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ORIGINAL CONTRIBUTION

Selective mutism and temperament: the silence and behavioral inhibition to the unfamiliar

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Abstract Behavioral inhibition (BI) is a suspected precursor of selective mutism. However, investigations on early behavioral inhibition of children with selective mutism are lacking. Children aged 3–18 with lifetime selective mutism ($n = 109$), social phobia ($n = 61$), internalizing behavior ($n = 46$) and healthy controls ($n = 118$) were assessed using the parent-rated Retrospective Infant Behavioral Inhibition (RIBI) questionnaire. Analyses showed that children with lifetime selective mutism and social phobia were more inhibited as infants and toddlers than children of the internalizing and healthy control groups, who displayed similar low levels of behavioral inhibition. Moreover, behavioral inhibition was higher in infants with lifetime selective mutism than in participants with social phobia according to the Total BI score ($p = 0.012$) and the Shyness subscale ($p < 0.001$). Infant behavioral inhibition, particularly towards social stimuli, is a temperamental feature associated with a lifetime diagnosis of selective mutism. Results yield first evidence of the recently hypothesized temperamental origin of selective mutism. Children at risk should be screened for this debilitating child psychiatric condition.

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Introduction

Children with selective mutism (SM) consistently fail to speak in specific situations despite speaking in others. In contrast to previous editions, DSM-5[1] assigns selective mutism to the anxiety disorders. The close link between selective mutism and childhood social phobia is widely recognized [2]. Social phobia, or previously avoidant disorder of childhood [3], is a hallmark of selective mutism with comorbidity rates of 97–100 % [4–8]. The prevalence of selective mutism varies from 0.7 to 2 % [9, 10] with a higher prevalence in immigrant populations [11]. Age of onset is around 2.7–4.2 years [4, 5, 12, 13]. Communication disorders are frequently (38–50 %) associated [12, 13] and language deficits are common in a subset of children with selective mutism [14–16]. Along with mute behavior, children with selective mutism may also appear inhibited or frozen and inactive in specific situations [17–20], which is why this phenomenon was also described as "selective inactivity" [21]. This inactivity is displayed in novel situations or when the child is in the focus of other's attention. In summary, children with selective mutism display various symptoms which are evocative of behavioral inhibition (BI), suggesting that behavioral inhibition constitutes an important etiological precursor of selective mutism.

Behavioral inhibition to the unfamiliar represents a temperamental trait, which was first described by Kagan and colleagues [22]. Behavioral inhibition is defined as an "initial tendency to withdraw, to seek a parent, and to inhibit play and vocalization following encounter with unfamiliar people and events" [23]. Other than the related concept of

shyness, behavioral inhibition additionally comprises inhibition in the presence of novel nonsocial objects, such as toys, games, and potentially dangerous or threatening stimuli and novel situations. Behavioral inhibition is considered to represent a lower threshold to limbic and sympathetic nervous system arousal including for instance a higher heart rate, reduced heart period variability under stress and an increased laryngeal muscle tension [22]. Behavioral manifestations of behavioral inhibition vary across development: Inhibited infants display negative affect and motor reactivity to novel stimuli. Toddlers tend to show reluctance to approach or withdraw from novel objects. They display little vocalization, eye contact and smiling with unfamiliar people and may cling to their mothers in unfamiliar situations [24]. Inhibited preschoolers become quiet and affectively subdued with unfamiliar persons and tend to remain solitary and watchful of others in kindergarten. Lack of spontaneous speech in the presence of an unfamiliar adult represents one of the most sensitive indices of behavioral inhibition in 5-year-old children [22]. Behavioral inhibition is found in 15 % of children and is moderately stable from toddlerhood through the early elementary school years with higher stability of extreme behavioral inhibition [25, 26]. In longitudinal studies early behavioral inhibition has been shown to pose a specific risk for anxiety disorders, particularly social phobia in early and middle childhood and adolescence [27–29]. A meta-analysis reported that children with behavioral inhibition have a seven times greater risk for developing social anxiety disorder (OR = 7.59, $p < 0.00002$) [30]. However, the homogeneity of the construct has been questioned [31]. A follow-up-study by Kochanska and Radke-Yarrow [32] identified how different qualities of inhibition affect later social behavior. In this study, inhibition towards *social* stimuli at the age 1.5-3.5 strongly predicted interactive behaviors such as staring, looking but not interacting, being unoccupied, and not conversing during interaction with an unfamiliar peer at age 5, whereas early inhibition towards *non-social* stimuli predicted the non-involvement in the group play with the unfamiliar peer. Non-social and social inhibition at the time of the first assessment was not correlated in this community sample. Another retrospective study found that social rather than non-social fearfulness accounted for the relationship between behavioral inhibition and symptoms of current anxiety disorder, social adjustment, and disability [33]. However, examining children with extreme behavioral inhibition might obscure such differences as inhibition towards both social and non-social stimuli tends to be high in such samples.

As the overlap of the phenomenology of selective mutism and behavioral inhibition is striking, the hypothesis that "selective mutism represents a severe and language based form of behavioral inhibition" [34] resulted. Other authors conceptualized selective mutism and social phobia as different stages in a developmental progression of behaviorally inhibited temperament [35] or suggested that selective mutism represents "the extreme end of a continuum of temperament and social behavior that has a biological basis" [5]. According to the model of Johnson and Wintgens [36] intense arousal of the sympathetic nervous system in novel situations during early childhood of children with selective mutism may lead to a freezing reaction which prevents active confrontation with feared stimuli and thus first shaping and then habituating an avoidant coping style in the form of muteness and inactivity.

However, evidence on abnormalities in sympathetic arousal in children with SM is scarce and inconsistent. One study comparing children with selective mutism, social phobia and controls did not find altered psychophysiological arousal in children with selective mutism [37], whereas another study showed evidence of atypical autonomic regulation during a physical, but not social exercise challenge [38]. The authors propose that atypical autonomic regulation may be involved in the auditory abnormalities previously described in children with selective mutism [39] and may also be responsible for potential laryngeal dysfunction as found in behaviorally inhibited children.

In spite of the significant link between selective mutism and social phobia on the one hand and behavioral inhibition and social phobia on the other hand, empirical data on the relation between selective mutism and behavioral inhibition and the etiological role of the latter for the development of selective mutism is lacking. Recently, a study investigating a community sample of $n = 57$ 3–6-year-olds showed a correlation between behavioral inhibition, social anxiety and selective mute features [40]. Remarkably, regression analysis correcting for social anxiety revealed that the contribution of behavioral inhibition to selective mutism was no longer significant indicating that the link between both entities is possibly mediated by social anxiety. However, conclusions are limited as the applied selective mutism questionnaire (SMQ) does not discriminate between selective mute and social phobic individuals [41].

Longitudinal studies have been proposed to determine the contribution of behavioral inhibition on the development of selective mutism [2, 42]. Given the small prevalence and the early onset of selective mutism, a prospective study would have to recruit a large sample of infants to detect true premorbid temperamental precursors of later selective mutism. Cross sectional studies with older children may only confirm the overlapping phenomenology of both entities already described.

Given that such a prospective investigation is difficult to conduct, the aim of the current study was to compare retrospectively infant behavioral inhibition as a potential etiological risk factor in children and adolescents with lifetime

selective mutism (ltSM), social phobia (SP), other internalizing conditions (INT) and healthy controls (CG) by a parent questionnaire.

We expected that children with selective mutism and social phobia alike were more behaviorally inhibited in their first years of life than those with internalizing disorders and healthy controls. This would be in line with previous findings concerning the relative specificity of behavioral inhibition as a risk factor for social phobia and the close relationship between selective mutism and social phobia. Excessive reticence and inhibition that exceeds those of just social phobic youths are well documented in the SM literature and in clinical practice. Against this empirical background, we expected behavioral inhibition scores shown in the first years of life by children with lifetime selective mutism to be higher than the levels shown by children affected by social phobia alone.

Materials and methods

Participants

Participants were part of a study on the psychopathology and diagnosis of SM conducted in University based Departments of Child and Adolescent Psychiatry, Psychosomatics and Psychotherapy in Germany. 334 children and adolescents aged 3–18 years participated in the study. Thirteen siblings, five families with three participating siblings each and a pair of twins were included in the study.

The sample includes participants with current SM ($n = 95$), SP ($n = 74$), internalizing disorders (INT, $n = 46$) and a healthy control group (CG = 119). A history of selective mutism was reported in further 13 participants with current social phobia and one healthy control participant, raising the number of individuals with ltSM to $n = 109$ and reducing the group of SP alone during lifetime to $n = 61$, and the CG to $n = 118$. In line with previous investigations, 95 % of the children with lifetime selective mutism also fulfilled criteria of social phobia. With respect to the focus of this study on early temperamental features as a potential etiological risk factor for lifetime psychiatric conditions, analyses were conducted according to lifetime diagnosis of selective mutism and not current diagnostic status.

The group INT included participants with major depression ($n = 14$), specific phobia ($n = 9$), obsessive compulsive disorder ($n = 8$), generalized anxiety disorder ($n = 4$), separation anxiety disorder ($n = 4$), panic disorder ($n = 3$), anxiety disorder NOS ($n = 3$) and adjustment disorder ($n = 1$).

Subjects with a prior diagnosis of pervasive developmental disorder were excluded from the study. Children who did not attend regular classroom activities and/or received special education programs were admitted if mental retardation (IQ < 70) was ruled out by a standardized test. Comorbid communication disorder as well as bilingualism and migration did not constitute exclusion criteria. Recommendations for the SM diagnosis in bilingual individuals [43] have been taken into account.

The gender ratio was balanced ($\chi^2 = 2.84$, $p = 0.418$) in the total sample (girls: 49.7 %) and within each group (ltSM: 54.1 %, SP: 41.0 %, INT: 52.2 %, CG: 49.2 %). The reported socioeconomic status (SES) relates to the International Socio-Economic Index of Occupational Status (ISEI) [44]. The SES of families of the CG was significantly higher ($p < 0.001$) compared to all other groups. The groups differed significantly in mean age ($p < 0.001$): The group INT (mean age 13.6 years) was significantly older than ltSM (mean age 9.8 years) and the CG (mean age 8.4 years). Moreover children with SP alone (mean age 11.5 years) were significantly older than the CG.

Procedure

The majority of SM, SP and INT participants were included at University Departments of Child and Adolescent Psychiatry; the CG was recruited through contacts to kindergartens and schools. According to protocols approved by the local ethic committee written informed consent was obtained from parents and adolescents. Following the consent to participate, families received the questionnaires. A structured diagnostic interview (Kinder-DIPS) [45] was conducted with mothers of children with selective mutism and social phobia additionally to the clinical diagnosis to ensure correct current and lifetime diagnoses of the child with regards to DSM-IV-TR [46] and to attain further socioeconomic and developmental information. Parents of the INT and CG, respectively, completed a screening form about SES, developmental risk factors, bilingualism and migration. Mothers completed the Retrospective Infant Behavioral Inhibition (RIBI) [47] measure.

Measures

Retrospective infant behavioral inhibition (RIBI): The RIBI is a retrospective parent report on behavioral inhibition in the first two years of life. The 20-item questionnaire covers the subscales Distress to Novelty, Fear and Shyness and displays a Total BI score. Answers were given on a 5-point Likert-scale, where a higher Total BI score indicating less inhibited behavior. Psychometric properties have been evaluated in two non-clinical samples [47]. Internal reliability was 0.91–0.92 for total BI in the two unselected samples. Inter-rater correlation of father and mother report reached $r = 0.71$. Principal Component Analysis supported a three-factor solution of the core features of behavioral

Table 1 Means (SD) of the retrospective infant behavioral inhibition (RIBI) as a function of group

Scale/subscales	ltSM mean value (SD)	SP alone mean value (SD)	INT mean value (SD)	CG mean value (SD)	*F*	*df*	*p* (η^2)	Post hoc Bonferroni
Total BI	32.9 (14.4)	39.8 (15.3)	53.9 (13.8)	55.0 (12.0)	59.65	3(323)	<0.001 (0.357)	ltSM > SP > INT = CG
Distress to novelty	17.8 (6.6)	17.8 (6.2)	21.8 (5.5)	22.8 (3.9)	21.59	3(323)	<0.001 (0.167)	ltSM = SP > INT = CG
Fear	5.9 (4.0)	7.4 (3.8)	10.2 (3.6)	9.8 (3.5)	25.58	3(323)	<0.001 (0.192)	ltSM = SP > INT = CG
Shyness	9.2 (7.1)	14.5 (7.9)	21.9 (7.2)	22.3 (7.7)	70.01	3(323)	<0.001 (0.394)	ltSM > SP > INT = CG

ltSM lifetime selective mutism, *SP* social phobia, *INT* internalizing group, *CG* control group

Lower RIBI scores indicate higher behavioral inhibition

inhibition. Scores of the RIBI were positively correlated with a parent-report temperament questionnaire and a laboratory-based test at age 14 months with the child.

329 of 334 RIBI questionnaires were completed by the mothers of participants and returned to us. All could be included in the analysis; the number of missing items did not exceed 5 % (one item missing). Psychometric properties were reevaluated, as the RIBI was used in a clinical sample for the first time. Internal consistency was good with Cronbach's $\alpha = 0.94$ for the Total BI scale and $\alpha = 0.84$ for the subscale Distress to Novelty, $\alpha = 0.79$ for Fear, and $\alpha = 0.94$ for Shyness). Unrestricted principal components analysis using Varimax rotation and Kaiser Normalization supported the previously described three-factor model.

Statistical analysis

The Statistical Package for the Social Sciences (SPSS 22) was used for data analyses. Pearson and Kendall-Tau-b correlations were used to identify correlations between age, socioeconomic status, gender and RIBI scores. We compared between-group-differences using MANCOVAs, controlling for confounding variables. Post hoc analyses were calculated using t-tests with Bonferroni correction for multiple testing. All *p* values are calculated as two-tailed.

Results

Retrospective infant behavioral inhibition

Before conducting the analysis of mean values and group differences, correlations between age, gender, SES and RIBI results were evaluated for the total sample.

SES ($r = 0.05$, $p = 0.353$) and age ($r = 0.092$, $p = 0.095$) did not correlate with total BI score. As there was a discrete age effect on the Shyness subscale ($r = 0.111$, $p = 0.044$), characterizing older children as less inhibited when confronted with social stimuli as infants and toddlers, we conducted separate analyses for each group. Age was correlated to BI scores only in the groups ltSM (Shyness: $r = 0.275$, $p = 0.004$) and SP alone (total BI: $r = 0.307$, $p = 0.016$; Shyness: $r = 0.307$, $p = 0.016$). No correlations between age and BI were found in the groups INT and CG ($p > 0.05$).

Kendall-Tau-b revealed correlations between gender and the Total BI score in the total sample (r = -0.094, $p = 0.040$) and the subscale Shyness ($r = -0.110$, $p = 0.016$), whereby girls were described as being more inhibited than boys. Subsequent analyses indicated a correlation with gender only within the ltSM group for Total BI ($r = -0.169$, $p = 0.036$), Fear ($r = -0.170$, $p = 0.039$) and Shyness ($r = -0.202$, $p = 0.013$), but not within the SP alone, INT or CG.

Means and standard deviations of the RIBI total score and subscales of the four groups and MANCOVA results, correcting for age and gender, are reported in Table 1. Relative RIBI scores of the groups are illustrated in the Box plot diagram in Fig. 1.

Between-group-differences were found for the Total BI score and all subscales. Post hoc analysis revealed that children and adolescents of the ltSM and SP alone groups were generally described as more inhibited in their first years of life than the group INT and CG. The latter two did not differ with respect to Total BI or any subscale score. Moreover, children with ltSM were perceived as more inhibited than those with SP alone according to the Total BI score ($p = 0.012$) and the Shyness subscale ($p < 0.001$).

As it may be hypothesized that parental judgment was biased by the current psychopathology of their child, in the sense that the parents retrospective memory of infant behavioral inhibition might be negatively affected by their child's current severe selective mutism, we additionally conducted analyzes referring to current diagnostic status to control for this effect. In this case, the difference between SM and SP groups should increase when the 13 currently social phobic individuals with a history of selective mutism were transferred from the ltSM group to the SP group. However, results contradicted this hypothesis and changed

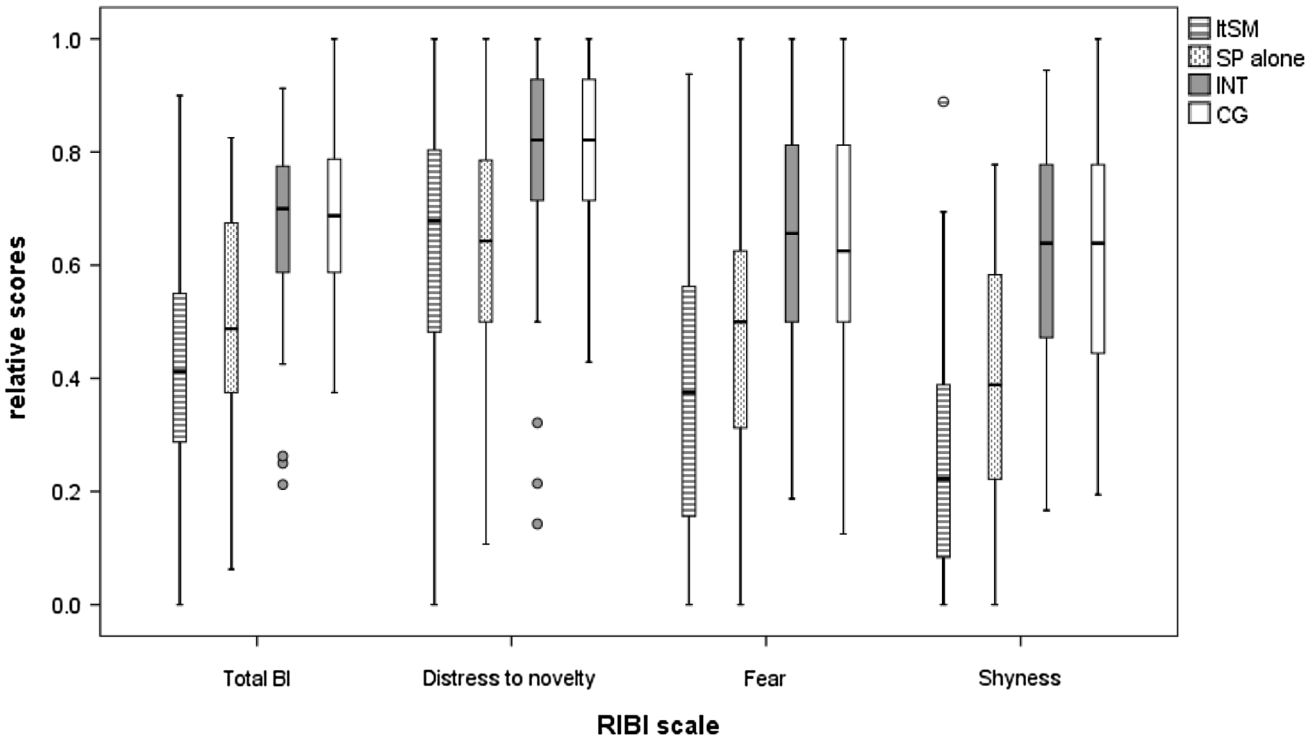

Fig. 1 *Box plot* of RIBI relative scores. *ltSM* lifetime selective mutism, *SP* social phobia, *INT* internalizing group, *CG* control group. Lower RIBI scores indicate higher behavioral inhibition

to the opposite direction: Significant differences between all groups in a MANCOVA persisted, but post hoc analysis demonstrated that the difference between current SM and SP groups was no longer significant (total BI score: $p = 0.458$; Shyness: $p = 0.062$).

Discussion

For the first time, our study reports empirical data regarding early behavioral inhibition preceding later selective mutism. The results provide evidence for a predisposing role of extreme infant behavioral inhibition for the development of selective mutism, similarly also for social phobia.

Congruent with prospective longitudinal studies that have highlighted the association of early behavioral inhinition with the development of social anxiety [27–29, 48], increased infant behavioral inhibition was only reported by mothers of children with lifetime selective mutism and social phobia alone, but not by mothers of children of the INT and CG. Moreover, as hypothesized, children with lifetime selective mutism had an even higher infant BI score than their social phobic counterparts. Intriguingly, the observed difference in the Total BI score derives particularly from a stronger inhibition when confronted with social stimuli. This may constitute another specific predisposing link to mute reactions in communicative situations and is in line with the follow-up-study of Kochanska and Radke-Yarrow [32], who found that early inhibition towards social stimuli rather than non-social stimuli predicted a failure to speak with unfamiliar peers at age 5. If different social interaction patterns of selective mutism and social phobia are elicited by strong behavioral inhibition to unfamiliar social cues in children with selective mutism, this factor might possibly also account for further discriminating characteristics since social inhibition and the more general behavioral inhibition are distinguished by a number of cognitive, behavioral and physiological characteristics [31].

In addition to the different aspects of behavioral inhibition, the intensity of behavioral inhibition may also influence not only the extent but also the quality of social fears. Stronger behavioral inhibition in general was shown to elicit more interactional than performance social fears and predicted earlier onset social phobia [49]. The clinical literature yields cues that highly interactional social communication usually causes more difficulties to children with selective mutism than prepared speech, e.g. giving presentations [36]. Again, as empirical literature is lacking, conclusions are speculative.

The correlation between gender and infant behavioral inhibition within the ltSM group was not found in two

unselected samples [47] and requires further investigation. On the contrary, parents of the community sample of the study by Muris and colleagues [40] rated girls as more inhibited than boys. Obviously, findings on the association of behavioral inhibition with gender are mixed. A gender specific interpretation of behavior may possibly influence parent's judgment more on the extreme range of children's behavior. Nevertheless, there is evidence that early behavioral inhibition is highly relevant for the development of adolescent social anxiety in females: In a study by Schwartz and colleagues [29] the predisposing link between behavioral inhibition at 21 months and social anxiety at age 13 years was stronger in females than in males. Another study yielded evidence, that parent-reported childhood shyness represents a stronger predictor of adolescent social anxiety in females compared to males [50].

Questions also arise on the negative correlation between age and RIBI scores in the more inhibited ltSM and SP alone groups. It might have been assumed that higher infant behavioral inhibition would be a predictor for a chronic course of selective mutism. However, parents of impaired adolescents reported less infant behavioral inhibition. This outcome may possibly result from recall bias. Alternatively, the finding may also be due to the decrease of the impact of early temperament on adolescent psychopathology, as the importance of individual and environmental risk factors increase across the life span.

Even if the clear link between extreme behavioral inhibition and selective mutism is not surprising from the view of child psychiatry, the nature of the relationship remains unknown just as between behavioral inhibition and social phobia, which is currently object of a controversial debate. Results may be interpreted as a confirmation of the previous hypothesis that selective mutism represents a severe and language based form of behavioral inhibition [34]. The remarkably early age of onset of selective mutism in the 3rd to 5th year of life, the high familial prevalence of selective mutism and social phobia (for overview see Muris and Ollendick [2]), and growing evidence for the inconspicuousness of environmental risk factors such as adverse parenting strategies [8, 51, 52] may argue in favor of such a strong temperamental and biological foundation of SM. Causative core biological mechanisms of behavioral inhibition and selective mutism might be the same. If so, the results reported here may also fuel the discussion, to which extent extreme behavioral inhibition represents a prodrome of clinical anxiety (see for survey of current debate: Perez-Edgar and Guyer [53]) or a "disorder in and of itself" [54].

Considering the finding of Muris and colleagues [40], that the link between behavioral inhibition and selective mutism is mediated by social anxiety and that children with selective mutism are rated as more impaired than those with social phobia [8, 37], together with our results of stronger inhibition to social cues in infants with later selective mutism, selective mutism may be also conceptualized as a severe form of early childhood social phobia. The latter is also more characterized by a fear of social unfamiliarity than of negative evaluation. However, in addition to symptom severity a clearly discriminating feature between both disorders is the fact that children with selective mutism fail to overcome unfamiliarity and remain mute and inactive even after acquaintance, maybe due to effective avoidance strategies.

Finally, as not all children with early behavioral inhibition develop clinical anxiety, behavioral inhibition as a temperamental trait and independent construct may function as a causal diathesis for social anxiety, which in turn is modulated by the presence of other additional risk factors. In the case of SM, bilingualism or communication disorders might act as such. Particularly language delay and communication disorders may limit the child's understanding and thus interact with its temperamental traits. However, this etiological concept alone does not sufficiently explain selective mutism in monolingual children without communication disorder.

A further elucidation of this yet unexplained association between early behavioral inhibition and selective mutism is of salient clinical interest to pediatric psychiatry as a sound basis for early prevention and treatment of this debilitating, often chronic psychiatric childhood condition.

There are several limitations which confine the conclusions of our study. First, the RIBI questionnaire has not yet been used and evaluated in clinical samples and by other authors before. A replication of findings in other clinical samples thus is clearly warranted. Our results further rely exclusively on retrospective parent ratings and not on additional examination in laboratory tests feasible in prospective studies. Retrospective reports are in principle prone to recall bias, which is particularly true for parent's judgment of older children. Furthermore, our retrospective design is not suited to elucidate causalities. To clarify a causal relation between behavioral inhibition and selective mutism, future longitudinal studies are clearly needed. Moreover, the mechanisms involved in the impact of social inhibition on specific peer interactions should be thoroughly investigated. We furthermore describe a clinical sample of referred participants, which may represent a more severely impaired subgroup. Additionally, the INT group included a number of heterogeneous disorders with a low n each. Overall group size of the INT was small.

However, extreme behavioral inhibition in infants and toddlers seems to precede the development of selective mutism as well as social anxiety. If future research, particularly prospective longitudinal studies, confirms these results, screening for extreme behavioral inhibition in early childhood would facilitate early diagnostic and intervention

in children at risk and prevent the frequently observed chronic course due to delayed psychiatric treatment.

Compliance with ethical standards

Conflict of interest On behalf of all authors, the corresponding author states that there is no conflict of interest.

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