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Family-Based Treatment of Early-Onset Obsessive-Compulsive Disorder

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ABSTRACT

Despite a meaningful common core of symptoms observed across the life span, there are particularly unique features of early-onset (prepubertal) obsessive-compulsive disorder (OCD) that make consideration of early presentation different from adolescent or adult onset and that may have important implications for treatment. This article will first review the unique features of early-onset OCD, focusing particular attention to the developmental and familial context of these children's symptoms. The literature on behavioral family interventions for other childhood disorders, specifically anxiety, as well as that on family processes (e.g., parent-child interactions) in families of children with OCD will be reviewed. The pediatric OCD cognitive-behavioral therapy (CBT) literature (CBT alone and CBT plus medication) will also be reviewed, focusing on current evidence-based treatment guidelines. Finally, a model of family-based treatment for young children with OCD and some preliminary pilot data will be presented.

INTRODUCTION

ALTHOUGH IT WAS ONCE THOUGHT that obsessive-compulsive disorder (OCD) did not occur in early childhood, onset of OCD has been documented as early as 3 years of age, with estimates of a mean age of 10 years (Hollingsworth et al. 1980; Swedo et al. 1989). The field to date has generally merged child- and adolescent-onset OCD into the term *juvenile onset* (Geller et al. 1998). Increasing evidence suggests that there is an early (prepubertal), pubertal, and late adolescent-adult onset of OCD and that there may be distinct phenomenologic and risk factors of each subtype. Despite a meaningful common core of symptoms observed across the life span (Rettew et al. 1992), there are particularly unique features of early-onset OCD that make consideration of early presentation different from adolescent or adult onset and that may have important implications for treatment. Additionally, comorbidity and risk factors for development of illness may be different.

This article will first review the unique features of early-onset OCD, focusing particular attention to the developmental and familial context of these children's symptoms. The literature on behavioral family interventions for anxiety disorders, specifically OCD, as well as that on family processes (e.g., parent-child interactions)

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in families of children with OCD will be reviewed. The pediatric OCD cognitive-behavioral therapy (CBT) literature (CBT alone and CBT plus medication) will also be reviewed, focusing on current evidence-based treatment guidelines. Of specific note, there are no evidence-based medicine treatment guidelines for young children with OCD. Finally, a model of family-based treatment for young children with OCD will be presented.

SYMPTOM PICTURE DURING EARLY CHILDHOOD

Gender difference

Children presenting with early-childhood-onset OCD (i.e., those with prepubertal onset) are more likely to be male, whereas the gender difference is reversed in adults (Swedo et al. 1989). Most studies note a male predominance in children (3:2), with the gender distribution becoming more equal in adolescence (Geller et al. 1998; Swedo et al. 1989). Cases with early onset are more likely to be familial than those with later onset (Nestadt et al. 2000; Pauls et al. 1995).

Comorbidity

Children with early onset of symptoms are more likely to have comorbid tic disorders, attention deficit hyperactivity disorder, and learning disabilities (Geller et al. 1996, 1998; Pauls et al. 1995). Family studies have established significantly elevated rates of comorbidity between OCD and tic disorders (Pauls et al. 1986). This finding is particularly strong for subjects with onset of OCD before age 9 years (Pauls et al. 1995). Individuals with early-onset OCD also appear to have a different pattern on single photon emission computed tomography scans from the late-onset OCD patients (Busatto et al. 2001).

Symptom expression

Early-onset cases have been recently identified as having an atypical pattern of symptom expression (Geller et al. 1996, 1998). In the young child, compulsions without obsessions are common, and the compulsive behaviors themselves may be different from those observed in adolescents or adults (Rosario-Campos et al. 2000; Swedo et al. 1989). In childhood-onset cases, children often involve family members in their ritualistic behavior, typically in the form of reassurance seeking (verbal checking; Rettew et al. 1992). These differences between children and adults may largely be due to developmental factors. Early childhood cognitive development is the most likely explanation for why obsessional thoughts are less prominent features in the symptom picture.

Embeddedness in the family

A striking difference between early-childhood and adult-onset OCD is the role of the family. Young children are "embedded" in a family context in a way that is meaningfully different from that of adults. Young children with OCD also are much more embedded in their families than are older children or adolescents. The dependence of children on their caregivers makes them vulnerable to multiple influences over which they have little control. Parental mental health, marital functioning, and family dynamics are just a few of the contextual factors that affect the nature and severity of impairment, treatment progress, and maintenance of treatment gains for children with psychiatric disorders (Kazdin 1995; Kazdin and Weisz 1998; Tharp 1991; Webster-Stratton 1985; Weisz and Weiss 1991). Further, the family and the related subsystems are also affected by the child's symptoms of OCD (Lenane 1989).

Parents are more likely to play an active role in young children's rituals (e.g., assisting with washing or checking; Lenane 1989). Empirical investigation of the possible effect of a family's social role in the development and maintenance of childhood OCD is limited. Clinical evidence suggests that families both affect and are affected by this disorder with regard to accommodation of and participation in rituals and avoidance behaviors (Lenane 1989, 1991; Pollack and Carter 1999; Steketee 1997). Patterns of family behavior, parent-child interactions, and parents' own interpretations of potentially anxiety-provoking stimuli also are likely to af-

fect their young children with OCD. For young children, it is impossible to understand their symptoms and their maintenance without approaching them from a family-based framework.

Implications for treatment

We are suggesting that there is an ongoing interactive cycle between the functioning of the child and the family. The presence of a child with OCD symptoms is likely to impact the functioning of the family unit and/or subsystems (parent-child, marital relationship). In turn, this compromised family functioning is likely to have an impact on the child and thus his or her symptoms. The resulting disequilibrium in the family needs significant attention in treatment, yet it is not typically a primary focus in the traditional individual treatment model. In this regard, it has been suggested that therapy during the early-childhood period is by necessity "de facto family context therapy," regardless of the theoretical underpinnings (Kazdin and Weisz 1998). Essentially, the child cannot be treated out of context of the family system.

The requisite cognitive component of CBT protocols for childhood OCD has limited utility at best during the early-childhood period. Young children have not yet reached the cognitive stage to fully comprehend, utilize, and benefit from cognitive therapy techniques (e.g., abstract thinking, cause and effect, understanding probability). Further, current approaches are based on an individual modality of treatment. Although adolescents may be able to attend a therapy session independently, understand and retain weekly assignments, and complete between-sessions homework (all integral steps in existing treatment protocols), young children cannot. In therapy with young children, caregiver involvement is essential, as they are often required to take on a supportive or even primary role in administering treatment. The individual therapy modality is not an optimal mode of treatment delivery for this age group.

Given the psychiatric comorbidity in this population, focusing on the OCD symptoms alone is often not sufficient. Younger children often have comorbid behavioral issues and are more apt to involve their family in their behaviors. Therefore, we propose that a family-based approach is required. This would involve teaching parents basic behavior management techniques not specifically related to the OCD per se, developing OCD-specific behavior modification plans, and teaching parents strategies to manage their child's anxiety and distress that may be crucial with this population.

It is our hypothesis that cognitive, developmental, and symptom differences, particularly embeddedness in the family context, play a significant role in understanding early-onset OCD and its treatment. A focus on OCD symptomatology alone, without considering involving the family system in treatment, may be insufficient for symptom amelioration and long-term improvement. We propose that the family context is an important vehicle for treatment development and delivery.

BEHAVIORAL FAMILY INTERVENTIONS FOR CHILDHOOD ANXIETY AND OCD

Behavioral family intervention has had a major influence in the field of psychopathology and has become an important paradigm in the treatment of childhood disorders (Sanders 1996; Taylor and Biglan 1998). The core components of behavioral family intervention involve teaching parents effective child management strategies (e.g., positive reinforcement, timeout) while at the same time teaching the family effective communication and conflict resolution strategies (Taylor and Biglan 1998). Behavioral family interventions have been empirically shown to benefit a wide range of disruptive behavior problems in children (e.g., attention deficit hyperactivity disorder, oppositional defiant disorder, conduct disorder). The success of behavioral family intervention has led to its extension to the treatment of other childhood disorders, including chronic illness (Sanders et al. 1994; Stark et al. 1990), developmental disabilities (Harrold et al. 1992), obesity (Graves et al. 1988), and anxiety (Barrett et al. 1996).

The empirical base supports the conclusion that when parents are trained to implement behavior change strategies, there is often a corresponding improvement in their children's behavior and adjustment. The underlying premise

of behavioral family intervention emphasizes the importance of involving parents, teachers, and significant persons as mediators or behavior-change agents to bring about lasting therapeutic change (Sanders 1996). The behavioral family intervention model considers the cognitive and affective dimensions of parent-child relationships as well as behavior as the targets of intervention, making it a cognitive-behavioral family intervention (Sanders 1996).

Although behavioral family intervention appears promising for the treatment of other childhood anxiety disorders (Barrett et al. 1996), there is a more limited empirical basis for this strategy in the OCD literature. Although ultimate treatment success is dependent on the child's cooperation, recent studies suggest the importance of including concurrent family intervention and individual treatment focused on removing parents from their children's rituals (Piacentini et al. 1994; Piacentini et al. 1997, unpublished manuscript). Interestingly, preliminary evidence also suggests that parents can play a role as co-therapists in the behavioral treatment and still not be overinvolved or enabling (Knox et al. 1996). Family involvement is particularly relevant to the treatment of children with OCD in the early childhood years, yet none of these studies has included children under the age of 8 years.

OBSERVATIONAL STUDIES OF FAMILY PROCESSES

Although we have clinical reason to believe that embeddedness in the family is a key to understanding better the nature of early-onset OCD, empirical investigation of the family's social role in the unfolding development and maintenance of childhood OCD is limited. Particularly given our interest in young children, it is critical to delineate the ways in which family processes (e.g., parent-child interactions that take place when children are anxious) may contribute to symptom development, severity, and maintenance. Using structured experimental paradigms, the following studies have observed parents and children in standardized anxiety-provoking interaction tasks and measured their affect and behavior.

Family processes in childhood OCD

Przeworski et al. (1999a, 1999b) examined family conflict and problem solving in OCD-relevant and non-OCD-relevant situations. In 29 mother-child dyads where the child had a current diagnosis of OCD, an OCD-relevant family conflict (e.g., child constantly asks family members for reassurance) and a non-OCD-relevant conflict were discussed and videotaped for 5 minutes each. Mothers and children were told to discuss each situation and to try to generate possible solutions; the discussions were later coded. Results indicated that family problem-solving style was generally consistent across OCD and non-OCD situations. Additionally, parent report of increased family accommodation was related to less agreement, fewer solution-based questions, and more anger during the experimental interactions.

In a recent study, Barrett et al. (2002) investigated whether families with children with OCD and families with other clinically diagnosed children and nonclinical children behave significantly differently during family interactions. The interactions consisted of two videotaped 5-minute family conversations. Both discussions were about ambiguous, hypothetical situations—one involving a physical threat and one involving a social threat. They observed and coded several behavioral concepts (i.e., control, warmth, doubt, avoidance, positive problem solving, confidence, and rewarding independence) in four different groups of families: those with children with OCD, those with anxious (generalized anxiety disorder, separation anxiety disorder, and social phobia) children, those with externalizing (oppositional defiant disorder, attention deficit hyperactivity disorder) children, and those with nonclinical children. Results demonstrated that parent and child behaviors during family interactions were substantially different in families with children with OCD from families with other clinical and nonclinical children. Specifically, parents of children with OCD showed less confidence in their child's ability, were less granting of autonomy, and used positive problem solving less often. In addition, children with OCD were less confident, showed their parents less warmth, and were less likely to use positive problem solving as well.

Expressed emotion (EE) in childhood OCD. The construct of EE consists of assessment of critical comments, emotional overinvolvement, hostility, warmth, and positive remarks by a relative about a family member/patient (Brown et al. 1972) and has been used as a predictor of course and relapse in severe psychiatric illness. Although EE has recently received more attention in adult OCD (Chambless and Steketee 1999; Steketee et al. 1998; Van Noppen 1999, unpublished manuscript), only three studies have examined EE in families of children and adolescents with OCD. Leonard et al. (1993) found that children with OCD living in high-EE families had poorer global adjustment at 2- to 7-year follow-up, but EE did not predict their OCD symptom severity. In another study, Hibbs et al. (1991) examined EE in families of children diagnosed with OCD and disruptive behavior disorders as well as in families of nonclinical controls. Parents in both psychiatric groups were more frequently rated as high EE, compared with the control group, although the OCD and disruptive behavior groups did not differ from one another. Przeworski et al. (1999a) examined the relation between EE and child OCD and found that parents were more likely to have high-EE profiles for their children with OCD than for non-OCD siblings. High EE was also associated with child OCD symptom severity and greater family accommodation. Thus, high EE may characterize family interactions and serve as a marker of the overall family distress that accompanies the diagnosis of an individual child with OCD.

CURRENT TREATMENTS FOR PEDIATRIC OCD

Currently, two treatment modalities have empirical support for effectively treating OCD: psychotherapy—specifically exposure with response prevention (E/RP)—and medication therapy (serotonin reuptake inhibitors [SRIs] and selective serotonin reuptake inhibitors [SSRIs]). In contrast to the adult literature, however, little research has focused on the treatment of pediatric OCD. This section reviews the literature regarding cognitive-behavioral treatments of OCD, specifically the efficacy of CBT and the combination of CBT and medication. A review of medication therapy alone will not be covered as it is beyond the scope of this article.

Psychotherapy trials

Empirical documentation regarding the efficacy of psychotherapy for children and adolescents with OCD has greatly lagged behind the adult literature (Piacentini 1999). Although CBT (specifically E/RP) has clearly demonstrated usefulness in the treatment of adults with OCD, it has not been well studied in children. Although some are currently under way, no large controlled trials of CBT with children or adolescents have been completed; however, clinical reports support its utility (March 1995; March et al. 1994).

Only three studies published to date have utilized an experimental design with more than one subject and well-documented, reproducible treatment programs. In the first of these, March et al. (1994) reported on a sample of 15, 8- to 18-year-olds treated with a structured CBT protocol for OCD. On average, children attended 10 therapy sessions over an 8-month period and were followed for 7.3 months. Results indicated significant pre- to posttreatment improvement in OCD symptoms for the sample, with a mean reduction in symptom severity of about 50%. Sixty-seven percent of the sample demonstrated better than 50% symptom improvement that was maintained at follow-up, and only 20% were nonresponders; 40% of the sample at posttreatment and 60% at follow-up were rated as asymptomatic. In addition, 40% of the sample were able to discontinue medication with booster CBT sessions. The development of a systematic CBT manual for children and adolescents with OCD (8–17 years of age) (March and Mulle 1998) has provided an important tool that could be exportable; however, it is not designed specifically for the very young child with OCD.

The results of the above report (March et al. 1994) suggest that as in the adult literature, CBT may be a useful treatment modality for youths with OCD. It is clearly the first study in the field to apply reliable and valid measures of treatment outcome to a large sample of patients; to assess patients before, during, and

following treatment; and to utilize a manualized treatment protocol. However, some specific issues limit the conclusions that can be drawn. All but one child in the study received adjunctive treatments including medication, family therapy, and supportive individual therapy. Further, duration of prior pharmacotherapy and dose adjustments during treatment were not specified and leave open the question as to which factors were responsible for treatment gains. There was no control or comparison condition. Also, it did not specifically target the very young child with OCD, similar to the ages discussed in this paper. The authors are involved in a large systematic trial of combined treatments (medication vs. CBT vs. medication + CBT), and those results will be important in evaluating the relative efficacy of the different treatment options.

Scahill et al. (1996) reported similar findings. These investigators treated seven children between the ages of 8 and 16 years with CBT by means of E/RP. Results indicated mean symptom improvement of 61% posttreatment, which decreased to 51% at a 3-month follow-up. Although this study did not have a control group, three children who declined CBT were followed for 3 to 6 months, and there was no improvement in their symptoms.

Franklin et al. (1998), in an open CBT study with 14 children and adolescents between 10 and 17 years of age, found similar results. Average symptom improvement at posttreatment and follow up was 67% and 62% respectively on the Children's Yale-Brown Obsessive Compulsive Scale. Further, this study found no difference in outcome measures between CBT applied intensively (i.e., daily sessions) versus weekly sessions.

In sum, although current evidence supports CBT in the form of E/RP for the treatment of OCD during late childhood and adolescence, and systematic manuals are now available, empirical documentation is needed. Despite the consistency in results across studies, there are numerous methodological limitations, including small samples, nonstandard application of treatment, ascertainment bias, treatment effects confounded by the presence of concurrent pharmacotherapy, and lack of randomly assigned control groups. None of the studies was conducted on children younger than 8 years of age. Although there are systematic studies in progress, none to our knowledge specifically focuses only on the early-childhood age group.

Combined treatments

Studies in adults with OCD suggest that combining CBT with medication enhances treatment efficacy (Cottraux et al. 1990; Marks et al. 1980). Expert consensus panel recommendations for treatment of juveniles and adults with OCD also support the combination of CBT and medication (King et al. 1998; March et al. 1997). Conversely, one could argue that experiencing and tolerating the anxiety is necessary for CBT to be effective and that for some patients CBT plus medication may be less effective than either alone as monotherapy. However, methodological limitations and lack of clearly specified control groups in previous work limit the strength of conclusions that can be drawn regarding this issue. As of yet, no large controlled studies have been published comparing psychotherapy, medications, or combined treatment approaches with children or adolescents, although a study is in progress.

In the only published, controlled study to date utilizing CBT for OCD in children and adolescents, de Haan et al. (1998) compared E/RP plus cognitive restructuring with clomipramine in 22 youngsters for a 12-week period. Results indicated that both treatments produced positive effects but that E/RP was significantly more effective in terms of both reducing symptom severity and increasing response rate. Thus, the efficacy of combined treatment approaches for children and adolescents remains an unanswered question. With children, concerns about medication efficacy and long-term use have prompted treatment guidelines to recommend CBT as first-line treatment, although severe cases and other individual clinical circumstances might suggest a role for medications in some cases (King et al. 1998; March et al. 1997). Thus, CBT would be considered first-line treatment for young children (5–8 years of age), for whom we are developing a family-based behavioral therapy treatment manual.

Summary of treatment outcome studies

In summary, two treatment modalities—psychotherapy (CBT with E/RP) and medication (SRIs/SSRIs)—have been studied with regard to treatment of OCD in children and adolescents. Evidence supports the efficacy of SRIs in targeting core symptoms of OCD; however, SRIs do not provide uniformly effective treatment, typically only produce partial symptom remission, and have a high rate of relapse following withdrawal. Although CBT is regarded by many as the psychotherapeutic treatment of choice for children and adolescents with OCD (particularly very young children), solid empirical documentation regarding its efficacy is lacking. Thus, we have little empirical knowledge about the treatment of OCD in young children.

PROPOSED TREATMENT APPROACH FOR EARLY CHILDHOOD OCD

Our specific interest in early-onset OCD has led our group to develop a cognitive-behavioral family intervention for the young child. Although modified from an existing OCD treatment manual (March and Mulle 1998), it differs fundamentally by teaching basic behavior management techniques (not specific to OCD), focusing on family accommodation and parental reinforcement of anxiety, and involving parents in implementation of the CBT program at home. Treatment consists of a multicomponent program combining psychoeducation, parent training, family treatment, and cognitive strategies (e.g., E/RP). Most treatment components are covered in every session.

The goals of the psychoeducation component are to educate parents about the neurobiology of OCD, correct misattributions about OCD, differentiate between OCD and non-OCD behaviors, and describe the treatment program in detail. The goals of the parent-training component are to teach parents basic behavior management techniques (e.g., positive reinforcement, attending to positive behaviors, timeout), develop a behavior modification plan that will be used throughout treatment to address both OCD and other behaviors, and teach parents strategies to manage their child's anxiety and distress. The goals of the family treatment component are to reduce family accommodation of child OCD symptoms, reduce criticism and hostility related to child OCD symptoms, promote positive family problem solving related to child OCD, and teach parents to understand the role of their own modeling of anxious interpretations and behaviors. The goal of the cognitive restructuring component is to provide children and parents with some basic and developmentally appropriate tools to cope with E/RP. These tools include learning how to externalize ("boss back") OCD, deep breathing, and using a fear thermometer to rate anxiety. The goal of the E/RP component is to have parents and children work together to develop a hierarchy and implement E/RP via a behavior modification sticker chart.

Modeled after the March and Mulle (1998) manual, our proposed treatment approach consists of 12 sessions delivered over the course of 14 weeks. The first 10 sessions are delivered weekly, with 2-week intervals between the last two sessions. The first two sessions (90 minutes) are conducted with parents alone, with the remaining sessions (60 minutes) conducted jointly with both parents and children. The proposed CBT has been piloted in four subjects (age range 4–11 years, mean = 6.75 years)—two boys (ages 6 and 11 years) and two girls (ages 4 and 6 years)—with a primary diagnosis of OCD. One patient had a secondary diagnosis of attention deficit hyperactivity disorder, one patient had a secondary tic disorder, and one patient had secondary separation anxiety disorder. The fourth patient had no comorbidity. All participants exhibited a decrease in OCD symptom severity as indicated by a decrease in Children's Yale-Brown Obsessive Compulsive Scale scores from pretreatment (mean = 26, SD = 4, range 20–43) to posttreatment (mean = 3, SD = 2, range 0–7) (see Fig. 1). A follow-up phone call at 3 months indicated that treatment gains were maintained.

There was no difficulty in recruiting patients. Subjects attended an average of 11.5 sessions. Mothers and fathers participated in treatment in all four families. None of the participants dropped out of the treatment, and all indicated

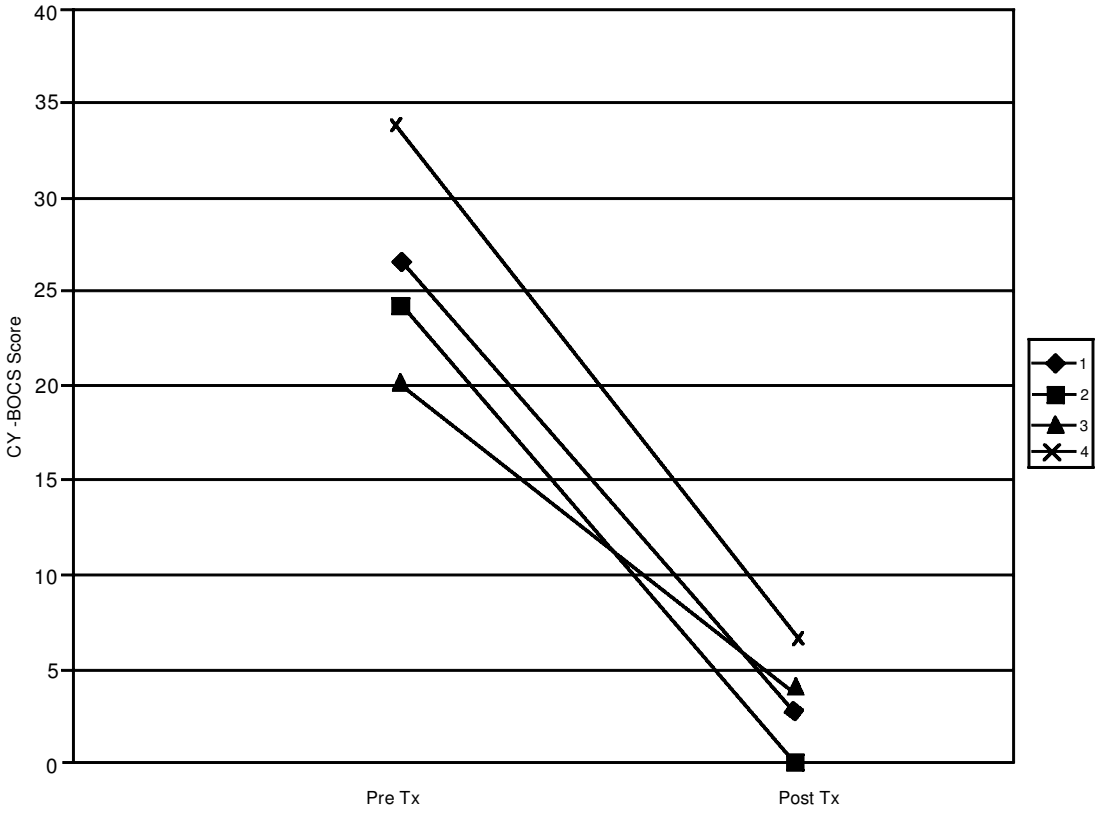

FIG. 1. Pilot treatment data of the four participants pre- and posttreatment. Tx = treatment.

their satisfaction with the treatment and their outcome. The team reviewed the treatment of each patient. The manual was acceptable to the children and their parents. The number of sessions was practical and doable. The parents were able to complete their components of the treatment. Although the findings are very preliminary, they provide an initial impression that the pilot manual can be developed and the intervention implemented.

CONCLUSION

Young children with OCD differ from older ones, both in their developmental status and in the phenomenology of symptoms. Intuitively, it makes sense to develop a behavior treatment therapy specific to young children that is safe and effective, practical, durable, and exportable. Although manualized treatments exist for older children with OCD, they cannot simply be extrapolated down for younger children.

Interestingly, work in the child psychology literature on the moderators and mediators of childhood anxiety, as well as cognitive-behavioral family intervention, has generally gone unnoticed in the child psychiatry field. Our interest in the development of a family-based CBT manual for young children with OCD has drawn upon the fairly large literature of family-based treatment for young children with non-OCD anxiety disorders. This article presents the preliminary description of the proposed components of a cognitive-behavioral family intervention for young children with OCD.

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