Doi: 10.5455/PBS.20220412092734

ORIGINAL RESEARCH

Peer Bullying, Anxiety, and Depression Levels Among Turkish Children with Childhood Onset Fluency Disorder (COFD): A single-Center, Case-Control Study

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Abstract

Objective: Childhood Onset Fluency Disorder (COFD) is associated with elevated levels of emotional/ behavioral problems and peer bullying. Studies on Turkish children with COFD are limited. We aimed to evaluate rates of peer bullying and its relationships with stuttering severity and symptoms of depression and anxiety among Turkish children with COFD.

Methods: Forty 8-12 years old children with COFD without comorbid neurological/ medical disorders and 36 age — and gender-matched controls were evaluated with self-report scales for depression, trait/ state anxiety and peer bullying. Stuttering severity was evaluated with Stuttering Severity Instrument-4. Multivariate and bivariate analyses were used in comparisons. P was set at 0.05.

Results: COFD group had significantly elevated depression and state anxiety scores while trait anxiety was elevated but at trend levels. 52.5 % of children with COFD was bullied while this rate was 27.8 % for controls (Odds Ratio= 2.9). Bullying was not related with gender, self-reported symptoms and stuttering severity.

Conclusion: Standardized trainings/practices should be introduced among Turkish speech and language therapists to screen and address peer bullying, depression, and anxiety among children with COFD in collaboration with child and adolescent psychiatrists.

Keywords: Stuttering, Children, Bullying, Anxiety, Depression

INTRODUCTION

Stuttering ("Childhood onset fluency disorder", COFD), listed among the communication disorders in DSM-5 (1) is characterized by developmentally incongruent speech dysfluency and temporal patterns leading to academic and social dysfunction. The speech dysfluency may present with sound/ syllable repetitions, sound prolongations, interjections, word disruptions, audible/ silent blocks, circumlocutions, excess physical tension during speech, whole word repetitions or their combinations. The DSM-5 allows the diagnosis in presence of speech-motor and sensory deficits but requires the speech dysfluency to be more than those typically associated with those deficits (1). The disorder is reported to develop mostly between

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Citation: Kilicaslan F, Cicek E, Gungoren S, Kutuk MO, Tufan AE. Peer Bullying, Anxiety, and Depression Levels Among Turkish Children with Childhood Onset Fluency Disorder (COFD): A single-Center, Case-Control Study. Psychiatry and Behavioral Sciences 2022;12(3):99-105. Doi: 10.5455/PBS.20220412092734

Received: Apr 12, 2022 **Accepted:** Sep 13, 2022

2-7 years of age and is thought to affect 8.0-11.0% of children and 1.0% of the general population (2). In most of the affected children (i.e., approximately four-fifth) the symptoms remit spontaneously within two to four years after onset. Like other neurodevelopmental disorders, COFD is commoner among males with a 2:1 ratio in childhood which may increase later in life (3). COFD commonly occurs with other neurodevelopmental disorders such as attention deficit hyperactivity disorder (ADHD) and specific learning disorders (SLD) (1,4,5). The relationships between stuttering and other developmental language disorders are still controversial (5,6). Other psychopathologies including post-traumatic stress, generalized anxiety, social anxiety and nocturnal enuresis may also accompany COFD (1,3,4).

Studies conducted among youth from Turkey as well as other countries suggest that COFD is associated with elevated levels of emotional and behavioral problems (7-9). Both those problems and the presence of COFD may lead to reduced sense of competence in communication and increased rejection in interpersonal relationships (10, 11). Turkish children without COFD as well as their parents and parents of Turkish children

with COFD may perceive children with this condition more negatively compared to those from the US (12-14). Those negative perceptions may lead to peer bullying and studies conducted up to now have found that children with COFD report elevated rates of peer bullying compared to their unaffected peers (15). Rates of peer bullying among children with COFD may vary between 27.3 - 83.0 % (16-18). Peer bullying among those children may increase psychological distress and anxiety, reduce academic achievement, selfesteem, and self – efficacy, and may continue to have deleterious effects over the long term (15, 19). Despite the importance of determining the rates, correlates and treatment implications of peer bullying among children with COFD, there are only four studies conducted on this phenomenon from Turkey (20-23). Kara and Karamete (2018) found that 85.0 % of Turkish adults with COFD reported peer bullying victimization during their school life and that verbal abuse was the most common form (22). Kayhan Akturk and Ozdemir (2021) found that even among preschool children with COFD peer rejection and peer violence were significantly elevated (20). Özgür and Gürbüz Özgür (2020) reported that peer rejection and bullying significantly reduced quality of life among children with COFD (23). Erim and Aydın Uysal (2022) reported that there were no standardized trainings/ practices among Turkish speech and language therapists on addressing peer bullying in COFD (21). According to their results, the therapists used mostly desensitization, problem solving/ coping strategies and involvement of the family/ teachers of children in addressing this problem. However, there are no controlled studies evaluating rates of peer bullying and its relationships with stuttering severity and symptoms of depression and anxiety among Turkish children with COFD.

Therefore, we aimed;

- a. to compare the rates of peer bullying among Turkish children with COFD and control children,
- b. to compare self-reported symptoms of depression and anxiety among Turkish children with COFD and control children,
- c. to evaluate correlates and predictors of peer bullying among Turkish children with COFD.

METHODS

Study Setting and Participants

The study was conducted at the Department of Child and Adolescent Psychiatry and Department of Pediatrics of a

hospital in Turkey between June 1st, 2019 and June 1st, 2020. Children who applied to the Department of Child and Adolescent Psychiatry with complaints of "stuttering" were evaluated by a Child and Adolescent Psychiatrist and a Speech Language Therapist independently for diagnosis and those fulfilling criteria for COFD (ICD-10 F80.81, DSM-5 315.35) and aged between 8 – 12 years were enrolled in the study. Chronic neurological and/or medical disorders (e.g., epilepsies, diabetes mellitus, etc.) and intellectual disabilities were exclusion criteria.

Davis and colleagues (24); reported that 10.7 % of 387 controls were victimized by bullies compared to 37.5 % of 16 children with COFD, leading to an odds ratio of 5.4 (95 % Confidence Interval= 1.9 – 15.5, Z= 3.1, p=0.002; 25). According to a priori power analysis for OR= 5.4 a proportion of approximately 0.40 and a p value of 0.05 (two-tailed) with equal allocation between groups, we aimed to collect 80 participants (40 with COFD and 40 controls) for 95.0 % power (26).

Convenience sampling was used to recruit participants. Within the study period 80 patients applied with the complaints of "stuttering" and 10 were excluded due to presence of chronic neurological/ medical disorders, 4 due to intellectual disability, 23 were younger than 8 years old, 1 had hearing impairment and 2 were diagnosed with Tourette's disorder leaving 40 children with COFD. Thirty age — and gender-matched children applying to the Department of Pediatrics for well-child visits and who had no speech/ hearing problems formed the control group. History of psychiatric/ neurological and/ or chronic medical disorders were criteria for exclusion from the control group.

Study Procedures

The parents of all participants completed the sociodemographic data form after study procedures and aims were explained to them and following their informed consent. The children completed the Olweus Bully – Victim Questionnaire – Revised Form (OBVQ-R), State-Trait Anxiety Inventory for Children (STAI-CH) and Children's Depression Inventory (CDI). As a part of the evaluations, the severity of stuttering was assessed by the speech and language therapist using Stuttering Severity Instrument-4 (SSI-4).

Ethical approval

All participant children provided verbal or written assent while the parents provided written informed consents before participation. The study was approved by Ethics Committee of Harran University on Clinical Research (No: 16773 – 16/04/2019).

Measurement Tools

Sociodemographic Data Form: This form was developed by the researchers and was used to collect sociodemographic data including age, gender, education status, medical history, age at onset of stuttering symptoms, previous treatments, number of siblings of children as well as educational/ vocational status of the parents and family history of psychiatric disorder.

The Olweus Bully/Victim Questionnaire (OBVQ-R): OBVQ was developed in 1983 by Olweus to evaluate peer bullying and was revised in 1996 (27). This self-report instrument includes 39 items and may group 8 – 16 years old children as "bully", "victim", "bully/victim" or "none" according to cut-off scores of items 24 – 33, 4-13 or both: respectively. The reliability and validity of the Turkish form was established by Dolek (28).

State-Trait Anxiety Inventory for Children (STAI-CH): STAI-CH was developed to evaluate trait and state anxiety in children via 20-item, likert-type questions for each (29). For both state and trait anxiety the scores vary between 20 – 60 with greater scores denoting increased levels of anxiety. The Turkish translation was found to be valid and reliable previously (30).

Children's Depression Inventory (CDI): This 27-item, likert-type instrument was developed to evaluate cognitive, affective, and behavioral symptoms of depression over a two-week period among children aged 7 – 17 years (31). The total scores may vary between 0 and 54 with a cut-off score of 19 for clinically significant levels of depression. The Turkish translation was previously found to be reliable and valid with the same cut-off score (32).

Stuttering Severity Instrument-4 (SSI-4): The SSI -4 was developed by Riley in 1972 to assess the severity of stuttering among school age children aged between 6 and 16 years old and was revised later (33). SSI-4 evaluates the frequency, duration, and physical concomitants of dysfluencies as well as the nature of individual's speech. The Turkish version was found to be reliable and valid previously (34).

Statistical Analysis

All statistical analyses were performed using IBM SPSS for Windows version 25.0 0 (IBM SPSS Inc, Armonk, NY, USA). The normality of data distribution was assessed using Shapiro-Wilk test, skewness, kurtosis, histogram, and Q-Q plot. Children's and their fathers' age did not

distribute normally according to Shapiro-Wilk test while maternal ages and self-report scales distributed normally (p < 0.05). Quantitative variables were summarized as means and standard deviations or medians and interquartile ranges depending on assumptions of normality. Categorical variables were compared using Chisquare test between groups. Fisher's exact test, Yates' correction, linear-by-linear association, and likelihood ratio tests were used as required. Quantitative variables among groups were compared with student's t test for independent group and Mann-Whitney U tests. Multiple dependent variables (i.e., depression, state, and trait anxiety) across two groups were compared with MANOVA with follow-up univariate ANOVAs to evaluate which of the dependent variables differ significantly across groups (35). Bivariate correlations were evaluated with Pearson's test. Logistic regression analysis (exploratory) was used to determine predictors of bullying victimization among children with COFD. p value < .05 was considered as statistically significant (two-tailed). Effect sizes for significant findings were also reported.

RESULTS

Sociodemographic Characteristics of Children with Childhood-Onset Fluency Disorder (COFD, Stuttering) and Controls and their Parents

The study included 40 children (85.0 % male) with a mean age of 10.0 (S.D.=1.4) years diagnosed with COFD and 36 control children (75.0 % male) with a mean age of 9.5 (S.D.=1.2) years. The groups were similar in terms of age and gender (p=0.114 and 0.471; respectively (Mann-Whitney U and $\chi 2$ test with Yates' correction, Table 1). Table 1 presents sociodemographic and parental characteristics of children with COFD and controls (Table 1).

The mean raw SSI-4 score among COFD group was 20.6 (S.D.=6.1). Accordingly, the symptoms were classified as being "mild" in more than half (n=23, 57.5 %). Twelve children (30.0 %) were classified as "moderately affected" while the rest (n= 5, 12.5 %) were judged to display "severe" symptoms. Mean age of onset for stuttering was 5.0 (S.D.= 1.7) years. Approximately one-tenth of the children (10.5 %) had received some speech therapy previously. Most children from COFD group had family history of speech disorders (n= 28, 70.0 %; COFD in 96.4 % and phonological disorder in 3.6 %).

Table 1. Sociodemographic and parental characteristics of Turkish children with Childhood Onset Fluency Disorder (COFD) and control children

Median (IQR), Mean (SD) or N (%)		COFD (n=40)	Controls (n=36)	p*	Effect size**
Birth order – First		15 (37.5)	15 (41.7)	0.892	-
Sibling number		3.0 (2.0)	3.0 (2.0)	0.780	
Maternal age (years)		34.9 (4.6)	35.9 (5.3)	0.396	-
Maternal	None	10 (25.0)	13 (37.1)	0.784	-
education	Primary	22 (55.0)	12 (34.3)		
	High school	6 (15.0)	3 (8.6)		
	University	2 (5.0)	7 (20.0)		
Maternal vocation	Housewife	38 (95.0)	28 (77.8)	0.043	0.33
	Other	2 (5.0)	8 (22.2)		
Paternal age (years)		39.3 (4.5)	43.2 (7.6)	0.010	0.63
Paternal education	None	2 (5.0)	9 (25.0)	0.495	-
	Primary	25 (62.5)	10 (28.6)		
	High school	8 (20.0)	10 (28.6)		
	University	5 (12.5)	6 (17.1)		
Paternal vocation	None	1 (2.5)	4 (11.1)	0.064	-
	Unskilled	36 (90.0)	24(66.7)		
	Skilled	3 (7.5)	8 (22.2)		

IQR= Inter-quartile range, SD= Standard deviation, $*\chi2$ test with Yates', Fisher's and Linear-by-linear corrections as required, Mann-Whitney U and t tests, **: Cohen's d for t tests, Phi and Cramer's v for $\chi2$ tests.

Comparison of Children with Stuttering and Controls by Peer Bullying, State-trait Anxiety, and Depressive Symptoms

According to OBVQ-R; twenty-five of children were victims (72.0 % from COFD) while six were both victims and bullies (50.0 % from COFD). There was only one child classified as bully only (from the COFD group). The groups did not differ significantly in terms of children who bully their peers. However, more than half of children with COFD were bullied by peers (n=21, 52.5 %) while 27.8 % of controls (n=10) reported peer bullying (p=0.050, Yates' correction). Accordingly, children with COFD were judged to be under almost three times elevated risk for bullying (O.R= 2.9, 95 % CI= 1.1-7.5, z=2.2, p=0.031, Table 2).

Table 2. Comparison of children with Childhood Onset Fluency Disorder (COFD, stuttering) and controls according to peer victimization

N (%)	COFD (n= 40)	Controls (n= 36)	P*	Effect size
Victims	18 (45.0)	7 (19.4)	0.07	-
Bullies	1 (2.5)	0 (0.0)		
Both victims and bullies	3 (7.5)	3 (8.3)		
Neither victim nor bully	18 (45.0)	26 (72.2)		

^{*}Chi Square test

Self-reported depressive, state and trait anxiety symptoms among groups are listed in table 3 and compared across

groups with MANOVA and follow-up univariate ANOVAs. Covariance matrices of the dependent variables were equal across groups (Box's M=10.2, p=0.137) and error variances did not differ significantly except state anxiety (Levene's test, p=0.003). Therefore Pillai's trace was used for comparisons. The groups tended to differ in terms of self-reported depressive and anxiety symptoms (F [3.0, 72.0] = 2.5, p=0.063, partial η 2=0.10).

Table 3. Self-reported depressive symptoms, state and trait anxiety of Turkish children with Childhood Onset Fluency Disorder (COFD) and control children

Mean (SD)	COFD (n=40)	Controls (n=36)	p*	Effect size**
Children's Depression Inventory	12.3 (6.6)	8.3 (7.2)	0.015	0.08
STAI-CH-State	32.6 (8.1)	29.5 (4.9)	0.046	0.05
STAI-CH – Trait	34.4 (6.7)	31.4 (6.6)	0.054	0.05

SD: Standard deviation, STAI-CH: State trait anxiety inventory for children, *: univariate ANOVA, **: partial $\eta 2$

Follow-up analyses revealed that children with COFD reported significantly elevated depressive and state anxiety symptoms while they tended to report elevated levels of trait anxiety.

Effects of Gender on COFD Symptom Severity, Depression, and Anxiety

According to SSI-4, the rates of females displaying "mild" (n=4,17.4%), "moderate" (n=1,8.3%) and "severe" (n=1,20.0%) symptoms of COFD did not differ significantly from males (p=0.714, Likelihood ratio). None of the female children bullied others and four out of eighteen children bullied were females (22.8%). There was no significant difference between genders for OBVQ-R categories (p=0.647, Chi square test). In MANOVA, there were no significant differences between genders in terms of depressive symptoms, state, and trait anxiety (F [3.0, 36.0] =0.9, p=0.442, partial η 2=0.07, Pillai's trace).

Comparison of Children with Stuttering According to Presence of Peer Victimization

Rate of bullying victimization in "mild", "moderate" and "severely" symptomatic COFD groups were 57.1 % (n=12), 23.8 % (n=5) and 19.0 % (n=4); respectively with no significant difference (p=0.374, χ 2 test). Rates of bullying peers in corresponding groups were 25.0 % (n=1), 50.0 % (n=2) and 25.0 % (n=1); respectively with no significant difference (p=0.374, χ 2 test). Bullying victimization did not significantly affect depressive symptoms, state, and trait anxiety (F [3.0, 35.0]= 1.4, p=0.269, partial η 2= 0.11).

Correlates and Predictors of Bullying Victimization Among Children with COFD

Symptom severity according to SSI-4 did not correlate with CDI, STAI-CH trait, and state anxiety. Logistic regression analysis with enter method was used to evaluate predictors of being bullied among children with COFD and gender, being first child of the family, maternal psychopathology, and age of onset of stuttering were entered as predictors. Although the model was a good fit for the data (χ 2 [8]= 4.2, p=0.839, Hosmer – Lemeshow test) it did not improve case detection significantly $(\chi 2 [49=5.7, p=0.221))$ and could explain 17.8 % of the variance in bullying victimization (Nagelkerke R2=0.178). At the outset the model could classify all children victimized by bullying correctly and none of the others were correctly classified (overall= 52.5 %). After the analysis 71.4 % of those victimized by bullying and 57.9 % of the rest could be correctly classified for an overall accuracy of 65.0 % (Table 4). Later age of stuttering onset was the sole significant predictor for reduced risk of bullying.

Table 4. Predictors of bullying victimization among Turkish children with Childhood Onset Fluency Disorder (COFD)

Variable	Odds Ratio	р	95 % Confidence Interval
Female gender	0.4	0.356	0.1-2.9
Later born sibling	0.5	0.330	0.1 – 2.1
Maternal psychopathology	5.0	0.266	0.3-86.7
Age of onset of stuttering	0.6	0.045	0.4 - 1.0

DISCUSSION

This single-center, case-control study aimed to evaluate rates, correlates and predictors of peer bullying and selfreported symptoms of depression and anxiety among Turkish children with COFD and age - and gendermatched control children. To this end we enrolled forty children with COFD and 36 controls. More than half of children with COFD had mild or moderate symptoms and 52.5 % reported peer bullying while 27.8 % of controls reported peer bullying. Accordingly, children with COFD were under almost three times elevated risk for bullying. Children with COFD also reported significantly elevated depressive and state anxiety symptoms while they tended to report elevated levels of trait anxiety. There were no effects of gender or bullying victimization in self-reported depression and anxiety symptoms among children with COFD while logistic regression, later age of stuttering onset predicted reduced risk of bullying.

Studies from various countries suggest that COFD is associated with emotional and behavioral problems (7-9). This association may be due to shared genetic etiologies, neuropsychological vulnerabilities, reduced self-efficacy and increased rejection in interpersonal relationships with age or the effects of internalized stigmatization due to negative stereotypes (1,10,14,36,37). Studies conducted up to date suggest that COFD is associated with anxiety disorders and to a lesser extent depression and that anxiety may lead to persistence of symptoms, negative communication attitudes and increased avoidance (11,38,39). Partially supporting those views, we found that children with COFD in our sample reported significantly elevated depressive symptoms and state anxiety while the difference did not reach significance for trait anxiety. This discrepancy may be due to limited sample size affecting power of our study. Indeed, a posthoc analysis for MANOVA with two groups and three response variables suggests that we have achieved 49.0 % power (26). Also, self-reports may be due to response, recall and reporting bias and the use of parent and teacher reports as well as semi-structured interviews for psychopathology (i.e. K-SADS-PL) may have enriched our results.

Children with COFD may be under elevated risk for bullying with rates between 27.3 - 83.0 % (15-18). The risks for bullying maybe associated with negative perceptions of children with this condition, associated emotional and behavioral problems and problematic parenting practices (12-14,40). Exposure to peer bullying among those children further increases their psychological distress and anxiety and reduces achievement, self-esteem, and self-efficacy (15, 19). Rates, correlates, and predictors among Turkish children affected by COFD have received relatively scant attention up to now. In a retrospective study, Kara and Karamete (2018) found that 85.0 % of Turkish adults with COFD recalled peer bullying during their education, of which verbal abuse was the most common form (22). Counterintuitively, Kayhan Akturk and Ozdemir (2021) found that peer rejection and violence among such children may start from the preschool period onward (20). Özgür and Gürbüz Özgür (2020) found that peer bullying and rejection significantly reduced quality of life on children with COFD (23). In this study we found that 52.5 % of children in the clinical group were bullied and that the children were under approximately three times elevated risk of bullying. Bullying in our study was not affected by gender, symptom severity and not related with self-reported depressive and anxious symptoms.

In exploratory analyses, there was a signal that later age of onset may reduce risks of bullying. The rate of bullying we found is broadly in accordance with those reported in the literature (15) and less than those reported by Kara and Karamete (2018) (22). This may be due to recall bias in the Kara and Karamete study (2018) or due to response and recall bias in our study (22). Also, it may be argued that OBVQ-R classifications may have limited ecological validity and use of teacher reports as well as sociometric evaluations may have led to increased accuracy of identification for bullying in our study (24). Also, depression and anxiety symptoms may be evaluated with multiple informants and/ or clinical interviews to better delineate their relationships with bullying. The fact that most children with COFD had mild / moderate symptoms may introduce ceiling effects and affect correlations with peer bullying. The lack of effect of gender on bullying may be due to limited number of females with COFD in our study because previous studies from Turkish samples consistently found male gender as a risk factor for exposure to peer bullying. Alternatively, OBVQ-R items may not adequately tap indirect forms of bullying more common among female children (27,28,41). Future studies with larger samples on children with COFD from both genders may use alternative methods of evaluations (e.g., teacher reports, parent reports, sociometry etc.) to better discern the effects of gender on peer bullying.

Limitations and conclusion

Our results should be evaluated within their limitations. Firstly, the results are valid only for the treatment seeking population at the study center and may not be valid for children in the community or seeking treatment from other centers. Secondly, use of self-reports for depression/ anxiety and peer bullying only may have introduced recall and reporting bias to our study and the constructs of state/ trait anxiety may not adequately cover the range of anxiety symptoms. Employment of multiple informants by using various methodologies (i.e., broad band parent/ teacher reporting scales, parent/ teacher reports of depression/ bullying and anxiety, peer nominations for popularity/bullying/victimization) and use of clinical interviews (e.g., K-SADS-PL) may have enriched our results. Third we did not evaluate the effects of comorbid psychopathologies among children with COFD. Lastly, the male preponderance in our study may limit our analyses on effects of gender and future studies should enroll larger samples of children with COFD from both genders. Regardless of those limitations

our study supports and extends the results of previous studies (15, 20, 22,23) suggesting that risk of peer bullying may be elevated in COFD regardless of gender and symptom severity. Standardized trainings/practices should be introduced among Turkish speech and language therapists to screen and address peer bullying, depression and anxiety among children with COFD in collaboration with child and adolescent psychiatrists as suggested previously (21).

Acknowledgement: We would like to acknowledge the Department of Pediatrics of Mehmet Akif İnan Education and Research Hospital who supported the control group participation in this research.

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