

The relationship between parental catastrophizing about child pain and distress in response to medical procedures in the context of childhood cancer treatment: A longitudinal analysis.

Caes Line, PhD^{1,*}, Goubert Liesbet, PhD², Devos Patricia, MA³, Verlooy Joris, MD³, Benoit Yves, PhD⁴ & Vervoort Tine, PhD²

¹Centre for Pediatric Pain Research, IWK Health Centre, Halifax, Nova Scotia, Canada

²Department of Experimental-Clinical and Health Psychology, Ghent University, Belgium

³Department of Pediatric Oncology/Hematology and Stem Cell Transplantation, Ghent University Hospital, Ghent, Belgium

⁴Department of Pediatric Oncology/Hematology and Stem Cell Transplantation, Ghent University Hospital and Ghent University, Ghent, Belgium

* Corresponding author: Line Caes, Centre for Pediatric Pain Research, IWK Health Centre, 5850/5980 University Avenue, P.O. Box 9700, Halifax, NS, B3K 6R8, Canada. Tel: 01 (902) 470 6759; Fax: 01 (902) 470-7118. Electronic mail may be sent to line.caes@iwk.nshealth.ca

Number of text pages: 25

Number of tables: 1

Number of figures: 3

Keywords: Pain, Parent Stress, Children, Longitudinal Research, Hematology

FUNDING:

Line Caes is currently a postdoctoral fellow funded by the Louise & Alan Edwards Post-Doctoral Fellowship in Pediatric Pain Research. At the time that this research was conducted, Line Caes was supported by the Fund for Scientific Research – Flanders (Belgium) (F.W.O.) as an Aspirant fellow. Tine Vervoort is a postdoctoral researcher funded by the F.W.O.

ACKNOWLEDGMENTS:

Line Caes and Tine Vervoort are both trainee members of Pain in Child Health: A CIHR Strategic Training Initiative. The authors would like to thank Julie Vermeirsch, Sophie Vandenbroucke, Katrien Apers, Ellen Van Geertsom, Geertrui Berghmans, Chloë Noppe, Marieke Van Schoors, Jozefien Vanhaeren, Lisa Roosen and Sarah Kindt for their help with the data collection and/or input of the data. Furthermore, we want to thank the research nurses, Véronique Van de Velde and Sophie Van Lancker, for their help with planning the studies and practical issues of incorporating the study within the daily operation of University Hospital Ghent. The authors also thank the children and parents who took part in this study. There are no conflicts of interest that may arise as a result of the research presented in this article.

The relationship between parental catastrophizing about child pain and distress in response to medical procedures in the context of childhood cancer treatment: A longitudinal analysis.

ABSTRACT

Objective: Children with leukemia frequently undergo invasive medical procedures, such as lumbar punctures (LP) and bone marrow aspirations (BMA). To date, cross-sectional evidence indicates that LP/BMA procedures continue to elicit distress over the course of treatment in children and parents. Methods: The current study used prospective analyses investigating, in 28 children diagnosed with leukemia, the course of parental and child distress when confronted with consecutive LP/BMA procedures and potential moderation by catastrophic thinking. Parent's level of catastrophic thoughts was assessed before the first treatment-related LP/BMA, while child and parent distress was reported on after each LP/BMA procedure. Results: Whereas parental distress decreased over time among low catastrophizing parents, LP/BMA procedures remained highly distressing for high catastrophizing parents. Child distress during LP/BMA procedures increased over time and was positively related with parental distress. Conclusion: These findings stress the importance of targeting child and parent distress as early as possible in treatment.

OBJECTIVE

Childhood cancer, with leukemia as the most common form, is a major health problem affecting not only the child but also their parents. Specifically, evidence has shown that a significant number of parents exhibit high levels of stress and uncertainty immediately following the diagnosis of child cancer (Kazak, 2005) and particularly during the subsequent treatment process (Fedele et al., 2013; Pöder, Ljungman, & von Essen, 2010). Specifically, repeated painful invasive procedures, such as lumbar punctures (LP) and bone marrow aspirations (BMA), have been identified as a major stressor for both the child and his/her parents (Conte, Walco, Sterling, Engel, & Kuppenheimer, 1999; Ljungman, Gordh, Sörensen, & Krueger, 2000). While evidence indicates that cancer-related pain in general and procedure-related pain (e.g., needle pricks, subcutaneous and intra-muscular injections) and associated parental distress responses in particular tend to be less problematic towards the end of treatment (Ljungman et al., 2000; Pöder et al, 2010), this does not seem to be the case for the experience of parental distress associated with LP/BMA procedures (Katz, Kellerman, & Siegel, 1980; Kazak et al., 1995). Specifically, parental distress in response to LP/BMA procedures was found to be equally high 21 months after diagnosis compared to 10 months after diagnosis, suggesting that parents may not habituate to LP/BMA procedures (Kazak et al., 1995). Due to the bidirectional nature of the parent-child relationship, persistent parental distress may, in turn, have a negative impact on their child's as well as their own adjustment to the treatment process (Fedele et al., 2013). Indeed, previous research has consistently demonstrated a strong relationship between parents' and child's experience of distress in response to medical procedures (Dahlquist, Power, Cox, & Fernabach, 1994; Jay et al., 1983; Kazak et al., 1995). This has often lead health care practitioners/hospitals preventing parents from being present during child medical procedures

despite preliminary evidence indicating that parents prefer to be given a choice whether or not to be present during medical procedures (Boudreaux, Franics, & Loyacano, 2002; Franck et al., 2004).

However, available research is not without limitations and there are a number of important gaps in the literature that remain to be addressed. While previous studies suggest that parental distress in the context of LP/BMA procedures remains substantially present throughout the treatment, these findings are limited by their cross-sectional nature (Katz et al, 1980; Jay et al., 1983; Kazak et al., 1995). Consequently, little is known on how families adjust to the treatment process *over time*. Furthermore, it is unclear which factors contribute to potentially persistent levels of parental distress in response to consecutive, invasive medical procedures such as LP/BMA procedures. A better understanding of the contributing factors might provide enhanced and early identification of parents at risk for persistent levels of distress throughout treatment. This is particularly important among parents of a child diagnosed with leukemia given earlier findings indicating persistent distress might be likely (Kazak, 2005). In accordance with individual differences on the role of perceived threat in the attenuation of general stress symptoms in response to childhood cancer (see e.g., Barakat et al., 1997), the extent to which parents perceive their child's LP/BMA procedures as threatening (i.e., having catastrophic thoughts) may influence how parental distress responses in the context of LP/BMA procedures evolve over time. Specifically, previous findings in parents of healthy schoolchildren and children with chronic pain indicated that parental catastrophic thoughts about child pain contribute to elevated feelings of distress and protective behavior in response to child pain (Caes, Vervoort, Eccleston, Vandenhende, & Goubert, 2011). However, whether parental catastrophizing contributes to persistence of distress when parents are confronted with multiple,

consecutive painful procedures in their child and how this affects their behavior (e.g., preference to be present) remains as yet to be addressed.

Accordingly, the current study, in children diagnosed with leukemia, employed a prospective design to investigate how parental distress in the context of child LP/BMA procedures evolves over the course of consecutive procedures throughout the child's treatment as a function of parental catastrophic thoughts about child procedural pain and whether parental distress persistence is associated with child distress over time. Within the present study, parents were, as part of the Hospital standard protocol, allowed to be present in the treatment room pre- and post-procedure phase but not *during* the actual LP/BMA procedure. Based upon empirical literature, we hypothesized that (1) parental distress would be persistent over time, particularly among parents endorsing high levels of catastrophic thinking about child pain and less so among parents with low levels of catastrophizing and that (2) parental and child distress would be positively associated over time. Additionally, in light of preliminary findings showing that most parents want to be present during medical procedures (see e.g., Boudreaux et al., 2002; Franck et al., 2004), yet were due to standard hospital procedure not allowed to within the present study, we also explored parental desire to be present during LP/BMA procedures and how this desire evolves over time and relates to catastrophizing.

METHOD

Participants

The present study reports on longitudinal data collected in the context of the "XX-study" that aimed at investigating the role of parental catastrophic thinking in understanding parental and child responses to invasive medical procedures during pediatric cancer treatment. Cross-sectional data describing XX *-to conform to double blind peer review procedure we have omitted*

the description of this data in the current version of the manuscript-have been reported elsewhere (XX). The present study reports on a unique phase of the study designed to prospectively investigate the associations between parental catastrophic thinking and parent and child level of distress. Participants were children recently diagnosed with leukemia at XX Hospital and their parents. Families were excluded 1) if they did not speak and write Dutch, 2) if their child had any pre-existing developmental delay or 3) had relapsed. Additionally, when children had received a bone marrow transplantation during their treatment, they were also excluded from further participation. Of the 38 families invited to participate in this longitudinal part of the XX-study, six families refused participation (response rate: 84.21%, 16 boys; 16 girls). Main reason for non-participation was being overwhelmed with the diagnosis. Further, four families were excluded due to non-completion of the questionnaires ($N=1$), parental absence during most of the LP/BMA procedures ($N=1$), child bone marrow transplantation early in treatment ($N=1$) or LP/BMA procedures not being performed in accordance with the standard protocol (i.e., the child being sedated during the LP/BMA procedures; $N=1$). The final sample size consisted of 28 participating families (15 boys, 13 girls). Most children were diagnosed with Acute Lymphoblastic Leukemia (ALL, $N=22$). The remainder of the children were diagnosed with Acute Myelogenous Leukemia (AML). For most families ($N=18$), both parents agreed to participate in the study, yet in most circumstances only one of the parents (i.e., mostly the mother) attended the preparations and aftercare of the LP/BMA procedure. Accordingly, for most procedures we obtained data from either the mother or the father (see Descriptives for more details). For the remainder of the children ($N=10$) only the mother provided consent to participate. The mean age of the children was 6.59 years ($SD=4.29$, range=0.6-15). None of the children were able to attend school during the period of the study. Mothers' mean age was 36.11

years ($SD=5.76$, range=23-47). The mean age of the participating fathers was 39.81 years ($SD=5.48$; range=32-50). Most parents were married or co-habiting (85.7%) and about half of them had received education beyond the age of 18 years (fathers; 50%, mothers; 59.3%). All participating children and parents were Caucasian of which 96.4% ($N=27$) had the Belgian and 3.6% ($N=1$) the Dutch nationality. Ethical approval was obtained from the Ethics Committee of XX Hospital.

Procedure

Families who met the inclusion criteria, and who were at least three days post being diagnosed but prior to the first treatment-related LP/BMA procedure (i.e., occurring approximately eight days after being diagnosed) were consecutively invited to participate in the present study. Consequently, participating children had received prior LP/BMA procedures but only for diagnostic purposes. Families were informed about the aim of the study (i.e., investigating factors influencing parental and child distress during treatment-related LP/BMA procedures). It was also emphasized that they were able to withdraw participation at any time. When families agreed to participate, children (older than 12 years) and their parents provided written consent.

Each LP/BMA procedure the child had to undergo as part of his/her *intensive* treatment (i.e., *induction* and *consolidation* phase in which the child receives intensive chemotherapy requiring frequent hospitalization) was consecutively included in the study. LP/BMA procedures to determine the diagnosis or performed during the *maintenance* phase of the child's treatment (i.e., involving less intense chemotherapy and less frequent hospitalizations) were excluded. Following standard clinical protocol of XX Hospital, an aneutectic mixture of local anesthetics lidocain and prilocaine is applied to the child's skin approximately one hour before the LP/BMA

procedure (Young, Schwartz, & Sheridan, 1996). During the LP/BMA procedure, children also receive a mixture of nitrogen peroxide-oxygen (i.e., 50% nitrous oxide/50% oxygen), which is a colorless gas that tranquilizes, but not anesthetizes the child and has an analgesic impact on the skin (Reinoso-Barbero et al., 2011). The LP/BMA procedure starts after inhalation of this mixture through a facial mask for at least 3 minutes. For most LP/BMA procedures, three staff members are present: a physician, a nurse, and a child life specialist. At the time of the study, the department employed three child life specialists. At the start of treatment, the child is allocated to one of the three child life specialists, who accompanies/supports the child during each painful medical procedure **and** administers the nitrogen peroxide-oxygen. Prior to the LP/BMA procedure, the child life specialist also explains the procedural aspects to the child and parents. Parents do not receive specific instructions on how to support their child in coping with these procedures. If the responsible child life specialist was not available at the time of the LP/BMA procedure, another child life specialist was briefed and attended the procedure. As part of XX Hospital standard protocol, parents are allowed in the treatment room during pre- and post-procedure phase but are asked to wait outside during the actual LP/BMA procedure.

All participating parents were requested to report on their catastrophic thoughts about child procedural pain before the first treatment-related LP/BMA procedure took place (i.e., the first LP/BMA procedure included in the study). Parents thus reported on their level of catastrophizing after their child had already received at least one LP/BMA procedure to confirm the diagnosis of leukemia. The first author (XX) or one of six research assistants was present in the treatment room pre-, during and post-procedure in order to record the duration of the LP/BMA procedure. Importantly, the researcher kept the interaction with the staff, parents and child during the different phases of the procedure to a minimum. After each LP/BMA procedure

the attending parent was requested, once the child and parent had returned to the child's room, to report on his/her felt distress and desire to be present during the procedure. Furthermore, since a large proportion of the children were too young to provide self-reports (i.e., 32% of the children ($N=9$) were younger than four years; Stinson, Kavanagh, Yamada, Gill, & Stevens, 2006), the child life specialist attending the procedure was asked to rate the child's distress during each LP/BMA procedure. The study protocol is available upon request from the first author.

Measures

Parental distress during LP/BMA procedures.

After each LP/BMA procedure, parents were asked to indicate how much distress they had experienced during the LP/BMA procedure. Based upon the work of Batson, Fultz, & Schoenrade (1987), parental distress was indexed by means of parental ratings of four emotion adjectives reflecting emotional distress ('worried', 'upset', 'anxious', 'sad') rated on an 11-point scale ranging from "not at all" (0) to "extremely" (10). A mean score of parental distress was calculated ranging from 0 to 10. The use of emotional adjectives is a reliable, valid, short and easy to complete methodology, which lends itself well to adapt in order to assess distress in response to a specific situation, such as a LP/BMA procedures (Caes et al., 2011). Reliability within the current study was excellent ($\alpha=.99$).

Parental desire to be present

After each LP/BMA procedure, parents were also requested to rate to what extent they wanted to be with their child during the LP/BMA procedure ("*To what extent did you want to be with your child during the LP/BMA procedure?*") using an 11-point scale ranging from "not at all" (0) to "a lot" (10).

Parental catastrophizing thoughts about their child's procedural pain.

Parents' catastrophic thoughts about their child's procedural pain were assessed with a state measure of the Pain Catastrophizing Scale for Parents (PCS-P-state; Goubert, Vervoort, Cano, & Crombez, 2009), which, compared with the trait measure of parental catastrophic thinking, is likely to be more compatible with the situation under investigation and thereby may have higher predictive value. Parents completed the PCS-P-state only once, prior to the first treatment-related LP/BMA procedure. In line with previous studies (see e.g. Caes et al., 2011), the state version of the PCS-P comprised one adapted item from each subscale of the PCS-P assessing catastrophizing thoughts about LP/BMA-related pain. (Rumination: "to what extent did you keep thinking about how painful the LP/BMA procedure is for your child?"; Magnification: to what extent did you think that, because of the pain, something serious might happen to your child?"; Helplessness: "to what extent did you think, that because of the pain of your child, you would not be able to stand the LP/BMA procedure?"). Parents were instructed to indicate, using an 11-point numerical rating scale (0=not at all; 10=a lot), to what extent they had experienced these thoughts during their child's previous LP/BMA procedure (i.e., the LP/BMA procedure performed to confirm the diagnosis of ALL or AML). A mean score of these three items was calculated, ranging from 0 to 10. Cronbach's alpha was .83 for mothers and .89 for fathers.

Child distress during the LP/BMA procedures.

The child life specialist was, after each LP/BMA procedure, requested to rate the child's distress ("*Specify how anxious you think the child was during the LP/BMA procedure*") using an 11-point rating scale ranging from 0 (not at all) to 10 (a lot).

Data analysis

The data of the present study are hierarchically nested. Specifically, parental/child distress and desire to be present during each LP/BMA procedure (level one) are nested within

individuals (parents/child; level two), which are in turn nested within couples (mother and father of a particular child; level three). Therefore, the data were analyzed by means of multilevel modeling (HLM version 6.01, Raudenbush, Bryk, & Congdon, 2004) as this method allows more precise parameter estimates compared to traditional statistical methods, such as repeated-measures analyses of variance (Kenny, Kashy, & Cook, 2006; Nezlek, 2001) and has the ability to handle missing data (Hox, 2010). Moreover, with this technique we were able to identify a mother and father of the same child as different participants while taking into account that they form a dyad as parents of the same child (Kenny et al., 2006). If only one parent attended the LP/BMA procedure, the data-point for that procedure was considered as missing data for the non-attending parent.

The following set of analyses was performed with 1) parental distress and 2) parental desire to be present as dependent variables. In a first step, a baseline model, without any predictors except “time” (i.e., the number of the LP/BMA procedure, e.g., the first=0, second=1, third=2, etc.), was run to calculate the level of variance in the dependent variables accounted for by the variables between dyads (level three) and within dyads (level two, between parents of the same dyad and level one, within parents). In the second step, we controlled for the duration of the LP/BMA procedure and child level of distress by including these variables in the first level. Third, parent sex, PCS-P state, and the interaction terms parent sex x PCS-P state, time x parent sex and time x PCS-P state were entered into the model as level 2 variables. Lastly, in order to control for the impact of child age, sex and type of diagnosis (AML or ALL), these variables were added to the third level. A similar analysis was performed with child distress as a dependent variable without adding parental catastrophic thinking and its interactions terms to the analysis. Full maximum likelihood estimation was applied and for each step in this build-up

strategy the likelihood ratio deviance test was used to determine whether the variables at each level were retained, using $p < .05$ as criteria. The likelihood ratio deviance test compares the deviance, or lack of fit between model and data, of two models to determine the best-fitting model. In addition, the final, most parsimonious model was determined by using the likelihood ratio deviance test to compare the best-fitting model derived from the build-up strategy with and without the non-significant control variables (i.e., t-test with $p < .05$) included in the model. To further obtain the most parsimonious model, the slopes of first level variables were fixed if the random error term was non-significant ($p < .05$; Nezlek, 2001). The slopes for the effect of the first and second level variables were fixed on the third level because dyads do not have enough lower-level units to allow for the slopes to vary (Kenny et al., 2006). All continuous variables were standardized and grand mean centered to allow comparison across parents and more coherent interpretations of the coefficients. The effect size r , with $r = .10$ a small effect, $r = .30$ a medium effect and $r = .50$ a large effect, was calculated for all significant effects (Kenny et al. 2006).

RESULTS

Descriptive statistics

A total of 242 LP/BMA procedures were observed with 10 LP/BMA procedures (range=4-14) on average over the course of the child's intensive treatment. Lumbar punctures were most frequent (60.1%), followed by a bone marrow aspiration (27.7%) and both procedures consecutively (6.1%). Mean duration of LP/BMA procedure was 8.12 minutes ($SD=5.80$, range=1.58- 36). On average, 62.1% of the LP/BMA procedures were attended by mothers, 15.2% by fathers and in 7.9% of the procedures both parents attended the procedure. Level of parental catastrophizing about child procedural pain (PCS-P-state) for fathers ($M=5.07$;

$SD_{\text{fathers}}=2.44$, range=1-9) and mothers ($M=4.40$; $SD_{\text{mothers}}=2.90$, range=0-10) was rather moderate and did not significantly differ from each other ($t(21)=1.26$, ns). The average level of distress experienced during LP/BMA procedures was 4.21 ($SD=2.57$, range=0-10) for parents and 4.10 ($SD=2.47$, range=0-10) for children. Parents expressed a relatively high desire to be present during the procedures ($M=7.24$, $SD=2.78$, range=0-10).

The influence of parental catastrophic thinking on parental feelings of distress

The intercept model indicated that 21.74% of the variance in parental distress was accounted for by variables on the third level (between parent-child dyads, i.e., child characteristics), 35.20% by variables on the second level (within parent-child dyads; i.e., parent characteristics) and 43.05% by first level variables (within parents, procedure-related characteristics). Adding the first and second level variables yielded significant likelihood ratio tests (model with first level variables vs. intercept model: $\chi^2(2)=89.17$, $p<.001$; model including first and second level variables vs. model only including first level variables: $\chi^2(5)=20.56$, $p<.001$), while including the third level variables yielded a non-significant likelihood ratio test ($\chi^2(3)=3.48$, $p=.32$). Consequently, only the first and second level variables were retained. The model excluding non-significant control variables (duration of procedure, parent sex, PCS-P state x parent sex and time x parent sex) revealed a significant likelihood ratio test when compared to the model including all first and second level variables ($\chi^2(4)=25.79$, $p<.001$). Therefore these variables were excluded from the final, best-fitting model. The final model revealed a significant negative effect of time ($\gamma_{100}=-0.08$, $t(167)=-2.85$, $p<.01$, $r=.16$) and a positive effect of parental catastrophic thinking ($\gamma_{020}=0.78$, $t(21)=2.75$, $p<.05$, $r=.41$). However, results also indicated a significant time x parental state catastrophizing interaction ($\gamma_{120}=0.07$, $t(167)=2.22$, $p<.05$, $r=.13$), indicating that whereas parents with low levels of catastrophic

thoughts show a slight decrease in their levels of distress over time, feelings of distress remain high throughout treatment among parents highly catastrophizing about their child's pain (see Figure 1). Furthermore, child distress ($\gamma_{300}=.17$, $t(167)=3.45$, $p<.001$, $r=.19$) made a significant contribution in explaining parental distress during LP/BMA procedures, indicating that child and parental distress are positively interrelated. Results for the final model are presented in Table 1.

The influence of parental catastrophic thinking on parental desire to be present

Large proportions of variance in parental desire to be present were explained by all levels in the model, specifically level 3 (child characteristic) explained 42.68%; level 2 (parental characteristics) 30.23% and level 1 (within parents or procedure-related characteristics) explained 27.08%. Similar as to the model for parental distress, only adding the first and second level variables yielded significant likelihood ratio tests (model with first level variables vs. intercept model: $\chi^2(4)=54.01$, $p<.001$; model including first and second level variables vs. model only including first level variables: $\chi^2(5)=15.74$, $p<.01$), thereby excluding the third level variables from the model ($\chi^2(3)=1.54$, $p>.50$). The model excluding non-significant control variables (child distress and parent sex) also fitted the data better compared to the model including all first and second level variables ($\chi^2(3)=47.21$, $p<.001$); therefore these variables were excluded from the final model. Analyses with the final model indicated a significant time x parental catastrophic thoughts about child procedural pain interaction ($\gamma_{120}=0.10$, $t(18)=2.36$, $p<.05$, $r=.41$; see Table 1). Specifically, parents reporting low levels of catastrophic thinking reported a decrease in their desire to be present during LP/BMA procedures. Conversely, parents endorsing high levels of catastrophic thoughts about child procedural pain during LP/BMA procedures showed an increase over time (see Figure 2). Furthermore, the duration of the procedure showed a significant, positive association with parental desire to be present

($\gamma_{200}=0.001$, $t(124)=2.34$, $p<.05$, $r=.22$), indicating that the longer the procedure took the higher parents' desire to be present during the procedure.

The course of child distress

Analyses indicated that a large proportion of the variance in child distress was due to child (50.63%) and parental characteristics (49.36%). Only 0.01% variance was accounted for by the variables on the first level (within parents or procedure-related variables). Significant likelihood ratio tests were found when adding the first and second level variables (model with first level variables vs. intercept model: $\chi^2(11)=910.36$, $p<.001$; model including first and second level variables vs. model only including first level variables: $\chi^2(2)=8.11$, $p<.05$). However, adding the third level variables revealed a non-significant likelihood ratio test ($\chi^2(3)=2.76$, $p>.50$), therefore these variables were excluded from the model. The model excluding non-significant control variables (duration of procedure, parent sex and time x parents' sex) fitted the data better compared to the model including all first and second level variables ($\chi^2(10)=60.26$, $p<.001$), therefore these variables were excluded from the final, best-fitting model. Findings revealed a significant effect of time ($\gamma_{100}=0.09$, $t(119)=2.07$, $p<.05$, $r=.19$), indicating increasing levels of child distress during LP/BMA procedures (as reported by the child life specialist) with increasing number of procedures undergone. **Parental level of distress also showed a significant contribution indicating a positive association between parental and child distress experiences** ($\gamma_{300}=0.17$, $t(27)=2.25$, $p<.05$, $r=.44$). Results of the final model are presented in Table 1.

CONCLUSION

The present study investigated, in a sample of children with leukemia and their parents, whether 1) parental distress during LP/BMA procedures persists over time, particularly among parents endorsing high levels of catastrophic thinking about child pain and whether (2) high

levels of parental distress are associated with increased levels of child distress. Additionally, we also explored the preference of parents to remain present during the LP/BMA procedures. The results of the present study were partially in line with expectations. First, findings indicated that parental distress in response to LP/BMA procedures changed over time depending upon parental catastrophizing about child procedural pain. Specifically, parental distress gradually decreased over the course of consecutive LP/BMA procedures among parents with low catastrophic thoughts but remained equally high among parents endorsing high levels of catastrophic thinking. A comparable pattern for high versus low catastrophic thinking was apparent for parental desire to be present during LP/BMA procedures. Further, child distress (as reported by the child life specialist) increased over the course of multiple LP/BMA procedures and was positively associated with parental distress.

The current findings are consistent with prior research in parents of healthy schoolchildren and chronic pain samples (Caes et al., 2011) yet extend the earlier results in several ways. First, to our knowledge, this study is the first to prospectively investigate the role of parental pain catastrophizing in the context of consecutive, invasive medical procedures. Second, while exploratory, the present study also provides insight into parental desire to be present during invasive medical procedures, how this evolves over time and relates to parental catastrophic thinking about child pain.

Findings indicated that parental catastrophizing about child pain influences how parents adapt to frequent, consecutively occurring painful medical procedures in their child. Specifically, whereas LP/BMA-related parental distress decreased over time among parents with low levels of catastrophic thinking, LP/BMA procedures remained equally distressing for parents with high levels of catastrophizing. While further research is needed, one potential explanation is that pain

catastrophizing affects emotion regulation capacity. Possibly, parents with low levels of catastrophic thinking are able to adopt adequate strategies to regulate distress in response to these consecutive, invasive procedures. A well-documented emotion regulation strategy in the context of pain is distraction, which has found to be associated with decreased pain aversiveness and increased pain tolerance (see e.g., Malloy & Milling, 2010). However, both adult and child literature suggests that distraction from pain is particularly effective among individuals with low levels of catastrophic thinking but may be counterproductive for individuals who endorse high levels of catastrophic thinking (Van Damme, Crombez, Nieuwenborgh - Van De Wever, & Goubert, 2008; Verhoeven, Goubert, Jaaniste, Van Ryckeghem, & Crombez 2012; Vervoort et al., 2011). Applied to our findings, it is plausible that only parents low in catastrophic thinking successfully employed distraction to regulate distress in response to the LP/BMA procedures.

While the proposed interpretation on effective use of distraction is speculative at present and requires further empirical investigation, our findings on parental desire to be present may also be interpreted in this light. Specifically, parental absence during invasive medical procedures such as LP/BMA procedures – which is often standard practice (Franck et al., 2004) – may allow parents to distract from child pain. Such explanation may account for our finding that desire to be present, like parental distress, decreased among parents with low levels of catastrophic thinking (i.e., for whom distraction likely works), but increased among parents endorsing catastrophic thoughts about their child's pain (i.e., for whom distraction is likely to be counterproductive). Yet, as our findings on parental presence are limited to one question, future research is needed on how best to ascertain parental preference to be present. **Furthermore, more research on factors influencing parental desire to be present**, when parental presence is less or more beneficial both for the parent as well as the child and why this is the case **is warranted**

(Boudreaux, Franics, & Loyacano, 2002; Franck et al., 2004). For example, parents' own experiences and discomfort with medical procedures might downsize parental desire to be present during their child's procedures and the beneficial effect of parental presence.

Interestingly, our findings also indicated that children do not habituate to the distressing experience of LP/BMA procedures. On the contrary, findings showed that children were perceived by the child life specialist as becoming increasingly distressed over time. Comparable, but cross-sectional findings have been reported by Katz and colleagues (1980) and Kazak and colleagues (1995), indicating that child anxiety and discomfort do not spontaneously diminish with repeated exposure. Furthermore, we found that parental and child distress were positively interrelated over the course of treatment. However our data do not allow for any conclusion on the direction of this association. Due to reporting on their distress experience after the post-procedure phase, parental ratings of their distress experience during the LP/BMA procedure might be influenced by their observation of child's level of distress during the post-procedural phase. Nevertheless, the absence of parents during the LP/BMA procedure does not preclude an influence on their child's responses. Heightened distress among parents with high catastrophic thoughts might also be apparent in anticipation of the procedure (Caes, Vervoort, Trost, & Goubert, 2012), thereby influencing subsequent child distress during the procedure, even in the absence of parents. Although more research is needed on the direction of this association, these findings further emphasize the importance of targeting both child as well as parent distress in early stages of treatment. To date, a lot of progress has been made in optimizing pharmacological as well as non-pharmacological interventions to reduce the child's pain and fear during painful medical procedures (Blount et al., 2009; Conte et al., 1999). However, it is as yet unclear how to assist parents in effectively coping with painful medical procedures nor is it standard practice to

do so (Chambers, 2003). Targeting parental catastrophizing by means of cognitive behavioral therapy (CBT) based interventions such as functional analysis and cognitive restructuring (Williams, Eccleston, & Morley, 2012) might prevent development of severe parental and associated child distress during these procedures over time (Dahlquist et al., 1994; Fedeles et al., 2013; Kazak, 2005). Alternatively, it may also prove functional to provide parents with adequate strategies to attenuate induced distress without targeting catastrophizing per se. Mindfulness and acceptance based approaches, which increase awareness of catastrophic thoughts and associated feelings of distress without judging but instead, accepting these inner experiences and thoughts, might constitute a potential pathway to cope with catastrophic thoughts about child pain and associated distress experience (McCracken & Gauntlett-Gilbert, 2011; Schütze, Rees, Preece, & Schütze, 2010).

A number of limitations deserve consideration, each of which point to directions for future research. First, the study sample was small and recruited in only one hospital, utilizing moderate sedation for LP/BMA procedures. Consequently, we may not have been able to detect small effects and generalization of the results, especially to situations where general sedation is used for LP/BMA procedures, might be limited. Related to this issue is the wide age range of our participants. Although child age did not impact the current findings, further investigation in different age groups is needed to explore similarities and differences across development. Second, as many children were too young to provide self-reports, the child life specialists provided ratings on child distress. Consequently, the results need to be interpreted with caution, as the longitudinal analyses might have been limited due to multiple persons providing ratings at different points in time. *Although the briefing they received prior to the procedure may have influenced the observation of child distress by child life specialists unfamiliar to the child, it*

would have been unethical to eliminate this briefing procedure. Future research is needed to replicate the current findings and investigate whether similar associations can be found when using other measures including child self-reported and observational assessment of child distress. Third, staff members' behavior during the LP/BMA procedures may have impacted child distress experience. Yet, this influence was not taken into account. Further, since parental state levels of catastrophic thoughts were assessed after the LP/BMA procedure to determine the diagnosis, child distress during this LP/BMA procedure might have influenced parents' report of catastrophic thinking. Future research might benefit from including assessment of parental catastrophic thinking before any LP/BMA procedures have taken place. Finally, although only six families declined study participation, the main reason for non-participation was being too overwhelmed with the diagnosis. Consequently, a self-selection of parents with high levels of distress might have taken place. These limitations notwithstanding, the current findings attest to the importance of assessing parental pain catastrophizing in the context of repeated painful LP/BMA procedure in order to advance understanding of associated parent and child distress. Future research is needed to replicate and explore alternative explanations suggested by the current findings.

FIGURE LEGEND

Figure 1: Impact of time and parental catastrophic thoughts on parental distress.

Figure 2: Impact of time and parental catastrophic thoughts on parental desire to be present.

REFERENCES

- Barakat, L.P., Kazak, A.E., Meadows, A.T., Casey, R., Meeske, K., & Stuber, M.L. (1997). Families surviving childhood cancer: a comparison of posttraumatic stress symptoms with families of healthy children. *Journal of Pediatric Psychology*, 22, 843-859.
- Batson, C.D., Fultz, J., & Schoenrade, P.A. (1987). Distress and empathy: two quantitatively distinct vicarious emotions with different motivational consequences. *Journal of Personality*, 55, 19-39.
- Blount, R.L., Zempsky, W.T., Jaaniste, T., Evans, S., Cohen, L.L., Devine, K.A., & Zeltzer, L.K. (2009). Management of pediatric pain and distress due to medical procedures. In Robers Editor, & Steele Editor (Eds.), *Handbook of Pediatric Psychology* (fourth edition, pp.171-188). New York: The Guilford Press.
- Boudreaux, E.D., Francis, J.L., & Loyacano, T. (2002). Family presence during invasive procedures and resuscitations in the emergency department: A critical review and suggestions for future research. *Annals of Emergency*, 40(2), 193-205
- Caes, L., Vervoort, T., Eccleston, C., Vandenhende, M., & Goubert, L. (2011). Parental catastrophizing about child's pain and its relationship with activity restriction: The mediating role of parental distress. *Pain*, 152, 212-222.
- Caes, L., Vervoort, T., Trost, Z., & Goubert, L. (2012). Impact of parental catastrophizing and contextual threat on parents' emotional and behavioral responses to their child's pain. *Pain* 153, 687–695.
- Chambers C.T. (2003). The role of family factors in pediatric pain. In McGrath Editor, & Finley Editor, (Eds.), *Context of pediatric pain: biology, family, culture*. (pp. 99-130). Seattle: IASP Press.

- Conte, P.M., Walco, G.A., Sterling, C.M., Engel, R.G., Kuppenheimer, W.G. (1999). Procedural pain management in pediatric oncology: a review of the literature. *Cancer Investigation*, 17, 448-459.
- Dahlquist, L.M., Power, T.G., Cox, C.N., & Fernabach, D.J. (1994). Parenting and child distress during cancer procedures: a multidimensional assessment. *Children's Health Care*, 23, 149-166.
- Fedele, D.A., Hullmann, S.E., Chaffin, M., Kenner, C., Fisher, M.J., Kirk, K., Eddington, A.R., Phipps, S., McNall-Knapp, R.Y., & Mullins, L.L. (2013). Impact of a parent-based interdisciplinary intervention for mothers on adjustment in children newly diagnosed with cancer. *Journal of Pediatric Psychology*, 38(5), 531–540.
- Franck, L.S., Cox, S., Allen, A., & Winter, I. (2004). Parental concern and distress about infant pain. *Archives of Disease in Childhood-fetal and Neonatal Edition*, 89, F71–F75.
- Goubert, L., Vervoort, T., Cano, A. & Crombez, G. (2009). Catastrophizing about their children's pain is related to higher parent–child congruency in pain ratings: an experimental investigation. *European Journal of Pain*, 13, 196-209.
- Hox, J.J. (2010). *Multilevel analysis: Techniques and Applications*. (2nd edition). New York: Routledge.
- Jay, S.M., Ozolins, M., Elliott, C.H., & Caldwell, S. (1983). Assessment of children's distress during painful medical procedures. *Health Psychology*, 2, 133-147.
- Katz, E.R., Kellerman, J., & Siegel, S.E. (1980). Behavioral distress in children with cancer undergoing medical procedures: developmental considerations. *Journal of Consulting and Clinical Psychology*, 48(3), 356-365.
- Kazak, A.E. (2005). Evidence-based interventions for survivors of childhood cancer and their

- families. *Journal of Pediatric Psychology*, 30, 29-39.
- Kazak, A.E., Boyer, B.A., Brophy, P., Johnson, K., Scher, C.D., Covelman, K. & Scott, S. (1995). Parental perceptions of procedure-related distress and family adaptation in childhood leukaemia. *Child Health Care*, 24, 143-158.
- Kenny, D.A, Kashy, D.A., & Cook, W.L. (2006). *Dyadic Data Analysis*. New York: The Guilford Press.
- Ljungman, G., Gordh, T., Sorensen, S., & Kreuger, A. (2000). Pain variations during cancer treatment in children: A descriptive survey. *Pediatric Hematology and Oncology*, 17, 211-221.
- Malloy, K.M. & Milling, L.S. (2010). The effectiveness of virtual reality distraction for pain reduction: A systematic review. *Clinical Psychology Review*, 30, 1011-1018.
- McCracken, L.M., & Gauntlett-Gilbert, J. (2011) Role of psychological flexibility in parents of adolescents with chronic pain: development of a measure and preliminary correlation analyses. *Pain*, 152(4), 780-785.
- Nezlek, J.B. (2001). Multilevel random coefficient analysis of event- and interval-contingent data in social and personality psychology research. *Personality and Social Psychology Bulletin*, 27, 771-785.
- Pöder, U., Ljungman, G., & von Essen, L. (2010). Parents' perceptions of their children's cancer-related symptoms during treatment: a prospective, longitudinal study. *Journal of Pain and Symptom Management*, 40, 661-670.
- Raudenbush, S.W., Bryk, A.S., & Congdon, R. (2004). *HLM 6: Hierarchical Linear and Nonlinear Modeling*. Lincolnwood, IL: Scientific Software International.
- Reinoso-Barbero, F. Pascual-Pascual, S.I., de Lucas, R. García, S., Billoët, C., Dequenne, V., &

- Onody, P. (2011). Equimolar nitrous oxide/oxygen versus placebo for procedural pain in children: a randomized trial. *Pediatrics*, *127*, e1464-e1471.
- Schütze, R., Rees, C., Preece, M., & Schütze, M. (2010). Low mindfulness predicts pain catastrophizing in a fear-avoidance model of chronic pain. *Pain*, *148*, 120-127.
- Stinson, J.N., Kavanagh, T., Yamada, J., Gill, N., & Stevens B. (2006). Systematic review of the psychometric properties, interpretability and feasibility of self-report pain intensity measures for use in clinical trials in children and adolescents. *Pain*, *125*, 143–157.
- Van Damme, S., Crombez, G., Nieuwenborgh - Van De Wever K., & Goubert, L. (2008). Is distraction less effective when pain is threatening? An experimental investigation with the cold pressor task. *European Journal of Pain*, *12*(1), 60–67.
- Verhoeven, K., Goubert, L., Jaaniste, T., Van Ryckeghem, D.M.L., & Crombez, G. (2012). Pain catastrophizing influences the use and the effectiveness of distraction in schoolchildren. *European Journal of Pain*, *16*(2), 256–267.
- Vervoort, T., Caes, L., Crombez, G., Koster, E.H.W., Van Damme, S., Dewitte, M., & Goubert, L. (2011). Parental catastrophizing about child's pain and selective attention to varying levels of facial pain expression in children: a dot-probe study. *Pain*, *152*, 1751–1757.
- Williams, A.C.D.C., Eccleston, C., & Morley, S. (2012). Psychological therapies for the management of chronic pain (excluding headache) in adults. *Cochrane Database of Systematic Reviews*, *11*, CD007407.
- Young, S.S, Schwartz, R., & Sheridan, M.J. (1996). EMLA cream as a topical anesthetic before phlebotomy office in children. *Southern Medical Journal*, *89*(12), 1184-1187.

Table 1: Final model for parental distress and desire to be present and child distress.

	<i>Parental distress</i>			<i>Parental desire to be present</i>			<i>Child distress</i>		
	<i>Coeff.</i>	<i>SE</i>	<i>T</i>	<i>Coeff.</i>	<i>SE</i>	<i>T</i>	<i>Coeff.</i>	<i>SE</i>	<i>T</i>
Intercept (γ_{000})	4.72	0.27	17.14***	6.89	0.42	16.32***	3.63	0.37	10.00***
Time (γ_{100})	-0.08	0.03	-2.85**	0.03	0.03	1.03	0.09	0.04	2.07*
Duration of procedure (γ_{200})	/	/	/	0.001	0.0004	2.34*	/	/	/
Child distress (γ_{300})	0.17	0.05	3.45***	/	/	/	/	/	/
Parent distress (γ_{300})	/	/	/	/	/	/	.17	0.07	2.25*
PCS-P state (γ_{020})	0.78	0.28	2.75*	-0.14	0.48	0.31	/	/	/
Parent sex*PCS-P state (γ_{030})	/	/	/	0.92	0.46	1.99	/	/	/
Time*PCS-P state (γ_{120})	0.07	0.03	2.22*	0.10	0.04	2.36*	/	/	/

Coeff. = Coefficient; PCS-P state=state version of the Pain Catastrophizing Scale – Parent version; * $p < .05$; ** $p < .01$; *** $p < .001$



