

**ORIGINAL ARTICLE: OUTCOMES**

Psychiatric comorbidity in children with psychogenic and functional breathing disorders

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Abstract

Background: The present study aims to assess psychiatric diagnoses in children with psychogenic and functional breathing disorders (PFBD), which consist of children with psychogenic cough, throat-clearing tics, and sighing dyspnea, and compare them to a control group without any diagnosis of chronic medical problems.

Methods: The participants consist of 52 children with PFBD and 42 children without any chronic medical problems. Psychiatric diagnoses were assessed via semistructured psychiatric interviews in both groups.

Results: The two groups did not differ on age (PFBD group 11.25 ± 2.61 , control group 11.17 ± 2.58 ; $t = 0.14$, $P = .88$) or sex (48.1% of the PFBD group were female, 61.9% of the control group were female; $\chi^2 = 1.79$, $P = .18$). 55.8% of the PFBD group and 28.6% of the control group had at least one psychiatric diagnosis according to the semistructured interviews ($\chi^2 = 6.99$, $P = .008$). The most common psychiatric diagnoses in the PFBD group were attention deficit hyperactivity disorder (ADHD; 17.3%), tic disorders, (15.4%), and specific phobia (15.4%). 11.5% of the cases in the PFBD group were diagnosed with somatic symptom disorder and more than half of the patients ($n = 27$ (51.9%)) showed clinical characteristics of tic disorders.

Conclusion: Psychiatric diagnoses are common in children with PFBD, and teamwork involving child psychiatrists may be essential for the management of children with PFBD.

KEYWORDS

clinical trials, epidemiology, evidence-based medicine and outcomes

1 | INTRODUCTION

Psychogenic and functional breathing disorders (PFBD) are chronic respiratory problems without an underlying disease. PFBD includes psychogenic cough, throat-clearing tics, and sighing dyspnea. Different terms are used to explain the conditions; sighing dyspnea, habit cough,¹ psychogenic cough,² somatoform respiratory disorders,³ somatic cough syndrome,⁴ and tic cough.⁵ There are some

controversial diagnostic criteria for PFBD. Some of the authors suggest that characteristics of the cough, such as honking and the barking nature of the cough, are essential for diagnosis. However, others propose that a lack of symptoms during sleep is a marked characteristic for diagnosis.⁶ Guidelines recommend diagnosis should take place after thorough physical evaluation and ruling out other possible causes. Due to insufficient evidence, diagnosing PFBD is a complex issue in which there is a lack of clinical consensus.

Children with PFBD undergo frequent examinations and different kinds of medication before the diagnosis can be made. Moreover, many children may skip school because of problems caused by the

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symptoms of PFBD.⁶ Early diagnosis may lead to a more favorable outcome. Treatment recommendations include suggestion and hypnosis. However, recent literature suggests that simple reassurance may be an adequate treatment option.⁶ A thorough diagnosis is important.

Psychiatric comorbid diagnoses of children with PFBD were investigated by numerous studies. Most of these studies were among children with psychogenic cough and the results were highly variable. However, none of these studies used semistructured diagnostic assessment tools. Additionally, none of the studies had a control group from a similar population. Studying psychiatric diagnoses may contribute to the elimination of diagnostic confusion as well as the improvement of treatment. On the contrary, psychiatric diagnoses in children with other psychogenic respiratory disorders (such as sighing dyspnea and throat-clearing) are not given any serious attention in the literature.

Another aim of the study is to examine somatic symptom disorders and tic disorders in children with PFBD. As far as we know, there are no studies which focus on these diagnoses after the CHEST guideline and expert panel report.⁵ The guideline proposed the diagnosis of tic disorder for habit cough and somatic symptom disorder for psychogenic cough according to the *DSM-5*. The evaluation of the criteria of disorders may shed light on the literature on that issue.

The aims of the present study are:

- To evaluate comorbidity (psychiatric diagnoses) in children with PFBD and compare them to an age- and sex-matched control group from a similar population.
- To examine somatic symptom disorders and tic disorders criteria (according to the *DSM-5*) which are proposed psychiatric diagnoses for psychogenic respiratory disorders according to The American College of Chest Physicians.

2 | METHODS

The study sample consisted of 52 children with PFBD and 42 children as the control group without any chronic medical problems. Among the children with PFBD, 12 patients had sighing dyspnea, 11 patients had throat-clearing tics and 29 patients had psychogenic cough. All of the participants were recruited from Bezmialem Vakif University Hospital pediatrics clinics. The subjects with psychogenic cough were examined and diagnosed by pediatric pulmonary specialists and child and adolescent psychiatrists. Children with PFBD were referred to the pediatric pulmonology clinic between January 2017 and November 2018. A total number of 58 children diagnosed with PFBD during the duration of the study. Four subjects refused to participate in the study and two subjects did not attend their psychiatry appointments. Chest X-Rays were performed on all of the PFBD groups, and a pulmonary function test was performed on children over the age of 7 (98.1% of the PFBD group). Additionally, a skin prick test, flexible bronchoscopy, and sweat chloride test were conducted on the selected patients whenever needed. A "suspected

PFBD" diagnosis was determined when physical examination and all other investigations were normal.⁵ Subjects were diagnosed as PFBD after psychiatric evaluation. The control group included age- and sex-matched children without any chronic health problems, who were recruited from pediatric clinics between July and August (2018). The children provided verbal assent, and written consent was obtained from their parents. The study was approved by Bezmialem University Ethics Committee (date: 22 October 2018; number: 16064).

2.1 | Measures

The sociodemographic information was collected via a form developed by authors of the study. The form consisted of questions about parental age, education, physical and psychiatric history of parents and siblings, marital status, and annual income. Parental education was measured by the highest level of schooling the parents had, family status was assessed by whether the children came from divorced or intact families and psychiatric history assessed whether the parents had previously consulted a child psychiatrist for their child or not.

The clinical characteristics of children with psychological breathing problems were evaluated by a form developed based on the current literature. The form evaluated types of problems, duration of problems, factors affecting the problems, secondary gains, and other clinical features as well as treatment variables.

2.2 | Kiddie schedule for affective disorders and schizophrenia present version

As a semistructured diagnostic interview based on *DSM-IV* criteria,⁷ and recently updated to *DSM-5*,⁸ the Kiddie schedule for affective disorders and schizophrenia present version covers a wide range of psychiatric conditions (mood disorders, schizophrenia, anxiety disorders, obsessive-compulsive disorder, tic disorders, elimination disorders, eating disorders, disruptive disorders, substance use disorders, adjustment disorders, and posttraumatic stress disorder). The psychiatric interview was performed with both parent and child. The interview began with questions about developmental history and functioning, thereafter, both parent and child answered screening questions about each diagnosis. If the child and parent both gave positive answers to the screening questions, a detailed evaluation was performed to determine whether the child fulfilled the diagnostic criteria of the disorder. The Turkish version of the instrument was used, which was found to be valid and reliable.⁹

2.3 | Statistical analysis

All the analyses were conducted using SPSS for Windows (v. 20; SPSS Inc, Chicago, IL). Descriptive variables included frequency, mean and standard deviation for each variable (SD). Categorical variables were compared with the χ^2 test. The Fisher exact test was used where needed. Continuous variables were compared with the Student *t* test or the Mann-Whitney *U* test. Test significance level was set at $P \leq .05$.

3 | RESULTS

The mean age of the PFBD group was 11.25 ± 2.61 years and the control group was 11.17 ± 2.58 years ($t = 0.14$, $P = .88$). 48.1% of the PFBD group and 61.9% of the control group were female ($\chi^2 = 1.79$, $P = .18$). Other sociodemographic variables did not differ between the two groups (Table 1).

3.1 | Clinical characteristics of children with PFBD

Amongst the PFBD group, there was sighing dyspnea in 12 subjects, throat-clearing in 11 subjects, and cough in 29 subjects. More than half of the group reported that their problems had continued for less than 6 months. Clinical characteristics of the children are displayed in Table 2. The duration of problems were categorized as follows: 0 to 1 months (12 [23.1%]), 1 to 3 months (15 [28.8%]), 3 to 6 months (8 [15.4%]), 6 to 12 months (3 [5.8%]), and more than 12 months (9 [17.3%]).

3.2 | Psychiatric disorders

55.8% of the PFBD group and 28.6% of the control group had at least one psychiatric diagnosis according to the semistructured interview and the difference was statistically significant ($\chi^2 = 6.99$, $P = .008$). The most prevalent disorders in the PFBD group were ADHD (17.3%), tic disorders (15.4%) and specific phobia (15.4%). Both internalizing and externalizing disorders were more common in the psychogenic cough group, but the difference was not statistically significant (Table 3).

3.3 | Tic disorders

Eight subjects had a diagnosis of tic disorder in the clinical examination, and a further three cases reported to have had a

lifetime tic disorder which was not present in the time of the examination. Additionally, suppressibility, distractibility, suggestibility, variability, and the presence of a premonitory sensation were present in 17 (34.7%), 19 (40.4%), 16 (30.8%), 32 (68.1%), and 27 (51.9%) patients, respectively. More than half of the patients ($n = 27$ [51.9%]) showed at least three of the clinical characteristics of tic disorders. However, only nine (17.3%) of the cases reported that their symptom had remained for more than 1 year which is one of the criteria for both Tourette's disorder and Persistent (Chronic) motor or vocal tic disorder. Therefore, provisional tic disorder may be the diagnosis of nearly half of the cases.

3.4 | Somatic symptom disorder

The PFBD group was assessed to determine whether they fulfill the criteria of somatic symptom disorders, according to DSM-5. The rate of the subjects fulfilling the 1, 2, and 3 criteria of the disorders was 100%, 11.5%, and 28.8%, respectively. Therefore, six (11.5%) of the cases had somatic symptom disorder, and only three (5.8%) of the patients were diagnosed with persistent somatic symptom disorder. Half of the patients who were diagnosed with a somatic symptom disorder were reported to have additional symptoms such as fatigue and lack of appetite.

4 | DISCUSSION

To the best of our knowledge, the current study is the first study comparing psychiatric disorders in children with PFBD to an age- and sex-matched control group from a similar population with a semistructured diagnostic interview. The results of the study showed that more than half of the subjects with PFBD had at least one psychiatric disorder which was significantly higher than the control

TABLE 1 Sociodemographic variables of PFBD and control group

	PFBD (n = 52)	Control (n = 42)	Statistics
Age (mean \pm SD, range), y	11.25 ± 2.61 (6-17)	11.17 ± 2.58 (6-17)	$t = 0.14$, $P = .88$
Sex (M/F)	27/25	16/26	$\chi^2 = 1.79$, $P = .18$
Paternal age (mean \pm SD), y	41.04 ± 5.32	39.80 ± 8.08	$z = 0.87$, $P = .38$
Paternal education (\geq high school), n (%)	28 (53.8)	24 (58.5)	$\chi^2 = 0.21$, $P = .65$
Maternal age (mean \pm SD), y	37.14 ± 5.66	36.69 ± 4.83	$z = 0.27$, $P = .79$
Maternal education (\geq high school), n (%)	20 (39.2)	20 (47.6)	$\chi^2 = 0.66$, $P = .42$
Income (\geq 6000\$/y), n (%)	19 (36.5)	17 (40.5)	$\chi^2 = 0.15$, $P = .70$
Family status (intact family), n (%)	50 (96.2)	42 (100)	$P = .50^*$
Number of children	1.81 ± 0.40	1.93 ± 0.26	$t = 1.68$, $P = .093$
Psychiatric referral of the child, n (%)	19 (36.5)	12 (28.6)	$\chi^2 = 0.67$, $P = .41$
Medical illness in family, n (%)	22 (42.3)	14 (33.3)	$\chi^2 = 0.79$, $P = .37$
Psychiatric disorder in family, n (%)	14 (26.9)	12 (28.6)	$\chi^2 = 0.32$, $P = .86$
Allergy in family, n (%)	20 (39.2)	17 (40.5)	$\chi^2 = 0.015$, $P = .90$

Abbreviations: F, female, M, male; PFBD, psychogenic and functional breathing disorders.

*Fischer's exact test.

TABLE 2 Clinical characteristics of children with psychogenic and functional breathing disorders

Problem of psychogenic breathing	Sighing dyspnea (N = 12)	Throat cleaning (N = 11)	Psychogenic cough (N = 29)	Total (N = 52)
Duration of problem (>6 m), n (%)	5 (41.7)	5 (45.4)	5 (17.2)	15 (28.8)
Lack of symptoms during sleep, n (%)	9 (75.0)	9 (81.8)	18 (62.1)	36 (72.0)
Less symptoms during school, n (%)	2 (16.7)	4 (36.4)	7 (24.1)	13 (25.0)
Less symptoms during a social activity, n (%)	4 (33.3)	6 (54.5)	4 (13.8)	14 (29.2)
Less symptoms during exercise, n (%)	4 (33.3)	5 (45.5)	23 (79.3)	32 (66.7)
School absence because of symptoms, n (%)	3 (25.0)	6 (54.5)	14 (50.0)	23 (46.9)
Secondary gains because of symptoms, n (%)	2 (16.7)	2 (18.2)	14 (48.3)	18 (36.0)
Suppressibility, n (%)	5 (41.7)	4 (36.4)	8 (27.6)	17 (34.7)
Variability, n (%)	5 (41.7)	6 (54.5)	21 (72.4)	32 (68.1)
Premonitory sensation, n (%)	6 (50.0)	8 (72.7)	13 (44.8)	27 (51.9)
Distractibility, n (%)	3 (25.0)	5 (45.5)	11 (37.9)	19 (40.4)
Suggestibility, n (%)	6 (50.0)	3 (27.3)	7 (24.1)	16 (30.8)
Fatigue, n (%)	3 (25.0)	7 (63.6)	10 (34.5)	20 (38.5)
Lack of appetite, n (%)	3 (25.0)	4 (36.4)	6 (20.7)	13 (25.0)
Headache, n (%)	4 (33.3)	2 (18.2)	9 (31.0)	15 (28.8)
Nausea, n (%)	3 (25.0)	0	6 (20.7)	9 (17.3)

TABLE 3 Psychopathologies in PFBD and control group

	Sighing dyspnea (N = 12)	Throat cleaning (N = 11)	Psychogenic cough (N = 29)	PFBD (n = 52)	Control (n = 42)	Statistics PFBD vs control
Int dis, n (%)	3 (25.0)	5 (45.5)	10 (34.5)	18 (34.6)	8 (19.0)	$\chi^2 = 2.81, P = .093$
Dep dis	1 (8.3)	2 (18.2)	2 (6.9)	5 (9.6)	1 (2.4)	$P = .22^*$
GAD	1 (8.3)	2 (18.2)	3 (10.3)	6 (11.5)	2 (4.8)	$P = .29^*$
SAD	1 (8.3)	1 (9.1)	4 (13.8)	4 (7.7)	3 (7.1)	$P = 1.00^*$
SoAD	0	1 (9.1)	3 (10.3)	6 (11.5)	3 (7.1)	$P = .73^*$
SP	1 (8.3)	2 (18.2)	5 (17.2)	8 (15.4)	4 (9.5)	$P = .54^*$
PD	0	2 (18.2)	2 (6.9)	4 (7.7)	0	$P = .13^*$
Anx OS	1 (8.3)	0	0	1 (1.9)	2 (4.8)	$P = .59^*$
OCD	0	1 (9.1)	1 (3.4)	2 (3.8)	0	$P = .50^*$
Elm dis, n (%)	1 (8.3)	0	2 (6.9)	3 (5.8)	1 (2.4)	$P = .63^*$
Enuresis	1 (8.3)	0	2 (6.9)	3 (5.8)	1 (2.4)	$P = .63^*$
Encopresis	0	0	0	0	0	0
Ext dis, n (%)	0	3 (27.3)	7 (24.1)	10 (19.2)	2 (4.8)	$P = .059^*$
ADHD	0	2 (18.2)	7 (24.1)	9 (17.3)	2 (4.8)	$P = .10^*$
ODD	0	1 (9.1)	2 (6.9)	3 (5.8)	0	$P = .25^*$
CD	0	0	0	0	0	0
Tic disorders, n (%)	2 (16.7)	1 (9.1)	5 (17.2)	8 (15.4)	2 (4.8)	$P = .18^*$
Total, n (%)	6 (50.0)	7 (63.6)	16 (55.2)	29 (55.8)	12 (28.6)	$\chi^2 = 6.99, P = .008$

Abbreviations: ADHD, attention deficit hyperactivity disorder; Anx OS, anxiety disorders other specified; CD, conduct disorder; Dep dis, depressive disorders; Elm dis, elimination disorders; Ext dis, externalizing disorder; GAD, generalized anxiety disorder; Int dis, internalizing disorders; OCD, obsessive-compulsive disorder; ODD, oppositional defiant disorder; PD, panic disorder; PFBD, psychogenic and functional breathing disorders; SAD, separation anxiety disorder; SoAD, Social anxiety disorder; SP, specific phobia.

*Fischer's exact test.

subjects. A study from India reported that the rate of comorbid psychiatric disorders was 62.5%, and the most prevalent disorder was conversion disorder (21.8%) according to ICD-10.¹⁰ A study from the United States of America reported that 11% of the children with

psychogenic cough had conversion disorder and 2% of them had anxiety disorders.¹¹ Our results indicated higher comorbidity with externalizing psychiatric disorders than other studies in the literature. More studies with structured interviews are required to

gain a proper understanding of psychiatric morbidity in children with psychogenic cough.

The clinical characteristics of the three PFBD groups showed some similarities and differences, although no distinctive pattern across different diagnostic groups was noted. There were subtle differences in clinical characteristics of symptoms across the subgroups of PFBD (psychogenic cough, throat-clearing tics, and sighing dyspnea). Associated functional symptoms were more common in our study than a recent study from the United Kingdom. However, school absence rates were similar in both studies.⁶ Lack of symptoms during sleep is lower in our sample than the aforementioned study. It should be noted that we simply asked the children and caregivers for nocturnal symptoms, and we did not perform an objective evaluation of a cough count at sleep. A case study showed a lower cough count at sleep than wakefulness, but our findings are in accordance with the case study¹² and the CHEST guideline.⁵

According to the literature on psychomorbidity among children with chronic cough, anxiety, and depression may be seen either as a result of the chronic cough^{13,14} or may exacerbate the symptoms.^{4,15} Interestingly, ADHD was the most prevalent disorder in the PFBD group, followed by tic disorders and specific phobias. Therefore, our results showed that there is an increased risk of psychomorbidity in children with PFBD, but the relationship is complex and not pointing to a specific disorder. However, there is a tendency for children with PFBD to have high rates of psychiatric disorders including both internalizing and externalizing disorders. It is unclear whether there is a causal relationship between psychiatric disorders and PFBD. Additionally, the control group did not consist of children with a known respiratory disorder such as asthma, which was shown to be associated with increased psychomorbidity.¹⁶ Thus the findings of the present study did not show a difference from a known breathing disorder and a functional/psychogenic breathing disorder. Future studies recruiting children with a known respiratory disorder as the control group may shed light on this issue. Longitudinal studies with larger samples are needed to show whether there is a causal relationship between psychiatric disorders and PFBD.

An interesting finding of the present study is the difference in psychiatric diagnoses across different subgroups of PFBD. Specifically, more than half of the children with psychogenic cough and throat-clearing had externalizing disorders. On the contrary, none of the children with sighing dyspnea had externalizing disorders. In the present study, we combined psychogenic cough and functional breathing disorders into a single group, similar to the work of Niggermann,¹⁷ because the studies of psychiatric disorders in children with PFBD are not yet sufficient to reach a consensus on the methodology. However, to overcome the heterogeneity of subgroups, separate columns in tables as functional and psychogenic breathing disorders were made. More research is needed to enlighten the difference across subgroups of PFBD further.

According to the clinical examinations, more than half of the patients showed at least three characteristics of a tic disorder which are suppressibility, distractibility, suggestibility, variability, and the presence of a premonitory sensation. Thus, our clinical examination

results were in line with the CHEST guideline.⁵ High lifetime diagnosis of tic disorders and rates of comorbid diagnosis of ADHD were in line with a diagnosis of tic disorder.¹⁸ However, higher rates of treatment of suggestion¹⁹ and hypnosis¹¹ are not the case in children with tic disorders.²⁰ High rates of symptom relief in 1 year may be in accordance with a diagnosis of a provisional tic disorder.⁶ Functional brain imaging studies may shed light on the involved brain areas.

More than one-tenth of the cases have fulfilled the diagnostic criteria of somatic symptom disorder. Half of those who fulfilled this criteria had additional symptoms such as fatigue, headache, and lack of appetite. It is well-demonstrated in the literature that children with somatic symptom disorder have more unexplained physical symptoms,²¹ and a lower level of functioning.²² Interestingly, most children did not show the second diagnostic criteria of somatic symptom disorder which is "excessive thoughts, feelings, or behaviors related to the somatic symptoms." Parents seemed to be more worried about the symptoms than their children. More studies are needed to show the differences in the clinical characteristics of the cases who have somatic symptom disorder.

The present study has several limitations. The sample size is one of the most considerable limitations. Validity was compromised due to the small sample size of subgroups, hence statistical analysis was not performed between subgroups. The cross-sectional nature of the present study decreases the ability to infer a causal relationship between variables. The sample was recruited from one center in Istanbul, which may deter generalizability of the findings. However, Bezmialem University Hospital is a referral center for patients from all counties of Istanbul as well as the surrounding cities. To the best of our knowledge, the present study is the first study to evaluate psychiatric disorders and characteristics of tic disorder and somatic symptom disorder according to a semistructured interview in a population of PFBD. More studies with different countries are needed to clarify the clinical characteristics of cases.

5 | CONCLUSIONS

The present study found that children with PFBD had more psychiatric disorders than the control participants. In this study, we suggest that different subgroups of PFBD may have a distinct psychiatric profile which needs further attention. The correct diagnosis may be provisional tic disorder in roughly half of the cases and somatic symptom disorders in a tenth of the cases. Further studies with larger samples and longitudinal methodology are needed to clarify the relationship between psychiatric diagnoses and PFBD.

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CONFLICT OF INTERESTS

The authors declare that there are no conflict of interests.

AUTHOR CONTRIBUTIONS

ACÖ, EE, HY, and EÇ contributed to planning, conducting, and reporting of the study. FUK and MAN contributed to conducting research.

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