balinese culture that protect individuals from developing mental health disorders (Djelantik et al., 2021). However, another explanation could be that the scale used to measure PGD was not culturally adapted from within the population and instead only used at etic approach. Therefore, the scale may not have captured PGD symptoms that are most distressing or representative in this population. It was recently suggested that PGD assessment across cultures would benefit from the inclusion of both etic and emic methods within an assessment tool, such as provided by the IPGDS (Kokou-Kpolou, 2021). Below is a summary of our research exploring the development and first applications of this combined methodology using both the etic 'standard scale' and the emic cultural supplement of the IPGDS in different cultural groups.



 Table 1

 IPGDS Cultural Supplement Items: Developing a Catalogue Culturally Relevant of Grief Symptoms

Cultural Supplement item	German- speaking sample	Chinese sample	Arabic migrant sample
I experience strong physical problems since the loss (e.g., headache, problems with appetite).			\square
I would do anything to feel close to the deceased (e.g., visit their grave everyday, sleep next to their picture).	\square		If I could, would do anything to feel close to the deceased (e.g., visit their grave everyday, sleep next to their picture). (slightly reworded item)
Since the loss my behavior has changed drastically in an unhealthy direction (e.g., excessive alcohol consumption).	\square		\square
The loss shattered my trust in life or faith in God/a higher spiritual power.	\square		The loss shattered my beliefs (i.e. my understanding of how the world should work, spiritual beliefs, religious beliefs).
It is impossible for me to focus.			\checkmark
My grief is so intense that I feel stuck in grief.			\square
I just can't seem to fall back into a rhythm.			
I feel paralyzed and disconnected, (e.g., as if I am not in my own body).			
I have no energy or desire to engage in activities.			
This life holds no meaning since the death.			\square
I want to die to be with the deceased.			
I don't feel close to other people or feel no satisfaction when being around others.	abla		
I feel like I have completely lost control.			I feel like I have completely lost control over my life or over myself.
I am searching for the deceased with the hope to find him/her.			\square



Cultural Supplement item	German- speaking sample	Chinese sample	Arabic migrant sample
I constantly look back upon the past relationship.			
I feel so helpless since I lost him/her.			
I feel he/she is beside me.			
I cry loudly when I think of the loss.			
I can't trust others since the loss.			
I feel disconnected from the new society I live in (e.g. the country I move to).			
Without a funeral (body, or other burial			\square
ritual) I cannot move on with my life.			TBC
Not knowing what happened to them is			\checkmark
the worst part.			TBC
I would rather know they are dead then			\square
face this uncertainty.			TBC
If I were in my home country I would			
have more support for my grief.			TBC
I feel so overwhelmed with grief that I			
cannot deal with all the changes in my new country.			TBC
There are so many things to worry			
about in my new country that I never			TBC
have time to grieve.			
When I talk about the loss no one			
understands me.			TBC
I am grieving for multiple loved ones at			\checkmark
the same time.			TBC

Note. The first column represents the items of the IPGDS cultural supplement. The subsequent columns indicate from which cultural group the item was developed and validated. The items with TBC indicate that these need to be validated in a large sample.

The cultural supplement emerged from a bottom-up qualitative approach. Items were developed from key informant interviews with German-speaking and Chinese-speaking health care workers (Killikelly et al., 2020; Stelzer, Höltge, et al., 2020; Stelzer, Zhou,



Merzhvynska, et al., 2020), bereaved migrants and refugees (Killikelly et al., 2021) and Japanese health care professionals (Killikelly et al., 2022). Currently there are several versions of the cultural supplement that are being validated in different cultural groups. The Chinese version has been psychometrically validated in a sample of n=325 Chinese bereaved (Killikelly et al., 2020), the migrant cultural supplement (for bereaved migrant individuals living in a host country) has recently been validated in 121 bereaved migrants (in preparation). A Japanese version and a version for Arabic speaking refugees experiencing ambiguous loss are currently under development.

Summary of Recent Findings From the Implementation of IPGDS Cultural Supplement

Below we outlined how the cultural supplement of the IPGDS has been used to explore culturally relevant symptoms in different cultural groups. To date the cultural supplement has been used in two main ways 1) to compare and contrast a wide range of possible PGD symptoms between different cultural groups 2) to identify new culturally relevant symptoms of PGD within a cultural group or context. The earliest results from the implementation of the cultural supplement in the Chinese bereaved sample show the value of the supplement. Firstly, an item specific analysis revealed that certain items were endorsed more strongly in the Chinese sample when compared to the German speaking sample. For example, the most strongly endorsed item in the Chinese sample was Item 15 (I constantly look back upon the past relationship) whereas for the German speaking sample it was Item 17 (I feel he/she is beside me). Additionally overall scores on the cultural supplement were higher in the Chinese sample than the German speaking sample, possibly indicating the items were more culturally relevant for the Chinese sample, as expected (Killikelly et al., 2020). Our recent study explored PGD in Germanspeaking and Chinese samples using a network analysis. We confirmed the presence of a core network of PGD symptoms consisting of yearning and emotional distress in both Swiss and Chinese participants (Stelzer, Höltge, et al., 2020). However, when culturally relevant items were included in the network this improved the predictability of the network for the Chinese sample only, possibly indicating that the cultural supplement yielded a better fit. Important network differences also revealed a strong connection between Item 11 (wish to die to be with the deceased) and Item 14 (searching for the deceased) for Chinese participants that was not found for German-speaking participants. We concluded that separation distress is a particularly relevant therapeutic target for Chinese participants.

In our latest study, 121 migrants to Switzerland completed the standard scale of the IPGDS and the culturally adapted 'Migrant version' of the cultural supplement. This version of the cultural supplement was developed from focus groups and interviews with Syrian migrants. New items were developed based on these interviews (e.g., Item 4: The

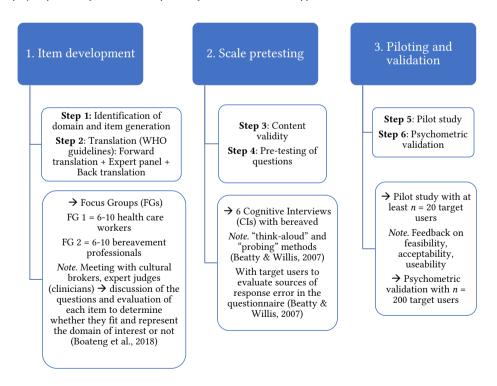


loss shattered my beliefs (i.e., my understanding on how the world should work, spiritual beliefs, religious beliefs), Item 13: I feel like I have completely lost control over my life or over myself and item: 19 I feel disconnected from the new society I live in (e.g. the Country I moved to). Each of these items must be answered in response to the loss of a loved one. To reduce the item list of the cultural supplement, a preliminary analysis of the response rates to each item revealed that the most endorsed items included Item 4 and Item 19. This potentially indicates that the inclusion of these culturally relevant items improved the sensitivity of the Migrant version of the cultural supplement.

Recommended Methods for Adaptation of the Cultural Supplement

Researchers and clinicians might be interested in developing a cultural supplement for the IPGDS based on their own community and context. In line with this we propose the following steps for adaptation (see Figure 1).

Figure 1
Step by Step Method for Cultural Adaptation of the IPGDS Cultural Supplement





However, first, in cross cultural research there are two important methodological caveats that should be transparently and forthrightly presented, particularly when comparing symptoms of mental disorder between different groups. First, the researchers definition of cultural group should be clearly stated and defined. Cultural group can be defined in different terms with a focus on different features (Ryder et al., 2011). In our research we define cultural group specifically in terms of the features that may intersect with mental disorder and PGD: a group of people who share a common language, regional history, beliefs, patterns of behaviour and values (National Center for Cultural Competence, 2001). This should be measured transparently and systematically through a simple questionnaire. Brief questions of cultural group and identity could be asked for example: what is your country of origin? Which culture influences you the most? How connected do you feel to Western culture? (Killikelly et al., 2020). Second, the effect of unmatched groups should be clearly presented to highlight any possible confounding factors. When possible, cultural groups should be matched in terms of characteristics that might affect the severity of PGD symptoms. For example, loss related characteristics should be similar across groups (e.g., relationship to the deceased, type of loss (natural or unnatural), time since loss). Demographic characteristics such as age, gender, and co-morbid mental health disorders should also be clearly documented.

The first step in the development of a new cultural supplement questionnaire is item development. A wide range of possible symptom items are gathered from cultural brokers or key experts such as clinicians or researchers that belong to the cultural group of interest and have key clinical knowledge. For example, 'free listing' is a technique used to elicit a large number of possible symptoms (Kumar, 1989; World Health Organisation, 2012). Focus groups may then reduce the number of items to the most highly endorsed and relevant. These items are then translated into the language of the group under study following the WHO's recommended translation process. The second step is scale pretesting (Boateng et al., 2018). The content validity of newly suggested symptoms can then be established via cognitive interviews with a representative sample of bereaved individuals (e.g., gender, age, type of loss, duration of loss all represented). For information on how to conduct cognitive interviews including the think aloud and probing technique see (Abi Ramia et al., 2018; Drennan, 2003; Prince, 2008). The aim of this step is to ensure that the format and nature of the questions are clear, concise and valid. Finally, the new questions should be piloted in a small sample of intended users. This can be followed by a larger scale psychometric validation study whereby standard psychometric properties of the scale (validity and reliability) are established (see Killikelly et al., 2020).

It should be noted that the cultural supplement should be used alongside the 'standard scale' i.e., the 14 items of the ICD-11 PGD definition. This is particularly important when establishing cross cultural prevalence rates, differences and similarities of symptom structure or establishing methodologically robust comparisons across different groups and contexts. The 'standard scale' provides the ICD-11 PGD symptom list and



could be used in clinical samples for diagnosis. This assumes the universalist approach to mental disorders. The cultural supplement can add to this list for purposes of exploring alternative PGD symptoms and supporting treatment planning. For example, somatic symptoms are not included in the ICD-11 PGD definition, however several different cultural groups have strongly endorsed physical symptoms following bereavement. After assessing with the IPGDS, a clinician may then offer interventions and techniques to alleviate somatic symptoms if these are most distressing symptoms indicated. These somatic symptoms may not be discovered without the wide range of questions covered by the cultural supplement.

Implications and Future Research

Since 2022 the new ICD-11 is used worldwide in clinics and research settings. Researchers are presented with a unique opportunity to document the impact of the inclusion of a new mental health disorder, PGD on patient experience and clinical outcomes. Additionally, the inclusion of the cultural caveat presents several challenges and opportunities. We invite clinicians and researchers to consider using the IPGDS and the cultural supplement in their clinical and research settings to add to the growing database of literature exploring prolonged grief disorder in different cultural contexts. There are many questions that remain unanswered about the relationship and importance of culture and mental disorder. One outstanding research question concerns the etiology of prolonged grief disorder and its' location on the universalist versus relativist spectrum. The development and testing of additional cultural supplements in different cultural groups worldwide could help identify which prolonged grief disorder symptoms are universal and which are culturally relative. One hypothesis could be that core symptoms of prolonged grief disorder are universal for example yearning and preoccupation with the deceased, while supplementary symptoms and examples of emotional distress may vary depending on cultural group. Although currently we recommend assigning a PGD diagnosis following the guidance of the IPGDS standard scale and the ICD-11 PGD definition, treatment planning may be enhanced with a person-specific approach. Therefore, the cultural supplement may be a valuable tool to improve clinician-patient relationship and treatment decision making. It is important to note that the cultural supplement is subject to some key limitations. For example, the definition of culture will be specific to the research group, the research question and the sampling method. Researchers and clinicians should provide thorough and transparent information on how they selected participants and the sampling method used to determine the cultural group or context. Additionally, it will be important to clearly document loss related variables, such as the type of loss, time since loss and the nature of the loss (sudden, violent etc.) as this may have a significant impact on the nature and severity of PGD symptoms particularly in different cultural contexts (Djelantik et al., 2020). In conclusion, the inclusion of PGD as



a mental health disorder opens the door for further robust, systematic research on the relationship between grief and culture.

Funding: The authors have no funding to report.

Acknowledgments: We would like to thank the participants for taking the time to complete our research surveys. We would like to thank the following Master's students for their work on this research programme: Alexandra Reymond, Olivia Gabban, Lea-Martina Christen, Hanzhang Xie.

Competing Interests: The authors have declared that no competing interests exist.

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Clinical Psychology in Europe (CPE) is the official journal of the European Association of Clinical Psychology and Psychological Treatment (EACLIPT).



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Announcements



Announcement of the Registered Report "Can a Variant of the Implicit Association Test Detect Nonsuicidal Self-Injury in a Clinical Population? A Registered Report"

Femke Cathelyn ^{1§} \bigcirc , Tilia Linthout ^{1§} \bigcirc , Pieter Van Dessel ¹ \bigcirc , Laurence Claes ^{2,3} \bigcirc , Jan De Houwer ¹ \bigcirc

[1] Department of Experimental Clinical and Health Psychology, Ghent University, Ghent, Belgium. [2] Faculty of Psychology and Educational Sciences, University of Leuven, Leuven, Belgium. [3] Faculty of Medicine and Health Sciences, University of Antwerp, Antwerp, Belgium.

Clinical Psychology in Europe, 2023, Vol. 5(1), Article e11499, https://doi.org/10.32872/cpe.11499

Published (VoR): 2023-03-31

Handling Editor: Cornelia Weise, Philipps-University of Marburg, Marburg, Germany

Corresponding Author: Tilia Linthout, Department of Experimental Clinical and Health Psychology, Ghent University, H. Dunantlaan 2, 9000 Ghent, Belgium. Tel: +32 479 19 31 25. E-mail: Tilia.Linthout@UGent.be

Editor's note: This is an announcement of a Registered Report which received In-Principal-Acceptance (IPA) to be published in "Clinical Psychology in Europe". The study protocol is publicly accessible at https://doi.org/10.23668/psycharchives.12576. In this announcement, a brief summary of the study protocol is presented.

Background

Nonsuicidal self-injury (NSSI) is a severe and prevalent mental health problem (Nock, 2010). Measures to detect which individuals are at risk for NSSI would be valuable for clinical practice. However, we still lack strong predictors of future NSSI behaviour, with the most notable exception being prior NSSI behaviour (Franklin et al., 2017; Griep & MacKinnon, 2022; Kiekens et al., 2018; Turner et al., 2013; Whitlock et al., 2013). Yet, the measurement of prior NSSI behaviour with self-report measures can be difficult because individuals may be motivated to conceal this harmful behaviour (Long, 2018; MacDonald et al., 2020; Simone & Hamza, 2020). To overcome this problem, an implicit measure has been developed that assesses automatic responding to statements about prior NSSI behaviour (i.e., the past nonsuicidal self-injury Implicit Association Test: P-NSSI-IAT;



[§]These authors contributed equally to this work.

Cathelyn et al., 2021). Previous studies tested the predictive utility of this measure in online studies with samples of at risk participants and produced promising results (Franklin et al., 2017; Sohn et al., 2021).

Aims

The main aim of this study is to validate the P-NSSI-IAT by assessing its ability to detect prior NSSI behaviour in a sample of clinical patients.

Method

We will target patients who receive outpatient treatment for various conditions. Participants will first complete the P-NSSI-IAT. Next, they will be asked how many times they have intentionally cut or carved their skin without intending to kill themselves in the past twelve months and the past 30 days and how likely they would be to intentionally cut or carve their skin without intending to kill themselves in the future.

Discussion

The registered study is the first to examine the clinical utility of a new implicit measure for prior NSSI behaviour (the P-NSSI-IAT). It will provide an answer to the question whether the P-NSSI-IAT allows detection of self-rated prior NSSI and future likelihood of NSSI in a sample of clinical patients.

Funding: This manuscript is supported by Ghent University grant BOF16/MET V/002 to JDH.

Acknowledgments: The authors have no additional (i.e., non-financial) support to report.

Competing Interests: The authors have declared that no competing interests exist.

Supplementary Materials

The study protocol for this Registered Report is publicly accessible via PsychArchives.org (see Index of Supplementary Materials below).

Index of Supplementary Materials

Cathelyn, F., Linthout, T., Van Dessel, P., Claes, L., & De Houwer, J. (2023). Supplementary materials to "Announcement of the Registered Report "Can a variant of the Implicit Association Test detect nonsuicidal self-injury in a clinical population? A Registered Report" [Pre-registration protocol]. PsychArchives. https://doi.org/10.23668/psycharchives.12576



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