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Examining the Relationship Between Parental Broader Autism Phenotype Traits, Offspring Autism, and Parental Mental Health

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ABSTRACT

Broader Autism Phenotype (BAP) traits may be present in parents of children on the autism spectrum. However, the prevalence and impact of these traits on parental mental health is poorly understood. We explore BAP traits and its relationship to mental health in 228 fathers and 261 mothers from a sample of 266 scholars from the EPINED study (Spain) grouped as follows: non-autism ($N=122$), autism traits ($N=93$: 38 subthreshold autism and 55 autism symptoms) and autism ($N=51$). BAP rates were higher in fathers (36.9%) than in mothers (26.1%). Moreover, BAP was significantly more frequent in fathers of autistic children (52.3%) than in fathers of children without autistic traits (28.0%), with no differences between autistic conditions groups. Mothers of autistic children exhibited higher psychological distress (36.0%) than mothers of comparison group (19.2%). Fathers with BAP obtained significantly higher scores in emotional problems than fathers without BAP. Multivariate analyses showed that, fathers' emotional problems were associated with their BAP traits, whereas in mothers they were associated with having a child with autism as well as the child's emotional dysregulation. Identifying BAP in parents of autistic conditions children can help professionals to provide specific strategies for improving the well-being of children and parents.

1 | Introduction

Understanding of autism has evolved significantly over the years, now extending beyond the obvious clinical manifestations to include a broader spectrum of associated traits and phenomena. In general terms, autism is defined by persistent differences in communication and social interaction and repetitive behaviors or restricted interests that usually manifest in early childhood (APA 2013). The influence of autism traits is observed across essential life domains, including social interactions, education

and family functioning (Kostiukow et al. 2019; Morales-Hidalgo, Voltas, and Canals 2021). With global and Spanish prevalences estimated at 1.18% and 1.53%, respectively (Morales-Hidalgo, Voltas, and Canals 2021; Talantseva et al. 2023), the study of autism is an ongoing area of research.

Autism has been considered a multifactorial condition with genetic and environmental factors that interact to determine its etiology (Arora et al. 2023). Although genetic contributions are essential and explain as much as 83% of its variability (Qiu

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Summary

- The study examined whether parents of children with autism and autism traits had more subthreshold autistic traits (termed Broader Autism Phenotype-BAP) compared to parents of children without neurodevelopmental disorders (NDD).
- We also studied the impact of BAP on parental mental health. Results indicated that fathers presented higher BAP rates than mothers, specifically fathers of autistic children.
- Additionally, mothers of autistic children reported more psychological distress than mothers of children without NDD, and fathers with BAP experienced more emotional problems compared to those without BAP.
- These findings underscore the need to identify BAP in parents to develop effective strategies for improving the well-being and quality of life of affected children and their families.

et al. 2022), environmental factors during the prenatal, perinatal and early postnatal periods have also been identified (Arafa et al. 2022; Kazantzidou et al. 2023; Nudel et al. 2022). This etiological complexity has contributed to the identification of a broader phenotype associated with autism and involved in the inheritance of several autism traits (De Groot and Van Strien 2017). The broad autism phenotype (BAP), introduced by Kanner in 1943, describes traits in first and second degree relatives of autistic individuals who do not meet the full criteria for autism but still exhibit some characteristics (Kanner, 1943). BAP provides insight into the heritability of autistic traits and aids in developing early interventions and support strategies, enhancing the well-being of both parents and children (Godoy-Giménez et al. 2021; Trevis et al. 2020). This extended phenotype can be manifested such as pragmatic language challenge (Jouravlev et al. 2020), social communication (Dučić, Kaljača, and Stojković 2022), and social interaction differences (Pruitt, Rhoden, and Ekas 2016), cognitive rigidity (Green et al. 2019), and repetitive behaviors (Rubenstein and Chawla 2018).

Research indicates that BAP is significantly more prominent in parents of children with autism (Bang et al. 2021; Dong et al. 2023; Tan et al. 2022). The prevalence of BAP presents significant variability, as was reported in a systematic review conducted by Rubenstein et al. in 2018. Prevalence varies between parents, with percentages ranging from 2.6% to 80% in fathers, from 3% to 52% in mothers, and from 5.3% to 56% in both parents (Rubenstein et al. 2018). These figures highlight the breadth of the presence of BAP in the relatives of autistic individuals and emphasize the diversity of subclinical expressions associated with this phenotype. Moreover, a higher presence of BAP has been observed in males compared to females (De Vries, Begeer, and Geurts 2023; Suzuki et al. 2017; Yousefvand et al. 2022). This is consistent with the global trend of male predominance in the prevalence of autism (Zeidan et al. 2022). To our knowledge, research on BAP has been conducted in the relatives of autistic individuals. Autistic children of parents with BAP have been found to be more likely to display repetitive behaviors (Nayar

et al. 2022) and mild language and motor delays with emotional dysregulation (Rubenstein et al. 2018) than autistic children of parents without BAP. These findings suggest that BAP in parents may be related to the inheritance and manifestation of autism in their children. Knowing the existence of parental BAP is therefore essential for understanding variability of the autism spectrum, identifying possible factors that increase the likelihood of certain manifestations, and recognizing the genetic contribution of autism (Nayar et al. 2021). It also helps to develop early interventions and support strategies for families affected by autism.

Another interesting aspect identified in research is the relationship between BAP and mental health. The literature shows that the occurrence of depressive and anxiety problems in parents of autistic children is high (Ault et al. 2021). However, few studies have specifically analyzed the relationship between BAP and parental mental health. Some authors have related the presence of emotional challenges to the presence of autistic traits in parents, especially mothers (Kulasinghe, Whittingham, and Mitchell 2021). Recent studies indicate that BAP may pose a risk factor for developing mental health problems. This may be either directly, if emotional problems are considered a feature of BAP (Kurtz et al. 2023), or indirectly, if autistic traits are considered a predisposing factor (Carpita et al. 2019) or a child's autistic traits acts as a stress factor.

The main aim of this study is to determine the BAP traits of (1) parents of autistic children, (2) parents of children with autism traits (i.e., subthreshold autism and autistic symptoms) and (3) parents of children in a comparison group without autism or autistic traits. We expected that fathers of children with any autistic condition would have significantly higher BAP levels than those of children in the comparison group. We also expected that fathers would present higher BAP than mothers. Since few studies have analyzed the relationship between BAP and emotional state, we also aimed to evaluate whether parents' psychological health is associated with the challenges associated to having a child with any autistic condition or whether it is associated with the presence of BAP. We expected that parents with higher levels of BAP and parents of children with any autistic condition would present more emotional challenges.

2 | Methods

2.1 | Study Design

This study is part of the Epidemiological Research Project on Neurodevelopmental Disorders (EPINED). This project consists of a two-phase cross-sectional study that applies screening and diagnostic procedures to the school population in Tarragona, Spain. One aim of the project is to describe the epidemiology of autism in two age ranges: preschoolers aged 4–5 years (PRE) and primary schoolchildren aged 10–12 years (SCH). Details of the sample as well as the screening procedure and methods have been extensively described in Morales-Hidalgo et al. (2018) and Morales-Hidalgo, Voltas, and Canals (2021). The EPINED protocol has been validated by the Ethics Committee of the Hospital Universitari Sant Joan in Reus, Spain (13-10-31/10proj5).

2.2 | Procedure

In the first phase (screening), psychological and sociodemographic information was collected from parents and teachers. The parents responded to the Spanish version of the Childhood Autism Spectrum Test (CAST; Morales-Hidalgo et al. 2017a; Scott et al. 2002), while teachers completed the EDUTEA questionnaire (Morales-Hidalgo et al. 2017b) to divide participants into those who are and those who are not highly likely to show symptoms of autism. Children who scored above the cutoff point on at least one of the tests were considered to have an increased likelihood for autism. The psychometric characteristics of the questionnaires used in this study were good (CAST: a sensitivity of 84%, a specificity of 93%, and a positive predictive value (PPV) of 0.63 for detecting autism; EDUTEA: a sensitivity of 87%, a specificity of 91%, and a PPV of 0.87 for detecting autism). Children with increased likelihood for autism (and children with prior diagnosis) and children without increased likelihood for autism (comparison group) made up the second-phase sample. These children and their families were assessed individually.

In the second phase, the parents completed the Child Behavior Checklist for preschool children (CBCL1^{1/2}-5; Achenbach and Rescorla 2000) and school-aged children (CBCL6-18, Achenbach and Rescorla 2001) to collect data on emotional, behavioral, social and attentional problems. Specialized and trained professionals also administered the Wechsler Intelligence Scales for Preschool and School-Aged Children (WPPSI-IV and WISC-IV according to age group) (Wechsler 2003, 2012). Autism diagnosis was made by taking into account DSM-5 criteria. Information for diagnosing was collected using the Autism Diagnostic Observation Scale (ADOS-2; Lord et al. 2012) administered to the children and the Autism Diagnostic Interview (ADI-R; Rutter, Le Couteur, and Lord 2003) administered to the parents. For diagnostic purposes, we considered the cut-off scores in all the ADI-R diagnostic algorithm domains and the ADOS-2 calibrated severity score (≥ 4) (according to criteria of Gotham et al. 2009) or when a consensus in diagnosis was reached by two researchers considering the information from the two instruments and observation in school environment. In addition, some diagnoses were previously performed by mental health professionals (early detection and stimulation centres, child and adolescent mental health centers, or centers specialized in diagnosing and treating ASD). A diagnosis of subthreshold autism was established when both screening questionnaires were positive, and the child was no more than 2 points below the cutoff of every ADI-R and ADOS-2 domain—indicating the presence of prominent traits of autism—and clinical consensus for an autism diagnosis was not reached between two researchers. Children with increased likelihood for autism in the screening questionnaires who did not meet the criteria for autism or subthreshold autism diagnosis were considered to have subtle manifestations of autism and conformed the group of autistic symptoms (Morales-Hidalgo et al. 2018; Morales-Hidalgo, Voltas, and Canals 2021).

2.3 | Participants

In the recruitment phase, 3727 parents of a total of 6921 children from representative schools in the province of Tarragona gave their informed consent (53.9% of the initial sample) and participated in

the study. In the second phase, 781 children were assessed individually for diagnosis. Exclusion factors for the current analysis were ADHD diagnosis and being adopted. Families who did not complete the BAP questionnaires were also discarded from the study. The number of participants therefore decreased to 266 children (PRE: 140; SCH: 126) and 489 parents (261 mothers and 228 fathers). Based on DSM-5 diagnostic criteria, the children were initially divided into four main groups: autism ($N=51$), subthreshold autism ($N=38$), autistic symptoms ($N=55$), and children without neurodevelopment conditions (NDC) (without intellectual disability, with non-autistic traits and not diagnosed with ADHD) ($N=122$). Subsequently, for the analysis of the results, the children with subthreshold autism and autistic symptoms were regrouped as autism traits, obtaining three categories: autism ($N=51$), autism traits ($N=93$), and non-autism ($N=122$). No autism diagnosis was reported in the parents (see Figure 1).

2.4 | Assessment Instruments

Sociodemographic Questionnaire: Sociodemographic and clinical data were collected from the children and their families using an ad hoc questionnaire to determine age, sex, parental relationship, ethnicity and family socioeconomic level (calculated by adapting the Hollingshead index; Hollingshead 1975).

Autism Spectrum Quotient (AQ-Short) (Hoekstra et al. 2007): The parents answered questions about themselves on the AQ-Short version validated for the Spanish population (Lugo-Marín et al. 2019). This version comprises 28 items with Likert-type responses (1 = “strongly agree”, 2 = “slightly agree”, 3 = “slightly disagree” and 4 = “strongly disagree”). Items 2, 4, 5, 7, 10, 13, 14, 15, 16, 22, 23, 25 and 26 were reverse scored. The same cut-off points as for the Spanish version were established (total score > 63) with a sensitivity of 0.98 and a specificity of 0.84. The five defined factors (Imagination, Switching, Routines, Social Skills and Numbers/Patterns) were maintained with their respective cut-off points (> 17 ; > 10 ; > 10 ; > 16 ; and > 10 , respectively) (Lugo-Marín et al. 2019). The cut-off points from the AQ-Short scale for the presence of BAP (total score) and difficulties in clinical features (factor scores) were preserved for the present study.

The General Health Questionnaire Short Form (GHQ-12) (Goldberg and Hillier 1979): The parents answered the 12-item General Health Questionnaire validated for the Spanish population (Del Pilar Sánchez-López and Dresch 2008). This version of 12 items with Likert-type responses from 0 to 3 (in positive items 0 = “always” and 3 = “never”; in negative items 0 = “never” and 3 = “always”) has a good level of reliability in Spain (Cronbach’s alpha 0.76). Determination of the GHQ factors has been much discussed. Some authors describe three factors (Coping Strategies, Self-Esteem, and Stress) (Del Pilar Sánchez-López and Dresch 2008), while others describe two (Anxiety/Depression and Social Dysfunction) (Cuéllar-Flores et al. 2014; Mayhew, Stuttard, and Beresford 2020). Still others assert that it is a unidimensional screening instrument (Rocha et al. 2011). Due to the aim of our study, we used two factors to explore our participants’ psychological problems: (1) those related to anxiety/depression (items 2, 5, 6, 9, 10, 11) and (2) those related to social dysfunction (items 1, 3, 4, 7, 8, 12). In both cases, high scores are associated with a greater risk of developing

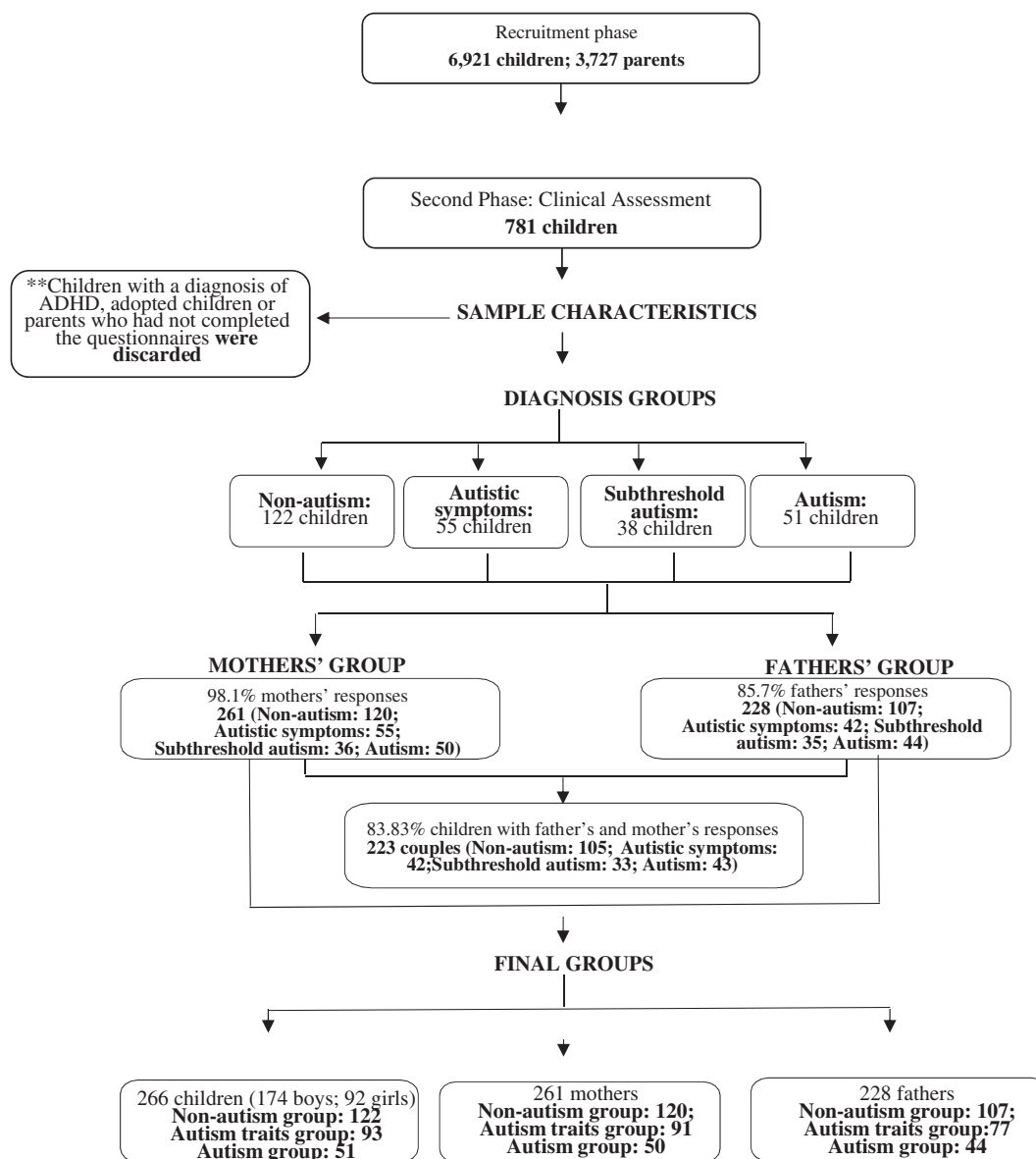


FIGURE 1 | Study design and distribution of participants flow chart.

psychological problems (Cuéllar-Flores et al. 2014). However, to determine the frequency of mental health problems in parents, a cut-off point of 12 was established in line with previous authors (Anjara et al. 2020; Rathore, Rana, and Chadha 2022).

2.5 | Statistical Analyses

Statistical analyses were performed with IBM SPSS 29.0 and EPIDAT 4.1. Chi-square and Z-tests were conducted to compare independent proportions and examine differences in the frequencies of AQ-Short total and factor scores, as well as GHQ-12 total scores. ANOVA and Bonferroni post hoc tests were used to examine differences in the means of AQ-Short total and factors and of GHQ-12 total and factors, respectively. These comparisons were made between parents of children in the autistic condition groups and children without NDC. Chi-square tests for frequencies and t-tests for means were conducted to compare the autistic conditions' groups with the non-autistic group.

All analyses were performed for both mothers and fathers. To explore which variables were associated with parental mental health, multiple regressions models were performed using the Enter method. We created dummy variables to facilitate comparisons between the effects of having an autism diagnosis or having autistic traits, relative to the comparison group. Potential predictors in the first model were: having autism or autism traits; the presence of BAP based on the cut-off point established in the AQ-Short (0: did not exceed the AQ-Short cut-off point; 1: did exceed the AQ-Short cut-off point); full intelligence quotient score; sex (1: boy; 2: girl); age (in months); and family socioeconomic level (1: high; 2: medium; 3: low). In the second model, the emotional dysregulation profile obtained through the CBCL was added to the previous regression. To the third model we added a variable resulting from the interaction between the child's autism and their emotional dysregulation profile.

A significance level <0.01 was used to control the increase in type-I error caused by multiple comparisons.

3 | Results

3.1 | Descriptive Data of the Sample

Table 1 shows the descriptive data of the sample by autistic condition groups. The mean age of the mothers was 38.3 (SD: 6.8) and the mean age of the fathers was 41.4 (SD: 6.8). Also, 13.9% of participants had a low family socioeconomic level (FSE), 63.5% had a medium level and 22.6% had a high level. The percentage of low FSE was higher in the autistic groups. The male gender was dominant in children belonging to the autism and subthreshold autism groups (174 boys and 92 girls). With regard to ethnicity, children from North Africa were classified mainly in the autistic symptoms group and children from Eastern Europe were classified mainly in the autism group. Scores on the Wechsler scales showed means within the normal range except for the autism group, which were a little lower. The ADIR-R, ADOS and CBCL scores increased as severity in the autistic condition groups increased (Table 1).

3.2 | Relationship Between Parental BAP and Autistic Condition Groups

Based on the cut-off point for the AQ-Short scale, 26.1% ($n=68$) of mothers and 36.9% ($n=84$) of fathers exhibited an expanded phenotype ($p=0.010$).

First, we analyzed the presence of BAP and mental health levels of the parents across the initial EPINED groups (autism, subthreshold autism, autism symptoms, and non-autism) (Table S1). No significant differences were found between the three groups with autistic conditions in either fathers or mothers, but significant differences were observed in fathers between the autism group and the non-autism group. Thus, we combined the autistic symptoms group and subthreshold autism group into a single “autism traits” group (Table 2) for the following analyses.

Analysis of BAP percentages by autism group, autism traits group and comparison group without autism showed no significant differences among mothers (20.0% vs. 29.7% vs. 25.8%), whereas the fathers of autism children displayed higher BAP frequencies than those of children in the comparison group (52.3% vs. 28.0%) (see Table 2). With regard to children whose mothers and fathers both answered the AQ-S ($n=223$; 118 parents of autistic conditions children and 105 parents of children without autistic traits), 14.4% ($n=17$) of parents of autistic conditions children and 12.4% ($n=13$) parents of controls had BAP ($p=0.658$).

With regard to AQ-Short factors (Table 2), only the mothers of children in the autism condition groups (autism and autism traits) displayed a tendency to present more differences in social skills than mothers of children in the comparison group (52.0% vs. 53.8% vs. 37.5%). The percentages of differences in social skills and routines were highest in the fathers of autistic conditions children (56.8% vs. 55.8% vs. 39.3% and 27.3% vs. 24.7% vs. 13.1% respectively), though differences were only significant for the imagination factor ($p<0.01$) between autism and non-autism group (70.5% vs. 43.9%). The mean scores on the AQ-Short questionnaire (Table 2) were similar to those observed

in the frequency analysis. Only in relation to fathers was the total score of AQ-Short significantly higher in the autism group (64.6) than in the comparison group (59.1). For AQ-Short factors, scores on routines and imagination were also significantly higher in the fathers of autistic children than in the fathers of controls.

3.3 | Relationship Between the Psychological Problems of Parents and the Presence or Absence of Autistic Traits

Based on the GHQ-12 scale cut-off point, the frequency of mothers with psychological problems was higher in the autism condition groups (autism and autism traits) than in the mothers of children in the comparison group (36.0% vs. 34.1% vs. 19.2%) (see Table 2). On the other hand, no significant differences were found between the fathers of children in the autism condition groups and the fathers of children in the comparison group.

With regard to the GHQ-12 mean scores, the mothers of children in the autism group scored significantly higher on the total scale ($M: 11.1$, $SD: 5.8$) than those of children in the non-autistic group ($M: 8.9$; $SD: 3.6$). However, in anxiety/depression, mothers of children in both autism condition groups scored higher ($M: 4.9$; $SD: 3.9$ and $M: 4.3$ $SD: 3.3$) than those of children in the non-autism group ($M: 3.1$; $SD: 2.7$). No significant differences were found in the fathers' groups.

3.4 | Psychological Problems of Parents by Presence of BAP

Table 3 shows that the frequency of psychological problems was not significantly different in relation to the presence or absence of BAP in fathers or mothers. However, fathers with BAP showed significantly higher GHQ-12 total scores ($M: 10.2$, $SD: 3.9$) and anxiety/depression scores ($M: 4.2$, $SD: 2.7$) than those without BAP ($M: 8.8$, $SD: 3.1$ and $M: 3.0$, $SD: 2.5$, respectively).

From Table 4, we see that the first linear regression model showed that the GHQ-12 total scores of mothers were positively associated with having an autistic child ($\beta 3198$, 1371–5026, $p<0.001$), while the scores for anxiety/depression were associated with having a child with autistic conditions (autism and autism traits) and the presence of BAP. When we included emotional dysregulation in the second and third model, it was significantly and positively associated with both GHQ-12 total score and the score for anxiety/depression. In the third model, which included the interaction covariate of children's autism condition \times emotional dysregulation, the data did not change in relation to the second model showed. For fathers, BAP was the only variable that was positively associated with GHQ-12 total score and anxiety/depression in all models.

4 | Discussion

This study has described the presence of BAP traits and their association with the mental health of parents of schoolchildren in two age groups (3–6 years and 10–12 years) with autistic

TABLE 1 | Characteristics of the sample ($N=266$).

	Autism traits ($n=93$)			Autism ^d ($n=51$)	<i>p</i>	Group difference
	Non-autism ^a ($n=122$)	Autistic symptoms ^b ($n=55$)	Subthreshold autism ^c ($n=38$)			
<i>Parents' characteristics</i>						
Age, years, <i>M</i> (SD)						
Mothers	39.2 (6.1)	35.6 (8.4)	38.1 (8.6)	38.6 (5.6)	0.025	ab*
Fathers	41.9 (5.6)	39.9 (8.9)	42.3 (8.6)	41.4 (4.9)	0.290	
Parents' relationship, % (<i>n</i>)						
Separated	12.3 (15)	0.0 (0)	18.4 (7)	9.8 (5)	0.145	
Family socioeconomic status, % (<i>n</i>)						
Low	8.2 (10)	18.2 (10)	21.0 (8)	17.6 (9)	0.142	
Medium	64.8 (79)	67.3 (37)	63.2 (24)	56.9 (29)		
High	27.0 (33)	14.5 (8)	15.8 (6)	25.5 (13)		
<i>Children's characteristics</i>						
Sex, % (<i>n</i>)						
Male	60.7 (74)	54.5 (30)	78.9 (30)	78.4 (40)	0.012	ac*, ad*, bc*, bd**
Female	39.3 (48)	45.5 (25)	21.1 (8)	21.6 (11)		ac*, ad*, bc*, bd**
Age group, % (<i>n</i>)						
Preschool-aged children	48.4 (59)	60.0 (33)	50.0 (19)	56.9 (29)	0.464	
School-aged children	51.6 (63)	40.0 (22)	50.0 (19)	43.1 (22)		
Ethnicity, % (<i>n</i>)						
Spanish	86.9 (106)	61.8 (34)	89.5 (34)	77.1 (37)	0.006	ad***, ad*, bc**
Asian	0.8 (1)	1.8 (1)	0.0 (0)	2.1 (1)		
Eastern Europe	3.3 (4)	3.6 (2)	2.6 (1)	10.4 (5)		
North Africa	5.7 (7)	25.5 (14)	7.9 (3)	8.3 (4)		ab***, bc*, bd*
South America	3.3 (4)	7.3 (4)	0.0 (0)	2.1 (1)		
Wechsler, <i>M</i> (SD)						
Total IQ	107.6 (12.9)	99.13 (13.8)	97.7 (18.9)	87.8 (19.7)	<0.001	ad***, bd**, cd*
CBCL, <i>M</i> (SD)						
Internalizing problems	53.1 (9.3)	62.0 (10.8)	64.9 (7.6)	68.4 (10.8)	<0.001	ac***, ab***, ad***, bd**
Externalizing problems	48.5 (9.4)	55.0 (10.0)	59.3 (11.8)	62.3 (13.0)	<0.001	ac***, ab***, ad***, bd**
Total problems	50.0 (10.1)	59.7 (11.7)	64.5 (9.1)	68.3 (11.3)	<0.001	ac***, ab***, ad***, bd***
Emotional dysregulation	161.9 (11.7)	174.7 (20.7)	186.2 (21.7)	189.4 (22.7)	<0.001	ac***, ab***, ad***, bc**, bd***
ADI-R, <i>M</i> (SD)						

(Continues)

TABLE 1 | (Continued)

	Autism traits (n = 93)			Autism ^d (n = 51)	p	Group difference
	Non-autism ^a (n = 122)	Autistic symptoms ^b (n = 55)	Subthreshold autism ^c (n = 38)			
Reciprocal social interaction	1.8 (2.1)	3.0 (2.4)	7.6 (4.0)	14.4 (6.0)	<0.001	ac***, ad***, bc***, bd***, cd***
Communication	1.3 (1.4)	1.9 (1.7)	6.2 (3.5)	10.9 (4.0)	<0.001	ac***, ad***, bc***, bd***, cd***
Restrictive behavior patterns	0.5 (0.7)	1.1 (1.3)	2.3 (1.1)	4.4 (2.5)	<0.001	ac***, ad***, bc***, bd***, cd***
Developmental alterations	0.1 (0.4)	0.6 (1.2)	1.5 (1.5)	3.2 (1.5)	<0.001	ab*, ac***, ad, bc**, bd***, cd***
ADOS-2						
Scales, M (SD)						
Social communication	0.3 (0.6)	0.6 (0.8)	1.5 (1.3)	2.1 (1.4)	<0.001	ac***, ad***, bc**, bd***, cb***, cd**
Social interaction	1.0 (1.7)	2.5 (2.2)	4.0 (2.0)	6.7 (3.6)	<0.001	ab***, ac***, ad***, bc*, bd***, cd***
Repetitive behavior	0.1 (0.3)	0.3 (0.6)	0.8 (1.0)	2.4 (1.3)	<0.001	ac***, ad***, bc**, bd***, cd***
Total score	1.4 (2.1)	3.3 (2.7)	6.2 (3.5)	11.2 (4.9)	<0.001	ab***, ac***, ad***, bc***, bd***, cd***
Severity levels, % (n)						
Minimal evidence	96.7 (118)	85.5 (47)	57.9 (22)	17.6 (9)	<0.001	ab**, ac***, ad***, bc**, bd***, cd***
Slight	0.8 (1)	12.7 (7)	26.3 (10)	27.5 (14)	<0.001	ab***, ac***, ad***
Moderate	1.6 (2)	1.8 (1)	13.2 (5)	47.1 (24)	<0.001	ac**, ad***, bc*, bd***, cd***
Severe	0.8 (1)	0.0 (0)	2.6 (1)	7.8 (4)	<0.001	ad*

Note: Comparisons have been made using ANOVA, chi-square test, and comparison of independent proportions. Significant differences in bold ($p \leq 0.01$).

Abbreviations: ADI-R, Autism Diagnostic Interview; ADOS-2, Autism Diagnostic Observation Scale—2; CBCL, Child Behavior Checklist; IQ, Intelligence Quotient; M, medium; SD, standard deviation.

* $p < 0.05$.

** $p \leq 0.01$.

*** $p \leq 0.001$.

^aNon-autism group.

^bAutistic symptoms group.

^cSubthreshold autism group.

^dautism group.

conditions in the province of Tarragona (Spain). To our knowledge, this is the first study to compare BAP levels in the parents of children with different levels of intensity of autism traits with those of parents of children without autistic traits while also analyzing the relationship between parental BAP and psychological state. We observed that BAP was more prevalent in fathers than in mothers and that these autistic traits in fathers were more frequent in fathers of autistic children. Our analyses also indicated that mothers of children in the autism condition groups presented higher levels of emotional problems than those in the comparison group. After adjusting for several covariates, our analyses indicated that BAP traits were associated with emotional problems in fathers; associated with emotional

problems in mothers, on the other hand, were children's emotional dysregulation and autism.

4.1 | Variability in BAP Traits Among Fathers and Mothers of Children in the Context of Autism

Our results suggest that the rate of BAP traits of fathers of children in the autism condition groups (52.3% in autism and 40.3% in autism traits) was higher than that of mothers (20.0% and 29.7%). This is consistent with data from previous studies (Rubenstein and Chawla 2018). However, to our knowledge, previous studies only compared parental BAP between children

TABLE 2 | Distribution of BAP and emotional problems by diagnostic group by AQ and GHQ-12 test.

		Mothers				Fathers			
		Non-autism ^a (n = 120)	Autism traits ^b (n = 91)	Autism ^c (n = 50)	p between groups	Non-autism ^a (n = 107)	Autism traits ^b (n = 77)	Autism ^c (n = 44)	p between groups
<i>AQ test</i>									
Total score	% (n)	25.8 (31)	29.7 (27)	20.0 (10)	0.601	28.0 (30)	40.3 (31)	52.3 (23)	0.015 ac**
	M (SD)	58.2 (6.7)	59.6 (6.6)	57.9 (6.8)	0.251	59.1 (7.6)	61.5 (7.3)	64.6 (8.8)	<0.001 ac***
Social skills	% (n)	37.5 (45)	53.8 (49)	52.0 (26)	0.039 ab*	39.3 (42)	55.8 (43)	56.8 (25)	0.038 ab*, ac*
	M (SD)	15.9 (2.5)	16.9 (2.7)	16.4 (2.6)	0.027 ab*	16.1 (2.7)	17.0 (3.1)	17.3 (3.4)	0.034
Routines	% (n)	21.7 (26)	15.4 (14)	10.0 (5)	0.157	13.1 (14)	24.7 (19)	27.3 (12)	0.056 ab*, ac*
	M (SD)	9.0 (1.6)	9.1 (1.4)	8.3 (1.7)	0.022 ac*	8.8 (1.5)	9.3 (1.7)	9.7 (1.7)	0.010 ac*
Switching	% (n)	15.8 (19)	19.8 (18)	18.0 (9)	0.755	32.7 (35)	33.8 (26)	45.5 (20)	0.306
	M (SD)	8.8 (1.6)	9.2 (1.7)	8.8 (1.8)	0.181	9.5 (2.0)	9.8 (2.0)	10.3 (2.1)	0.112
Imagination	% (n)	54.2 (65)	48.4 (44)	50.0 (25)	0.689	43.9 (47)	58.4 (45)	70.5 (31)	0.008 ac**
	M (SD)	17.5 (3.2)	17.3 (2.7)	17.6 (2.6)	0.831	17.4 (3.1)	18.1 (2.9)	19.5 (3.3)	0.001 ac***
Numbers/ patterns	% (n)	6.7 (8)	4.4 (4)	8.0 (4)	0.657	6.5 (7)	9.1 (7)	13.6 (6)	0.372
	M (SD)	6.7 (2.1)	7.0 (2.0)	6.8 (2.1)	0.481	7.1 (2.1)	7.2 (2.3)	7.8 (2.7)	0.235
<i>GHQ-12 test</i>									
Total score	% (n)	19.2 (23)	34.1 (31)	36.0 (18)	0.012 ab*, ac*	23.4 (25)	35.1 (27)	27.3 (12)	0.217
	M (SD)	8.9 (3.6)	10.2 (4.4)	11.1 (5.8)	0.005 ac**	8.8 (3.1)	10.0 (3.5)	9.4 (4.1)	0.084
Anxiety/ depression	M (SD)	3.1 (2.7)	4.3 (3.3)	4.9 (3.9)	<0.001 ab*, ac**	3.0 (2.5)	4.0 (2.7)	3.8 (2.7)	0.023 ab*
	M (SD)	5.8 (1.7)	5.8 (1.7)	6.0 (2.2)	0.798	5.8 (1.3)	6.0 (1.4)	5.6 (1.9)	0.232
Social dysfunction									

Note: Cut-off scales: AQ-Short total > 63; Social skills > 16; Routines > 10; Switching > 10; Imagination > 17; and Numbers/patterns > 10; GHQ-12 total > 12. Comparisons have been made using the chi-square test, ANOVA, Student's *t*-test and comparison of independent proportions. Significant differences in bold: ($p \leq 0.01$). The shadows in the tables were to differentiate the lines containing percentages vs. the lines containing averages

* $p < 0.05$.

** $p \leq 0.01$.

*** $p \leq 0.001$.

^aNon-autism group.

^bAutism traits group.

^cautism group.

diagnosed with autism and those without autism. An exclusive feature of our study, on the other hand, was that we recruited participants with different levels of intensity of autism traits (from children diagnosed with autism to children with less pronounced autistic traits) and analyzed these levels of intensity in relation to parental BAP traits. As our results found no significant differences between the groups of autism condition in relation to BAP traits, this highlights the importance of considering

subthreshold conditions and supports the existence of an autism spectrum continuum and the genetic transmission of its features. Although they do not full fill all the DSM-5 diagnostic criteria for autism, the children with subthreshold autism present socio-communicative, emotional and behavioral differences that can also interfere with their functioning (Morales-Hidalgo et al. 2018). Moreover, autistic traits may manifest differently in each individual and the intensity of the condition may not be

TABLE 3 | Psychology problems by the presence of BAP.

		Mothers			Fathers		
		BAP (<i>n</i> = 68)	Without BAP (<i>n</i> = 193)	<i>p</i>	BAP (<i>n</i> = 84)	Without BAP (<i>n</i> = 144)	<i>p</i>
GHQ-12 total	% (<i>n</i>)	33.8 (23)	25.4 (49)	0.181	35.7 (30)	23.6 (34)	0.050
	<i>M</i> (SD)	10.4 (4.6)	9.5 (4.4)	0.111	10.2 (3.9)	8.8 (3.1)	0.006
Anxiety/depression	<i>M</i> (SD)	4.7 (3.2)	3.6 (3.3)	0.021	4.2 (2.7)	3.0 (2.5)	<0.001
Social dysfunction	<i>M</i> (SD)	5.7 (2.0)	5.9 (1.7)	0.471	5.9 (1.8)	5.8 (1.2)	0.421

Note: Cut-off scales: GHQ-12 total > 12. Comparisons have been made using the chi-square test and Student's *t*-test. Significant differences in bold ($p \leq 0.01$). Abbreviation: BAP, broader autism phenotype.

the sole determinant in the expression of these traits (Dell'Osso et al. 2023; Sharp et al. 2023; Talli et al. 2022).

The ratio in favor of fathers (40.3% for the autism traits group and 52.3% for the autism group) is consistent with figures reported by authors from several countries (Rubenstein and Chawla 2018). However, our rate in fathers is higher than that reported in a similar study conducted in Istanbul, which found that 25.2% of fathers and 17.8% of mothers of autistic children presenting BAP (Bora et al. 2016). Similar rates of parental BAP (33% of fathers and 23% of mothers) were reported by Wheelwright et al. (2010) in Cambridge, UK. Maxwell et al. (2013) in Philadelphia, on the other hand, found that 21% of fathers and 10% of mothers of autistic children presented BAP, figures which are less than half the rates found in our study. Despite the variability in the ranges of BAP traits reported in the literature (Rubenstein and Chawla 2018), fathers of autistic children consistently present more BAP than mothers, which may support the existence of a genetic predisposition to autism. Although the precise reasons for this sex difference are not fully understood, one possible explanation relates to higher rates of autism diagnosis in males than in females (Talantseva et al. 2023; Zeidan et al. 2022). Fathers may be more likely to transfer genetic mutations that increase likelihood of autism (Rubenstein et al. 2018). Mothers, on the other hand, tend to show a protective genetic pattern for autism in their children (Zhang et al. 2020) as well as higher levels of social competence (Key et al. 2022), which could make it more difficult to identify BAP traits or even diagnose autism through questionnaires that have not been specifically designed to identify the female phenotype of autism.

4.2 | Exploration of the Association Between Parental Mental Health With BAP and the Autistic Conditions of Children

Our data have shown that a significantly higher percentage of mothers in the autism condition groups (autism and autism traits) scored above the cut-off point on the GHQ-12 than the mothers of control children. Also, both fathers and mothers of children in the autism condition groups scored significantly higher in anxiety/depression than parents of the comparison group peers. Parents of autistic children are also known to present higher levels of stress, anxiety and depression than parents of typical-development children, which implies a greater risk of the parents developing emotional disorders (Nunnally et al. 2023;

Papadopoulos 2021). The responsibility of caring for autistic children, the intensity level of autistic traits, stigmatization, and a lack of social understanding and support have been associated with the possibility of presenting mental health (Manda et al. 2023; Porter et al. 2022; Solaiman et al. 2023; Stankova et al. 2023; Sulaimani and Mursi 2022). However, although similar levels of psychopathology have been found in fathers and mothers of autistic children, multiple studies agree that it is mothers who experience higher rates of psychological problems such as stress, depression and anxiety (Schnabel et al. 2019). On the other hand, when BAP traits are considered, only fathers with BAP scored significantly highly on GHQ-12 total scale and anxiety/depression. Previous evidence has been found of the relationship between BAP and the presence of emotional difficulties (McDonnell and Nuttall 2018; Rea et al. 2018), whereby individuals with BAP traits tend to present higher levels of anxiety (Lau et al. 2014; Kurtz et al. 2023) and depression (Ingersoll and Hambrick 2011; Kulasinghe, Whittingham, and Mitchell 2021; Pruitt, Rhoden, and Ekas 2016). These data are consistent if we bear in mind that autistic traits are also associated with increased emotional problems (Robinson and Conner 2022; Bonete, Molinero, and Ruisanchez 2023) and challenges in emotional regulation (McDonnell and Nuttall 2018; Rea et al. 2018). Moreover, prior literature also indicates that adults with autistic traits experience numerous stressful and victimization situations and less effective coping skills (Trundle et al. 2023) Having BAP could therefore be considered a risk factor for presenting anxiety and depression problems, especially in fathers (Kurtz et al. 2023; Pelton et al. 2023). Multiple linear regression models have supported these differences between fathers and mothers. The only variable consistently associated with the emotional health of fathers were BAP traits, whereas for mothers having a child with autism and the emotional dysregulation characteristics of the child were more important. We found that children in the autism condition groups had more emotional dysregulation than non-autism children, and specifically, those with autism and with subthreshold autism were the groups with more emotional dysregulation. When we included emotional dysregulation in the multiple regression model, it was the only significant variable that was associated with emotional health in the mothers. Considering that emotional dysregulation is more present in the autism condition groups, when we included the interaction covariate (autistic conditions \times emotional dysregulation) in the third model we found that the effect of emotional dysregulation was independent of the autistic condition. Previous studies have shown that emotional regulation problems are very frequent

TABLE 4 | Associations between BAP and the mental health of the parents: Multiple linear regression models.

	GHQ total scale			Anxiety/Depression		
	Mothers	Fathers	Mothers	Fathers	Mothers	Fathers
	β (95% CI)	β (95% CI)	β (95% CI)	β (95% CI)	β (95% CI)	β (95% CI)
<i>Model 1</i>						
Autism dummy	3.198 (1.371, 5.026)***	0.183 (-1.365, 1.731)	2.261 (0.928, 3.593)***	0.528 (-0.670, 1.727)		
Autism traits dummy	1.175 (-0.061, 2.410)	0.931 (-0.104, 1.966)	1.079 (0.179, 1.980)*	0.796 (-0.005, 1.596)		
Parents BAP	1.101 (-0.120, 2.323)	1.258 (0.298, 2.217)**	1.051 (0.161, 1.941)*	1.074 (0.331, 1.817)**		
Total IQ	-0.005 (-0.043, 0.033)	-0.028 (-0.060, 0.004)	-0.008 (-0.036, 0.020)	-0.018 (-0.043, 0.006)		
Sex	0.724 (-4.53, 1.901)	0.292 (-0.687, 1.270)	0.307 (-0.551, 1.165)	0.243 (-0.514, 1.001)		
Age	0.001 (-0.014, 0.017)	-0.003 (-0.016, 0.009)	-0.003 (-0.014, 0.008)	-0.005 (-0.014, 0.005)		
Family socioeconomic status	0.398 (-0.568, 1.364)	-0.664 (-1.474, 0.146)	0.424 (-0.281, 1.128)	-0.430 (-1.057, 0.197)		
	$R^2 = 0.085; F_{238,7} = 3.142$	$R^2 = 0.078; F_{206,7} = 2.489$	$R^2 = 0.106; F_{238,7} = 4.050$	$R^2 = 0.093; F_{206,7} = 3.006$		
	$p = 0.003$	$p = 0.018$	$p < 0.001$	$p = 0.005$		
<i>Model 2</i>						
Autism dummy	1.434 (-0.480, 3.349)	-0.005 (-1.724, 1.714)	0.912 (-0.496, 2.319)	-0.018 (-1.342, 1.306)		
Autism traits dummy	0.139 (-1.136, 1.414)	0.821 (-0.320, 1.961)	0.263 (-0.674, 1.201)	0.445 (-0.434, 1.323)		
Parents BAP	0.889 (-0.282, 2.060)	1.171 (0.199, 2.143)*	0.860 (-0.001, 1.721)*	1.004 (0.255, 1.752)**		
Total IQ	-0.006 (-0.043, 0.030)	-0.029 (-0.060, 0.003)	-0.008 (-0.035, 0.018)	-0.019 (-0.043, 0.006)		
Sex	0.760 (-0.368, 1.888)	0.259 (-0.727, 1.245)	0.397 (-0.433, 1.226)	0.295 (-0.465, 1.054)		
Age	0.003 (-0.011, 0.018)	-0.003 (-0.016, 0.010)	-0.001 (-0.011, 0.010)	-0.003 (-0.013, 0.007)		
Family socioeconomic status	0.209 (-0.715, 1.133)	-0.678 (-1.490, 0.133)	0.292 (-0.387, 0.972)	-0.457 (-1.082, 0.168)		
Emotional dysregulation	0.066 (0.037, 0.095)***	0.008 (-0.019, 0.035)	0.050 (0.028, 0.071)***	0.020 (-0.001, 0.041)		
	$R^2 = 0.160; F_{236,8} = 5.626$	$R^2 = 0.080; F_{204,8} = 2.214$	$R^2 = 0.181; F_{236,8} = 6.503$	$R^2 = 0.108; F_{204,8} = 3.089$		
	$p < 0.001$	$p = 0.028$	$p < 0.001$	$p = 0.003$		
<i>Model 3</i>						
Autism dummy	2.261 (-0.236, 4.758)	0.162 (-2.100, 2.423)	1.373 (-0.465, 3.211)	0.004 (-1.737, 1.746)		
Autism traits dummy	2.378 (-2.148, 6.904)	1.254 (-2.718, 5.227)	1.512 (-1.819, 4.843)	0.503 (-2.557, 3.562)		

(Continues)

TABLE 4 | (Continued)

	GHQ total scale			Anxiety/Depression		
	Mothers	Fathers	Mothers	Fathers	Mothers	Fathers
	β (95% CI)	β (95% CI)	β (95% CI)	β (95% CI)	β (95% CI)	β (95% CI)
Parents BAP	0.935 (-0.240, 2.109)	1.173 (0.199, 2.147)*	0.886 (0.022, 1.750)*	1.004 (0.254, 1.754)**		
Total IQ	-0.005 (-0.042, 0.031)	-0.029 (-0.060, 0.003)	-0.008 (-0.035, 0.019)	-0.019 (-0.043, 0.006)		
Sex	0.824 (-0.311, 1.959)	0.270 (-0.723, 1.262)	0.432 (-0.403, 1.267)	0.296 (-0.468, 1.061)		
Age	0.003 (-0.012, 0.018)	-0.003 (-0.016, 0.009)	-0.001 (-0.012, 0.010)	-0.003 (-0.013, 0.007)		
Family socioeconomic status	0.196 (-0.729, 1.120)	-0.682 (-1.497, 0.132)	0.285 (-0.395, 0.965)	-0.458 (-1.085, 0.169)		
Emotional dysregulation	0.072 (0.041, 0.103)***	0.009 (-0.019, 0.038)	0.053 (0.030, 0.076)***	0.020 (-0.002, 0.042)		
Interaction between autism conditions and emotional dysregulation	-0.005 (0.015, 0.005)	-0.001 (-0.010, 0.008)	-0.003 (-0.010, 0.004)	0.000 (-0.007, 0.007)		
	$R^2 = 0.164; F_{235,9} = 5.116$	$R^2 = 0.80; F_{203,9} = 1.964$	$R^2 = 0.183; F_{235,9} = 5.837$	$R^2 = 0.108; F_{203,9} = 2.733$		
	$p < 0.001$	$p = 0.045$	$p < 0.001$	$p = 0.005$		

Note: Autism dummy: autism (1) and autism traits, comparison group (0); Autism traits dummy: autism traits (1) and autism, comparison group (0).

Abbreviations: BAP, broader autism phenotype; IQ, intelligence quotient.

* $p \leq 0.05$.

** $p \leq 0.01$.

*** $p \leq 0.001$.

in autistic children (Conner et al. 2020; Davico et al. 2022) and are manifested as intense behavioral challenges (Northrup et al. 2022) that may lead to greater stress for their caregivers (May and Williams 2022). We think that while mothers may play a more active role in the parenting of their children than fathers, the presence of BAP traits in fathers may also influence the emotional health of mothers.

5 | Limitations and Future Directions

Several limitations should be considered in relation to our study. This is the first study to analyze BAP traits in mothers and fathers of children with and without autistic condition in Spain and to examine the relationship between those traits and parental mental health. However, our sample is not representative of the whole population and cultural factors have not been taken into account which limits the generalizability of the results. Moreover, although the AQ-Short has been validated in the Spanish population and enabled us to obtain total and factor scores that report specific clinical characteristics, unlike BAPQ this instrument does not assess BAP specifically (Hurley et al. 2006). Another limitation is that the mean IQ scores of all groups, including autism, are within the average range our findings should not be generalized to parents of children with autism and cognitive delay. Our results provide valuable information about the association between the BAP traits of parents and their mental health when they have a child with autistic conditions. They also provide further information about parental BAP and suggest that the parents of children with autistic traits as well as those of children diagnosed with autism present BAP traits. Moreover, having BAP also appears to be associated with emotional challenges, which supports the importance of thoroughly investigating BAP. Therefore, in future studies it could be interesting to explore longitudinal data to assess changes in parental mental health over time. A detailed understanding of BAP and its relationship to emotional well-being can provide valuable information for designing interventions and support measures that are adapted to the specific needs of families of autistic children. Early identification of these characteristics and early provision of psychological support may also be crucial to improve family mental health.

6 | Conclusion and Clinical Implications

Research on the prevalence of the BAP in relatives of autistic individuals has so far been limited. This study has explored BAP traits using the AQ-Short scale and reported that the presence of BAP was higher in fathers (36.9%) than in mothers (26.1%). BAP was more frequent in fathers of children on the autism spectrum than in those of children without autistic traits, which supports the genetic predisposition of autism. The parents of children in the autism condition groups (autism and autism traits) reported greater emotional problems but this association presents differences between the two progenitors. While emotional problems in fathers appear to be related to their BAP traits, in mothers they may be more related to emotional dysregulation and the presence of autistic conditions in their children. Our findings not only expand knowledge about the prevalence and impact of BAP traits of each parent, but also offer practical guidance for

more effective interventions. Recognizing the presence of BAP in parents facilitates the design of emotional support programs and strategies to manage the impact of BAP traits on family dynamics, thus improving the well-being of children and their parents. Additionally, evidence from these phenotypic data within the family environment contributes to the understanding of the etiopathogenesis of autism and may be useful in future research into this condition.

Conflicts of Interest

The authors declare no conflicts of interest.

Data Availability Statement

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

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Supporting Information

Additional supporting information can be found online in the Supporting Information section.