



Research Article

Two Years of COVID-19 Pandemic: What Psychological Impact in Primary Ciliary Diskinesia? A Prospective Study of an Italian Center

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Abstract

Pre-existing chronic disease consisted of a highly vulnerable condition during the COVID-19 outbreak. The aim of this prospective longitudinal study is to investigate the long-term impact on psychological well-being of a cohort of PCD patients from an Italian center compared to healthy subjects. The Psychological General Well-Being Index and Parenting Stress Index-Short questionnaires were filled out by PCD patients and their parents, respectively, in May 2021 and in May 2022. The percentages of PCD patients and their parents showed not significant difference in the different categories of distress level compared to healthy controls. The psychological well-being and parental stress of PCD population did not get worse during two years of the COVID-19 pandemic, unlike healthy subjects. The impairment expressed in areas of 'depression', 'self-control', 'general health' and vitality' was higher in the control group than in PCD patients, one year after the onset of COVID-19 pandemic. The COVID-19 pandemic did not have a negative impact on psychological well-being of PCD patients and their parents. These data could be explained by the constant elevated psychological distress with the ability to employ strategies to cope the stressful conditions, together an early vaccination campaign in PCD population.

Keywords: COVID-19 pandemic; severe acute respiratory syndrome due to coronavirus 2; primary ciliary dyskinesia; mental health; parental stress; psychological burden

Abbreviations:

- **PCD:** Primary Ciliary Dyskinesia
- **SARS-CoV2:** Severe acute respiratory syndrome due to Coronavirus 2

- **COVID-19:** Coronavirus Disease 2019
- **PSI-SF:** Parenting Stress Index – Short Form
- **PD:** Parental Distress
- **P-CDI:** Parent-Child Dysfunctional Interaction
- **DC:** Difficult Child
- **DEF:** Defensive Responding

- **PGWBI:** Psychological General Well-Being Index

Introduction

The Coronavirus disease 2019 (COVID-19) pandemic, caused by the novel coronavirus SARS-CoV-2, is the first global pandemic of the twenty-first century [1]. The COVID-19 pandemic had relevant economic and social implications, as well as being a public health emergency. In the absence of an effective biological cure or vaccine against the virus, social distancing and hygiene were the main strategies to avoid contagion. However, the lack of social interaction in addition to the fear of infection, the unemployment, the stress, and the mass panic, exacerbated many determinants of poor mental health.

The psychological impact of the pandemic on the general population has been increasingly reported in the scientific literature. A global increase in prevalence of both anxiety and major depressive disorders, with an additional 76.2 million and 53.2 million cases, respectively, was estimated [2].

Despite the mild physical illness of pediatric SARS-CoV-2 infection [3], studies about the mental health of children and adolescents during the first year of the COVID-19 pandemic, reported higher prevalence of depressive and anxiety symptoms in comparison with pre-pandemic estimates, as well as an increased prevalence of suicidal ideation, suicide, and non-suicidal self-injury [4]. However, data on the effects of the pandemic on children's and adolescents' mental health are limited and contradictory, and long-term effects remain to be clarified.

Primary Ciliary Dyskinesia (PCD) is a rare disease of motile cilia dysfunction, with wide genetic and clinical heterogeneity [5, 6]. The progression of the disease is highly variable, with some patients having good lung function, relatively few respiratory infections and good quality of life in adulthood, while in other cases progressive decline in lung function and eventually respiratory failure are reported [7]. PCD is a chronic disorder with potential psychological effects on the intra-familial relationships because of the frequent medical consultations, the patients' perception of being sick and the possible effects of negation and rejection of disease by the patients and/or their parents.

In a previous study, we evaluated the psychological impact of the COVID-19 pandemic on both children and adult patients with PCD, during the first 2 months of the COVID-19 outbreak in Italy (March 2020– May 2020), over the general lockdown period [8]. Among PCD parents, only 20% showed high levels of parental stress. Compared with healthy subjects, the PCD population did not show a significantly different psychological burden or parental stress during quarantine [8].

However, the COVID-19 pandemic lasted for more than two years with relevant economic and social implications. This prospective longitudinal study aimed to investigate the long-term

impact on psychological well-being of a PCD population from an Italian center and to assess whether the patients continued to maintain an adequate level of mental health.

Materials and Methods

A prospective-longitudinal study was conducted at the Department of Translational Medical Science, Pediatric section, University “Federico II”, Naples – Italy. The same group of PCD patients, and their mothers, included in our previously published report [8], were enrolled.

All participants signed informed consent. The study was conducted in accordance with the Declaration of Helsinki for Human Research and approved by the Ethics Committee of the Federico II University of Naples (Protocol No. 275/20).

The primary outcome was the assessment of psychopathological well-being over a two-years period, namely in May 2021 (defined as T1) - May 2022 (defined as T2), one year and two years since the onset of outbreak, respectively. The results were compared with those obtained from our previous study in May 2020 (defined as T0) [8].

The inclusion criteria were a) PCD diagnosis, according to the European Respiratory Society guidelines [9], b) active follow-up for at least 12 months, 3) participation in the previous study conducted at our center [8], 4) adherence to the study protocol after informed consent signature. The exclusion criteria were a) PCD diagnosis made in 2020, b) concomitant chronic diseases or psychiatric/neurodevelopmental disorders, c) incomplete informations availability, d) inability to give informed consent. PCD patients were remotely enrolled through telephone contact.

A questionnaire for assessing psychological stress level during COVID-19 pandemic was sent to PCD patients, or their parents, by mail. The participants were divided in two group, Group A and Group B, according to the division of our previously published report [8]: the subjects which were part of Group A in our previously study [8] were enrolled in the same group, regardless of age (six patients in the Group A turned 15 years old during the study time). Therefore, all subjects' mothers, who were part of Group A in our previous study [8], filled out the Parenting Stress Index-Short Form questionnaire (PSI-SF) [10], adapted to the Italian [11]; in the same way, all PCD patients, who were previously part of Group B [8] filled out the Psychological General Well-Being Index questionnaire (PGWBI) [12], Italian version.

Age- and sex-matched healthy controls, who were part of previous study [8] were enrolled. They filled out the PGWBI and PSI-SF questionnaires according to the previous division [8].

For the description of the questionnaires, the psychometric characteristics, and the methods of interpretation of their results, see our previous article [8].

Data Analysis

Statistical analyses were made with Graphpad Software, version 6.0.1 (San Diego, CA, USA). Fisher's test was used to assess longitudinal difference in psychometric characteristics, investigated with PGWBI and PSI-SF questionnaires, in both study group and healthy controls as well as between the two groups. Categorical characteristics were showed as mean \pm SD and compared by unpaired t-Test, p-value was two-sided and $p < 0.05$ was considered statistically significant.

Results

The Group A consisted of 10 PCD patients [60% males, mean age 14.2 years (SD 2.3)] and the Group B consisted of 17 PCD patients [53% males, mean age 29.4 years (SD 12.04)]. Within the control population, the PSI-SF was filled out by the mothers of 10 healthy subjects [70% males; mean age 10.8 years (SD 5.3)] and PGWBI was filled out by 17 healthy subjects [65% males; mean age 35.2 years old (SD 15.45)]. All participants completed the questionnaires at T1 and at T2 time.

Parental report of Psychopathological Well-Being by PSI-SF

At T1, 4/10 patients of Group A (40%) presented 'non-pathological stress' (score (s) $<$ 50th percentile), 5/10 (50%) showed 'symptomatic stress' (50th $<$ s $<$ 85th percentile) and only 1/10 case (10%) 'high stress' (s $>$ 85th percentile). In the healthy controls, 7/10 cases (70%) presented 'symptomatic stress' (50th $<$ s $<$ 85th percentile). In the remaining cases, 3/10 subjects of control group (30%) presented 'non-pathological stress' (s $<$ 50th percentile). When the percentages of PCD patients (Group

A) and control group subjects for each stress categories (high; symptomatic stress; non-pathological stress) were compared, no significant statistically differences were found.

At T2, 3/10 patients of Group A (30%) presented 'non-pathological stress' (s $<$ 50th percentile); the remaining cases, 5/10 (50%) showed 'symptomatic stress' (50th $<$ s $<$ 85th percentile) and 2/10 patients (20%) 'high stress' (s $>$ 85th percentile). In the healthy controls, 5/10 subjects (50%) presented 'symptomatic stress' (50th $<$ s $<$ 85th percentile), 4/10 (40%) 'high stress' (s $>$ 85th percentile) and only one subject 1/10 (10%) showed 'non-pathological stress' (s $<$ 50th percentile). The percentage of Group A patients and healthy controls with different categories of stress levels (high; symptomatic stress; non-pathological stress) were not significantly different.

The raw scores of PSI-SF subscales (PD, P-CDI, DC and DEF) and PSI-SF total stress score showed no significant differences between PCD patients' mothers (Group A) and controls at T1 and at T2.

In Group A, comparing the PSI-SF subscales and total stress scores obtained at T0, T1 and T2, no significant differences were found (Figure 1). Otherwise, in the healthy controls, statistically significant differences were found in PD subscale score between T0-T2 ($p = 0.02$); in P-CDI subscale score between T0-T1 ($p = 0.04$) and between T0-T2 ($p = 0.0005$); in DC subscale score between T0-T1 ($p = 0.005$) and between T0-T2 ($p = 0.002$); in DEF subscale score between T0-T2 ($p = 0.0213$), and in total stress score between T0-T1 ($p = 0.009$) and T0-T2 ($p = 0.0009$) (Figure 1).

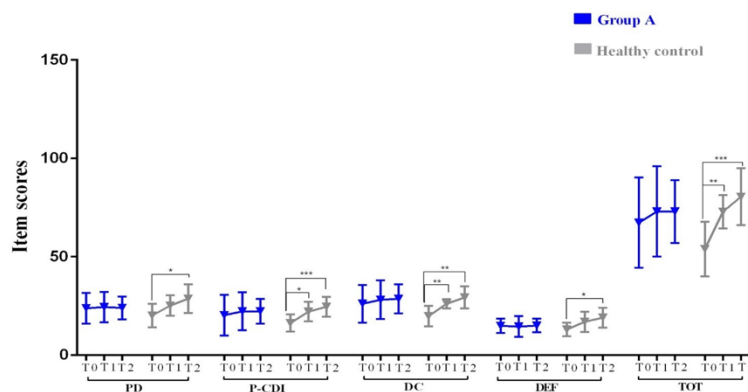


Figure 1: Distribution of PSI-SF mean scores of subscales and total stress score in PCD mothers and healthy controls in the different periods of the COVID-19 pandemic. The parenting stress index-short form questionnaire (PSI-SF) is organized into four subscales: parental distress (PD); parent-child dysfunctional interaction (P-CDI); difficult child (DC); defensive responding (DEF) and Total Stress score (TOT). The PSI-SF questionnaire was filled out by the Group A patients' mothers (n = 10) and by the mothers of healthy controls (n = 10). After the first assessment (May 2020 - defined as "T0"), the evaluation was performed in May 2021 (defined as "T1") and in May 2022 (defined as "T2"), after 1 year and 2 years of pandemic, respectively. Data are expressed as mean and Standard Deviation. Each triangle (blue and gray for Group A patients and controls, respectively) represents the average scores obtained for each period (T0; T1; T2). Paired t-test was performed. * $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$.

Subjective psychological well-being by PGWBI.

At T1, 8/17 Group B patients (47%) presented ‘no distress’ ($s > 72$), 6/17 (35%) showed ‘moderate distress’ ($60 < s < 72$) and 3/17 patients (18%) ‘severe distress’ ($s < 60$). In the healthy controls, 3/17 subjects (17%) presented ‘no distress’ ($s > 72$), while 9/17 (52%) and 5/17 (29%) showed a total score indicating ‘moderate distress’ ($60 < s < 72$) and ‘severe distress’ ($s < 60$), respectively. Percentages of PCD patients and healthy controls with different categories of PGWBI total scores (i.e., severe; moderate; no distress) were not significantly different (Table 1).

At T2, the results of the PGWBI questionnaire showed that 11/17 Group B patients (65%) showed ‘no distress’ ($s > 72$), and 2/17 (12%) ‘severe distress’ ($s < 60$). In the healthy controls, 9/17 subjects (53%) presented ‘no distress’ ($s > 72$), while 5/17 (29%) and 3/17 (18%) showed ‘moderate distress’ ($60 < s < 72$) and ‘severe distress’ ($s < 60$), respectively (Table 1). Percentages of PCD patients and healthy controls with different total scores of PGWBI (i.e., severe; moderate; no distress) were not significantly different (Table 1).

	T1			T2		
	Group B	Healthy control	<i>p</i>	Group B	Healthy control	<i>p</i>
Severe distress ($s < 60$)	3 (18) [†]	7 (41) [†]	0.26	2 (12) [†]	3 (18) [†]	0.60
Moderate distress ($60 < s < 72$)	6 (35) [†]	8 (47) [†]	0.73	4 (23) [†]	5 (29) [†]	1.0
No distress ($s > 72$)	8 (47) [†]	2 (12) [†]	0.1	11 (65) [†]	9 (53) [†]	1.0
<i>PGWBI scores</i>						
Anxiety	16.3 (5.2) [*]	16.5 (3.2) [*]	0.90	17.7 (5.7) [*]	15.9 (3.6) [*]	0.10
Depression	12.3 (2.0) [*]	10.6 (0.9) [*]	0.002	12.7 (2.0) [*]	12.1 (1.8) [*]	0.28
Well-being	11.8 (4.0) [*]	9.8 (2.1) [*]	0.08	12.5 (2.8) [*]	10.7 (3.3) [*]	0.12
Self-control	11.0 (3.1) [*]	8.0 (3.4) [*]	0.01	12.1 (3.0) [*]	10.7 (2.3) [*]	0.34
General Health	11.3 (2.3) [*]	8.9 (1.6) [*]	0.002	12.1 (3.0) [*]	11.6 (1.6) [*]	0.45
Vitality	13.6 (3.6) [*]	9.8 (2.5) [*]	0.001	13.7 (3.3) [*]	11.2 (4.2) [*]	0.07
PGWBI Total score	76.2 (16) [*]	66.0 (13.2) [*]	0.009	80.8 (16) [*]	73.3 (14.7) [*]	0.07

Table 1: Results of PGWBI questionnaire filled out by PCD patients and healthy controls during COVID-19 pandemic. 17 PCD patients (Group B, the same studied subjects in our previously published report [8]) filled out the Psychological General Well-Being Index questionnaire (PGWBI) in May 2020 (T0); in May 2021 (T1) and in May 2022 (T2). We compared the PGWBI total and subscale scores between the three studied times (T0-T1, T0-T2 and T1-T2). Data are expressed as mean and Standard Deviation (in parenthesis). A *p*-value < 0.05 was considered significant. † Expressed as number of patients or controls and percentage in parenthesis. * Expressed as mean and Standard Deviation in parenthesis. Unpaired t-test was administered. Abbreviations: PGWBI, psychological general well-being index.

When we considered the percentile score of PGWBI subscales, at T1, the number of PCD patients (Group B) for the subscale of ‘depression’ was statistically different than the number of healthy controls ($p=0.006$) (Figure 2), with a major number of healthy subjects that presents clinically significant symptoms of depression compared to PCD. Otherwise, at T2, statistically significant differences were found in the number of PCD and healthy subjects for ‘anxiety’ and ‘self-control’ subscales ($p=0.0184$ and $p=0.044$ respectively), with a major number of healthy subjects that presents clinically significant symptoms of anxiety and a worst sense of self-control compared to PCD.

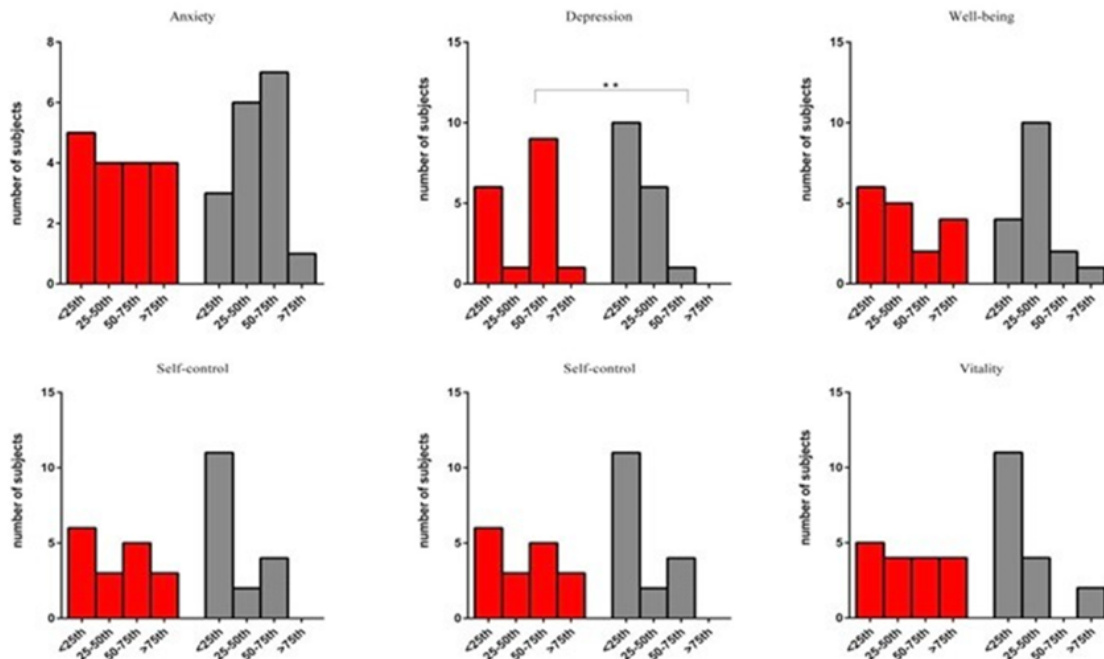


Figure 2: Distribution of Group B patients and healthy controls in the classes of percentile of PGWBI subscales at T1. The Psychological General Well-Being Index (PGWBI) questionnaire was filled out by Group B patients ($n = 17$) and healthy controls ($n = 17$) in the different periods of COVID-19 pandemic. The assessment of psychopathological well-being was evaluated in May 2021 (defined as ‘‘T1’’), after 1 year of pandemic. The PGWBI is organized in six subscales: anxiety; depression; well-being; self-control; general health; vitality. Classes of percentile were defined for subscales, according to gender and age, as follows: <25th percentile: ‘severe distress’; between 25th and 50th percentile: ‘moderate distress’; between 50th and 75th percentile: ‘no distress’; >75th percentile: ‘positive well-being’. Red columns indicate Group B patients and gray columns indicate healthy controls. Fisher’s test was administered. ** $p < 0.01$

We compared raw subscales and total score of the PGWBI questionnaire completed by 17 patients with PCD with those of 17 healthy control subjects at time T1 and T2. We showed that at T1 time, the healthy controls had worst total score ($p=0.009$) and subscale scores exploring ‘depression’ ($p = 0.002$), ‘self-control’ ($p=0.01$), ‘general health’ ($p=0.002$), and ‘vitality’ ($p=0.001$) than Group B. The scores of the remaining subscales (‘anxiety’ and ‘well-being’) were not statistically significant different (Figure 3) (Table 1). At T2 time, no significant differences of subscale and total scores PGWBI were found in PCD patients (Group B) compared to the controls (Figure 3) (Table 1).

In Group B, comparing the PGWBI total and subscales scores obtained at T0-T1-T2, not statistical significantly differences were found (Figure 2). Otherwise, in healthy controls, statistically significant differences were found in the subscale’s ‘depression’ between T0-T1 ($p = 0.0007$) and T1-T2 ($p = 0.01$), ‘self-control’ between T0-T1 ($p = 0.0001$) and between T1-T2 ($p = 0.02$), ‘general health’ between T0-T1 ($p = 0.001$) and between T1-T2 ($p = 0.0006$) and ‘vitality’ between T0-T1 ($p = 0.001$) (Figure 3).

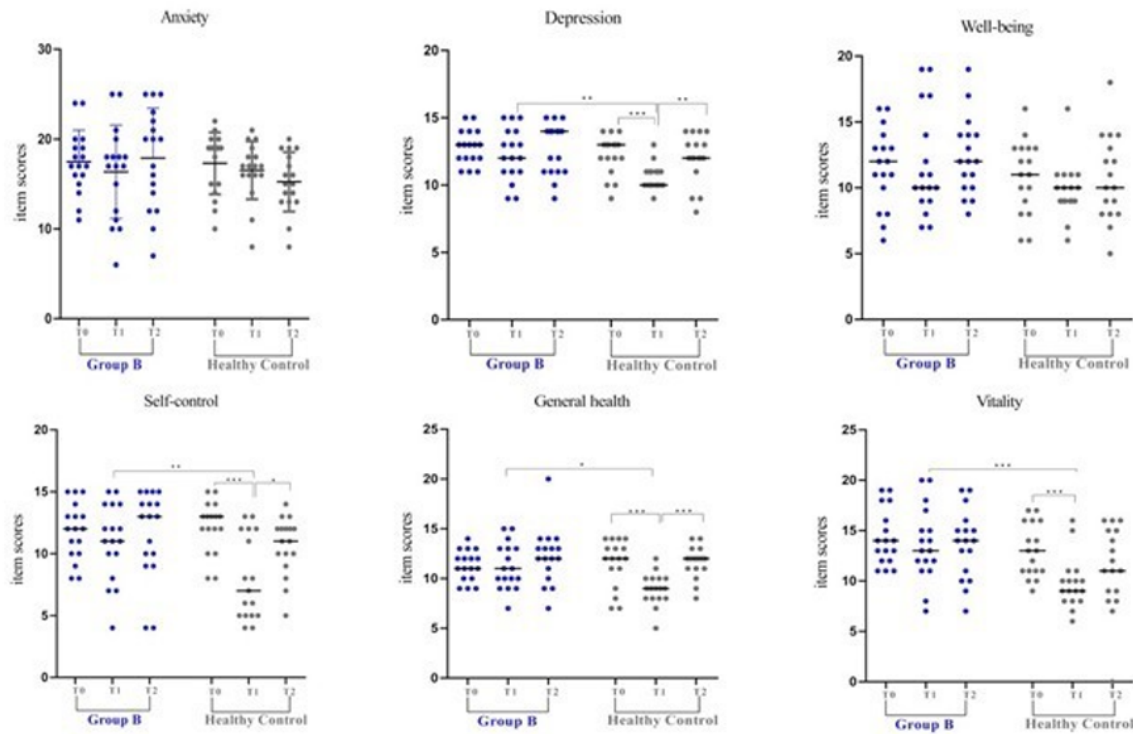


Figure 3: Distribution of PGWBI subscales scores in PCD patients and healthy control in the different periods of COVID-19 pandemic. The Psychological General Well-Being Index (PGWBI) questionnaire was filled out by PCD patients - Group B (n = 17) and healthy controls (n = 17). The PGWBI is organized in six subscales: anxiety; depression; well-being; self-control; general health; vitality. Each point (blue and gray for PCD patients and controls, respectively) represents the raw score obtained by a single subject. Paired t-test and unpaired t-test were performed. * $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$.

Discussion

The emergence of the COVID-19 pandemic brought about an environment where many crucial aspects of poor mental health are exacerbated [2]. Patients with pre-existing chronic medical conditions had to face the increased risk of severe illness by SARS-CoV-2 infection [13], on the one hand, and the fear to going to the hospital and the difficulties in routine medical treatments due to shortages of medicines and human resources in hospitals, on the other [14]. Hence, patients with chronic diseases consisted of a highly vulnerable group during the COVID-19 outbreak with the risk of the most increased negative psychological impact compared to healthy population. During the two-year period, we showed that the percentage of PCD population reporting increased emotional and stress burden was not statistically significantly different from healthy population. Moreover, the mental well-being of PCD patients and their parents did not show a worsening

over time. Supported by previously published data [15, 16], we hypothesized that these results may be explained to the constant elevated psychological distress of the PCD patients and their parent caused by the limitations related to disease. Furthermore, with the decree dated March 12, 2021, our country adopted the new national strategic plan for the prevention of SARS-CoV-2 infections with the execution of the national vaccination campaign, which redefined the population categories to be vaccinated and the priorities [17]. Patients with PCD were included in the category of “highly frail patients” due to the respiratory implications typical of the disease. Therefore, in April 2021, patients followed at our center for PCD over the age of 16 years were summoned to our facility to be vaccinated for SARS-CoV2. In PCD patients younger than 16 years old, vaccination was not yet recommended, and their parents were vaccinated. We speculated that this vaccination campaign reduced the fear of the COVID-19 infection and prevented a further worsening of perceived psychological distress.

The PSI-SF scores did not differ between PCD parents and control group. However, in Group A, stress levels did not change significantly over time (T0, T1, T2), whereas in healthy subjects' parents, stress levels showed a statistically significant increase over the different period of COVID-19 pandemic (T0, T1, T2), in agreement with previous data [18].

The literature data were ambiguous on defining parental stress levels of patients with chronic disease during the pandemic, with some studies reporting a higher stress level [19], others a lower stress levels [20], compared to the general population. We supposed that this difference could be explained by the different chronic conditions analyzed, their usual implications in daily life and the support strategies provided to families.

Regarding the evaluation of the psychological well-being during the different periods of COVID-19 pandemic (T0; T1; T2), we identified clinically significant differences between Group B patients and the control group. Comparing the scores of each PGWBI subscales, a higher impairment was expressed in areas of 'depression', 'positivity', 'well-being', 'self-control', 'general health' and vitality' in the control group than in PCD patients, one year after the onset of COVID-19 pandemic (T1). These results confirmed the data of the literature, showing that the level of depression and psychological burden increased in general population due to the epidemic related concerns [21, 22].

Early data identified adults and children with chronic disease as especially vulnerable to mental health impact of the COVID-19 pandemic [23-27]. More recent studies did not confirm this data. A longitudinal study in inflammatory bowel diseases patients showed that health-related quality of life was not related to restrictions of the COVID-19 pandemic, but rather to the course of the disease, in relation to its activity phases [28]. Moreover, an Italian study of patients with respiratory chronic disease, showed that after a year from the pandemic beginning, depression and anxiety scores were like rates described for this population before the pandemic [29]. Our results were consistent with these published data. We speculated the children with chronic respiratory disease, such as PCD, are more accustomed to care, hygienic measures and to staying at home as a protective factor for their illness. They may be better able to employ strategies to cope the stressful conditions than the healthy population and paradoxically affect their parents' levels of psychological well-being [30, 31].

Conclusions

To our knowledge, this is the first study to examine the psychological well-being and parental stress in PCD population

of an Italian center compared with healthy subjects, during the two –year COVID-19 pandemic period. In our PCD population, psychological burden did not change during the different periods of the COVID-19 pandemic, unlike the healthy control population: This data can be partly explained by the earlier start of the vaccination campaign in PCD population, falling into the category of 'frail patients', compared to healthy pediatric population. Moreover, PCD patients have a long history of managing their health-related stressors, so this stress tolerance may have facilitated their adaptation to this global threat.

It has been hypothesized that children may be particularly susceptible to the psychological effects of the COVID-19 pandemic. In this context, parents play a particularly important role in mitigating the harmful psychological effects. The literature showed that children relied on trusted adults for protection and as a reference for assessing danger and attributing meaning to events [32]. Therefore, it can be particularly destabilizing for a child to perceive that the parent is distressed and unable to prevent a traumatizing event. PCD parents maintain a stable level of stress during pandemic, conversely in general population parent stress tends to increase, underling a close link between parent-reported and child-reported distress [33]. Effective management of distress in parents can reflect positively on their children's mental health [34]. The presence of other physical health problems such as diabetes, cerebrovascular disease, heart disease and other chronic conditions represent risk factors associated with mental health problems during COVID-19. However, not all chronic conditions present the same trajectors and implications on well-being during pandemic. It is possible that PCD patients and their parents are more able to adapt to the restrictive measures dictated by the pandemic and have a greater knowledge of infection prevention measures through removal measures, frequent hand washing and the use of personal protective equipment than the general population. The studies regarding the mechanisms of induction of psychological burden, of the protective factors and of the risk factors still result scarce and show inconsistencies in literature and across reviews. Repeated assessments of mental health in general population and in vulnerable individuals are recommended to respond to current and future health crises [35].

Author Contributions: M.P.R., F.S. and M.B. designed the study. F.S., M.P.R. and M.B. drafted the manuscript. V.D.C. and C.Bi. collected and provided the patient data. M.A.M. performed the statistical analysis. The whole scheme was planned and supervised by F.S., M.B., A.C. and C.Br. F.S., M.P.R., A.C. and M.B. critically revised the manuscript. All authors have read and agreed to the published version of the manuscript.

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Institutional Review Board Statement: The study was conducted in accordance with the Declaration of Helsinki for Human Research and approved by the Ethics Committee of the Federico II University of Naples (Protocol No. 275/20).

Informed Consent Statement: Signed informed consent was obtained from all participants by email.

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Conflicts of Interest: The authors declare no conflict of interest.

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