Children with selective mutism: an examination of the attentional and psychophysiological mechanisms underlying the failure to speak and the situational factors that trigger symptomatology

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Januar 2022

A Dissertation

Submitted to the Faculty of Psychology and Sport Science

In Fulfilment of the Requirements of the Degree of Doctor of Natural Science

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1 Introduction

Various prominent models of anxiety have in common that they propose that threatening stimuli processed by an organism lead to a fear response [1–3]. This simple sequence of how a fear response evolves consists of the components of a threat, attentional processing of the threat, which is subsequently accompanied by processing in brain circuits, and the subsequent fear response [2]. The fear response can further divide into a subjective feeling of fear; an overtly displayed behavior such as avoidance behavior, and a physiological component, for example, a reaction of the autonomic nervous system [2, 4]. Alterations in these processes of locating attention to threat (e.g., bias of attention) and the manner of responding to threat (e.g., autonomic overarousal) are considered mechanisms for underlying symptomatology of anxiety disorders [5, 6]. All three components of the sequence (threat, attention processing, fear response) are essential to understand how symptoms of anxiety disorders occur and are considered in the treatment of anxiety disorders [2]. Accordingly, both transdiagnostic and disorder-specific models of anxiety include the psychophysiological features[1] of altered attention processing and altered autonomic activity as factors contributing to explain phenomenology, etiology, and maintenance of the disorder [5–7]. When it comes to the treatment of anxiety disorders, one can address both the component of attention processing of threat (e.g., by attention training) [8, 9] and the component of fear response (e.g., exposure therapy) [10]. Knowledge of disorder-specific threatening stimuli is also important for the selection of appropriate stimuli, for example, for the creation of fear hierarchies in the context of exposure therapy [11]. Against this background, it is particularly remarkable that these aspects have scarcely been researched yet for the anxiety disorder, selective mutism (SM). Children with SM can't speak in certain social situations, while they can speak in others [12]. Considering that SM is a severely debilitating and typically long-lasting anxiety disorder [13, 14], it seems crucial to include children with SM to examine these fundamental features of anxiety disorders. A better understanding of these aspects could be the basis for developing targeted therapeutic approaches (e.g., training of attention allocation) and improving already established interventions (e.g., selected appropriate stimuli for exposure) for children with SM. In addition, the question arises whether and how SM differentiates from other anxiety disorders, especially social anxiety disorder (SAD), with which SM has numerous overlaps [15]. Identifying whether and how attention processing as well as the autonomic fear response differs between the two disorders could have implications for the understanding of the relation between SM and SAD.

2 Theoretical Background

2.1 Selective mutism as an anxiety disorder

SM is an anxiety disorder characterized by a consistent and predictable failure to speak in certain social situations that include; an expectation for speaking behavior, while speech production is unimpaired in

[1] Consistent with previous research [3], both autonomic nervous system activities and processes of attention are referred to as psychophysiological features in the project at hand

others [12]. Thus, children may talk to their parents at home but not to the children and teachers at school. The disorder has a typical age of onset during early childhood [16] and is associated with social and academic functioning impairments [17–19]. Epidemiologic studies indicate an average prevalence of about 1% of children and adolescents meeting diagnostic criteria for SM [16]. A study that screened a representative sample of elementary school students for symptoms of SM showed that 2.6% of all children showed marked SM symptoms [20]. It suggests that there might be a high number of unreported cases. It also shows that SM is associated with long-lasting communication difficulties, lower levels of functioning, and the presence of other anxiety disorders and symptoms of SM in adulthood [14, 13]. With the introduction of the DSM-5 [12], SM was, for the first time, classified under the anxiety disorders category. It is therefore implied that failure to speak predominantly occurs due to anxiety. Although features of other disorder categories such as oppositional behaviors or developmental delays also seem to be relevant to SM and refinement of the classification is discussed in this context [21], it is undisputed that anxiety/fear is a central feature of SM [15, 22]. A substantial overlap of phenomenological, etiological, and therapeutic characteristics between SM and other anxiety disorders [15] supports the classification of SM as an anxiety disorder. The overlap between SM and social anxiety disorder (SAD), characterized by a marked fear of social evaluation, is particularly strong [15]. Phenomenologically, children with SM report predominantly social fears as the cause of silence [23], experience elevated levels of fear in social situations [24], and are rated by clinical observers as highly anxious when expected to speak [18, 25, 26]. Children with SM exhibit other anxiety disorders on average in 80% and comorbid SAD in 69% of cases across different studies [22]. Etiologically similar factors extensive in SM are also central to other anxiety disorders [15]. Among other factors such as a genetic influence [27] or migration background [28], the temperamental trait of behavioral inhibition (BI), which is a risk factor of anxiety disorders [29] and is manifested by high levels of shyness, distress to novelty, and features of fear as early as infancy [30], has been identified as prominent in children with SM [31–33]. When it comes to treatment, there is preliminary evidence that fundamental intervention techniques (e.g., exposure therapy) that are established and highly effective in reducing the level of fear/anxiety also lead to symptom reduction in children with SM [34, 35].

Based on the extensive overlap between SM and SAD, there is a discussion about how SM and SAD are related [15, 26, 24, 36, 18, 37]. In this context, it was assumed that children with SM show an higher level of fear than children with SAD, and failure to speak occurs either through the extreme anxiety level or an avoidance mechanism associated with a reduction of intense arousal [36, 38, 24, 26]. Studies that have examined anxiety levels of children with SM compared with children with SAD; have been predominantly based on behavioral observation [18, 36] or subjective reports [24, 38]. These studies suggest that both disorders exhibit equal levels of trait social anxiety and state anxiety in non-speech-demanding social situations [24, 18, 26, 38]. Only in speech-demanding social settings are children with SM characterized by higher anxiety levels than children with SAD, both subjectively and in behavioral observations [24, 18, 26]. It suggests that anxiety levels do not differ fundamentally

between children with SM and SAD but depend on the situation. It is, however, problematic that central aspects, which are crucial for the occurrence of the symptomatology of anxiety disorders such as anxiety triggers, attention processing or the physiological fear response, have hardly been investigated in children with SM, nonetheless in the comparison between SM and SAD. Regarding the fear response in SM, there are findings for subjective reports [24, 32, 15] and behavioral observation [18, 36], but the physiological dimension is mostly unexplored. At the level of attention processing, no study exists in children with SM to date, making it impossible to assume possible differences between children with SM and SAD in this regard. An investigation of attention processing and the physiological fear response would be salient to clarify the relationship between SM and SAD. Moreover, there is currently little empirical research on stimuli or situations that trigger anxiety and thus failure to speak in children with SM. Given that the DSM-5 describes disorder-specific triggers for most anxiety disorders, including SAD [12], it would be essential to identify the triggers for SM.

2.2 Potential triggers of anxiety in children with SM

The definition of anxiety disorders in the DSM-5 states that: different anxiety disorders can differentiate in terms of the situations and objects perceived as a threat [12]. These triggers, for most anxiety disorders, are clearly defined in the DSM-5 (e.g., different types of social situations for SAD) and implemented in their diagnostic criteria (e.g., agoraphobia requires that a marked fear occurs in at least two of five described situations) [12]. In contrast, SM is the only anxiety disorder in which triggers of anxiety have not yet been described in DSM-5 [12]. That is, presumably, because there has been very little research on situational factors that trigger fear and thus the failure to speak in SM. Given the scarce research on anxiety triggers in SM, there appear to be different approaches to potential anxiety trigger in children with SM. Phenomenologically, SM largely overlaps with SAD [15] and can be considered a developmental variant of BI [31]. Accordingly, the circumstance that social fear is a central phenomenon for most children with SM suggests that social situations especially might be a predominant trigger of anxiety in SM. However, not all children with SM exhibit elevated levels of social anxiety [22], so other triggers may also be present for children with SM. Here, for example, research suggests the potential importance of; triggers related to reduced tolerance for mistakes [23] as well as triggers related to agoraphobia [37] or separation anxiety [37] in children with SM. In addition, it seems reasonable to assume that new and unfamiliar situations, which are known to induce distress in children with high levels of BI [39, 40, 30], also cause anxiety in children with SM. Seeing how children with SM are retrospectively described by high levels of BI in infancy [31], and BI is the strongest predictor of SM symptomatology during preschool age [33], this trigger might be of great relevance. Because of the extreme anxiety children with SM experience during verbal social situations [24], situations with a demand for speech can also be a primary trigger of anxiety in SM. Clinically, three situational factors that trigger the symptomatology of SM are considered in clinical manuals [41, 42]. These are; the *places* where the silence occurs, the *persons* with whom the child can't speak, and the *activities* during which the child fails to speak [42, 41]. Empirically, there are mixed findings regarding the identification of

these three factors. While a factor analysis of one diagnostic instrument could find exactly this factor structure with the factors; place, person, and activity [43], this was not the case based on another instrument [44]. Moreover, few studies to date have directly investigated which situational factors trigger the symptomatology of SM. These studies revealed that predominantly places and persons outside of the home, such as school and teachers, public or new situations, and persons not part of the core family induce failure to speak in SM [45–47]. In addition, stressful activities and activities in which children are the focus of attention have been found to induce symptoms of SM [47]. Despite the clinical relevance and initial evidence of the importance of these three major situational factors of anxiety triggers in SM, they have never been investigated systematically together in a single study. Moreover, recent studies investigated these only based on non-validated items with a closed-ended answer format.

2.3 Altered attention processing of threat and its potential relevance in children with SM

2.3.1 Altered attention processing of threat

In addition to the presence of a trigger, attention processing of the threat is critical to whether a fear response occurs according to models of anxiety [5]. While efficient attentional processing of threat is presumed to be adaptive, alterations or biases in attention processing of threat are considered a feature of pathological anxiety [5, 48]. In this context, three components of biased attention to threatening stimuli that occur at different stages of attentional processing are postulated by different models of anxiety [5]. These are a) an early occurring bias towards threat associated with a facilitated detection of threat (also called hypervigilance) and, at later stages of attentional processing, b) a delayed disengagement when a threat is detected or c) an avoidance of threat [5]. Early attentional bias describes; an orientation of attention towards a threatening stimulus, delayed disengagement, a prolonged fixation of attention on and avoidance and a turning of attention away from the threat [5]. While the early attention bias towards threat is viewed by different theories as an automatic threat detection mechanism [49–51], the two components that occur later are viewed more as a strategic process associated with deficits in regulatory abilities [49, 50]. Hence, most theories assume that individuals with high levels of anxiety initially detect a threat faster and subsequently have difficulties disengaging attention from it and/or avoiding it [2][5]. Attentional avoidance is considered especially relevant to the maintenance of anxiety disorders, which is supported by, for example, intervention studies showing that a reduction in attentional avoidance leads to a reduced anxiety level [52]. In children and adolescents with anxiety disorders, there is evidence at the transdiagnostic level both for the presence of an early bias toward threatening stimuli [53] and for greater avoidance of them [54], but not for difficulties to disengage from threat [54, 53]. Whether or not a stimulus elicits an attentional bias depends on content-specificity [55], meaning that certain stimuli elicit anxiety and an attentional bias only in certain anxiety disorders. It is,

[2] Because simultaneously delayed disengagement and avoidance are mutually exclusive, it depends on the paradigm and measurement method used to determine which of the two components, or whether both, are found sequentially at later stages of processing in individuals with anxiety disorders [5]

therefore, necessary to look at findings on a disorder-specific level. For example, In Albon et al. (2010) demonstrated in an eye-tracking study that children with separation anxiety focused longer on presented pictures of separation situations in an early phase of attentional processing and avoided them more in a late phase than children with TD [56]. Remarkably, these attentional biases decreased in the children with separation anxiety disorder after completing cognitive behavioral therapy, which may argue for the disorder maintaining the relevance of the biases [57]. In SAD, experimental studies suggest that direct eye contact in particular, and thus the eye region of the social counterpart, plays a central role in the induction of attentional bias [58–64, 48]. Studies with homogeneous samples of children with SAD consistently show an early attentional bias toward the threatening stimulus (e.g., the eye-part of the social counterpart) [48, 64, 63, 58], but are inconsistent regarding later stages of attention processing. At later stages, evidence found that children with SAD compared to children with TD had difficulty disengaging from threat [63] avoided it more strongly [65]or did not show any difference [64, 58]. However, only Keil et al. (2018) [58] examined gaze behavior specifically toward the eye region of the social counterpart as a threatening stimulus, whereas the other studies in children with SAD analyzed attentional biases only, unspecifically toward faces as such. These studies have used static stimuli, criticized for their lack of social relevance [54]. Recent studies in adults with dynamic stimuli suggest that the attentional bias associated with social anxiety also occurs at later stages of attentional processing, especially when the displayed stimuli are of high social relevance [66, 67]. Therefore, paradigms used in previous studies in children with SAD do not appear to have been well suited for examining biases occurring in later phases.

In addition to a bias of the direction of attentional focus on threatening stimuli, recent research suggests that the extent of visual exploration (the extent an individual looks around in the environment) might also be altered while experiencing fear/anxiety [68–71]. While the extent of visual exploration has not been studied in children with anxiety disorders, results in adults are mixed. Some studies suggest that adult individuals with SAD engage in more visual exploration during social interactions than healthy individuals, which has been interpreted as an indicator of hypervigilance in terms of increased scanning for threatening stimuli in the environment [69, 72]. In contrast, some studies suggest that threatening situations are associated with lower levels of visual exploration, which has been considered an indicator of reduced oculomotor activity and thus of a state of attentive freezing (see section 2.4.1) [68, 73, 71]. While the former studies have demonstrated increased visual exploration in free-viewing tasks [69, 72], the latter have identified reduced visual exploration as a function of the avoidability in aversive stimuli. Here, attentive freezing occurred particularly in situations considered to be unavoidable [73, 71]. Given that these studies were conducted exclusively with adults, it is thus far completely unclear how the extent of visual exploration in threatening situations is affected in children.

2.3.2 Potential relevance of altered attention processing in children with SM

In light of the content-specificity of attentional biases described above [55], the question arises as to which stimuli elicit fear and thus an attentional bias in children with SM. Given that most children with

SM report social fear [23] and similar anxiety levels in social-evaluative situations as children with SAD [24], it seems reasonable that threatening cues can be considered immanent to social situations, for most children with SM. As described above, for individuals with SAD, direct eye contact with the social counterpart, in particular, induces attentional bias in social situations [58, 60–62]. It can, therefore, be assumed that eye contact from the social counterpart also represents a threatening stimulus in social situations for those children with SM, for whom social anxiety is a central phenomenon. Consistently, several clinical case reports describe that children with SM avoid eye contact with their social counterparts [74–76], suggesting that an attentional bias related to the eye region can be found in children with SM. This potential attentional bias in children with SM might be even more prevalent in speech-demanding social situations, as indicated by higher levels of anxiety in children with SM in verbal situations, than in children with SAD [24]. Despite the theoretical and empirical foundation that biased attention processing plays a crucial role in the context of pathological anxiety, its potential significance in children with SM has never been investigated experimentally.

2.4 Altered autonomic activity in response to threat and its potential relevance in children with SM

2.4.1 Altered autonomic activity in response to threat

Similar to attention processing of threat, an efficient fear response is considered adaptive [77]. An immediate fear response mediated by the autonomic nervous system, allows for a swift adaptation to the threat [78]. Alterations in the fear response and the underlying autonomic processes are considered a feature of pathological anxiety [6]. The activity of the autonomic nervous system consists of a sympathetic and parasympathetic strand and can be subdivided into two different components [79]. On the one hand, tonic activity describes the basic level of arousal both during and without a threat [79]. High sympathetic (equivalent to a low parasympathetic) tonic arousal is associated with the low responsiveness of the autonomic nervous system [80]. Contrastingly, phasic activity describes the physiological response to a threat [79]. Anticipation of [81]; and confrontation with a threat leads to increased sympathetic arousal (equivalent to a decrease in parasympathetic activity), called reactivity. Arousal[3] recovers after the threatening situation has passed, referred to as recovery [82]. Both; autonomic basic arousal levels and the change of autonomic arousal from non-threatening to threatening and back are critical correlates distinguishing autonomic activity between individuals with high levels of anxiety and typically developing individuals [77, 78]. Transdiagnostic models of anxiety propose increased tonic arousal reflecting reduced responsiveness in combination with a blunted fear response (lower reactivity and slower recovery) to be characteristic of individuals with high anxiety levels [77]. This activation pattern is also called restricted autonomic flexibility [78, 77], illustrated in Fig. 1. The rationale behind this theory is that a reduced responsive autonomic nervous system that mediates a blunted and thus inefficient response does not allow for a fast and active adaptation to the threat (for

[3] In the following, high autonomic arousal means high sympathetic arousal, which is equivalent to low parasympathetic arousal

example, through flight or fight), whereas a quick, strong fear response does [77, 78]. Restricted autonomic flexibility is thus considered a marker of inefficient regulation of fear/anxiety.

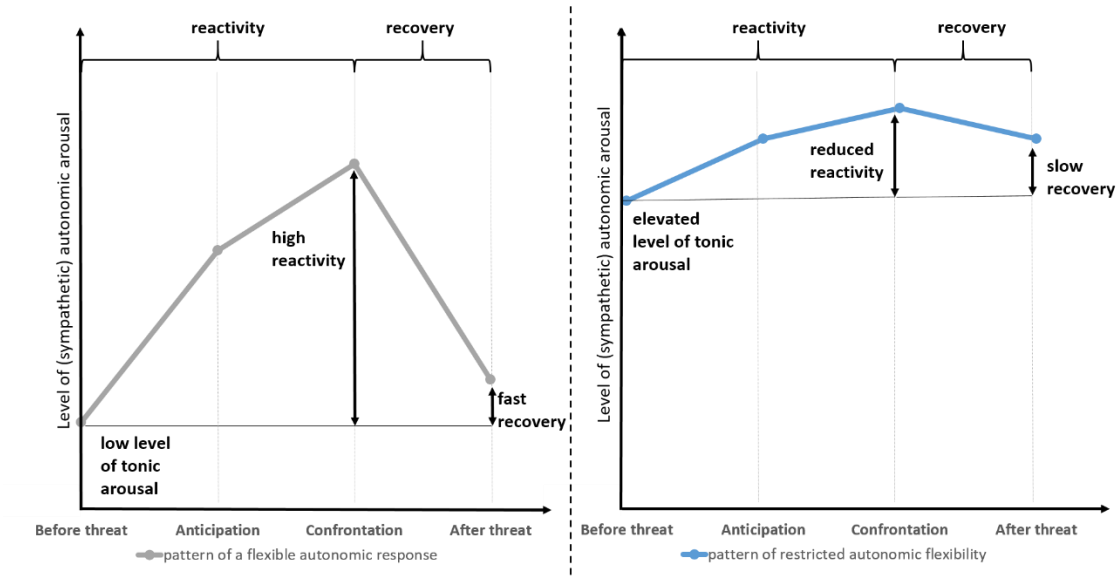

Fig. 1: Tonic and phasic characteristics of a flexible (left) and inflexible (right) autonomic response.

Overall, both a blunted phasic fear response and tonic increased arousal (reduced responsiveness) have been empirically shown to be characteristic of individuals with high levels of anxiety, including children with different anxiety disorders [82–86]. For example, Dieleman et al. (2015) showed that a sample of children with mixed anxiety disorders had higher tonic autonomic arousal than children with TD [83]. Furthermore, children with SAD and high levels of social anxiety consistently exhibited restricted autonomic flexibility [82, 84, 85]. In these studies, a fear response was measured continuously before, in anticipation of, and during evaluative stress tasks in which children had to retell a story and count out loudly in front of an unfamiliar experimenter in a laboratory setting. Subsequently, the children's autonomic fear response was measured during a thirty-minute recovery period, which was this long because the paradigm also measures the slow-responding stress hormone cortisol [85]. Children with SAD and elevated social anxiety showed tonically increased arousal as well as reduced reactivity and slowed recovery [82, 85, 84]. Remarkably, increased tonic arousal at rest in a familiar environment in children with SAD could also be found, thus without the influence of social stress potentially induced by an unfamiliar laboratory setting [86]. Moreover, the activation pattern of restricted autonomic flexibility does not appear to differ fundamentally between most anxiety disorders [6, 4].

Before an active adaptation to a threat mediated by the autonomic nervous system occurs, there seems to be a freezing reaction also mediated by the autonomic nervous system [87]. Freezing is understood to be a broader fear response, which is associated with autonomic changes (e.g., drop in heart rate) and inhibition of motor activity, including speech production [87, 88] and eye movement [68]. Even though the phenomenon of freezing in human individuals is still poorly understood and little empirical research exists on it [87], research suggests that freezing typically occurs as an adaptive and

initial fear response that facilitates a subsequent reaction (e.g., fight or flight) [68]. Therefore, in individuals without an anxiety disorder, freezing is a short initial reaction of orientation followed by a switch towards an active mode [87]. On the contrary, the few studies on individuals with anxiety disorders suggest a more severe or prolonged freeze response. Here, individuals remain passive due to overarousal [89, 90].

2.4.2 Potential relevance of altered autonomic activity in response to threat in children with SM

The only two studies on autonomic activity in children with SM revealed mixed findings regarding autonomic flexibility in children with SM [25, 91]. Both studies used a non-standardized speech-demanding social stress paradigm in which children had to speak to unfamiliar audiences. In the study of Heilman et al. (2012), autonomic activity was also measured before, in anticipation of, during the stress task and a two-minute phase of recovery [91]. Children with SM showed, compared to children with TD, increased tonic arousal during the baseline condition in a laboratory setting but did not show a reduced reactivity or recovery in response to the speech-demanding situation. Unfortunately, this study did not include a group of children with SAD, so based on this study; no conclusion can be drawn about differences between SM and SAD in terms of autonomic activity. Because a recommended minimum five minutes for phases to reliably measure autonomic measures is needed [92], the two-minute recovery phase applied in this study is too short. Young et al. (2012) compared the autonomic activity before and during the stress task [25] in the second study. They did not find any differences between children with SM, SAD, and TD during a baseline measurement in a laboratory setting. Although, children with SM had lower autonomic arousal during a speech-demanding situation than children with SAD and comparable arousal to children with TD [25]. The lower autonomic arousal of children with SM, compared to children with SAD, was interpreted in terms of silence operating as an avoidance or emotion regulation mechanism leading to a reduction in arousal [25], thus maintaining silence in the long run [93, 94]. However, it is mentionable that the study did not include a nonverbal fear-inducing situation. Therefore, it remains unclear whether the reduction in arousal does not occur in situations in which children with SM are not expected to speak. Unfortunately, Young et al. (2012) did not investigate the course, and thus no arousal changes across different phases of the stress task, making it impossible to conclude reactivity or recovery [25]. Neither study examined tonic arousal in the absence of social stress, so no conclusion can be drawn about whether tonic arousal was increased or only due to the presence of strangers in an unfamiliar laboratory environment. However, given that children with high levels of anxiety, including children with SAD [83, 86, 85, 82, 84], can be characterized by restricted autonomic flexibility as described above, it seems highly likely that restricted autonomic flexibility might also be present in children with SM. In addition, indirect evidence exists in the literature based on questionnaires, clinical case reports, and behavioral observations suggesting that overarousal and an associated prolonged freezing response may be relevant in children with SM. These are, for example, elevated levels of the temperamental feature BI in children with SM [31–33], behavioral tasks in which children with SM showed a longer latency to move [18], reports from a subset of children describing

paralyzing fear as a cause of silence [23], and clinical descriptions of children with SM as "frozen with fear" [95]. However, psychophysiological markers associated with the broader fear response of freezing, such as reduced visual exploration [68], have not yet been studied in children with SM.

2.5 Aims

This dissertation aims to gain a better understanding of the different features: anxiety triggers, attention processing and autonomic activity, which are crucial to symptomatology of anxiety disorders [2], in children with SM. Each of the individual components is mainly unexplored in children with SM; however, all are important in understanding and treating the disorder. First, it aims to identify situational factors that trigger anxiety in children with SM. Given that anxiety triggers are disorder-specific [12] and are mainly unexplored for SM, it aims to investigate these based on an open response format in a sample of children with SM. Thereby, it intends to prevent important triggers from being neglected a priori. Second, it aims to investigate; whether and in which manner; attentional processing of threat alters in children with SM during fear-inducing social situations. While biases in attentional processing of threats have been found in children with other anxiety disorders [56], including children with SAD [58], they have not yet been studied in children with SM. For this purpose, attention processing of; direct eye contact with a social counterpart at different processing stages, and the extent of visual exploration, will be investigated based on an eye-tracking paradigm in children with SM. Third, the project aims to examine the physiological dimension of the fear response in children with SM. While several studies already exist on the dimensions of subjective fear and overt behavior in children with SM [15], the physiological dimension of the fear response is largely unexplored. Thus, the autonomic fear response of children with SM in response to fear-inducing situations is investigated. This could shed light on psychophysiological mechanisms underlying failure to speak. Beyond that, it aims to compare attentional processing and autonomic activity between children with SM and children with SAD. A comparison of these features could contribute to the question of how SM and SAD are related, considering that previous evidence is largely based on questionnaire data or behavioral observations. Accordingly, children with SM examined in studies two and three and children with TD, are compared to children with SAD. Overall, a better understanding of anxiety triggers, attentional processing of threatening stimuli, and autonomic fear response could expand the understanding of SM as an anxiety disorder. It might have important implications for future research, diagnostics, and clinical practice.

3 Studies I, II, and III

3.1 Study I

Although SM is classified as an anxiety disorder and is characterized by predictable failure to speak in certain situations [12], triggers that induce anxiety in children with SM and thus failure to speak have rarely been studied. The identification of disorder-specific anxiety triggers is important to differentiate anxiety disorders from each other and informs the diagnosis and treatment of anxiety disorders. Given that the scarce previous research has examined anxiety triggers in SM in a closed-response format [45,

47], it is questionable whether important triggers of person, place and activity may have been missed. Furthermore, the three major factors have not yet been investigated together so far.

3.1.1 Aims

The study [96] aimed to investigate anxiety triggers for the three major factors: place, person, and activity in children with SM. Given that situational triggers of symptoms in SM is a scarcely explored aspect that has not yet found its way into the description of SM in the DSM-5, it is aimed to examine triggers based on an open-ended response format.

3.1.2 Methods

In total, N = 91 parents of children aged 3 to 17 years and elevated level of SM symptomatology completed an online questionnaire asking about characteristics of place, person, and activity that trigger anxiety in their children. Symptomatology of SM was assessed using the evaluated parent questionnaire Frankfurt Scale for the Assessment of Selective Mutism (FSSM). Open-ended responses were analyzed for the three major factors using a qualitative content analysis, and interrater reliability was assessed based on an independent and blinded rating of responses.

3.1.3 Results

Parents reported characteristics that induce fear in their children for all three situational factors. Most striking for the factor person was a *lack of distance* (reported by 45% of the parents), for the factor place *unknown places* (56%), and the factor activity *new activities* (47%). Other reported anxiety triggers for the factor person were authority characteristics (36%), low familiarity (33%), external characteristics (25%), little child-focused (22%); for the factor place: crowds (44%), places with negative experience (21%), high volume (13%); for the factor activity: motor activities (27%), failure (25%), the focus of attention (22%), activities with speech demands (19%).

3.2 Study II

While research has identified restricted autonomic flexibility associated with reduced adaptability to threatening situations in children with other anxiety disorders including SAD [83, 82], it is as yet unclear whether children with SM also exhibit an inflexible fear response. Furthermore, psychophysiological mechanisms underlying failure to speak in children with SM are largely unclear to date. Although it is proposed that failure to speak functions as an avoidance mechanism leading to a reduction in intense fear arousal [26], psychophysiological studies in children with SM are scarce.

3.2.1 Aims

The study [97] aimed to assess whether children with SM, like children with SAD [82], can also be characterized by a restricted autonomic flexibility consisting of a tonically increased autonomic arousal in absence of a threat [86] and a blunted fear response in threatening situations [84, 82]. Therefore, it was hypothesized that both children with SM and children with SAD would show increased arousal in the absence of social stress and reduced reactivity and slower recovery in response to social stress. In addition, to investigate possible psychophysiological mechanisms associated with failure to speak, it

was aimed to measure autonomic activity in both a fear-inducing verbal and a fear-inducing non-verbal situation. The rationale behind this aim was that in contrast to nonverbal social stress tasks, verbal situations can be avoided through failure to speak.

3.2.2 Methods

In a total of N = 96 children aged 8-12 years (SM: n = 31, SAD: n = 32, TD: n = 33), autonomic activity was measured during a baseline measurement at rest and during two social stress paradigms containing either a verbal task or a nonverbal task. The resting baseline measurement took place in familiar surroundings and without the presence of a stranger, so that possible social stress was reduced to a minimum. In the verbal stress task, the children had to retell and continue a story, and in the nonverbal stress task, the children had to draw a picture story based on a story previously read to them. Autonomic activity was measured over different phases before, during and after the stressful social situations to investigate to what extent children show restricted reactivity as well as slow recovery.

3.2.3 Results

Children with SM showed a reduced reactivity in nonverbal situations compared to children with typical development (TD). However, not in verbal situations. There was no evidence for a slow recovery of autonomic arousal in children with SM. In addition, for the first time, it showed that children with SM exhibit tonically increased arousal even in non-social situations absent of threat compared to children with TD and children with SAD. Therefore, the results demonstrate that children with SM show indications of restricted autonomic flexibility. However, the study did not replicate findings from research on children with SAD of an increased tonic autonomic arousal in the absence of stress and restricted autonomic flexibility during a social stress paradigm compared to children with TD. There were also no differences regarding autonomic responses to the stress paradigms between children with SM and children with SAD. Furthermore, it appeared that not all children with SM actually failed to speak in the verbal stress task.

3.3 Study III

Alterations in attention processing of threatening stimuli are a feature of individuals with anxiety disorders [5]. Attentional biases regarding threat have already been identified in children with various anxiety disorders including SAD [58]. Although attentional biases represent a promising target in the treatment of anxiety disorders [9], they have not yet been studied in children with SM. Apart from the direction of attentional bias; altered levels of visual exploration in threatening situations also appear to be related to pathological anxiety [68, 69]. The literature suggests that both attentional avoidance of eye contact [74] and motor inhibition due to a freezing response [95] may be relevant in children with SM. Neither has yet been studied based on psychophysiological measures in a quasi-experimental research design.

3.3.1 Aims

The study [98] aimed to investigate attentional processing of threat in children with SM compared to children with SAD and children with TD. It has been hypothesized that children with SM, like children

with SAD, also exhibit an early attentional bias toward the eye region of the social counterpart compared to children with TD. Given the heterogeneity of findings regarding the later occurring components of attentional bias (avoidance and difficulties to disengage) in children with SAD [58, 64, 63] as well as regarding the extent of visual exploration in adults [69, 68], these variables were investigated in an exploratory manner. In order to be able to suitably investigate the later occurring components of attention processing, it seems vital to use dynamic stimuli with greater social relevance.

3.3.2 Methods

In a total of N = 84 children aged 8-12 years (SM: n = 28, SAD: n = 28, TD: n = 28), attentional processing was examined using eye tracking while the children watched videos with different types of social situations. In these videos, a social counterpart either asked a question, gave a social evaluation, or made a neutral statement. Early attentional bias was measured in accordance with previous research by fixation time on the eye region of the social counterpart during the first 500ms of the video. Attentional avoidance and difficulties to disengage were assessed by fixation time during the course of attention processing. Extent of visual exploration was measured by the length of the scanpath in pixels.

3.3.3 Results

No group differences in fixation time, at early stages of attentional processing or later stages during the course of attention processing of threat, were found. It demonstrated that symptom severity of SM is related to less fixation time on the eye region and thus attentional avoidance of the social counterpart's gaze across all conditions. Previous findings that children with SAD showed an early attentional bias towards threatening stimuli; could not be replicated. Neither were differences found between children with SAD and children with TD, nor differences between children with SAD and children with SM. There were also no differences between the groups with regard to fixation time over the course of attention processing and thus no indication of a late-occurring attentional bias. Evidence of reduced visual exploration during social situations in children with SM has been found. There was no difference in the extent of visual exploration depending on the content of the social situation (whether the actor asked a question, stated a social evaluation, or a neutral statement). An exploratory regression based on the entire sample further revealed that the extent of children's freezing in social situations, measured by items from a parent-rated questionnaire on SM, predicted the extent of visual exploration (the higher the freezing, the less visual exploration).

4 Discussion

4.1 Summary of study results

The present dissertation identified interesting new insights into anxiety triggers, attention processing and autonomic activity in children with SM. The first study revealed; which situational factors trigger anxiety and thus failure to speak in children with SM. Triggers were identified for all three factors of place, person, and activity in a single analysis. Second study showed the first evidence for restricted

autonomic flexibility in children with SM. For the first time, autonomic responses for verbal and nonverbal social stress have been differentially investigated in children with SM. Differentiating between social stressors with and without a speech demand showed that the autonomic fear response in children with SM differs between these situations. The third study, following previous studies in children with SAD, is the first that investigated a possible attentional bias toward the eye region of a social counterpart in children with SM. Here, contrary to expectation, neither children with SM nor children with SAD showed an early- or late-occurring attentional bias in the dynamic social situations. It was found that the higher the level of SM symptomatology, the stronger the attentional avoidance of the eye region. The finding that the higher the level of the freezing items of the FSSM, the lower the visual exploration in the whole sample, indicates an association between visual exploration and freezing. Reduced visual exploration regardless of video condition was found in children with SM, suggesting a potential relevance of freezing in SM. The significance of these findings; for the relationship between SM and SAD and the extent to which these aspects are related to failure to speak is discussed below. Furthermore, implications for therapy and research can be derived from the findings.

4.2 Discussion in light of current literature

Regarding anxiety triggers, triggers for all three major factors: person, place, and activity were identified. Generally, keep in mind that the open response format requires reported fear triggers to be freely recalled, and thus triggers that are less evident or too obvious are probably not recorded. Triggers; to be expected due to the phenomenological proximity of SM to BI [31] and SAD [15]; were identified. Most strikingly, triggers associated with unfamiliarity (unknown persons; unknown places; new activities) for all three major factors were. Infants with high BI levels often show a fear response towards new and unfamiliar situations [39, 31], described by the subdimension of BI distress to novelty [30]. Given the high relevance of BI during infancy in children with SM [31] and a strong association between BI and SM-symptomatology during the age of preschool [33, 32], this finding is in line with expectation. In addition, the reported trigger of authority or activities while being the center of attention corresponds to typical anxiety triggers of SAD [12, 99]. Surprisingly, activities with speech demand were reported as anxiety triggers in only 19% of the cases. In addition, the other reported triggers of the factor activities (new activities, motor activities, failure, focus of attention) are not explicitly limited to verbal situations. At first glance, this seems surprising because the core symptom of SM (silence) can inherently only occur in verbal situations. However, this result might be explained by the substantial overlap between SM and SAD and BI [15, 31] and the fact that symptoms of SAD and characteristics of BI also occur in nonverbal social situations [12, 30]. As expected, anxiety triggers not primarily related to social anxiety constructs or BI also became evident. As such, activities possibly associated with failure and the reduced tolerance for mistakes in children with SM assumed in the literature [23] have been reported. In addition, crowds were reportedly anxiety triggers for the place factor, consistent with findings that agoraphobia frequently occurs in adolescents with SM [37]. The reported triggers persons with lack of distance, including persons that put the child under pressure and places with negative experiences, are not yet

explicitly described in the literature for children with SM but are already indirectly considered in the treatment of SM. In this context, the defocused communication technique is used in various SM therapy approaches [42, 34, 100, 101], in which communication with the child takes place without pressure or expectation to speak. This approach also takes account of the concern that children with SM should not have the unpleasant experience of not being able to talk at the beginning of therapy. The assumption that the experience of failure to speak contaminates places makes speaking in that place less likely [102] seems to be supported by this result.

Regarding attention processing of threat, both potential early and late occurring attentional biases have been investigated in study II in children with SM across different types of social situations. Because this was the first study to examine attention processing in children with SM, there are no studies in the context of SM to compare the results. Contrary to the assumptions, the results of the project at hand suggest that children with SM do not detect the eye region of the social counterpart in an early phase of a dynamic social situation faster than children with TD. As suggested by various theories of anxiety [5] and meta-analyses in adults and children with various anxiety disorders [53, 49], faster detection of a threat is a typical feature of anxiety disorders. Given that an early bias towards threat is already present in children with high BI [103, 104], considered to be children at risk for SM [31], and consistently shown in children with SAD [58, 63, 64, 48], it seems rather unlikely that an early attentional bias is not present in children with SM. Furthermore, probably for methodological reasons to be considered, this consistently shown early attentional bias could not be replicated, in this project, for children with SAD. Here, differences between applied paradigms in the studies of children with SAD and the project at hand might account for the inconsistent findings. While this project examined attentional bias toward dynamic social stimuli, previous studies have contrasted stimuli of different fear-inducing intensity and examined which stimulus is more likely to be detected or avoided [48, 64, 63, 58]. Although the analysis of gaze behaviors; in more naturalistic dynamic social situations; entails a higher ecological validity [66], the study of attentional biases in children with SM using established paradigms would be important for a better comparison of these clinical groups. Regarding late-occurring attentional biases, the association between severity of the symptomatology of SM and attentional avoidance of the social counterpart's eye area is consistent with descriptions from case reports that children with SM avoid eye contact [74]. It was, however, not detected in the group comparisons, possibly due to low statistical power for group comparison in the study at hand (power for group-comparison ranged between .65 and .81, whereas dimensional analyses had sufficient power, see study III). The finding, nevertheless, indicates that SM is related to a bias at a late stage of attentional processing that is possibly associated with strategic processing [49, 50]. According to theories of anxiety, attentional avoidance of threatening stimuli might contribute to reduced anxiety levels and thus to the maintenance of the anxiety disorder in the long term [52]. Given that children with SM experience intense levels of fear in social situations [24], and the assumption is that children with SM might not be able to speak due to an overwhelming fear [45], gaze avoidance might represent a maladaptive strategy

to reduce intense levels of fear in social situations. Given the relation found between SM symptomatology and attentional avoidance across different social situations, this might be a mechanism in SM operating in social situations in general. In light of the findings that attentional avoidance appears to play a role in the maintenance of anxiety disorders [52, 57] and can be reduced by CBT [57], attentional avoidance might be important to consider in the therapy of SM as well (see clinical implications). No evidence regarding the third possible attentional bias, the difficulty of disengagement, was found in this project. Thus, children with SM do not seem to be captured by the eye region of the social counterpart. Consistently, there is no evidence for this bias on the transdiagnostic level in children with anxiety disorders [53, 54]. In children with SAD, only one study found evidence that children with SAD have difficulties shifting attention away from threat [63]. However, the authors also found that the bias was stronger in children with SAD the higher the children's symptom severity [63]. Compared to this study, in which children with SAD had 2SD higher social anxiety than children with TD, children with SM and SAD both had only 1SD higher social anxiety in the current project. In addition, the current project does not indicate increased exploration and thus no hypervigilance in children with SM and children with SAD, as suggested by studies of adults with SAD [69, 72]. This result seems surprising because, in this project, a free-viewing task with dynamic stimuli was applied similar to the studies with adults with SAD that found increased exploration [69, 72]. A possible explanation for this surprising finding could be developmental differences between children and adults, which are described for strategic attentional processing in general in the literature [53]. Given that the extent of visual exploration has not yet been studied in children with anxiety disorders, one can only speculate regarding possible differences between children and adults with anxiety disorders. One possibility might be that adult individuals with SAD have learned hyperscanning as a strategy to scan their environment for social threats, whereas children have not yet done so. Further research with different age groups or a prospective design would be needed to investigate this further. The reduced visual exploration found in children with SM (compared to TD) instead indicates inhibition of eye movement, which might be related to the phenomenon of (attentive) freezing (see the section below) [68]. The aforementioned is supported by the finding that; the higher the FSSM freezing items' score, the lower the extent of visual exploration across the whole sample.

Regarding the (autonomic) fear response, children with SM showed features of the typical activation pattern of restricted autonomic flexibility found transdiagnostically in individuals with various anxiety disorders, including children with SAD [77, 78, 83, 86, 82]. The typical pattern (Fig. 1), consisting of increased tonic autonomic arousal and a blunted fear response, was partially found in children with SM in the third study of the project at hand. While increased tonic arousal and reduced reactivity were found in children with SM, they did not show a prolonged recovery of arousal. It was the first study to measure tonic baseline arousal in children with SM in a familiar environment, as previous studies took place in a laboratory setting [91, 25]. It suggests that autonomic arousal in children with SM, similar to children with SAD [86], is already elevated in the absence of stress. Even though

tonically increased arousal is technically not part of the fear response because it is already present before a threatening situation, it contributes to an inflexible fear response because it is associated with reduced responsiveness [80]. According to psychophysiological theories, reduced responsiveness of the autonomic nervous system is associated with a fundamentally restricted ability for a rapid and profound reaction to a threat [77]. The resulting blunted and thus unsuitable response does not allow for a fast adaptation to the stressor [77]. Therefore, the dissertation provides evidence that children with SM might also have a limited ability to adapt to threatening/stressful nonverbal situations. Notably, only differences between children with SM and TD were evident, but there were no differences in autonomic activity between children with SM and SAD. It indicates disorder-unspecific autonomic alterations in children with SM, which are also found in other anxiety disorders [83, 82]. While previous studies showed that children with SAD also display slowed recovery after a social stress task [82], it was not the case for children with SM in the current project. It is important to emphasize that the recovery phase following the stress paradigms in this project was shorter than in previous studies examining autonomic flexibility in children with SAD [84, 82, 85]. Although the 5-minute phases applied in this project correspond to general methodological recommendations for measuring autonomic processes [92], they might not be suitable to identify slowed recovery from the social stressor in children with elevated social anxiety. In this context, it was found that individuals with high social anxiety continue to ruminate in the social situation's aftermath, which could further maintain arousal [105]. Therefore, based on the present results, it is not possible to rule out the possibility that arousal recovery is not altered in children with SM over a longer period. While autonomic activity in a nonverbal social stress task has never been studied before in children with SM, the results on autonomic flexibility regarding the verbal situation in children with SM are consistent with a previous study [91]. In both the previous and current study, there was no difference between children with SM and children with TD in the verbal stress task. In this context, the question arises why children with SM showed indications of restricted autonomic flexibility in nonverbal, but not in verbal situation. It might be explained by a function of silence or reduced speech production and the integration of findings from all three studies (see the section below).

The current project suggests that various triggers, not limited to verbal situations, can induce anxiety in children with SM. Furthermore, children with SM already appear to enter social situations with heightened tonic autonomic arousal, making an inflexible fear response more likely. Hence, it is unsurprising and consistent with studies based on the subjective report [24] and behavioral observation [18] that children with SM show elevated fear and thus restricted autonomic flexibility in nonverbal social situations. Given this background, it seems surprising that children with SM, by contrast, do not seem to show the maladaptive fear response in verbal situations. Despite the intent of the paradigm of study III, the speech demand of the verbal stress task did not result in all children with SM failing to speak. Even so, the number of words spoken during the verbal task (see study III) shows that children with SM spoke significantly fewer words on average than children with SAD and children with TD. Given that silence has been hypothesized to function as maladaptive emotion regulation or avoidance

mechanism that reduces anxiety in children with SM [25, 93, 94], this could also apply to reduced speech production. Thus, although speculatively, it seems reasonable that the lower number of spoken words was associated with reduced anxiety. Given that the level of subjective anxiety is associated with the level of autonomic inflexibility [106] and those verbal situations induce a high level of anxiety in children with SM [24], children with SM might have regulated their level of anxiety and thus the autonomic inflexibility of fear response in the verbal situation, albeit not deliberately, by reduced speech production. Because there is no requirement for speech in nonverbal social situations, a reduction of speech production might not be available as a regulatory mechanism in the yet fear-inducing nonverbal situations, resulting in an inflexible fear response. In line with this, children with SM in Study II showed reduced visual exploration, which can also be considered a maladaptive fear response (prolonged freezing), although speech was not required. Unlike the verbal situation of Study III, in Study II, children were not expected to speak, thus reduced speech production/silence was not available as a possible regulatory mechanism. However, it is important to consider that in addition to avoidance, freezing is also discussed as a mechanism of failure to speak [95]. It is essential to emphasize that the paradigms used in the project are not optimized to study the function of silence and possible freezing responses and disentangle these potential mechanisms of silence (see section future directions).

The dissertation might also contribute to answering how SM and SAD are related to each other. The results of studies II and III indicate that children with SM and children with SAD do not differ concerning the investigated psychophysiological processes. Thus, the project at hand suggests that phenomenological differences between SM and SAD are unlikely to be due to differences in attention processing and the autonomic fear response. Instead, children with SM seem to show unspecific transdiagnostic alterations in autonomic activity (i.e. restricted autonomic flexibility) that characterize children with other anxiety disorders, including SAD [83, 82]. Also, concerning attention processing of threat, there is evidence, at least in a late phase of processing, that SM is associated with alterations (i.e., attentional avoidance), which has been found transdiagnostically in children with various anxiety disorders [54]. Regarding the assumption that SM represents an extreme variant of SAD [45], findings that children with SM do not show acute expression compared to children with SAD concerning these psychophysiological processes might argue against this assumption. Children with SM did not display faster threat detection, more extreme restricted autonomic flexibility, or firmer/longer freezing of visual exploration than children with SAD. It seems consistent with the findings that children with SM do not show fundamentally higher levels of anxiety on a subjective level and in behavioral observations than children with SAD [22, 15, 18, 26, 24]. The project at hand provides evidence that this seems to be the case on the psychophysiological level as well. Therefore, it is reasonable to conclude that children with SM and children with SAD might be differentiated not by basic psychophysiological mechanisms, but in particular, by their specific anxiety trigger. This conclusion is consistent with the definition of anxiety disorders in the DSM-5 [12], which emphasizes the relevance of anxiety triggers in differentiating various anxiety disorders. Because the project at hand focused on the identification of possible triggers

in SM in an exploratory approach, triggers were not compared between SM and SAD. Therefore, based on the results of the project at hand, it is not possible to conclude which triggers can differentiate between SM and SAD. However, triggers assigned to the spectrum of social anxiety / BI and those that seem to be independent of it, and thus possibly disorder-specific for SM, were identified.

4.3 Clinical implications and future directions

Clinical implications

The dissertation might also have implications for the treatment of children with SM. Both the intrapersonal characteristics and the situational triggers studied in the project at hand might be critical to consider in the therapy of SM. For example, it might be crucial for therapists to consider that children with SM may already start the therapy session with increased fear arousal. Fear arousal is likely to increase when entering the session, as the unfamiliar surroundings and the unfamiliar therapist are strongly fear-inducing for many children with SM, according to the findings of this project. Accordingly, it seems promising to counteract this heightened level of anxiety by techniques such as defocused communication [101] or relaxation techniques. As already suggested by some therapy manuals for SM [42], the presence of motor inhibition in children with SM might emphasize the importance of implementing activation exercises to counteract this state of freezing. The finding that failure to speak appears to be an avoidance mechanism in (some) children with SM, which might be involved in the maintenance of the disorder, emphasizes the importance of exposure in therapy for children with SM, which is already a central intervention of most approaches to treat SM [42, 41, 34, 100]. Additionally, non-verbal social situations should also be explicitly considered in exposure therapy, as these children with SM seem to display restricted autonomic flexibility and thus probably a reduced ability to adapt to them. Also, the chance that the fear response is seemingly inflexible in children with SM might be important to consider in exposure therapy. Because children with SM might not respond swiftly to the anxiety trigger, it might be important to wait a sufficient amount of time between using staged exposure until a child with SM feels comfortable with the step. Seeing how children with SM seem to experience eye contact as aversive and avoid it, it appears prudent to make as little direct eye contact as possible at the beginning of therapy and to include holding eye contact as a goal during the exposure therapy. While in children and adolescents with high levels of anxiety, attention bias modification (ABM) appears to be promising as an intervention that directly targets attentional processing to reduce the level of anxiety [9, 107], the project at hand does not provide evidence that this would also be useful in children with SM. ABM primarily targets early attentional bias because it trains to direct attention away from a threat, whereas the current project does not suggest a need to modify early attentional bias in children with SM. Initial research in adolescents with high levels of anxiety that appears promising [54] suggests using visual search tasks where threatening stimuli have to be detected in a set of non-threatening distractors to address the avoidance of threat stimuli.

Future directions

The findings that both autonomic activity and attentional processing are altered in children with SM underscore the importance of further investigating these features and elucidating their etiological influence. Considering how the current project represents; the first study of attentional bias in children with SM and is the first to use dynamic stimuli in children with high levels of social anxiety; a replication of the results and examination of early attentional bias using established paradigms would be paramount. Given that encoding of social cues, including attention-processing, is considered only the first stage of social information processing [108] and research indicates the importance of alterations of additional stages in children with SAD [108], these might also be promising targets for research in SM. A question arises in this context as to whether and how additional stages of the transition from potentially biased attention to the behavioral outcome of failure to speak may be significant for the symptomatology of children with SM. Similar to children with SAD, it is conceivable that additional stages of social information processing alter in children with SM: the interpretation of the social situation (e.g., others laugh at me when I speak), the repertoire of possible responses to the social situation (e.g., failure to speak), and the evaluation of possible responses (e.g., expectation that others might not laugh, when I remain silent) might be promising to investigate in future studies. Because the eye-part of the social counterpart seems not to be a threatening stimulus for all children with SM (see study I), it would be noteworthy to examine attentional processing in children with SM towards other anxiety triggers as well. According to the identified anxiety triggers, it would be promising to investigate whether and how the gaze behavior of children with SM changes, e.g., in unfamiliar environments, towards unknown persons, and near people. Although the current project provided promising evidence for the potential mechanisms of failure to speak investigated here, modifications of the paradigms used would be sensible. A freezing response involves a broad activation pattern that includes markers of the autonomic nervous system, motor activity, including gaze motor activity, and occurs over a period of several seconds [87]. Therefore, in contrast to the paradigms used here, future studies should examine the interaction of all markers (autonomic response and gaze behavior) at a temporal resolution of seconds within a single paradigm. To be able to draw a valid conclusion regarding potential mechanisms of failure to speak and to link the psychophysiological measures to the occurrence of failure to speak, it would be essential that children are expected to speak in the applied paradigms. Additionally, a comparison of speech-demanding situations in which children with SM fail and do not fail to speak would be necessary (e.g., through successive increases in speech demand throughout a paradigm). In addition, it would be important to investigate a possible altered recovery of the autonomic fear response after a social stressor in children with SM over a longer period as in previous studies in children with SAD. Overall, an extension to younger age groups and children at risk for SM (high BI) would be necessary to examine whether the assumed mechanisms of failure to speak are present early in these children or develop during the course of the disorder. Additionally, the use of bigger sample sizes to investigate whether subgroups of SM can be characterized by different mechanisms that potentially need

different therapy approaches appears to be promising for future research. Following the identification of external situational factors in the current project using a qualitative approach, a quasi-experimental approach or an approach based on ambulatory assessment would be the next step to investigate these triggers quantitatively. To derive promising intervening approaches for SM and to guide further quasi-experimental research on SM, an etiological model based on these psychophysiological features, as is already the case for children with SAD [108], seems to be of great importance. While specific mechanisms derived from disorder-specific cognitive models of SAD are already addressed in therapy for children with SAD [109], this is not yet the case for children with SM.

4.4 Conclusion

In conclusion, the dissertation provides insights in triggers of anxiety, alterations in attentional processing of threat as well as the autonomic fear response of children with SM. All of these aspects have been largely unexplored, making this dissertation an important addition to the scarce psychophysiological research of SM. As has been shown in other anxiety disorders, including SAD, the project demonstrated that children with SM show signs of restricted autonomic flexibility and are thus likely to respond less adaptively to social stress. In addition, the project provides the first empirical evidence that attentional processing may be altered in children with SM, in that SM symptoms are associated with avoidance of eye contact. The alterations found do not appear to reflect specific psychophysiological processes of SM because these features occur transdiagnostically in children with anxiety disorders. Accordingly, the project's results suggest that children with SM and SAD do not differ concerning psychophysiological characteristics of attentional processing and autonomic activity. However, the identified anxiety triggers show various triggers can induce anxiety in children with SM. This, in contrast to psychophysiological features, could represent a possible starting point for differentiating SM from other anxiety disorders. In the project at hand, there was also initial evidence for the presence of the potential mechanisms of failure to speak, attentive freezing, and avoidance. Given that the applied paradigms were not designed for differentiating between possible mechanisms of failure to speak, current findings represent only initial evidence on which further research on mechanisms of SM should be built.

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Acknowledgments

Throughout the writing of this dissertation, I have received a great deal of support and assistance.

I would first like to thank my supervisor, Professor Dr. Christina Schwenck, for her valuable support through the whole process and her belief in my work. She not only inspired me to research on selective mutism, but also gave me a lot of freedom in the development and completion of my projects. I would also like to thank Dr. Angelika Gensthaler, who gave me valuable advice early on about selective mutism and always very helpful feedback on my manuscripts. Also thanks to Prof. Dr. Julian Rubel, who dedicates his time for reviewing my thesis. I'd also like to acknowledge the effort of Prof. Dr. Joachim Stiensmeier-Pelster and the rest of my dissertation committee members.

I would also like to extend my deepest gratitude to all the children and their parents who agreed to take part in our studies. These studies could not have been conducted without your help. I would also like to express my gratitude to the student research assistants who put a lot of time and effort into recruiting the families and collecting the data.

I am also extremely grateful to my (former) colleagues Daniela, Anne, Arleta and Anett, who have accompanied and supported me through the ups and downs of the doctorate.

I would also like to express my sincere thanks to my friends: Julian and Andre, for your constant company and longtime friendship; Jonas, for being such a good friend for so many years; Johannes, for the really great time we had together in Giessen; Philip, for the fun adventures together; Julian, for the evenings together in R-studios and at the chessboard; Florian, for keeping me fit; Jens, for the great hikes to clear the mind; Julie, for always believing in me.

I whole-heartedly thank Salomé for your support and consideration and for always being there for me.

Finally, I would like to thank my parents and my sister, who have always followed my path to a doctorate with interest, even though my mother was unfortunately unable to accompany it to the end. I would like to dedicate this work to her.

Publications

Vogel, F., Gensthaler, A. & Schwenck, C. Frozen with Fear? Attentional Mechanisms in Children with Selective Mutism. *Cognitive Therapy and Research* (2022).

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Schwenck, C., Gensthaler, **A., Vogel**, F., Pfeffermann, A., Laerum, S., & Stahl, J. (2021). Characteristics of person, place, and activity that trigger failure to speak in children with selective mutism. *European child & adolescent psychiatry*, 1-11.

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European Child & Adolescent Psychiatry
https://doi.org/10.1007/s00787-021-01777-8

ORIGINAL CONTRIBUTION

Characteristics of person, place, and activity that trigger failure to speak in children with selective mutism

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Received: 2 February 2021 / Accepted: 7 April 2021

Abstract

Selective Mutism (SM) is an anxiety disorder with predictable and circumscribed situations in which children remain silent while they speak unaffectedly in others. However, core features of anxiety inducing stimuli have rarely been studied so far. Parents of children with elevated SM symptomatology participated in an online-based study and answered open ended questions about specific characteristics of a person, place, and activity that elicit failure to speak in their child. The final sample consisted of $n = 91$ parents with children aged between 3 and 17 years ($M = 8.02$ years, $SD = 3.94$). Answers were analyzed by qualitative content analysis. Characteristics of a person were assigned to five categories with lack of distance as the most frequently reported feature. With respect to a place, the majority of parents mentioned unknown places as a silence trigger. The most frequently mentioned feature of an activity that was designated to be associated to silence was new activity. There were only few associations between the designation of these features, age, and gender. For the first time, anxiety inducing triggers related to person, place, and activity were comprehensively assessed in children with SM. This allows a differentiated and deeper understanding of an understudied disorder. The majority of characteristics can be associated with proposed etiological factors such as increased behavioral inhibition, conditioning processes, social anxiety, and a strong need for control. Implications for effective treatments are discussed.

Keywords Selective mutism · Qualitative research · Online study · Triggers for failure to speak

Introduction

Selective Mutism (SM) is a mental disorder with a typical onset in early childhood. According to the fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5), it comprises symptoms of consistent failure to speak in certain social situations that include an expectation for speaking behavior [1]. These situations must be circumscribed and predictable. At the same time, children with SM show unimpaired speaking behavior in other situations. Epidemiological studies indicate a mean prevalence rate of about one percent [2]. Therefore, the disorder cannot be described as rare, as it occurs at least about as often as autism spectrum disorder. However, compared to other mental disorders in childhood and adolescence, SM is highly underdiagnosed and understudied. Higher prevalence rates have been found in bi-/multilingual children [3, 4]. The disorder is associated with the child's severely impaired psychosocial functioning, and both, social and educational development are typically influenced considerably [5]. Additionally, the few longitudinal data available so far indicate that SM is not a temporary state that resolves by itself [6, 7]. In combination with the severe impairments caused by this disorder, a need for scientific findings that provide insights into the disorder and from which adequate treatment approaches can be derived becomes evident.

In DSM-5, SM was classified among anxiety disorders for the first time. This decision was based on a significant number of studies indicating anxiety as a central phenomenon of SM, a common etiology between SM and other anxiety

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Published online: 24 April 2021

disorders, and results from initial treatment studies [4, 8, 9]. High comorbidity rates between SM and other anxiety disorders, in particular with social anxiety disorders (SAD), have been found [9–11]. Etiologically, behavioral inhibition (BI) was found to display a significant role for anxiety disorders in general and for SM in particular. This temperament trait includes features of shyness, distress to novelty, and fear [12]. Early BI has been found to be a strong predictor for later SAD [13–15]. According to a retrospective evaluation by their parents, children with SM were rated as to be more behaviorally inhibited than children with SAD, children with internalizing disorders and typically developing children [16]. Other etiological factors discussed for SM are classical and operant conditioning [2] and reduced auditory processing in a smaller subgroup of children with SM [17–20]. Lastly, the few randomized controlled trial treatment studies available so far indicated promising results for behavioral/cognitive-behavioral treatments [2]. Interestingly, some of the successful treatment approaches added aspects of defocused communication and child directed interaction to the core features of behavioral treatments [21–24]. Defocused communication is characterized by taking direct attention away from the child, e.g., having the therapist sit next to the child instead of opposite from him, and establishing joint attention. In child directed interaction, the therapist lets the child take the lead, praises appropriate behavior, reflects what the child is doing, or imitates appropriate play.

Despite the definition of SM by the DSM-5, which requires the predictability of silence, the core features of anxiety inducing stimuli for children with SM have rarely been studied empirically yet. On the contrary, SM is the only anxiety disorder for which the DSM-5 does not specify anxiety eliciting triggers [1]. Clinically, three major factors influencing speaking behavior of children with SM have been identified, specifically the *person* with which the child interacts, the *place* where the interaction happens and the *activity* that is undertaken. Because a social situation is usually influenced by all three factors an interaction between the three components can be assumed and children are usually confronted with more than one anxiety eliciting stimuli [25]. The three factors were confirmed by a factor analysis of a screening questionnaire [26] although another screening tool has found only one factor on which all items loaded [27]. Empirical data on talking patterns of children with SM are sparse yet, but there are some single research results that indicate proof for the existence of such factors that have an influence on the speaking behavior of children with SM.

With respect to the *person*, unfamiliar people seem to trigger silence in the vast majority of children with SM [7, 28]. Some children with SM remain silent in the contact with other children in general, while others only cannot talk to specific children [7]. Additionally, the teacher frequently displays a person with whom children with SM cannot speak [29], and children with SM were found to remain silent more frequently in contact with adults compared to other children [28]. On the contrary, most children with SM speak with the members of their core family like parents and siblings, and familiarity has a positive influence on speaking behavior [7, 30]. With regard to the *place* of speaking behavior, most evidence indicates that children with SM remain silent in the school setting and day care while they usually speak unimpaired at home [7, 28–30]. Furthermore, public and new settings, family gatherings, and social events were found to be associated to muteness in children with SM [30]. Regarding the *activity*, being the focus of attention and being involved in stressful activities were found to have a negative influence on the ability to speak of children with SM [30].

In a quasi-experimental study that compared children with SM, SAD and typically developing children aged eight to 18 years, different categories of video scenes around the school setting were presented. Results indicated that children with SAD experience social evaluative situations as more fear eliciting as situations with speech demands, while children with SM rate both type of situations alike [31]. However, the situations presented in this study contained interacting triggers of person, place, and situation. It is therefore impossible to conclude which specific triggers were experienced by participants as causing anxiety. In a qualitative study children and adolescents aged 8 to 18 years were asked for the fear content and related cognitions responsible for remaining silent. Here, participants mentioned social fears regarding negative evaluation by others, fears of mistakes and language/voice related fears to be responsible for their muteness [20].

Despite of this insight in factors that have an influence on speaking patterns in children with SM, it must be noted that the majority of studies that addressed this question so far assessed the factors with closed question format and therefore might have missed important aspects of person, place and situation that were not asked for. Furthermore, no study assessed all three aspects at the same time allowing for a concise overview. The two studies that addressed the question more comprehensively only included children from the age of eight years. Because the mean age of onset for SM lies in preschool years, it remains unclear whether developmental aspects have an influence on the triggers for mute behavior. Therefore, the aim of the current research project was to assess characteristics of person, place, and activity with open ended questions and the adoption of qualitative evaluation of the answers given. With the help of an online survey, parents of children with elevated SM symptomatology participated in the current study, resulting in a comparably huge sample size and wide age range. The data available to date indicate familiarity of person, place, and activity to have an influence on speaking behavior in children with SM. Furthermore, an influence of circumscribed conditioned

places with negative experiences such as school on silence has been described. Regarding the activity, activities which contain attention and evaluation by others, activities with speech demands, and challenging activities with the possibility to fail were suggested to influence the speaking behavior of children with SM negatively. Due to the inductive approach of the qualitative analysis, no concrete hypotheses were formulated.

Materials and methods

Sample

Initially, $n = 224$ parents started the online-survey, of which $n = 127$ finished it. Of these 127 participants, $n = 36$ did not exceed the cut off for SM in a diagnostic measure for the screening of SM [27]. Therefore, our final sample consisted of $n = 91$ parents of children and adolescents ($n = 68$ female) with elevated SM symptomatology (SM). Inclusion criteria comprised an age of the child below 18 years as well as a score above the threshold for SM symptomatology.

Procedure

All parents participated in an anonymous online-based study conducted with the help of UNIPARK software. The survey was advertised through different media such as mental health professionals, inpatient and outpatient clinics, newspapers, online-forums, and schools. Initially, parents were informed about the study, and informed consent was given by button press. All parents answered a questionnaire about general information and standardized questionnaires. Then, parents were asked open ended questions that were anxiety eliciting for their child and would lead to silence. The study was approved by the local Ethics Committee of the Faculty of Psychology and Sports Science of the University of Giessen (Germany).

Assessment

Frankfurt Scale of Selective Mutism (FSSM) [27]

The Frankfurt Scale of Selective Mutism (FSSM) is a parent-rated questionnaire developed to screen for and evaluate Selective Mutism in children aged between three and 18 years. So far, the scale is freely available in German, English, Norsk, and Suomi (forward–backward translated). There are different versions for kindergarteners aged 3 to 7 years, schoolchildren aged 6 to 11 years, and teens aged 12 to 18 years. The questionnaire consists of a Diagnostic Scale with ten yes–no questions about the children's overall speaking behavior. The Severity Scale comprises 41 or 42 questions regarding the specific speaking behavior at kindergarten/school, in public, and at home that are answered on a 5-point-Likert-Scale. An evaluation of the FSSM revealed excellent reliability scores ($\alpha = 0.90$–0.98) and validity with a one-factor solution for the Severity Scale [27]. The diagnostic scale differentiates excellently between children with SM and a combined group of children with SAD, internalizing disorders and control children with sensitivities of 94–97% and specificities of 90–95% (AUC = 0.97–0.99) [27]. In the current study, we applied the sum score of the Diagnostic Scale (range 0–10) and the Severity Scale's relative score (range 0–1). The age-appropriate version (FSSM 3–7, FSSM 6–11 or FSSM 12–18) for each participant was applied. Only data sets were included in the study if the cut-off value for SM in the respective age group was exceeded in the diagnostic scale (see Gensthaler et al. [27]).

Open ended questions

All parents were introduced to the relationship between fear and mute behavior and then presented with three open-ended questions about the characteristics of a person, a place, and an activity that causes anxiety and mute behavior in their children (e.g. *"Please indicate which characteristics of a person/place/activity your child finds anxiety-provoking."*). Parents could answer these questions in a text field without word limitation and indicate as many answers as they wanted. No examples were given so as not to influence parents' response behavior.

Data analysis

A qualitative content analysis (QCA) was applied to each of the three open ended questions with the aim to classify answers into content categories [32, 33]. Following this procedure, all answers were read to get an overall impression in a first step. Then, text units that contained aspects of significant characteristics of a person/place/activity were highlighted and headings for these passages were formulated. These headings were then grouped into higher order categories, and a description of each category was formulated. Text units were then assigned to the different categories. Frequencies with which aspects of the different categories were named were calculated, and for each participant it was coded whether a certain category had been mentioned or not (1/0 coding), because parents could indicate as many aspects as they wanted. Finally, to assess inter-rater-reliability, an independent and blind researcher assigned all answers to the prescribed higher-order categories.

The statistical package for the social sciences (SPSS 24) was used to assess statistics about sample characteristics. Furthermore, Pearson correlations were calculated for the relationship between categories and age as well as severity

of SM symptomatology. If the categorial variable is coded with 0 versus 1, Pearson correlation equals point-biserial correlation. For the relationship between categories and bi-/multilingualism (0 = no, 1 = yes) and gender (0 = male, 1 = female) Phi-coefficient is indicated. Because of the exploratory nature of correlation analysis correction for multiple testing was not undertaken.

Results

Sample characteristics

All children were aged between three and 17 years ($M = 8.02$ years, $SD = 3.94$, $MD = 6.00$ years), 42.9% attended nursery school, 54.9% school, and 2.2% neither nor. Seventy-four children had been diagnosed with SM before, 15.4% were currently receiving psychotherapeutic care, and $n = 2$ children were treated with selective serotonin reuptake inhibitors. The sample comprised $n = 12$ children who were raised bilingual, and 67% of these bilingual children were born in Germany. In 20.9% of all families there was one parent with migration background, and in 3.3% of the families both parents were born in a country other than Germany. With 47.3% the majority of participants reported to have a monthly net family income between 3.000 and 4.999€, 38.5% indicated to have between 1.000 and 2.999€. Thus, an upper medium socioeconomic status of the sample can be assumed on average. The mean sum of the Diagnostic Scale of the FSSM was $M = 8.93$ ($SD = 1.124$), and the mean relative score of the Severity Scale was $M = 0.704$ ($SD = 0.122$).

Open ended questions

Overall, 86.8% of the participants gave at least one codable answer across all three questions. There were no differences in age, gender, or SM symptom severity between parents who gave a codable response or those who did not. Within the sample, participants differed with respect to the number of indicated categories of anxiety inducing characteristics related to a person ($M = 1.22$, $SD = 0.917$, Min = 0, Max = 4), place ($M = 0.90$, $SD = 0.746$, Min = 0, Max = 2) and activity ($M = 0.98$, $SD = 0.830$, Min = 0, Max = 4).

Anxiety inducing characteristics of person

Answers from $n = 69$ parents were categorized, the remaining $n = 22$ parents had either not answered the question or had given an answer that did not match the question ("Avoidance of eye-contact") or that reflected an unawareness of characteristics ("I don't know"). Within the answers of the 69 parents, 151 codable units were identified. With the help of QCA, five broader categories were extracted which are demonstrated in Table 1.

Of all parents who gave a codable answer, 45% indicated a characteristic of a person that fell into the category *lack of distance*. This category comprised descriptions of people that do not show sensitivity to the child's need for distance physically or psychologically, who address the child too directly or put pressure on the child by demands and expectations. About one third (36%) of the parents gave at least one answer that fell into the category *authority characteristics*. This category summarized behaviors and characteristics of a person that are usually perceived as authoritarian or aggressive. Also, about one third of the parents described features of *low familiarity* (33%), such as strangers and people who are difficult to assess or unpredictable to the child. Lastly, about one fourth of the parents gave an answer that fell into the category *external characteristics* (25%) and *little child-focused behaviors* (22%). The first category comprised visual or audible external characteristics which usually are perceived as neutral by the general population such as height, hair color, or high-pitched voice. The latter category included features of people that show little sensitivity in the sense of too much distance, clumsy contact with the child or people that do not try to connect emotionally with the child.

Inter-rater reliability of these categories was high with Kappa coefficient of $\kappa = 0.914$.

Anxiety inducing characteristics of place

With respect to anxiety inducing characteristics of a place, answers from $n = 61$ parents were categorized. Within the answers of these parents, 92 codable units were identified. With the help of QCA, four broader categories were extracted which are demonstrated in Table 2.

More than half of the parents (56%) gave at least one answer that fell into the category *unknown places*. This category comprised places that the child is unfamiliar with or which are associated with uncertainty with regard to procedures and little foreseeability in order how to behave correctly. Furthermore, 44% of parents indicated that places with a lot of people (category *crowds*) were anxiety inducing. About a fifth of the parents (21%) stated that *places with negative experiences* displayed places of fear. The category comprised places where the child had already had negative experiences or where it expected to meet certain people or expectations to talk. Lastly, 13% of the parents indicated that places with *high volume* or lots of noise caused fear in their children.

Inter-rater reliability of these categories was high with Kappa coefficient of $\kappa = 0.935$.

Table 1 Categories of reported anxiety inducing person-characteristics

Category	Description	Examples	Participants who reported this characteristic*	Characteristic compared to all reported characteristics**
Lack of distance	People who do not keep the distance, who get too close to the child physically; too directly address the child or demands and expectations that put pressure on the child; show little sensitivity to the child's need for distance	"Demanding people" "People who do not keep enough distance" "People who put her under pressure"	45%	28%
Authority characteristics	Behavior and characteristics of a person usually perceived as authoritarian or aggressive or group belonging to authorities	"Strict persons" "Loud voice" "Dominant persons" "Medical doctors"	36%	25%
Low familiarity	Strangers whom the child does not know and who are difficult for him to assess and unpredictable	"Strangers" "Not seen for a long time" "When it hardly knows the person"	33%	16%
External characteristics	Externally visible or audible per se neutral characteristics of a person	"Old age" "Male" "Tall"	25%	21%
Little child-focused	People who are not very focused on the needs of the child and show little sensitivity in the sense of too much distance or clumsy contact with the child, who do not try to get access to the child or who are insensitive to contact	"Closed people" "Unrelaxed and stiff" "Unfriendly"	22%	11%

*Only participants who had reported any codable characteristic ($n=69$); **$n=151$ codable units

Table 2 Categories of reported anxiety inducing characteristics of place

Category	Description	Examples	Participants who reported this characteristic*	Characteristic compared to all reported characteristics**
Unknown places	Unknown places that the child does not yet know or only knows a little, which are associated with uncertainty with regard to procedures and little behavioral safety	"Places that are new for my child" "If this is the first time anywhere" "Unknown place"	56%	46%
Crowds	Places with a lot of people	"When there are too many people in one place" "Many people in little space" "Many people"	44%	29%
Places with negative experience	Places where the child has already had negative experience or expects to meet or talk to certain people	"Medical practice" "Places where she is expected to speak" "Negative experience at this or similar place"	21%	16%
High volume	Places with high volume or much noise	"Volume dominates" "Noisy environment" "Loud noises"	13%	9%

*Only participants who had reported any codable characteristic ($n=61$); **$n=92$ codable units

Anxiety inducing characteristics of activity

With respect to anxiety inducing characteristics of activities, answers from $n = 64$ parents were categorized. Within the answers of these parents, 89 codable units were identified. With the help of QCA, five broader categories were extracted which are demonstrated in Table 3.

Of all parents who gave a codable answer, 47% indicated that a *new activity* would induce anxiety to their child. This category included activities that the child does not yet know and where the child does not know what to expect. About a fourth of the parents indicated that *activities associated with failure* (25%) and *motor activities* (27%) were anxiety inducing to their child. The first category comprised activities that the child cannot do or is afraid of failing and has not yet mastered, while the second category included motor activities and activities that could be potentially dangerous. Furthermore, 22% of the parents mentioned activities where the child could be the *focus of attention*, and 19% indicated that *speech demanding activities* would be anxiety provoking for their child.

Inter-rater reliability of these categories was acceptable with Kappa coefficient of $\kappa = 0.841$.

Correlations with child characteristics

For each of the three open ended questions, the fit of each category was correlated with age, SM symptom severity, bi-/multilingualism, and gender of the child. Results can be seen in Table 4.

With respect to characteristics of a person, there was only a negative trend between age and the category *little child focused* ($r = -0.203$, $p = 0.095$) indicating that the older a child with SM was, the less frequently caregivers identified distant and clumsy behavior of a person as an important feature for their child's silence. With regards to characteristics of a place, there were negative correlations between age and the categories *unknown places* ($r = -0.279$, $p = 0.029$) and *high volume* ($r = -0.268$, $p = 0.037$), and a positive correlation with *places with experiences* ($r = 0.312$, $p = 0.014$). Furthermore, there was a trend for a correlation between the category *high volume* and bi-/multilingualism ($\varphi = 0.249$, $p = 0.052$). The same category correlated negatively to gender ($\varphi = -0.272$, $p = 0.034$) indicating that high volume is more relevant for boys with SM and less for girls. Finally, regarding characteristics of an activity, there was a negative correlation between the category *new activities* and age ($r = -0.320$, $p = 0.010$) and a positive trend with

Table 3 Categories of reported anxiety inducing characteristics of activity

Category	Description	Examples	Participants who reported this characteristic*	Characteristic compared to all reported characteristics**
New activities	Activities that the child does not yet know, where he/she does not know what to expect and where the consequences are unforeseeable	"Everything that is new" "What he does not know and cannot judge" "The unknown activity"	47%	29%
Motor activities	Motor activities to be learned, activities that require courage or could be potentially dangerous	"Climb up somewhere" "Movements/activities considered to be dangerous" "Swimming, skating and other activities where he could lose control"	27%	17%
Failure	Activities that the child cannot do or is afraid of failing and has not yet mastered	"When she has to do something and is not sure if she can or can't do it" "Has previously had negative experiences with it" "When she feels overwhelmed"	25%	21%
Focus of attention	Activities where the child could be the focus of attention	"When many people are watching" "When she is observed by strangers" "If she attracts the attention of others in the process"	22%	18%
Activities with speech demands	Activities associated with talking to other people	"Something to talk about" "Speech required" "If you want her to speak in front of others"	19%	15%

*Only participants who had reported any codable characteristic ($n = 64$); **$n = 89$ codable units

Table 4 Pearson correlations/Phi-coefficients between age, SM symptom severity, bi-/multilingualism, gender and fit of category

Target	Category	Age	SM symptom severity (FSSM)	Bi-/multilingualism	Gender
Person	External characteristics	0.104	0.122	− 0.081	− 0.025
	Authority characteristics	0.182	− 0.021	− 0.054	0.178
	Low familiarity	− 0.087	0.142	− 0.136	0.075
	Little child-focused	− 0.203^{+}	− 0.047	− 0.061	− 0.148
	Lack of distance	− 0.196	0.062	0.179	− 0.089
Place	Unknown places	− 0.279*	− 0.166	− 0.188	− 0.061
	High volume	− 0.268*	0.131	0.249^{+}	− 0.272*
	Crowds	− 0.048	0.093	0.188	0.141
	Places with negative experience	0.312*	0.067	0.122	− 0.022
Activity	New activities	− 0.320**	− 0.100	− 0.146	0.224^{+}
	Activities with speech demands	0.079	− 0.007	0.014	0.077
	Failure	− 0.128	− 0.119	− 0.050	− 0.106
	Focus of attention	0.151	0.240^{+}	0.189	0.025
	Motor activities	− 0.027	0.212^{+}	− 0.064	− 0.085

FSSM Frankfurt Scale of Selective Mutism

$^{+}p < 0.10$; $*p < 0.05$; $**p < 0.01$

this category and gender ($\varphi = 0.224$, $p = 0.073$) indicating that parents of girls tended to mention this category more frequently than parents from boys. Furthermore, there were positive trends for the association between SM symptom severity and the categories *focus of attention* ($r = 0.240$, $p = 0.056$) and *motor activities* ($r = 0.212$, $p = 0.093$). All effect sizes are considered small except from medium effect sizes for the associations between age and *places with experiences* and age and *new activities*.

Discussion

The current study was designed to identify characteristics of the variables person, place, and activity that have an influence on the speaking behavior in children with elevated SM symptomatology. We chose a qualitative research strategy to ask parents about specific characteristics of these variables in an online survey.

In line with past research results regarding characteristics of a *person* as an interaction partner we had expected familiarity [7, 28] to display a crucial role for speaking behavior of children. Indeed, *low familiarity* was reported to be an important characteristic that prevents children with SM symptomatology from speaking. Behavioral inhibition has been found to display an important role for the etiology of SM [16], and distress to novelty is one core aspect of this temperament feature [12], which might explain for the strong reactivity of children with SM towards low familiarity. However, less than a quarter (22%) of the parents mentioned this feature spontaneously and it represented only 11% percent of all codable units. In contrast, the most frequently mentioned category was *lack of distance* with almost half of the parents indicating this feature to be associated with their child's silence. This characteristic has not been directly described to be associated with mutism in children yet. However, defocused communication and child directed interaction have been found to be important aspects in successful behavioral treatments [21–24]. These specific procedures take away any pressure to talk from the child and allow him to maintain the necessary distance and control over what is happening throughout the therapeutic process. Thus, they specifically address the increased need of many children with SM for control and less demanding social interaction. This finding might also explain the fact that some children with SM verbally interact with selected children or adults while they remain silent in interaction with others [7]. This supports the clinical experience, that being an unfamiliar person even may play into the clinicians' favor. This is true at least, if the clinician is able to connect with the child in a way that is completely stripped off any pressure to speak at first. Children with SM tend to rigidly and consistently divide their world into the people, places, and activities that are associated with either being able to talk or not being able to talk. Once the speaking-pattern is established and a person is assigned the role of a non-talking-person, it is more difficult for the child to overcome the silence than to start talking with a new person whom the child does not have a history of silence with. People on the non-talking side are said to be "contaminated". Lack of distance in the behavior of the interaction partner could still cause the child to feel that his scope for action is limited and

that the situation is inescapable. Such situations can cause a freeze response which is a passive coping strategy [34]. Among others, freeze response is expressed in immobility, including motor and vocal inhibition [35], which are typical symptoms in children with SM.

In line with some data indicating that many children with SM have difficulties talking to their teacher [29], and that they less frequently talk to adults than to children [28] we found *authority characteristics* to display an important role for silence with 36% of the parents mentioning this category. Again, this finding may reflect the fear component of the temperament feature behavioral inhibition [16]. Lastly, about a quarter of the parents mentioned certain external characteristics like an old age or body size and little child focused behavior like low interest in the child or hesitant and stiff interaction to be a reason for their child's silence. Even though the external characteristics mentioned comprised a wide range of very different and contradictory features between children, they are constant within children and therefore predictable as required by DSM-5 [1]. This finding may also indicate classical conditioning to display an etiological mechanism in SM. Since presenting little child focused behavior seems to play a role for a quarter of children, it would be reasonable to review formal requirements, e.g. the child needing to be present during the processing of formalities that are more likely to be carried out with the caregivers. During the diagnostic interview the clinician and the caregiver will usually speak about the issues a child has around his talking behavior. Instead of building up a playful and meaningful relationship with the child, the first contact with the child is stamped with little child focused behavior. Interestingly, there were no correlations of either category with age, SM symptom severity, bi-/multilingualism, or gender. Therefore, categories with respect to person do not seem to be specific for certain subgroups or sensitive to development and experience with the disorder.

With regard to the *place* of speaking behavior, most studies so far described the school setting and day care as locations where children with SM remain silent [7, 28–30]. Therefore, we had expected to find circumscribed places with negative experience to have an influence on speaking behavior of children with SM symptomatology. However, only a fifth of the parents (21%) indicated that *places with negative experiences* are crucial for the silence of their children, and these places not only comprised the school setting but also other places like medical practice or any place where the child had experienced speech demands in the past. The school setting may have been too evident for some parents to mention explicitly, and the result underlines the importance of trans-situational measurement of the symptomatology which is in line with previous studies. It also indicates that previous experiences make it more difficult for the child to overcome silence, a fact that displays an important indication for the treatment of children with SM as initial silence in the therapy setting should be avoided in order not to contaminate this place with silence. Here, people with whom the child speaks unaffectedly like usually caregivers could serve as co-therapists to enhance the likelihood of speaking behavior. In accordance with our assumption, more than half of the parents (56%) described *unknown places* to be significant for their child's silence. Again, this may demonstrate the strong behavioral inhibition and distress to novelty in children with SM [16]. The fact that the category unknown places was negatively correlated to age while the category places with negative experiences was positively associated to age could indicate a change in important mechanism in the course of development in children with SM. While behavioral inhibition plays a major role in younger children, classical conditioning takes on an increasing role with age and social experience. This result could also comprise important indications for successful intervention for children with SM. It seems to be important to expose young children with a variety of new experiences but at the same time be aware of the child to make positive experience and not to "contaminate" situations with silence. Therefore, gradual exposure in small steps with flanking measures to carefully encourage speaking behavior in children with SM such as defocused communication and child directed interaction [21–24] might be promising treatment approaches. Interestingly, places with a lot of people like *crowds* were mentioned to be an important factor for SM symptomatology by parents with 44%. It has only been described before that SM is linked to places with many people like family gatherings or social events [30]. However, in a study about the comorbidity profile of children with SM compared to children with SAD, it has been found that children with SM show an elevated rate of agoraphobia in adolescence with 27% as compared to 10% in those with SAD [36]. Therefore, agoraphobic tendencies appear to be present in a large proportion of children with SM at all age groups on a subclinical level and some of them may develop into agoraphobia in adolescence. Another aspect of crowds is that they contain publicity without much control over the people that can observe speaking behavior of the child [30]. The aspect of few control over a situation with lots of people may also be reflected in the fact, that none of the patients with SM and comorbid agoraphobia also presented with panic disorder in the study of Gensthaler et al. [16]. Clinically, crowds should be considered as part of exposure therapy. Lastly, a smaller category mentioned by parents were places with *high volume*. Although this characteristic of a place accounted for only 9% compared to all reported categories, it is noticeable that this percentage fits in well with the findings from research on reduced auditory processing in a small subgroup of children with SM [17–20]. Because this category was negatively correlated to age, it might be assumed

that developmental delay in auditory processing displays a role in some children with SM characteristics. However, this conclusion is speculative and longitudinal data on the development of children with SM in comparison to typically developing children are needed to draw a concise conclusion on this topic. Furthermore, the naming of this category was associated with the sex of the child and was reported more frequently by parents with boys. So far, there are no data on gender differences in specific SM symptoms available, but developmental delays are more frequently found in boys compared to girls in general, which might also explain for the gender difference here.

In line with past research [31] we had expected *activities with speech demands* and *attention by others* to have an influence on children's silence. Both categories were mentioned; however, only 19 and 22% of participants, respectively, named these characteristics of an activity as significant for their children's silence. Past research mainly focused on this kind of situations given the logical link to speech demanding situations and the high comorbidity rate between SM and SAD. This may have led to other important characteristics not being asked in other studies with a closed answering format. The majority of parents (47%) indicated *new activities* to be associated with their child's silence which again could be linked to an elevated distress to novelty due to high behavioral inhibition [36], and again a negative correlation with age was found for this category. About a fourth of the parents also mentioned *motor activities* (27%) and activities that the child associates with *failure* lead to SM symptomatology. This result fits well with our assumption that challenging activities are associated with the child's silence and other research that found increased fears of mistakes [20] in children with SM and the influence of stressful activities [30] on their speaking pattern.

Taken together, with the help of a qualitative research design and open-ended questions we were able to confirm findings from past research, and also identified new factors that were previously only indirectly associated with SM. Thus, we were able to replicate already known results and also gain new insights, which we see as a strength of this study. Another strength of the current study is the comparably huge sample size. However, our study also has several limitations to consider. All and above, the anonymous online-based study design comprises some disadvantages. A comprehensive diagnosis with a clinical interview was not possible here, and the inclusion was instead based on a screening questionnaire. We had chosen this approach because it contains a lower threshold for participation and therefore a higher representativeness regarding children with SM symptomatology. Also, the FSSM was proved to be an excellent measure to differentiate almost perfectly between children with SM and children with other disorders as well as typically developing children. However, a comprehensive diagnostic that also includes comorbidities would be desirable and should be the aim of future research. Such an approach would also rule out the possibility that the results were due to comorbidities rather than SM. With our approach, we can also not entirely rule out that other people than parents have filled in the questionnaires. Therefore, an online-based qualitative study is a good starting point to gain some first insights in an understudied phenomenon such as SM and makes it possible to derive hypotheses for further research, but definitely should be completed by laboratory-based hypotheses testing research approaches. Furthermore, girls were overrepresented in our study, and compared to the population, and children with migration background were underrepresented. Therefore, due to disproportionate distribution, some of the correlations between these variables and categories may have failed to meet significance and compared to epidemiological data our sample might not be representative for children and adolescents with SM. It is also possible that the procedure of an online survey specifically targeted parents who have an affinity for the Internet, which may have led to bias. Also, not all parents gave codable answers which might have limited the representativeness as well.

Another criticism is related to the qualitative methodology we used in our study. The process of category building in qualitative content analysis is to some extent influenced by the researchers' subjective views and the categories we defined might not have been perfectly distinct from each other. However, we followed the well-established guidelines and standards for qualitative content analysis to guarantee a maximum of intersubjectivity and trustworthiness for our analysis, which is also indicated by the high inter-rater-reliabilities which are in the range of "almost perfect" according to Cohen.

Conclusions

To conclude, there are several important aspects of a person, a place, and an activity that should be considered in the individual diagnostics and treatment of children with SM. Children and/or their parents should not only be explored regarding the obvious SM-typical situations such as speech demands and evaluation by others but also with respect to situations that contain other characteristics such as crowds, high volume, or lack of distance. Graduated exposure in small steps may be the treatment of choice to overcome unfamiliar situations and at the same time guarantee experiences of success rather than creating new experiences of perceived failure and contamination with silence. Behavioral treatment approaches that comprise little demands and scaled focus of attention such as defocused communication and child directed interaction could be promising against this

background. Future research should also consider subgroups with respect to age and gender, because different aspects of the factors found varied in their relation with these variables.

Funding Open Access funding enabled and organized by Projekt DEAL.

Declarations

Conflict of interest All authors have no financial relationships to disclose and declare that they have no conflict of interest.

Ethical approval The current study was approved by the local Ethics Committee of the Faculty of Psychology and Sports Science of the University of Giessen (Germany).

Informed consent All participants gave their informed consent prior to their inclusion in the study.

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Child and Adolescent Psychiatry and Mental Health (2021) 15:81
https://doi.org/10.1186/s13034-021-00430-1

Child and Adolescent Psychiatry and Mental Health

RESEARCH ARTICLE

Open Access

Psychophysiological mechanisms underlying the failure to speak: a comparison between children with selective mutism and social anxiety disorder on autonomic arousal

Felix Vogel* and Christina Schwenck

Abstract

Background: Selective mutism (SM) has been conceptualized as an extreme variant of social anxiety disorder (SAD), in which the failure to speak functions as an avoidance mechanism leading to a reduction of intense fear arousal. However, psychophysiological studies in children with SM are scarce and physiological mechanisms underlying the failure to speak are largely unknown. In contrast, children with SAD are characterized by a combination of a chronically elevated physiological arousal and a blunted physiological fear response to social stress. Due to the large overlap between SM and SAD, similar mechanisms might apply to both disorders, while differences might explain why children with SM fail to speak. The aim of our study is to investigate psychophysiological mechanisms of the failure to speak in children with SM.

Methods: We assessed in a total of N = 96 children [8–12 years, SM: n = 31, SAD: n = 32, typical development (TD): n = 33] resting baseline arousal in absence of social threat and the course of physiological fear response in two social stress paradigms, differing in terms of whether the children are expected to speak (verbal task) or not (nonverbal task).

Results: Children with SM were characterized by increased tonic arousal compared to the other two groups, and by a more inflexible stress response in the nonverbal but not in the verbal task compared to TD-children. Further analyses revealed that children with SM who did not speak during the verbal task already demonstrated reduced arousal in anticipation of the verbal task.

Conclusion: The increased tonic arousal generalized to non-social situations in SM could indicate a long-term alteration of the autonomic nervous system. Furthermore, the differential physiological stress response may indicate that silence acts as a maladaptive compensatory mechanism reducing stress in verbal social situations, which does not function in nonverbal situations. Our findings support the idea that the failure to speak might function as an avoidance mechanism, which is already active in anticipation of a verbal situation. Treatment of SM should take into account that children with SM may suffer from chronically elevated stress levels and that different mechanisms might operate in verbal and nonverbal social situations.

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Keywords: Selective mutism, Social anxiety disorder, Social phobia, Social stress, Cognitive variables, Physiological response

Introduction

Selective mutism (SM) is a mental health disorder in which affected children fail to speak in certain social situations where they are expected to, but their speech remains unaffected in other situations [1]. The disorder is associated with a chronic course of increased psychopathological symptoms persisting into adulthood and resulting in impairments in academic and socio-emotional development [2–4]. Previous research suggests a prevalence of approximately 1% [5], although the occurrence of SM is probably underestimated [6]. With the introduction of the DSM-5 [1], SM was classified among anxiety disorders for the first time. The reason for this was evidence that SM shares numerous similarities with other anxiety disorders, particularly social anxiety disorder (SAD) [5]. SAD is characterized by a marked fear of being evaluated by others in social situations as well as physiological symptoms [1]. The overlap between SM and SAD is, for example, reflected in high comorbidity rates up to 100% of SAD in children with SM [7] and findings that social fear is a central phenomenon in children with SM [8]. Additionally, etiological similarities between both disorders exist such as the increased risk of the temperament of Behavioral Inhibition (BI) during early childhood [9].

In light of this extreme overlap, hitherto an unresolved question is why children with SM fail to speak in certain social situations, whereas children with SAD do not, despite both clinical groups experiencing marked social fear. In this context, SM has been conceptualized as an extreme variant of SAD with an extreme fear and overarousal in social situations, in which failure to speak functions as an avoidance mechanism leading to a reduction in fear arousal [10–15]. While the assumption of SM as an extreme version was based on findings that children with SM are rated by clinicians as more anxious in social situations than children with SAD [11, 14], comparable levels of self-reported social trait anxiety between both groups speaks against this notion [11, 14, 16]. However, recent studies on children with SM suggest that it is important to differentiate between social situations in which children are expected to speak (verbal) and situations which do not require speech (nonverbal) [10, 13, 17]. During nonverbal social situations, children with SM and SAD report comparable levels of state anxiety [13] and are rated as comparably anxious by parents [17], teachers [17] and clinicians [10], but still show increased levels of fear compared to children with typical development (TD) [10, 13, 17]. In verbal social situations, children with SM are rated as more anxious by clinicians [10] and report higher levels of fear than children with SAD [13]. Therefore, research supports the assumption that children with SM show an extreme level of fear compared to children with SAD, even if this only applies to verbal situations. This suggests that failure to speak is related to extreme fear in verbal situations, which implies that it is part of a dysfunctional fear response. Consequently, differences in fear responses between SM and SAD may provide insights into mechanisms of failure to speak. However, support mainly comes from studies using data based on self-report or behavioral observations, and few quasi-experimental studies exist, which use objective measures of fear such as physiological processes. When faced with a fear-inducing situation, a fear response occurs in order to cope with the threat. Immediate fear responses to threat in the form of fight, flight or freeze are mediated by the autonomic nervous system [18], which consists of a sympathetic and a parasympathetic part. The presence of threat leads to the activation of the autonomic nervous system, which results in an increase in sympathetic and decrease of parasympathetic activity [18]. This change of arousal in response to a fear-related situation already initiates in anticipation of the threat [19, 20] and is called reactivity (in experimental designs defined as change from baseline to confrontation with threat). Reactivity is associated with a higher sympathetic activity indicated by an increase in skin conductance level (SCL) and heart rate (HR) and a reduced parasympathetic activity indicated by a decrease of the respiratory sinus arrhythmia (RSA) [18, 21]. Recovery from threat leads to decrease of SCL and HR and increase of RSA. Aside from the phasic change in physiological arousal in a fear-inducing situation (reactivity or recovery), the basic level of arousal in the absence of stress is called tonic autonomic arousal [22]. Disorder-specific models of other anxiety disorders [23], as well as psychophysiological theories of anxiety in general, propose physiological mechanisms to be involved in the symptomatology of anxious individuals [22, 24, 25]. In contrast, no empirically based model exists in SM and mechanisms of failure to speak are largely unclear, as psychophysiological research in children with SM is scarce. The assumption that failure to speak occurs due to more intense fear and associated overarousal in children with SM compared to children with SAD implies that children with SM have *higher autonomic reactivity* during an expectation to speak. At the same time, disorder-specific

models of SAD suggest that individuals with SAD exhibit increased reactivity compared to non-anxious individuals [23]. However, the few existing studies on autonomic activity in SM [14, 26] do not support the notion of a higher reactivity in SM compared to SAD as a mechanisms of failure to speak. In this context, Heilman et al. [26], who did not include a group of children with SAD, found no difference in reactivity during a non-standardized verbal social stress task between children with SM and typical development (TD). Young et al. [14] did not examine the change of autonomic arousal from the baseline to a non-standardized speech demanding task, but only investigated the level of arousal during both phases. Therefore, no conclusions about reactivity of the autonomic arousal can be drawn from this study. Also against the assumption of SAD models, most psychophysiological studies do not show a higher reactivity in children with SAD compared to children with typical development (TD) during social situations [17, 18, 21–24].

In contrast to the higher reactivity suggested by disorder-specific theoretical assumptions, psychophysiological theories of anxiety propose a *lower reactivity* to be indicative of a pathological fear response [18, 22]. In line with the idea that a fear response is an adaptation to the fear-inducing situation, it is assumed that strong and flexible reactivity is functional, as long as the arousal recovers quickly after the situation. Instead, they propose the combination of a *tonic elevated autonomic arousal* (in the absence of stress) and a blunted response of the autonomic nervous system to stress, consisting of a *lower reactivity* (lower increase of arousal) and *slower recovery* to be indicative of pathological anxiety [18, 22]. Tonically increased autonomic arousal is associated with reduced responsiveness of the autonomic nervous system [22]. The combination of high tonic arousal and blunted response is also known as restrictive autonomic flexibility, which can transdiagnostically be found in anxious individuals and has been interpreted as chronic dysregulation of the autonomic nervous system [27, 28]. While autonomic flexibility has not directly been studied in children with SM, research in children with SAD consistently indicates a restricted autonomic flexibility during social stress. Both an increased tonic arousal [27, 28] as well as a blunted response to social stress, with lower physiological reactivity and a subsequent slow reduction of arousal can be found in children with SAD [28–30], while the restriction of autonomic flexibility is positively related to the level of state anxiety [31]. However, physiological studies in socially anxious children have been criticized for being limited in indicating whether arousal is actually tonically increased or whether arousal is already influenced by the unfamiliar laboratory setting (e.g., presence of strangers) that induces stress in this clinical group [32]. For this reason, Asbrand et al. [32] examined resting arousal in a familiar environment and the absence of social stress, and found that children with SAD showed higher autonomic resting arousal here as well. This tonically increased arousal, even in non-stress inducing situations, can already be observed in toddlers with a high level of the temperamental BI feature [33–35], which is a precursor for both SM and SAD.

Given that inflexibility of the fear response, rather than high reactivity, is indicative of pathological anxiety, and that children with SM experience an extreme fear in verbal social situations compared with SAD, SM might be associated with an even stronger inflexibility of the fear response. However, psychophysiological studies focusing on autonomic activity in children with SM have not yet examined tonic autonomic arousal in the absence of stress, nor do they include direct analyses of both reactivity and recovery over the course of a stressful situation [14, 26]. Young et al. [14] found no significant differences in level of autonomic arousal between children with SM, SAD, and TD during a baseline in a laboratory setting, whereas statistical power was limited due to the very small sample size. The authors did not examine arousal after the verbal stress task and did not analyze differences between phases, so no conclusions about reactivity or recovery can be drawn from this study. Heilman et al. [26] examined baseline arousal in a laboratory setting in combination with physiological responses during and after a verbal social stress task. Although the authors examined reactivity to the verbal stress task, they did not examine recovery. The authors found a higher HR as well as a lower RSA in children with SM compared to children with TD during a baseline condition. Children with SM showed lower RSA after the task, indirectly suggesting a slower recovery from the stressor. However, the authors did not correct for differences in level of arousal between groups (baseline correction), did not use a standardized social stress task and did not control for speech production, which is known to influence physiological responses [36]. Taken together, these findings also seem to indicate a restrictive autonomic flexibility in children with SM, although not all components in SM have yet been investigated. Furthermore, baseline measurements of both studies were performed at an unfamiliar place and in the presence of strangers, both of which induce symptoms in children with SM. In this respect, it is questionable whether the findings of Heilman et al. [26] indicate a chronically increased arousal in children with SM or are a consequence of the factors inducing symptoms in SM. Complementary to the assumption that SM is associated with extreme fear, it has been proposed that failure to speak is an avoidance mechanism [5, 12, 14, 37]. Here, the assumption is that the intense fear experienced

by children with SM during a verbal social situation is reduced by the failure to speak [14]. While this is supported by evidence from a qualitative study in which individuals with SM report in some cases that failure to speak is associated with the experience of safety [8], there is also initial evidence from the above-mentioned psychophysiological studies. Children with SM showed a comparable level of autonomic arousal as children with TD during verbal tasks of both above-mentioned physiological studies and a lower arousal than children with SAD in the study of Young et al. [14]. The lower arousal compared to children with SAD has especially been interpreted in favor of considering failure to speak as avoidance mechanism. However, the design of both studies [14, 26] is not completely suitable for testing this assumption due to two reasons: (1) they did not examine a detailed course of the fear response. While an increase in autonomic arousal already starts in anticipation of a fear-inducing situation, it is implicit in the assumption of the avoidance mechanism that arousal decreases again as soon as the expectation to speak sets in and the child fails to speak. Therefore, it is important to capture the course of anticipation of the fear-inducing situation and the situation in which the child does not speak. (2) Both studies did not control for all important confounding variables. In this context, it is especially important to control speech production during the verbal stress task, as it has an impact on the physiological response. Additionally, in order to investigate a possible avoidance mechanism, it would be important to consider a nonverbal social stress task, which also induces fear in children with SM but cannot be avoided by failure to speak. Given that previous studies have not done this, a comparison of fear responses between these situations could provide insight into whether reduction happens only in the verbal situation that can be avoided by failure to speak.

Therefore, the current study aims to examine the responses of the autonomic nervous systems of children with SM compared to children with SAD and TD. In addition to tonic autonomic arousal during rest, the fear response (reactivity and recovery) is investigated with regard to both verbal and non-verbal social standardized stress situations. Additional to autonomic flexibility, we examine the course of the fear response comparing both types of situations in order to capture a potential reduction of arousal through failure to speak in SM. In order to gain detailed insight into the course of the fear response, we analyze arousal in different phases of the fear-inducing situations (baseline, anticipation, performance, recovery). While a story is to be retold in the verbal task, the nonverbal task consists of drawing a previously heard story, so that it cannot be avoided by failure to speak. To induce social stress, both tasks take place in front of an unfamiliar experimenter and children had been told that other children will later evaluate the recorded tasks. Furthermore, we analyze and control for possible baseline differences as well as confounding variables such as age, gender, and the number of spoken words for the analysis of physiological response. Furthermore, we take into account the level of BI retrospectively assessed for toddlerhood, as this has to be shown to have an influence on the tonic arousal level.

We aim to address the following hypotheses:

1. Autonomic flexibility: We aim to shed light on the fear response according to the concept of restricted autonomic flexibility in children with SM:
   - 1.1. Resting Arousal:
     - a. We expect children with SM and SAD will show higher tonic autonomic arousal (indicated by elevated HR and SCL and reduced RSA) during a rest period in a non-threatening environment compared to TD children.
     - b. To investigate whether the higher arousal is rooted in the temperament BI, which is a precursor to both disorders, we test whether the level of BI can predict arousal beyond the symptomatology of SM and SAD.
   - 1.2. Reactivity and recovery during nonverbal social stress: We expect that children in the clinical groups will show a lower reactivity and a slower recovery on sympathetic and parasympathetic markers during nonverbal social stress compared to TD children (SM = SAD > TD).
   - 1.3. Reactivity and recovery during verbal situations:
     We further expect the children with SM will show a lower reactivity and slower recovery during verbal stress compared to both children with SAD and children with TD and we expect children with SAD will show lower reactivity and slower recovery compared to children with TD (SM > SAD > TD).
2. Avoidance: We aim to investigate the failure to speak as a potential disorder-specific mechanism in SM. In order to address limitations of previous physiological studies in SM [14, 26], we aim to investigate the course of the fear response in children with SM and compare it between a verbal and a nonverbal social stress situation. Hereby, we want to identify whether a reduction of the physiological arousal occurs with

the onset of an expectation to speak after it increases in anticipation of the fear-inducing situation.

2.1. Reduction of arousal: We hypothesize that autonomic arousal in children with SM will increase (increase of HR and SCL and decrease of RSA) in anticipation of the speech demanding situation and will decrease (decrease of HR and SCL and increase of RSA) during the speech demand (performance of verbal task).

2.2. Comparison of paradigms: We hypothesize that a reduction from anticipation to performance of the stress task in children with SM will only occur in a verbal task with the expectation to speak, but not in a nonverbal social stress task that cannot be avoided by failure to speak. We therefore assume that the difference in autonomic arousal between anticipation and performance of the verbal social stress task (performance–anticipation) in children with SM will be larger than in the nonverbal situation.

Methods

Sampling and recruitment

Individuals were recruited throughout the state of Hessen via psychiatric clinics, psychotherapeutic outpatient clinics, schools, speech therapists, advertisements, mailings to households, and from an existing database. Compensation in the form of a €20 voucher was offered for participation in the main part of the study. The local ethics committee of the University of Giessen approved the study. All interested families were provided with a link with information about the study procedure and invited to participate in a brief online questionnaire in order to screen for symptoms of SM (using the Frankfurt Scale of Selective Mutism) and SAD (based on the DSM-5 criteria). Potential mental health disorders were diagnosed later in the study based on a comprehensive clinical interview (Diagnostic Interview for Mental Disorders in Children and Adolescents; Kinder-DIPS) conducted with the parents. BI was assessed through parent-report based on the Retrospective Infant Behavioral Inhibition Scale (RIBI) and social anxiety through self-report based on the Social Phobia and Fear Inventory for Children (SPAI-C). Informed consent was obtained from all caregivers both before the brief online questionnaire and before the main part of the study. From n = 159 caregivers of children between 8 and 12 years who participated in the online questionnaire, a total of n = 106 families agreed to participate in the main part of the study, during which the social stress tasks and a clinical interview for the assessment of diagnoses were conducted. This appointment took place at the families' homes, so as to ensure the acquisition of a resting measurement in a non-threatening environment [32], and to facilitate participation due to the familiarity of the environment. Due to technical difficulties in the physiological measurement, complete physiological data of n = 6 subjects were not available and were therefore excluded from the present study. In addition, n = 4 individuals were excluded, as they displayed other mental illnesses than SM or SAD during further clinical assessment and therefore could not be assigned to the TD group or either of the two clinical groups. The final sample consisted of N = 96 individuals (age: M = 9.48, SD = 1.28), of whom n = 31 were assigned to the SM group, n = 32 to the SAD group and n = 33 to the TD group, based on the conducted clinical interview. Of the n = 31 subjects with SM, n = 24 (80%) also fulfilled the diagnosis of SAD, whereas the precondition for assignment to the SAD group was the absence of comorbid SM. None of the children with SAD had a lifetime diagnosis of SM. The children of the TD group did not fulfill the criteria for any mental health disorder. It is important to emphasize that there is a risk to overdiagnose SAD in children with SM, as clinicians tend to interpret the failure to speak of children with SM as anxiety [16]. Given that the clinical interview used in the study at hand considers both verbal and nonverbal social situations for diagnosing SAD, we might have reduced the risk of overdiagnosing SAD in the SM group. An overview of the comorbidities is included in Additional file 1. Groups did not differ with respect to age and gender and clinical groups did not differ regarding the number of comorbidities. Table 1 shows an overview of the sample characteristics.

Procedure

After completing the online questionnaire, the families were visited in their homes by two experimenters. At the beginning of the appointment, parents and children were again informed that the aim of the study was to investigate bodily reactions of stress in children with selective mutism in different situations. Both were informed that the participating child would perform various tasks, either simply lying down, drawing something, or retelling a story, and that meanwhile, body responses would be measured. After clarification of the study contents and the confirmation of the informed consent of parents and children, electrodes were applied to the children in order to conduct the physiological measurements (see section below). Then, a measurement was performed in a resting state. For this, the individuals were instructed to lie down on a mat for 5 min without talking to the caregiver present in the room. During the resting measurement, the

Table 1 Sample characteristics

	SM	SAD	TD	*P*	Post-hoc
n	31	32	33		
Age	9.07 (1.15)	9.50 (1.19)	9.79 (1.41)	0.075	–
Gender (f/m)	19/12	18/14	18/15	0.889	–
FSSM-DS	7.71 (1.66)	4.44 (2.59)	0.70 (1.21)	< 0.000	SM > SAD > TD
FSSM-SS (z-score)	0.63 (.89)	0.23 (0.70)	− 0.81 (0.28)	< 0.000	SM > SAD > TD
SPAI-C sum score	18.86 (9.42)	18.92 (10.12)	9.24 (5.97)	< 0.000	SM = SAD > TD
RIBI sum score	39.06 (13.59)	41.91 (15.99)	23.84 (12.86)	< 0.000	SM = SAD > TD
Number of words	93.09 (86.43)	155.00 (65.46)	217.76 (81.55)	< 0.000	SM < SAD < TD

FSSM-DS Frankfurt Scale of Selective Mutism-Diagnostic Scale, *FSSM-SS* Frankfurt Scale of Selective Mutism-Severity Scale, *SPAI-C* social phobia and anxiety inventory for children, *RIBI* Retrospective Infant Behavioral Inhibition Scale, *SM* selective mutism, *SAD* social anxiety disorder, *TD* typical development

experimenters were not present in order to reduce possible stress caused by the presence of strangers. Before and after the resting measurement, the children were asked to rate their anxiety level on an anxiety thermometer and to assess the extent of their arousal using an analog scale adapted from the arousal dimension of the self-assessment manikin. Both the anxiety thermometer and the self-assessment manikin analog scale are well-established scales used in research to assess state anxiety and arousal in children with anxiety disorders [38, 39]. Subsequently, one experimenter together with the caregiver went to a separate room, where a structured clinical interview (Kinder-DIPS) was performed with the caregiver. The experimenters who conducted the interview were graduate students of psychology who were trained and supervised by a clinical psychologist. Meanwhile the other experimenter performed a social stress paradigm with the child alone (without a caregiver present). The child went through a standardized experimental protocol (see Fig. 1) consisting of a verbal and a non-verbal social stress paradigm, with the order being randomized (see "Materials" section). Importantly, the participating children did not know whether the experimenter was aware of their difficulties in speaking, so the children may have been less influenced by a possible expectation of the experimenter. Physiological data were continuously recorded during the stress paradigm. During the verbal task, the number of spoken words was counted. After both performance phases the children completed a short questionnaire on attentional focus, which is not relevant for the aim of the present study. The paradigm was followed by an eye-tracking paradigm and assessment of cognitive variables, which are published elsewhere.

Materials

Social stress paradigm

The stress paradigm of the current study was adapted from the Trier Social Stress Test for Children (TSST-C), which has been proven to be highly effective for stress induction [40] and is most commonly used in psychophysiological research in socially anxious children [28–30]. We had to adapt this paradigm because for our hypotheses, a differentiation between a verbal and non-verbal stress task is required and no validated procedure exists for this yet. Differences between the paradigm of the current study and the TSST-C as well as the rational behind each adaptation are displayed in Additional file 1: Table S5. For the construction of our paradigm, we created a standardized protocol (see Fig. 1) and considered stress-inducing elements of the TSST-C such as the use of a video recorder or a cover story with standardized phrases to elicit social stress (see description below). Children were seated at a table opposite the experimenter and also remained seated for the entire duration of both stress paradigms, which were conducted consecutively without any time break. Both the verbal and non-verbal

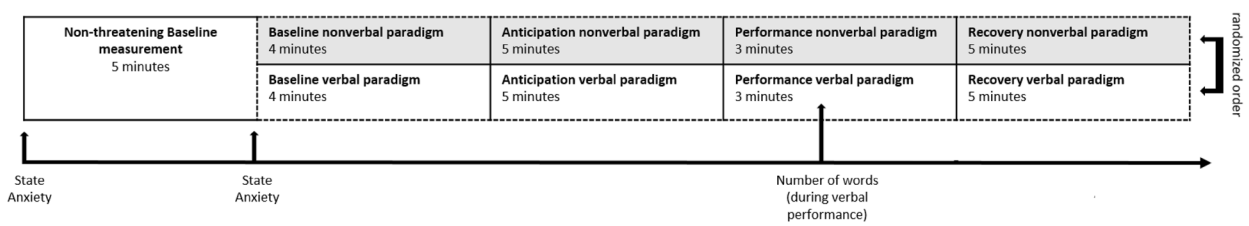


Fig. 1 Procedure of social stress paradigm and measured constructs

paradigms were intended to be as comparable as possible and began with a baseline phase (4 min) in which relaxing nature videos were shown on a screen. Subsequently, the children were informed that they now had time to prepare (anticipation: 5 min) for retelling and continuing a story presented to them in picture form (verbal) or painting a story read to them (non-verbal). Furthermore, the children were told that they were supposed to complete the task in a few minutes, each time in front of the experimenter and a video camera. A cover story was used for both paradigms, suggesting that the children would be recorded on video and that other children of the same age would see the video later and evaluate it. After the preparation time, the experimenter turned on the video camera and instructed the child to start telling the story or paint the picture story (performance: 3 min). For this purpose, TSST-C standardized phrases such as "other children will evaluate you afterward, so make an effort" were used to induce social-evaluative stress. During the verbal paradigm, if the child had not spoken after 20 s or had finished telling the story before the time was up, the child was notified in a standardized manner that "there is still time left". During the recovery phase (5 min), the child again watched relaxing nature videos. Relaxing nature videos during both the baseline and recovery phases were introduced with the instruction that children could relax while watching the videos. The baseline phase of the second paradigm followed directly after the recovery phase of the first paradigm had ended.

Psychometric measures

The *Diagnostic Interview for Mental Disorders in Children and Adolescents* (Kinder-DIPS) [41, 42] is a structured clinical interview used to diagnose mental health disorders in children and adolescents. An evaluation is possible according to both DSM-5 and ICD-10. The interview has a good to very good inter-rater reliability [43] and has a high acceptance by both interviewers and respondents [43, 44]. In the present study, the Kinder-DIPS was used to diagnose mental health disorders or to rule out the presence of such a disorder. Individuals who met the DSM-5 criteria for SM were assigned to the SM group, regardless of whether they also met the criteria for SAD. Individuals who met only the DSM-5 criteria for SAD but not for SM were assigned to the SAD group, and individuals who did not meet the criteria for any mental health disorder were assigned to the TD group.

The *Frankfurt Scale of Selective Mutism* (FSSM) [45] is a questionnaire for assessing the symptoms of SM in children and adolescents aged 3–18 years, based on a parental judgment. The FSSM comprises a diagnostic scale (DS) consisting of ten dichotomous items (yes/no) related to the child's general speech pattern and is available in three developmentally adapted versions (kindergarten children from 3 to 7 years old, school children from 6 to 11 years old and individuals from 12 to 18 years old). The versions used in the present study for children between 6 and 11 years of age and adolescents between 12 and 18 years of age have a cut-off value of 7 and 6 respectively, indicating the presence of SM. Additionally, the FSSM includes a severity scale (SS) that can be used to assess the symptom severity of SM. This consists of either 41 or 42 questions on speaking behavior in various situations, depending on the developmentally adapted version. Questions are answered on a 5-point Likert scale and a total sum score can be calculated. In the present study, we formed z-scores of SS to integrate the sum scores of the different developmentally adapted versions into a common total score. The ROC analysis performed by the authors confirms a very good differentiation between children with SM ($M = 8.2$, $SD = 1.5$), SAD ($M = 3.6$, $SD = 2.5$) and children without a mental health disorder ($M = 0.5$, $SD = 0.8$) and shows excellent reliability (Cronbach's $\alpha = 0.90–0.98$.). For our sample, the reliability for the FSSM was also excellent ($\alpha = 0.951–0.959$).

We used the German version of the *Social phobia and fear inventory for children (SPAI-C)* [46, 47], which measures symptom levels of social anxiety via self-report. The questionnaire consists of 26 items with a 3-point Likert scale on different social situations and an overall score of 52 can be reached. Both reliability (Cronbach's $\alpha = 0.92$) and validity are considered high [47–49]. Beidel et al. [46] reported a cut-off score of 18, which is able to differentiate between children with SAD and non-socially anxious children. For our sample, reliability was also excellent ($\alpha = 0.959$).

The *Retrospective Infant Behavioral Inhibition Scale (RIBI)* [45, 50] is a questionnaire based on a retrospective parental report to assess the child's behavioral inhibition (BI) for the first two years of life. The RIBI has a 3-factor structure and includes the subscales Distress to Novelty, Fear, and Shyness in addition to the total score scale for the expression of BI. Parents answer 20 items on a 5-point scale (0 = Yes, 1 = more likely Yes, 2 = partly, 3 = more likely Not, 4 = Not). The test has excellent reliability (> 0.90) and convergent validity in the form of positive correlations with questionnaires as well as laboratory tests for BI at 14 months of age [50]. In our sample, the reliability was also excellent ($\alpha = 0.909$).

Psychophysiological measures

All physiological measures were recorded with an acquisition sample rate of 1000 Hz simultaneously using the BIOPAC system MP160 and the portable bionomadix modules for ECG, respiration, and electrodermal activity. A 3-point ECG and two 2Ag/Cl electrodes for

measurement of electrodermal activity were installed on the children's thoraxes and index and middle fingers of the non-dominant hands. For the calculation of RSA, respiration was recorded using a respiratory belt on the child's chest. The physiological data were pre-processed using the Acqknowledge software (BIOPAC). All data sets were visually examined for artifacts, missing or obsolete QRS complexes were corrected and HR was calculated in beats per minute (BPM) based on an ECG channel. The electrodermal activity data was adjusted in Acqknowledge via linear regression for the increasing trend of SCL. The RSA was calculated using multi-epoch spectral analysis.

Data analysis

Data reduction and statistical analysis

All statistical analyses were performed in SPSS 26.0. We compared groups on age, symptom scores (FSSM, SPAI-C), temperament (RIBI), and the number of spoken words using univariate ANOVA and Bonferroni-corrected post-hoc tests. Number of words were counted from a tape recording, which was conducted during the performance phase of the verbal task. The reason for considering the number of words is that speech production itself can have an impact on autonomic processes [36]. Gender distribution was compared based on a chi-square test.

From the pre-processed physiological data, mean values for HR, SCL and RSA were calculated for the individual phases of both paradigms (verbal: BL_{verb}, Ant_{verb}, $Perf_{verb}$ Rec_{verb}; nonverbal: $BL_{nonverb}$ $Ant_{nonverb}$ $Perf_{nonverb}$ $Rec_{nonverb}$) and the initial resting phase. Outliers (z-value ± 3 SD) were excluded for the physiological variables, according to previous research [29]. For HR, $n = 0$ cases were excluded; for SCL $n = 4$ cases (SM: $n = 2$; SAD: $n = 2$); for RSA $n = 4$ cases (SM: $n = 2$; SAD: $n = 2$).

Resting arousal (H1.1 a and b) Initially, we checked whether the groups differed in terms of their subjective anxiety levels during the resting measurement in the absence of stress. This is important to rule out that possible differences in physiological resting arousal are not due to higher subjective anxiety. The mean values of the state-anxiety and experienced arousal before and after the resting measurement were z-standardized and integrated into an anxiety measure consisting of an emotional and bodily component and then compared between groups using independent sample *t* tests. To counteract the risk of alpha error accumulation due to multiple testing, autonomic arousal during rest was compared between groups using a multivariate analysis of variance (MANOVA) including all three measures (HR, SCL, RSA) and group (SM, SAD, TD). Significant group differences were decomposed using Bonferroni-corrected post-hoc tests. To test whether potentially increased resting arousal was due to the level of BI, we first calculated correlations between BI and resting arousal. Given that BI was not significantly correlated with any of the three measures during rest, the multiple regressions were not performed.

Reactivity and recovery during nonverbal and verbal social stress (H1.2 & H1.3) In order to minimize the accumulation of alpha errors by multiple testing, we largely based the statistical calculation of the physiological fear response as a function of group and phase on the established procedure in research on children with SAD [29, 30]: (1) to control baseline differences between the groups, we used a MANOVA including all measures (HR, SCL, RSA) and groups (SM, SAD, TD) to test whether autonomic arousal during baselines for the verbal and nonverbal paradigms differed between groups. Furthermore, a baseline correction was performed by subtracting the baseline means of the corresponding paradigm from the remaining phase means (verbal: $Ant_{verb} - BL_{verb}$, $Perf_{verb} - BL_{verb}$, $Rec_{verb} - BL_{verb}$; nonverbal: $Ant_{nonverb} - BL_{nonverb}$, $Perf_{nonverb} - BL_{nonverb}$, $Rec_{nonverb} - BL_{nonverb}$). This was done to control expected level differences between groups for phase analysis. (2) We performed a MANOVA with all physiological measures (HR, SCL, RSA) for each paradigm (verbal, nonverbal) preceding univariate analyses to keep alpha error accumulation at a minimum. The physiological measures were not combined into a common parameter, but were included as independent parameters. In both MANOVAs as well as in subsequently conducted mixed ANOVAs, we included group (SM, SAD, TD) and phase (baseline, anticipation, performance, recovery) as factors. Because we were interested in examining whether groups differ in terms of the course of their fear response (reactivity and recovery), we checked based on the MANOVAS whether there was a group × phase interaction with respect to all physiological measures per paradigm. (3) Significant effects of MANOVA on group × phase were then decomposed according to previous research [29]: in order to analyze reactivity (defined as the *change from baseline to performance*) and recovery (defined as the *change from performance to recovery*) we conducted separate mixed ANOVAs (group × phase) for reactivity (baseline, anticipation, performance) and recovery (performance, recovery) per measure and conducted Bonferroni-corrected post hoc tests for comparison of single phases between groups. MANOVAs and ANOVAs were Greenhouse–Geiser corrected. In addition, to control for possible confounding variables, we correlated reactivity (performance–baseline) and recovery (recovery–performance) of each measure with age and gender. Furthermore, we correlated the number of words with the physiological

measures during the performance phase of the verbal paradigm to control for a possible influence of speech production. None of these variables correlated with the physiological measures, so we did not include them as control variables for the analyses.

Avoidance (H2.1 & H2.2) To examine the first hypothesis (H2.1) on failure to speak as an avoidance mechanism, we compared autonomic arousal of children with SM between phases (baseline, anticipation and performance) of the verbal stress paradigm using paired *t* tests. To check whether a possible reduction occurs only in the verbal and not in the nonverbal paradigm (H2.2), we compared the phases also in the nonverbal paradigm with paired *t* tests. Furthermore, to compare the reduction of anticipation to performance between the two paradigms (H2.2), we calculated the difference between anticipation–performance for HR, SCL and RSA for both paradigms. Because the use of score differences has been criticized and their reliability mainly depends on the reliability of the underlying measures [51], we checked the internal consistencies of all three physiological measures per phase. These ranged between $\alpha = 0.876$ and 0.995, which implies that the difference scores are reliable as well [51]. We compared the differences using paired *t* tests. To adjust for alpha error inflation due to multiple group comparison regarding analyses of hypotheses 2, we performed a Bonferroni correction and used $\alpha = 0.003$ as the significance level for paired *t* tests.

During the conduct of the experimental study, we noticed that a proportion of children with SM spoke ($n = 23$) during the verbal performance phase, although with significantly fewer words than children with SAD or TD. A smaller group of children with SM did not speak at all ($n = 8$). Because we aimed at investigating whether a *failure to speak* represents an avoidance mechanism, we conducted a further analysis comparing children with SM who have spoken and who have not spoken (mixed ANOVA with phase: $BL_{verb,}$ $Ant_{verb,}$ $Perf_{verb}$ $Rec_{verb,}$ and group: spoken vs. not spoken with Bonferroni-corrected post-hoc tests). To investigate possible differences in clinical characteristics between the two subgroups, we compared them in terms of the questionnaires used in the study, age, and gender using a MANOVA.

Power analysis and paradigm check

We conducted an a priori power analysis for mixed ANOVA prior to the study. Given effect sizes of $f = 0.24$–0.26 reported in previous studies on phase × group interaction as well as group differences between socially anxious and healthy children in physiological measures [29], we calculated a minimum size per group of n = 31 in order to detect an effect with a statistical power of 90%. This requirement was met in our study.

To test whether the (a) randomization of the sequence of paradigms worked and were (b) protected from sequence effects, we (a) used a chi-square test to test whether the three groups differed in terms of the number of individuals who started with the verbal or nonverbal paradigm and (b) used a repeated measurement MANOVA to test whether the physiological responses per phase averaged between paradigms differed between children who started with the verbal ($n = 48$) or nonverbal paradigm ($n = 48$). Since (a) the groups did not differ in the distribution of the order of the paradigms X^2 (2, $N = 96$) = 0.762, $p = 0.683$) nor did the MANOVA show a group difference ($F(1, 96) = 2.443$, $p = 0.122$, $\eta_p^2 = 0.028$) or a group × phase interaction ($F(5.2, 2.03) = 0.545$, $p = 0.652$, $\eta_p^2 = 0.006$), randomization seems to have protected against possible sequence effects.

Results

Sample characteristics, psychometrics and confounding variables

The three groups did not differ with respect to age and gender (see Table 1). Children with SM and children with SAD showed higher scores than the TD group, consistent with previous research, on the psychometric measures of symptomatology of SAD and SM and level of BI. Children with SM showed a higher level on FSSM than children with SAD. As expected, children with SM spoke the fewest words during the verbal stress paradigm, followed by children with SAD and TD. The number of spoken words was not related to HR ($p = 0.396$), SCL ($p = 0.374$), and RSA ($p = 0.850$) during the performance phase of the verbal stress paradigm in which speech production was required. The correlations between reactivity and recovery of each physiological measure and age, gender, and the psychometric measures are presented in Additional file 1 for both paradigms. During the verbal stress paradigm, no significant correlations were found. During the nonverbal paradigm, the SM symptom score was related to a lower HR reactivity, while the extent of BI was related to the slower recovery of SCL.

Resting arousal (H1.1 a and b)

Regarding the measurement at rest without the presence of a stranger, a significant group difference was found in the MANOVA ($F(6, 172) = 2.889$, $p = 0.010$, $\eta_p^2 = 0.092$). There was a significant group difference for HR ($F(2, 2194.66) = 4.282$, $p = 0.017$, $\eta_p^2 = 0.090$), but not for SCL ($F(2, 59.56) = 0.733$, $p = 0.483$, $\eta_p^2 = 0.017$) and RSA ($F(2, 9.20) = 1.220$, $p = 0.300$, $\eta_p^2 = 0.027$). Bonferroni adjusted post-hoc tests indicated that children with

SM had a significantly higher HR at rest (SM: $M = 88.28$, $SD = 12.71$; SAD: $M = 79.42$, $SD = 9.05$; TD: $M = 79.97$, $SD = 9.76$) than children with SAD ($p = 0.005$) and children with TD ($p = 0.008$). There was no difference between children with SAD and TD ($p = 0.826$). Regarding subjective anxiety, there was no group difference before ($p = 0.311$) or after ($p = 0.192$) resting measurement, so the differences found here are unlikely to be due to group differences in actual anxiety levels. We did not find any correlation between level of BI and level of arousal during rest across all N = 96 children (HR: $p = 0.809$, SCL: $p = 0.752$, RSA: $p = 0.183$).

Reactivity and recovery during nonverbal and verbal social stress (H1.2 & H1.3)

(1) The MANOVA on the difference between groups in autonomic arousal during the baseline phases of the verbal and nonverbal paradigms was not significant ($F(12, 0.225) = 1.732$, $p = 0.064$, $\eta_p^2 = 0.112$). (2) MANOVAs on group × phase with all physiological measures showed a significant effect for phase in both the nonverbal ($F(316.93, 1.20) = 23.414$, $p < 0.001$, $\eta_p^2 = 0.220$) and the verbal paradigm ($F(393.65, 2.10) = 19.669$, $p < 0.001$, $\eta_p^2 = 0.192$), indicating that both paradigms induced a physiological fear response. For the nonverbal stress paradigm the MANOVA showed a significant group (SM, SAD, TD) × phase (baseline, anticipation, performance, recovery) interaction ($F(67.25, 4.00) = 2.484$, $p = 0.046$, $\eta_p^2 = 0.056$), however not for the verbal stress paradigm ($F(40.135, 4.19) = 0.982$, $p = 0.421$, $\eta_p^2 = 0.024$). Therefore, we decomposed the group × phase interaction using individual ANOVAS for reactivity (baseline, anticipation, performance) and recovery (performance, recovery) only for the nonverbal paradigm. For visualization, the physiological responses of the individual measures for both paradigms are shown in Fig. 2. Descriptive statistics of physiological variables of all individuals per phase are displayed in Additional file 1.

Regarding reactivity in the nonverbal stress paradigm, the individual mixed ANOVAs showed significant group × phase interactions for HR ($F(211.35, 3.07) = 4.174$, $p = 0.007$, $\eta_p^2 = 0.082$), but not for SCL ($F(4.70, 3.59) = 2.291$, $p = 0.069$, $\eta_p^2 = 0.052$) or RSA ($F(4.91, 3.70) = 1.002$, $p = 0.405$, $\eta_p^2 = 0.023$). Accordingly, reactivity of the nonverbal paradigm differed between groups only for HR. Bonferroni-corrected post hoc tests showed that children with SM and SAD did not differ in terms of their reactivity in the nonverbal paradigm (lower increase from baseline to performance) ($p = 0.172$). Children with SM showed significantly lower reactivity compared to children with TD ($p = 0.023$). Children with SAD did not differ in reactivity from children with TD ($p = 0.303$), but showed a lower increase from baseline to anticipation ($p < 0.001$).

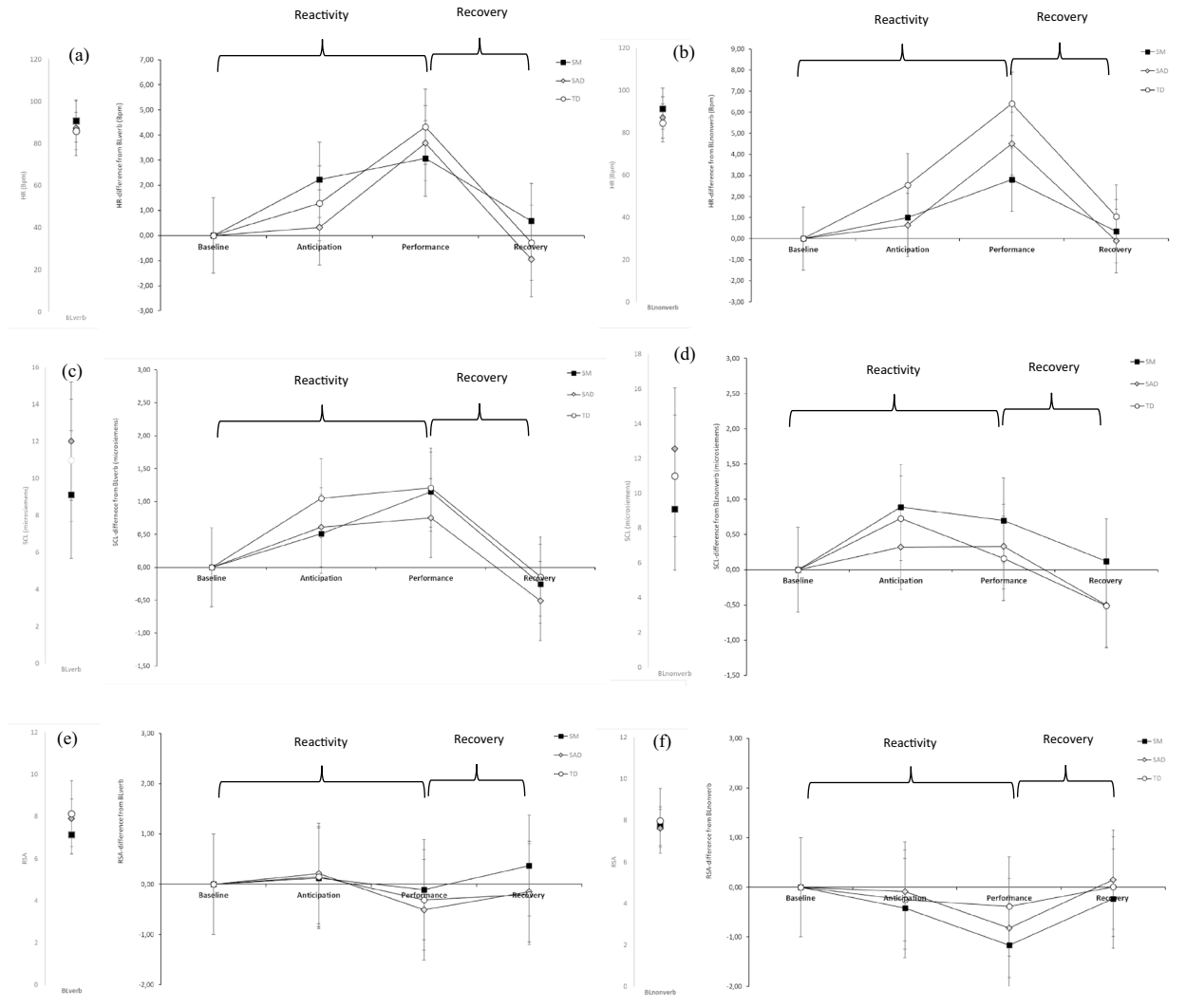
The individual mixed ANOVAs for recovery did not show a significant group x phase interaction for any of the measures. Therefore, no further Bonferroni correction was necessary at this point.

Avoidance (H2.1 & H2.2)

Based on the Bonferroni-corrected alpha level of $\alpha = 0.003$, none of the measures showed a significant increase (or decrease in case of RSA) from the baseline to anticipation to speak (HR: $p = 0.008$, SCL: $p = 0.005$, RSA: $p = 0.895$). Moreover, we did not find a reduction from the anticipation of the speech demand to the performance in which speech was required (HR: $p = 0.362$, SCL: $p = 0.005$, RSA $p = 0.857$). The courses of the fear response of HR and SCL for the verbal paradigm for children with SM are displayed in Fig. 2a, c. Furthermore, for the nonverbal paradigm, there was a significant increase from baseline to anticipation for SCL ($p < 0.001$), but not for HR ($p = 0.036$) or RSA ($p = 0.340$). Consistent with our expectation, no reduction from the anticipation phase to the performance phase occurred in the nonverbal paradigm based on the Bonferroni-corrected alpha level (HR: $p = 0.072$, SCL: $p = 0.196$, RSA: $p = 0.031$). The comparisons of the difference (anticipation—performance) between the two paradigms also showed no significance, considering the corrected alpha level (HR: $p = 0.444$, SCL: $p = 0.005$, RSA: $p = 0.051$).

Regarding the further analysis of children with SM who spoke vs. children with SM who did not speak, we found a significant group difference in HR ($F(1, 29) = 4.434$, $p = 0.044$, $\eta_p^2 = 0.133$) between the two subgroups. We did not find an interaction between group and phase ($F(3, 87) = 1.097$, $p = 0.355$, $\eta_p^2 = 0.036$). A post-hoc comparison showed that children with SM who did not speak had a significant lower HR during anticipation phase (ant_{verb}: $p = 0.037$; not spoken > spoken) but not during the performance phase ($perf_{verb}$: $p = 0.106$). Regarding SCL or RSA, we did not find any group differences (SCL: $p = 0.610$; RSA: $p = 0.817$). Results are displayed in Fig. 3. To test whether this is a result specific to situations with speech demand, we also examined whether there were differences between the two groups on physiological arousal in the nonverbal paradigm. There was no difference in the nonverbal paradigm between groups for any of the measures (range of p-values = 0.508–0.794), indicating that children with SM who fail to speak only show reduced arousal in anticipation of a speech-demanding situation.

A post-hoc comparison between subgroups of SM on social anxiety suggests that children with SM who did



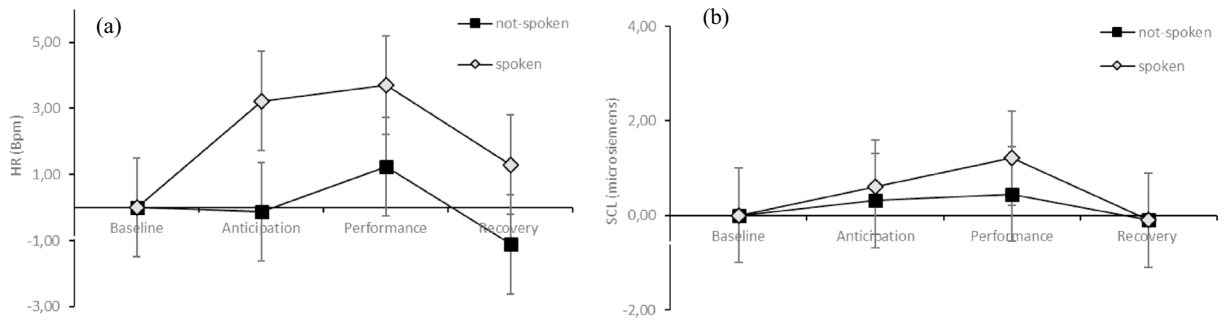
Note: RSA = respiratory sinus arrhythmia, HR = heart rate, SCL = skin conductance level, SM = selective mutism, SAD = social anxiety disorder, TD = typical development, BLverb = baseline phase during verbal stress paradigm, BLnonverb = baseline phase during nonverbal stress paradigm

Fig. 2 Mean baseline-values and baseline-corrected values of phases on **a** HR of verbal stress paradigm, **b** HR of nonverbal stress paradigm, **c** SCL of verbal stress paradigm, **d** SCL of nonverbal stress paradigm, **e** RSA of verbal stress paradigm and **f** RSA of nonverbal stress paradigm for SM-, SAD- and TD-group. *RSA* respiratory sinus arrhythmia, *HR* heart rate, *SCL* skin conductance level, *SM* selective mutism, *SAD* social anxiety disorder, *TD* typical development, BL_{verb} baseline phase during verbal stress paradigm, $BL_{nonverb}$ baseline phase during nonverbal stress paradigm

not speak may be an extremely socially anxious subgroup (SPAI-C: SM-not-spoken: $M = 25.54$, $SD = 6.86$; SM-spoken: $M = 16.32$, $SD = 9.33$, SAD: $M = 18.92$, $SD = 10.12$; SM not-spoken > SM spoken: $p = 0.014$; SM not-spoken > SAD: $p = 0.036$).

Discussion

The purpose of the present study was to investigate tonic autonomic arousal during rest as well as responses of the autonomic nervous system during a verbal and a nonverbal social stress situation in children with SM compared to children with SAD and children with TD. Hereby we aimed at gaining more insight into psychophysiological mechanisms underlying the failure to speak in SM. We expected that children with SM and children with SAD would have a restricted autonomic flexibility, consisting of a (H1.1) higher resting tonic arousal in the absence of social stress and a (H1.2) blunted response (lower reactivity and slower recovery) during social stress, compared to children with TD. We further assumed (H1.3) children with SM to show an even lower reactivity and a slower recovery during the verbal social stress task than children with SAD. Finally, we assumed that (H2.1) failure to speak in children with SM is associated with the



Note: HR = heart rate, SCL = skin conductance level, not-spoken = children with selective mutism who did not speak during performance phase of verbal stress paradimg, spoken = children with selective mutism who speak at least one word during performance phase of verbal stress paradigm

Fig. 3 Baseline-corrected values of verbal stress paradigm for **a** HR and **b** SCL for children with SM who have spoken and who did not. *HR* heart rate, *SCL* skin conductance level, *not-spoken* children with selective mutism who did not speak during performance phase of verbal stress paradimg, *spoken* children with selective mutism who speak at least one word during performance phase of verbal stress paradigm

reduction of arousal in response to a speech-demanding situation, after arousal has increased in anticipation of the situation with expectation to speak. We expected that (H2.2) the reduction of the arousal would not occur in a nonverbal situation that is unavoidable by failure to speak.

Restricted autonomic flexibility

Our study is the first to look at all components of autonomic flexibility (tonic autonomic arousal, reactivity and recovery) during different types of social stress in children with SM. Psychophysiological theories on autonomic flexibility assume that a tonically elevated autonomic arousal and a blunted fear response are markers for psychopathology [18, 24]. The assumption that children with SM showed restricted autonomic flexibility, as has already been demonstrated for children with SAD [29] and children with other anxiety disorders [52], could be partially confirmed by our results. More specifically, children with SM showed increased tonic arousal and reduced reactivity in the nonverbal stress situation, but there was no evidence that children with SM showed slower recovery. The results on tonic autonomic arousal during rest and blunted fear response are discussed below.

Tonic autonomic arousal during rest

On the one hand, the presence of a restricted autonomic flexibility in children with SM is indicated by the finding of increased autonomic arousal (higher HR) during rest in the absence of social stress in children with SM compared to children with TD, which partially confirms hypothesis 1.1. Contrary to hypothesis 1.1 and findings from previous research [52, 53], there was no difference in autonomic arousal during rest between children with SAD and children with TD. Previous studies on autonomic activity in children with SM have focused primarily on social situations [10, 14], so that methodological artifacts due to situational fear-inducing factors cannot thereby be ruled out. The study at hand is the first that suggests that altered physiological processes may be generalized to non-stress-inducing situations, similar to what has already been shown for children with SAD [32]. It is important to emphasize that we took numerous precautions to minimize social stress for the children. During the measurement, no stranger but a caregiver was present and the measurement took place in a familiar environment. The minimization of stress was also reflected in finding that the subjective anxiety level did not differ between groups either before or after the measurement. Therefore, this suggests that the elevated autonomic arousal found in children with SM is not a methodological artifact induced by higher subjective fear during measurement, but actually indicates chronic dysregulated autonomic activity [52]. Tonic autonomic arousal is considered an indicator of autonomic nervous system general responsiveness, with tonically elevated autonomic arousal being associated with a more inflexible physiological response [22]. One explanation for the finding of higher tonic arousal in children with SM might be the concept of allostatic load [54] which assumes that chronic stress is associated with chronic changes in the psychophysiological balance [32, 52]. Children with SM experience increased stress in social situations and are exposed to social stress in many different places such as in public or school [15]. Similar to what Asbrand et al. [32] already assume for children with SAD, this constant and everyday experience of chronic stress could have a lasting effect on the autonomic nervous system of children with SM, resulting in increased tonic arousal.

Complementing the assumption that chronic stress is associated with increased tonic arousal, there is also evidence in the literature that infants with high BI already exhibit increased tonic resting arousal [33–35]. Although we did not find a correlation between BI and tonic resting arousal across all children, level of BI and recovery of SCL were associated (see Additional file 1). This supports the idea that altered autonomic nervous system activity is already predisposed by BI temperament. Because we could only perform correlational analyses across all children due to the sample size, we cannot draw conclusion about the association between BI and physiological measures within the SM group. The extent to which physiological changes in children with SM are more likely to represent a developmental pathway linked to BI temperament, a consequence of chronic stress exposure, or a combination of both should be examined in longitudinal studies involving young children at risk for SM and SAD. Regarding the SAD group in our study, we were not able to replicate the finding of Asbrand et al. [32] that children with SAD show increased HR in a non-threatening environment. One possibility for the divergent results, though speculative, could be the rather low average severity (SAD: M = 18.92, clinical cut-off value = 18) of the children with SAD in our sample (see Table 1). Therefore, a chronically altered arousal at rest only might occur with a higher severity of the disorder.

Fear response (reactivity and recovery)

On the other hand, the presence of restricted autonomic flexibility in children with SM is supported by the finding of a lower reactivity of HR during the nonverbal social stress situation compared to children with TD. Since we found no difference in reactivity between children with SAD and children with TD and the groups did not differ in terms of recovery, hypothesis 1.2 is only partially confirmed. Contrary to hypothesis 1.3, we could not identify any group differences on the verbal stress paradigm and thus no more restricted autonomic flexibility in children with SM in response to a speech demand. The finding that a reduced reactivity in children with SM compared to children with TD occurs in nonverbal situations can be considered consistent with findings indicating higher levels of fear in children compared to TD children even in nonverbal situations [10, 13, 17]. The increased level of fear, which in these studies was primarily measured by questionnaires or by behavioral observation, also seems to manifest in restricted autonomic flexibility. In this context, interestingly, Milic et al. [17] showed that children with SM in a nonverbal behavioral observation task, in which no speech but gestures were expected, had a significantly longer latency to initiate behavior compared to children with TD and children with SAD. According to psychophysiological theories, restricted autonomic flexibility is associated with reduced and slowed adaptation to the threatening situation [18, 22], implying that reduced autonomic flexibility in children with SM is associated with inhibited behavioral coping with the stressful situation. The inhibition of initiation of behavior found by Milic et al. [17] in behavioral observations of children with SM, which might also be in line with findings that BI is a central phenomenon in children with SM [9], could thus be rooted in an inflexible response of the autonomic nervous system. However, it remains unclear why we did not find restricted autonomic flexibility in children with SM in verbal situations. This finding is inconsistent with evidence that children with SM experience an elevated anxiety level compared to children with SAD and TD during verbal situations [13]. Based on the finding that restricted autonomic flexibility is associated with the level of anxiety [31], we assumed that an inflexible fear response should occur especially while children with SM are expected to speak. However, these findings might be explained by a possible function of failure to speak. In this context, the literature suggests that a failure to speak may function as a maladaptive emotion regulation [37] or an avoidance mechanism that is associated with a reduction of anxiety in speech-demanding social situations [14]. Given that the level of subjective state anxiety is related to the inflexibility of the fear response, one could speculate that children with SM have regulated their anxiety level and thus their inflexible fear response in the verbal situation, albeit not deliberately, by failing to speak. Since the failure to speak is not available as a regulatory mechanism in nonverbal situations, no regulation of anxiety takes place. Consequently, a more inflexible stress response occurs that cannot be compensated for. This is also consistent with the only study in children with SM that investigated reactivity during a verbal stress task and did not find a difference between children with SM and TD [26]. Contrary to our assumptions in hypotheses 1.2 and 1.3, we did not find slowed recovery in children with SM or children with SAD compared to children with TD for either stress paradigm. In this respect, it remains an unresolved question whether all components of restricted autonomic reactivity, including reduced recovery from stress, can also be found in children with SM. Here, it is important to emphasize that the recovery phase in our study was significantly shorter than in previous studies of children with SAD [28–30], which have found a slower recovery for this clinical group. Due to the shorter recovery phase, we may not have detected possible differences between groups regarding the recovery of more slowly changing autonomic processes (e.g. SCL). Therefore, in future physiological studies in children with SM, it would be important to include recovery

phases of approximately 30 min in the paradigm, as has been the case in previous studies on children with SAD, which found differences in recovery of autonomic activity [29]. Furthermore and contrary to our hypotheses, we were unable to replicate the results found in previous studies that children with SAD show a blunted autonomic response to social stress. Here, methodological differences to previous studies are also a possible explanation. In particular, this could be due to the fact that we were not able to use the validated TSST-C, which is conducted in a non-familiar environment and with multiple adults as experimenters, which in consequence is likely to lead to a stronger anxiety induction. Although both paradigms in our study also induced stress and resulted in a physiological response in all three groups, our stressor may not have been strong enough to elicit a pathological stress response in children with SAD. In consequence, although speculative, this could mean that children with SM are more sensitive to social stress and already show a dysfunctional physiological response to even a small stressor. Future research may examine this using the validated TSST-C.

Avoidance

Contrary to our assumption, we did not find a reduction in the arousal of children with SM when confronted with the speech demand, after arousal initially increases in anticipation of the verbal situation. Moreover, the course of arousal did not differ between the verbal and the nonverbal social stress task in children with SM, so we had to reject both hypotheses on failure to speak as an avoidance mechanism (2.1 and 2.2.). However, it is important to emphasize that not all children with SM in our sample were completely mute during the verbal stress task. This limits the analyses regarding hypotheses 2.1 and 2.2, in which we aimed at investigating whether the *failure to speak* is associated with a reduction in the arousal and thus represents an avoidance mechanism. However, the further analysis we performed to check whether the course of the autonomic fear response differs between children with SM who spoke during the verbal stress task and those who did not speak supports the assumption of the avoidance mechanism. Here, for children with SM who did not speak, the reduction in autonomic arousal, as expected in hypothesis 2.1 for all children with SM, was also not shown at the beginning of the speech demand (during the performance phase). Instead, children with SM who did not speak already had lower arousal during the anticipation phase (see Fig. 3). During the anticipation phase, the children already knew that they would soon have to speak, but no speech was yet required. This suggests failure to speak, at least in a subgroup of children with SM, functions as an avoidance mechanism, which appears in anticipation of the actual speech demand and, following this logic, is accompanied by reduced fear of expectation. Thus, the results may support the assumption in the literature that failure to speak is an avoidance mechanism [11, 14] that reduces fear effectively in the short term but is maladaptive and maintains the disorder in the long term. Alternatively, this finding could be explained by another mechanism, described in the literature as *freezing*. Freezing is a fear response that is characterized by a pattern of different physiological responses such as motor inhibition, including voice production and a decrease in heart rate [55, 56]. In our study, children with SM who failed to speak in the situation had a lower heart rate but no lower skin conductance than the SM group who spoke, which would be consistent with the physiological freezing response [57]. Furthermore, this assumption would be consistent with the descriptions in the clinical literature of some children with SM being "frozen with fear" [58]. In addition, recent research shows that a freezing reaction can occur in preparation for a subsequent fear reaction [59]. Therefore, the reduced HR in our study during the anticipation phase in mute children with SM could also be explained by freezing. Further experimental research involving a sufficiently large sample of children with SM is needed to disentangle the underlying mechanisms of avoidance and freezing and to identify possible subgroups. Since only some of the children with SM have failed to speak in our study, whether the discussed mechanisms only occur in a (extreme socially anxious) subgroup of children with SM or whether these mechanisms are inherently related to SM, but can only be observed if failure to speak actually occurs, is still questionable. Here, it would be particularly interesting to investigate whether the children with SM who spoke in our verbal stress paradigm show similar mechanisms in the situations in which they fail to speak (e.g., at school), or whether there are different subgroups with specific mechanisms.

Clinical implications

Our study has several implications for clinical practice and future research. The finding that children with SM may show increased tonic arousal even in familiar surroundings in the absence of social threat might highlight the importance of defocused communication and anxiety reduction during therapy even in supposedly non-anxious situations. The indication that children with SM show restricted autonomic flexibility, especially in non-verbal social situations, could be an indicator for a generalized pathological fear response beyond verbal situations. In this respect, exposure, which is a central element of CBT in SM [60], should not only be performed in speech-demanding situations to address the

failure to speak but should also be explicitly generalized to non-verbal social situations in order to counteract the pathologically-altered fear reaction. Previous research suggests that non-verbal social situations induce a comparable level of anxiety in children with SM to that in verbal situations [13, 17]. Because different mechanisms may be present in children with SM in verbal and non-verbal situations, a differentiation in therapeutic interventions may be important for both types of situations. In verbal situations, it might be promising to start with therapeutic interventions (e.g., relaxation techniques) in the anticipation of a speech-demanding situation, since a dysfunctional fear response seems to start even before failure to speak occurs. In nonverbal situations, where a restricted autonomic flexibility to stress and thus a delayed adaptation to the social situation seem to be of great relevance, it seems to be promising to use therapeutic techniques that counteract the inhibition of behavior (e.g., motor activation) and create a comfortable atmosphere (e.g. through playful elements). Further research should investigate the underlying mechanisms of SM in different situations and large sample sizes to derive differentiated therapeutic interventions for different potential subgroups.

Limitations

There are methodological limitations, which confine the conclusions of our study. First, the stress paradigm used in the study is standardized but not validated. However, instructions and procedures were closely aligned with the TSST-C and there was a main effect phase with respect to physiological responses. Therefore, it can be assumed that the paradigms successfully induced social stress. Secondly, the recovery phases in our paradigm were shorter than in previous studies in children with SAD, so that we cannot make any assumption regarding the long-term recovery of the arousal. Thirdly, we have some heterogeneity in our SM group, as we included children with SM and comorbid SAD as well as non-socially anxious children with SM. Although the rate of SAD found in our SM group (80%) is consistent with rates reported in the literature, this heterogeneity did not allow for a clear differentiation between physiological processes in children with both SM and SAD, children with SAD only, and children with SM only and thus disorder-specific conclusions are limited. However, because we performed additional analyses within the SM group and correlational analyses based on symptom scores, we nonetheless also obtained disorder-specific insights based on our results. To fully disentangle disorder-specific mechanisms, future studies should include larger samples with the aforementioned subgroups. Fourthly, it is important to note that we were not able to diagnose autism-spectrum-disorder, speech or language disorders or assess information regarding developmental history based on the clinical interview used in the current study. Furthermore, we also did not capture cognitive abilities of the participating children. Fifthly, given that we chose a rather narrow age range of 8–12 years old due to the influence of age on physiological responses reported in the literature, our results cannot be generalized to other age groups. Therefore, the physiological findings found here and derived explanations for the function of failure to speak cannot be applied to other age groups (e.g. children at the preschool age or adolescents with SM). In order to be able to draw valid conclusions regarding etiological mechanisms of SM, studies with younger children and a longitudinal design would be of great relevance. Sixthly, although there was no experimenter in the room during the measurement at rest, the children may still have experienced participation in the study itself as a socially evaluative setting. However, because the groups did not differ in the subjective level of anxiety during the measurement at rest, it can be assumed that a possibly induced stress level was reduced to a minimum. Seventhly, it is important to emphasize that although we had sufficiently large statistical power for the main analyses, the further analyses of children with SM who spoke and who did not speak were based on only very small sample sizes. Future studies should more closely examine the mechanisms identified in the present study using larger sample sizes.

Conclusion

In conclusion, this is the first study that examined psychophysiological responses in children with SM differentiated for verbal and nonverbal social stress situations and a non-threatening resting measurement in familiar surroundings. We have addressed various methodological limitations of previous psychophysiological studies with children with SM by carefully controlling for confounding variables such as age, gender, and speech production; used a standardized albeit not validated stress paradigm; considered possible baseline differences; and looked at a detailed course of physiological arousal. We identified increased tonic arousal during a non-threatening resting measurement and restricted autonomic flexibility during nonverbal social situations in children with SM, indicating a pathologically altered physiological response in affected children. Furthermore, we showed that a subgroup of children with SM who did not speak during the verbal stress task showed a reduced physiological response already in anticipation of the speech demand. These results combined may suggest that failure to speak acts as an avoidance mechanism that can inherently function only

in verbal situations and counteracts a dysfunctional fear response, whereas in nonverbal situations a proper compensation is absent.

Supplementary Information

The online version contains supplementary material available at https://doi.org/10.1186/s13034-021-00430-1.

Additional file 1: Table S1. Correlation analysis between age, gender, symptom scores of SM, SAD, RIBI and physiological reactivity and recovery during verbal paradigm. **Table S2.** Correlation analysis between age, gender, symptom scores of SM, SAD, RIBI and physiological reactivity and recovery during nonverbal paradigm. **Table S3.** Descriptive statistics of physiological variables. **Table S4.** Comorbidities of clinical groups. **Table S5.** Comparison of the paradigm of the current study and the TSST-C regarding different aspects.

Acknowledgements
We thank all of the families who participated in our study.

Authors' contributions
Both authors were involved in planning the study design and recruiting the sample. FV was the major contributor in analyzing the data and in writing the manuscript. Both authors read and approved the final manuscript.

Funding
Open Access funding enabled and organized by Projekt DEAL.

Availability of data and materials
Not applicable.

Declarations

Ethics approval and consent to participate
The study was approved by the local ethics committee of the Department of Psychology of the University of Giessen and therefore has been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments. All persons who were included in this study gave their informed consent prior to the begin of their participation. Informed consent was obtained from all individual participants included in the study and written informed consent was obtained from the parents.

Consent for publication
Not applicable.

Competing interests
The authors declare that they have no competing interests.

Received: 4 August 2021 Accepted: 10 December 2021
Published online: 28 December 2021

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Publisher's Note

Cognitive Therapy and Research
https://doi.org/10.1007/s10608-021-10289-3

ORIGINAL ARTICLE

Frozen with Fear? Attentional Mechanisms in Children with Selective Mutism

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Accepted: 22 December 2021

Abstract

Background Children with selective mutism (SM) are consistently unable to speak in certain social situations. Due to an overlap between SM and social anxiety disorder (SAD) in children, similar mechanisms could apply to both disorders. Especially biased attentional processing of threat and fear-induced reduced visual exploration (referred to as attentive freezing) appear promising in SM.

Methods A total of N = 84 children (8–12 years, SM: n = 28, SAD: n = 28, typical development (TD): n = 28) participated in an eye-tracking paradigm with videos of a social counterpart expressing a question, a social evaluation or a neutral statement. We investigated gaze behavior towards the social counterpart's eye-region and the extent of visual exploration (length of scanpath), across conditions.

Results There were no group differences regarding gaze behavior on the eye region. Neither gaze behavior with respect to the eye region nor visual exploration were dependent on the video condition. Compared to children with TD, children with SM generally showed less visual exploration, however children with SAD did not.

Conclusion Reduced visual exploration might be due to the mechanism of attentive freezing, which could be part of an extensive fear response in SM that might also affect speech-production. Interventions that counteract the state of freezing could be promising for the therapy of SM.

Keywords Selective mutism · Social anxiety disorder · Eye-tracking · Freezing · Attention

Introduction

Selective mutism (SM) is an anxiety disorder in which affected children are consistently unable to speak in certain social situations, while their speech production is not impaired in other situations, such as with close family and friends (American Psychiatric Association, 2013). Situations typically associated with the inability to speak include, for example, unfamiliar places or the presence of strangers (Schwenck et al., 2021). The disorder typically occurs between 2 and 5 of age (Muris & Ollendick, 2015; Remschmidt et al., 2001; Steinhausen et al., 2006), severely interferes with everyday life functioning (Milic et al., 2020; Schwartz et al., 2006) and is associated with mental and communicative problems in adulthood (Remschmidt et al., 2001; Steinhausen et al., 2006).

SM and (Social) Anxiety

SM was first classified as an anxiety disorder in *Diagnostic and Statistical Manual of Mental Disorders* (5th ed.; DSM–5, American Psychiatric Association, 2013) due to an overlap with other anxiety disorders, particularly social anxiety disorder (SAD) (Muris & Ollendick, 2015). SAD is an anxiety disorder which is characterized by a marked fear of being evaluated by others in social situations (American Psychiatric Association, 2013). A recent meta-analysis demonstrates that 69% of children with SM have SAD (Driessen et al., 2020) with even higher rates, up to 100%, in most studies on SM (Gensthaler et al., 2016b; Oerbeck et al., 2014; Yeganeh

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Published online: 08 January 2022

et al., 2006). Despite the central importance of social anxiety for both SAD and SM (Gensthaler et al., 2016b; Muris & Ollendick, 2015; Schwenck et al., 2019; Vogel et al., 2019), it remains largely unclear why children with SM are unable to speak in certain social situations, whereas children with SAD do not. In this regard, evidence indicates that SM is associated with a more extreme fear in speech-demanding social situations than SAD (Schwenck et al., 2019) and affected children are unable to speak because they are overwhelmed by their anxiety (Black & Uhde, 1995; Muris & Ollendick, 2015). This is also supported by findings that show that children with SM are evaluated by teachers and clinicians to be more anxious than children with SAD in speech-demanding social situations, but not in nonverbal social situations (Poole et al., 2020; Yeganeh et al., 2003; Young et al., 2012). Consistently, the level of social anxiety in general (Muris & Ollendick, 2015) and level of fear in nonverbal or embarrassing social situations does not differ between the two clinical groups (Schwenck et al., 2019). Further support for the assumption that the inability to speak in certain situations in SM is caused by an extreme fear during verbal situations comes from findings that children with SM show higher levels of the temperamental style Behavioral Inhibition (BI) than children with SAD. This is especially the case for the subscale shyness, which indicates BI with regard to social situations (Gensthaler et al., 2016a). In a longitudinal study, this early inhibition to social stimuli in particular was found to predict a later inhibition of language in social interactions (Kochanska & Radke-Yarrow, 1992). In addition, a proportion of children with SM actually report a paralyzing anxiety during speech-demanding situations (Vogel et al., 2019), and are described as "frozen with fear" in the clinical literature (Anstendig, 1999; Yeganeh et al., 2003). However, this theory is based only on questionnaire data and clinical observations, and experimental studies investigating fear-related mechanisms are almost non-existent. Given the severe impairment of affected children with SM, it is surprising that there is minimal experimental research in SM so far and no disorder-specific model based on psychophysiological mechanisms such as attention processing exists. In contrast, disorder-specific models of SAD contain mechanisms that have proven to be successful in explaining symptomatology of both affected adults (Clark & Wells, 1995; Wong & Rapee, 2016) and children (Schäfer et al., 2012) and are therefore key targets of evidence-based therapy (Clark et al., 2006; Heeren et al., 2012).

Attention Processing in Anxiety Disorders

Direction of Biased Attention

One such mechanism is biased attention processing, which is considered to play a role in the development and maintenance of anxiety disorders (Dudeney et al., 2015; Mogg & Bradley, 1998). Three components of biased attention processing in anxiety disorders have been identified, each of which makes assumptions about the direction of bias and the stage of processing (Cisler & Koster, 2010). First, facilitated attention to threat is an early and automatic bias *towards* threat and thus is associated with a faster detection of threatening stimuli. Second, delayed disengagement from threat describes a prolonged attentional focus on threatening stimuli after a threat has been detected. Third, attentional avoidance is a strategic and late-occurring focus of attention *away* from the threat.

While there is strong evidence of an early attentional bias *towards* threat in adults with different anxiety disorders compared to healthy individuals (Bar-Haim et al., 2007), there are considerably fewer studies and mixed findings regarding the direction of attentional bias in anxious children (Dudeney et al., 2015; Lisk et al., 2020). While one meta-analysis that included both eye-tracking studies and studies using reaction time-based paradigms (e.g. dot-probe-task) found an early bias toward threat (Dudeney et al., 2015), another meta-analysis that included only eye-tracking studies found no such bias (Lisk et al., 2020). It is noticeable that most of the studies investigating attentional bias in anxious children used a transdiagnostic approach and included samples of mixed anxiety disorders (Dudeney et al., 2015; Lisk et al., 2020). However, research indicates a significantly greater effect size of attentional bias towards disorder-congruent threatening stimuli (e.g. socially relevant stimuli in SAD) compared to incongruent stimuli (Pergamin-Hight et al., 2015), which is also in line with the idea that specific fears underlie each anxiety disorder (American Psychiatric Association, 2013) and which could explain the mixed findings in samples of children with mixed anxiety disorders. For the later-occurring components, there are also mixed findings in anxious children and adolescents. At a transdiagnostic level, studies based on reaction time measures combined with longer stimulus presentations tend to indicate delayed disengagement (Dudeney et al., 2015), whereas eye-tracking studies point to attentional avoidance (Lisk et al., 2020). Given that the exact time of the onset of the later components as well as the time course of attentional disengagement is largely unclear, Lisk et al. (2020) point out the importance of differentiated analysis in different time windows for future studies.

Visual Exploration

In addition to the components regarding the direction of attentional bias, the extent of visual exploration that reflects oculomotor activity in the presence of a threat has relevance to attention processing in the context of fear (Löw et al., 2015; Rösler & Gamer, 2019; Wendt et al., 2017). Here, a reduced visual exploration, which is also called attentive

freezing, is considered part of a biologically-driven defense cascade that also includes other psychophysiological fear responses such as a reduction of body movement and vocal inhibition (Kozlowska et al., 2015; Löw et al., 2015; Roelofs, 2017; Rösler & Gamer, 2019; Wendt et al., 2017). While there are few studies on the extent of visual exploration in adults with mixed findings (Chen et al., 2015; Horley et al., 2004; Löw et al., 2015; Rösler & Gamer, 2019; Toh et al., 2011; Wendt et al., 2017; Wermes et al., 2018), there are no such studies on children yet. Given that children with SM are considered to be even more inhibited during verbal social situations than children with SAD, (Poole et al., 2020; Young et al., 2012) and are described as frozen during social situations (Anstendig, 1999), (attentive) freezing might be a potential mechanism in SM.

Biased Attention Processing in Children with SAD

On a disorder-specific level, there are only three studies in children with homogenous samples of SAD based on eye-tracking (Keil et al., 2018; Schmidtendorf et al., 2018; Seefeldt et al., 2014) and one based on discrete reaction time measures (Waters et al., 2010) investigating the direction of attentional bias. While all four studies indicate the presence of an early attentional bias *towards* threat in children with SAD compared to healthy individuals, they are contradictory regarding the later occurring components of biased attention processing (Keil et al., 2018; Schmidtendorf et al., 2018; Seefeldt et al., 2014; Waters et al., 2010). While only children with a low symptom severity of SAD showed an avoidance of threatening faces in the study of Waters et al. (2010), the results of Seefeldt et al. (2014) indicate that children with SAD show difficulties in disengaging from threat. Schmidtendorf et al. (2018) could not replicate the finding of a relocation of attention to threat at later stages of attention processing in children with SAD and Keil et al. (2018) found a shorter fixation time to the eye-region in children with SAD compared to healthy children in an early phase. However, there were no differences at later stages of attention processing in the latter study.

Possible reasons for the mixed findings regarding the later-occurring components might be methodological flaws and variations such as differences in the symptom severity of SAD across studies or that studies differed in terms of used threatening stimuli. While only the study by Keil et al. (2018) used the eye area of a social counterpart as a threatening stimulus, the other studies defined the entire face as social threat. In adult SAD patients, however, there are several studies indicating the direct gaze of a social counterpart as especially fear-inducing in affected individuals, which suggests that it has great relevance when investigating attentional processing in SAD (Judah et al., 2019; Langer & Rodebaugh, 2013; Moukheiber et al., 2010; Rigato & Farroni, 2013; Weeks et al., 2013, 2019; Wieser et al., 2009). Another critical methodological aspect with respect to these previous studies on attention processing in children with SAD, is that all previous studies used static stimuli to induce fear. In these studies, angry faces were paired as threatening stimuli with other social or neutral stimuli and attentional bias was measured in contrast to the two stimuli. Even though it is an established and standardized procedure, the use of static and thus less naturalistic social stimuli is increasingly criticized (Lisk et al., 2020). In this regard, dynamic social situations that have a high social relevance seem to be of particular importance for the creation of an anxiety-driven attention bias in individuals with high social anxiety (Lisk et al., 2020; Risko et al., 2016; Rubo et al., 2020) and thus are better proxies for real social situations. Furthermore, no study has examined the extent of visual exploration in children with anxiety disorders, although theories suggest that this may also be an important part of the fear response.

Biased Attention Processing in Children with SM

It is striking that no studies to date have investigated attention processing in children with SM. Given that social anxiety also lies at the heart of SM, which leads to high rates of comorbid SAD in SM and inability to speak occurs during an expectation to speak (APA, 2013), it can be assumed that both social-evaluative as well as speech-demanding situations are disorder-congruent in SM. Empirical evidence also supports this assumption (Schwenck et al., 2019). Given that clinical case reports suggest that some children with SM avoid eye contact in social situations (Kovac & Furr, 2019; Muris & Ollendick, 2021; Wong, 2010), direct eye-contact might also be a disorder-congruent threat in SM.

Current Study

This is the first study to investigate the attention processing of threat in children with SM. We also aim to investigate whether and how components of the attentional bias differ between children with SM and children with SAD in order to identify disorder-specific mechanisms of SM. According to the research outlined above (Pergamin-Hight et al., 2015; Schwenck et al., 2019; Weeks et al., 2013), we applied dynamic video-stimuli of high social relevance, which included the three conditions: questions, social-evaluative and neutral statements. As the dependent variable, we examined *fixation time on the eye region* of the social counterpart during different phases of attention processing as well as the *amount of visual exploration*. Both variables were measured using eye tracking. The following three research questions will be examined:

1. Given that social anxiety lies at the heart of both SM and SAD and given that previous studies indicate an early attentional bias *towards* disorder-congruent threat in socially-anxious children, we predicted that children with SM and children with SAD would show an early attentional bias *towards* a social counterpart's eye-region in disorder-congruent situations compared to typically developing (TD) children.
   1.1: Given that questions are disorder-congruent in children with SM, as they show a stronger fear-inducing effect in children with SM compared to children with SAD, who in turn experience higher levels of fear when asked a question than children with TD (Schwenck et al., 2019), we expect a stronger bias towards threat in children with SM compared to the other groups and a stronger bias in children with SAD compared to children with TD (SM > SAD > TD).
   1.2: We predict that both children with SM and children with SAD would demonstrate a bias towards social-evaluative threat (SM = SAD > TD).

2. Both the investigation of delayed disengagement and avoidance will be exploratory as studies of later-occurring components of attentional bias in socially anxious children produced mixed results.
3. Given that there has been no previous study of the extent of eye movements in anxious children, we investigate the length of scanpaths in the presence of threat in an exploratory manner. In this context, we aim to compare the scanpath length between the three groups (SM, SAD, TD). Because possible reduced visual exploration is related to the concept of attentive freezing and given that BI, which is a key risk factor for SM and SAD, is associated with the inhibition of motor function, we also want to investigate whether the level of BI predicts visual exploration.

Materials and Methods

Sample

Children aged 8 to 12 years old with either SM or SAD or both were recruited in rural and urban areas throughout the state of Hesse (Germany) via in-patient and out-patient clinics, speech therapists, schools and via communications with households in Giessen (Germany). The participation was compensated with a voucher worth €20 (approx. $24). Children of the TD group were recruited from an existing database, via online advertisements and newsletters. Initially, a total of $n = 159$ caregivers of children took part in an online questionnaire on the platform UNIPARK to screen for symptoms of SM and SAD. Regarding the screening instruments, 44 children met both criteria for SM and SAD, 29 met criteria exclusively for SAD, 11 children met criteria exclusively for SM and 75 children did not meet criteria for either SM or SAD. A total number of 95 caregivers gave written consent and took part in the experiment. The remaining 65 of the initial 159 individuals did not agree to further participate in the experiment and decided not to continue with the study. We visited the families at home in order to conduct the Kinder-DIPS with caregivers and the experimental paradigm with the children. We conducted the experiment at the families' homes because we hoped to reach families in a larger area and to be able to include individuals with more severe symptoms, as children with SM and SAD have higher levels of anxiety in unknown places and tend to avoid them. Due to missing data caused by technical problems, four children had to be excluded. A number of seven of the 91 remaining individuals had a tracking ratio of less than 50% during the experiment, so that these children were also excluded according to previous research (Hartmann & Schwenck, 2020). The children excluded based on the tracking ratio did not differ from the final sample in age ($p = 0.247$), gender ($p = 0.399$), or SM symptomatology ($p = 0.840$) and SAD symptomatology ($p = 0.714$), so this is unlikely to be a selective drop-out. The final sample consisted of 84 children, from which 28 met the primary diagnosis of SM and were assigned to the SM group. Children who met the criteria for SM were assigned to the SM group regardless of whether they had a comorbid SAD or not. Of the 28 childrenwith SM, 25 (89.3%) also met the criteria for SAD**,** which is in line with previous research. According to the DSM-5 criteria, a child with SM does not additionally meet the criteria for SAD if, for example, he or she does not show anxiety towards other children, as this is a prerequisite for the SAD diagnosis. Twenty-eight ($n = 28$) children solely met the diagnosis of SAD and thus were assigned to the SAD-group and 28 showed no mental disorder and thus were assigned to the TD**.** All diagnoses, and thus assignment to groups, were based on DSM-5 criteria using a structured clinical interview (Kinder-DIPS) with parents. Experimenters who have conducted the Kinder-DIPS were adequately trained and were either psychologists or advanced students of psychology. A list of comorbidities is provided in the supplements. There was no significant difference regarding comorbidities between the SM and SAD groups. The mean age of our sample was $M = 9.71$ ($SD = 1.25$). Sample characteristics are displayed in Table 1.

Materials

Diagnostic Interview for Mental Disorders in Children and Adolescents (Kinder-DIPS)

The Kinder-DIPS (Margraf et al., 2017; Schneider et al., 2017) is a structured clinical interview enabling the

diagnosis of frequent mental disorders of childhood and adolescence according to DSM-5 and ICD-10. The interview has a high acceptance by both interviewers and interviewees (Neuschwander et al., 2017) as well as good to very good interrater-reliability (Neuschwander et al., 2013). In the present study, Kinder-DIPS was used to diagnose the mental disorder or to rule out the presence of such a disorder. Individuals who met the DSM-5 criteria for SM were assigned to the SM group independently of whether they also fulfilled the criteria for SAD. Individuals who only met the DSM-5 criteria for SAD but not for SM were assigned to the SAD group and individuals who did not meet any mental disorder criteria were assigned to the TD group.

Frankfurt Scale of Selective Mutism (FSSM)

The FSSM (Gensthaler et al., 2020b) is a parent-rated questionnaire assessing symptoms of SM in children and adolescents aged 3–18 years. The FSSM includes a diagnostic scale (DS) consisting of ten dichotomous items (yes–no) on the child's general speech pattern, with a cut-off value of 6 or 7 indicating the presence of SM, depending on the developmentally adapted version. Developmentally adapted versions are available for kindergarten age, school-age children between 6 and 11, and adolescents from 12 to 18. In addition, the FSSM provides a Severity Scale (SS) that can be used to dimensionally assess the symptom severity of SM for each version. Depending on the developmentally adapted version, this comprises either 41 items for kindergarten age or 42 items for the other two versions on speech behavior in different situations, taking into account the factors of verbal content, person and place. The questions are answered on a 5-point Likert scale and a total sum score can be calculated. In the present study, we formed z-scores of the SS in order to integrate sum scores of the different developmentally adapted versions to a joint total score. For this purpose, we z-standardized the mean SS scores of the different developmentally adapted versions for each child and created a new variable with the comparable z-score. Receiver operating characteristics analysis, which assesses the balance between sensitivity and specificity of a diagnostic instrument, indicated very good differentiation between children with SM, SAD and children without a mental disorder in the original sample of the FSSM (Gensthaler et al., 2020b). The authors of the questionnaire report high validity, as the SS of the FSSM correlates significantly with clinicians' symptom ratings (Gensthaler et al., 2020b). Previous reports also indicate excellent reliability (Cronbach's $\alpha = 0.90–0.98$) for the FSSM (Gensthaler et al., 2020b). The reliability was excellent in our sample as well ($\alpha = 0.951–0.959$).

Social Phobia and Anxiety Inventory for Children (SPAI-C)

We adopted the German version of the SPAI-C (Beidel et al., 2000; Melfsen et al., 2011), measuring self-reported symptoms of social anxiety. The questionnaire consists of 26 items with a 3-point Likert scale concerning different social situations. Scores ranged from 0 to 52. Validity is considered as high (Kley et al., 2012; Melfsen et al., 2011). Previous reports also indicate excellent reliability (Cronbach's Alpha = 0.92) for the SPAI-C (Kley et al., 2012; Melfsen et al., 2011). The reliability was excellent in our sample as well ($\alpha = 0.959$). Beidel et al. (2000) reported a cut-off score of 18, which differentiates well between children with SAD and non-socially anxious children.

Retrospective Infant Behavioral Inhibition Scale (RIBI)

The RIBI (Gensthaler et al., 2013; Gensthaler et al., 2020a) is a questionnaire assessing the child's BI regarding the first two years of life based on a retrospective parent report. The RIBI includes the subscales Distress to Novelty, Fear and Shyness and is summed up to a total score of BI. Items are answered on a 5-point scale (0 = Yes, 1 = more likely Yes, 2 = partly, 3 = more likely Not, 4 = Not). The test has excellent reliability ($\alpha > 0.90$) and convergent

Table 1 Sample characteristics

	SM	SP	TD	*p*	Post-hoc
n	28	28	28		
Age	9.39 (1.23)	9.61 (1.17)	10.14 (1.27)	.067	–
Gender (f/m)	18/10	16/12	12/16	.261	–
FSSM—DS	7.54 (2.25)	4.11 (2.83)	.79 (1.44)	< .000	SM > SAD > TD
FSSM-SS (z-score)	.72 (.98)	.17 (.78)	− .89 (.34)	< .000	SM > SAD > TD
SPAI-C sum score	19.26 (9.81)	18.05 (10.11)	8.88 (5.94)	< .000	SM = SAD > TD
RIBI score	39.64 (14.46)	41.43 (16.68)	25.14 (15.63)	< .000	SM = SAD > TD
Tracking-ratio (%)	71.08 (13.55)	72.78 (12.38)	73.23 (13.85)	.815	-

FSSM-DS Frankfurt Scale of Selective Mutism—Diagnostic Scale, *FSSM-SS* Frankfurt Scale of Selective Mutism—Severity Scale, *SPAI-C* Social Phobia and Anxiety Inventory for Children, *RIBI* Retrospective Infant Behavioral Inhibition Scale, *SM* Selective Mutism, *SAD* Social Anxiety Disorder, *TD* = Typical Development

validity indicated by positive correlations with questionnaires assessing BI as well as the behavioral observation of BI at 14 months of age. The reliability was excellent in our sample as well (α = 0.909).

Video Task

The self-constructed video task consists of a set of 39 trials (13 trials × 3 conditions) containing a fixation cross (randomly presented for 2–4 s) followed by a video-sequence (2–6 s) and a free viewing task (4 s). We created two identical sets of 36 videos with one female and one male adult amateur actor each (both were in their mid-20 s). In each video, the actor formulates either (1) a question ("How are you feeling today?"), (2) a negative social-evaluative statement ("You don't look good today!") or (3) a neutral statement ("I feel pretty good today."). During the free-viewing task, we presented the actor's face as a static image for 4 s after his/her question or statement was finished. The rationale here was that we could thus study gaze behavior both during dynamic interaction and in response to a question or a neutral or evaluative statement (after the question or statement was expressed). The actors looked into the camera throughout the sequences as well as the free-viewing task as if they were addressing the children directly. For the purpose of the standardization of the actors' position, the size of the face (55% in relation to the background), facial expression and clothes were kept constant throughout all videos. The length of the videos including the free-viewing task varies between 6 and 10 s (M = 7.90, SD = 0.94). Between conditions, on average the videos did not differ in length. To check whether the different lengths (6, 7, 8, 9 and 10 s) of the videos have an influence on gaze behavior, we investigated this for scanpath and fixation time in a mixed ANOVA (length x group). There was neither a main effect for length nor an interaction. To test whether the measured variables fixation time (in ms) and scanpath length (in px) were reliable in the present paradigm, we calculated Cronbach's alpha for both variables across the 13 trials per condition (3 × 13 trials). Fixation time exhibited excellent reliability scores for the whole video (α = 0.948–0.955) as well as for the 4 s free viewing task (α = 0.915–0.922) and good to very good reliability scores for the first 500 ms of the whole video (α = 0.756–0.808). The scanpath length had very good reliability scores for the whole video (α = 0.800–0.873) as well as for the 4 s free viewing task (α = 0.814–0.820).

Procedure

After parents participated in the online questionnaire to complete the FSSM, two experimenters visited the families at home to conduct the Kinder-DIPS with caregivers and the experimental paradigm with the children simultaneously but in separate rooms. While one experimenter conducted the interview with the parents, the other experimenter conducted various experiments with the child. Prior to the eye-tracking experiment published here, a physiological measurement was performed, which will be published elsewhere. The child had a standardized 5-min break before the eye-tracking experiment began. To run the experimental paradigm, the experimenters brought a laptop with an eye-tracker and installed it at a table, in front of which the child was seated with a chair. Each child was placed in a standardized position at a distance of 60 cm from the front of the screen (DELL Precision M4800, 17 inch) on which a remote eye tracker was mounted (SMI RED 250 mobile). The sound of the videos was played through the headphones that the children wore during the experiment. Children were instructed by the experimenter to sit as calmly as possible in front of the screen to avoid motion artifacts. They were also informed that they were about to watch some videos in which a person would speak to them. They were instructed to watch the videos and not answer back. The stimuli were presented using the SMI Experiment Center and eye-tracking data was recorded continuously. A 5-point calibration was performed with a tolerated deviation of 0.5 degrees followed by a validation step with an identical tolerated deviation. Trials were presented in a randomized order, whether the video was presented with a female or male actor was also chosen at random. After finishing the video task, children had time to complete the SPAI-C. The whole study session at the families' home lasted about two hours. The Local Ethics Committee of the Department of Psychology of the University of Giessen approved the study.

Data Preparation and Statistical Analysis

Data Preparation and Pre-analysis on Whole Video Sequences

According to previous studies using direct gaze as threatening stimuli (Weeks et al., 2013), we created an area of interest (AOI) around the eye region using BeGaze SMI-Software. All statistical analyses regarding components of attentional bias and visual exploration have been conducted in SPSS 26 using an alpha level of 0.05 and Bonferroni correction for multiple testing. We calculated correlations between the dependent variables and the score of BI as well as symptom scores of SAD and SM in the whole sample ($N = 84$). For both the investigation of possible attentional bias and the extent of visual exploration, we first (a) performed analyses for the complete video sequences independently of the conditions. We did this to investigate whether the eye contact of the depicted social counterpart, which represents a threatening stimulus for the clinical groups, is associated with an altered *fixation time in ms* or the *extent*

of visual exploration across all videos. As a marker of the *extent of visual exploration* or eye-movement, we analyzed the *length of the scanpath in pixels* according to previous studies (Horley et al., 2004; Toh et al., 2011).

Early Attentional Bias Towards Threat

Regarding an *early attentional bias towards the eye region* directly when the social counterpart is displayed on the screen, we (b) analyzed according to previous research (Waters et al., 2010) the *fixation time in ms* on the eye-region during the first 500 ms of the whole video sequences. We further investigated (c) the first 500 ms of the 4 s free-viewing time window following the video sequences. Here, we wanted to examine an early attentional bias towards the eye-region in response to each condition (question, evaluation, neutral statement) as formulated in hypothesis 1.

Late-Occurring Attentional Biases Towards Threat: Avoidance and Delayed Disengagement

To assess the potential late-occurring attentional components *delayed disengagement* and *avoidance* in reaction to the videos and because analyzing the entire video might blur the effect, we examined (d) *fixation time in ms* for the second half of the free-viewing task (the last 2 s). For a higher resolution, we additionally divided the 4-s sequence into eight blocks of 500 ms each, and (e) we examined the course of attention over the time intervals according to previous studies (Lisk et al., 2020; Schmidtendorf et al., 2018).

Extent of Visual Exploration

For the *extent of visual exploration* or eye-movement in response to the three conditions, we (f) investigated the length scanpath in pixels for 4 s of the free-viewing task. Given that the extent of visual exploration is associated with the construct of attentive freezing (Rösler & Gamer, 2019), we calculated correlations between length of the scanpath and freezing-related items (item 3: "Is your child incapable in certain situations of shaking his/her head, of nodding or of pointing to something when asked to?", item 4: "Do his/her movements seem slow or frozen-like to you in certain situations?" and item 5: "Does your child's facial expression appear less vivid or even expressionless and "frozen" in certain situations?") of the DS of FSSM (Gensthaler et al., 2020b). Due to the limited sample size, it was not possible to test if these items' load on a joint factor using an exploratory factor analysis. However, the three items showed substantial correlations among themselves ($r = 0.640–0.347$), in contrast to the other items of the DS.

Performed Analyses

a & b: In order to investigate the *fixation time on the eye-region during the whole video*, the *fixation time on eye-region during the first 500 ms of the whole video sequence* and *length of the scanpath during the whole video sequence,* we conducted a multivariate analysis of variance (MANOVA) comparing groups regarding the above-mentioned dependent variables.

c & d: In order to investigate *fixation time on the eye-region during the first 500 ms of 4 s-free viewing task* and the *fixation time on the eye-region during the second half of the 4 s-free viewing task*, we conducted 3 (group) × 3 (condition) repeated measures analyses of variance (ANOVA) for each of these dependent variables. Group (SM, SAD, TD) served as between-subject independent variable and condition as within-subjects variable.

e: Regarding the analysis of the 4 s-sequence in 500 ms time windows, we calculated a 3 (condition) × 3 (group) × 8 (8 time intervals a 500 ms) repeated measures ANOVA for *fixation time in ms*.

f: In order to investigate the *length of the scanpath during the 4 s free viewing task*, we conducted a 3 (group) × 3 (condition) repeated measures analyses of variance (ANOVA). Group (SM, SAD, TD) served as between-subject independent variable and condition as within-subjects variable.

The MANOVA (analyses: a & b) as well as ANOVA performed for the scanpath and non-exploratory fixation time variables (analyses: c, d and f) met all assumptions as the variables follow a normal distribution and Mauchly's tests for sphericity were not significant. Given that the Mauchly test for the ANOVA performed for exploratory analyses of the time course (e) was violated, a Greenhouse–Geisser adjustment was used to correct violations of sphericity.

Results

Descriptive Statistics and Correlational Analyses

Demographics and mean scores on psychometric measures for all three groups are presented in Table 1. Groups did not differ significantly regarding age, gender or quality of eye-tracking data. In line with previous studies, we found group differences regarding diagnostic scale as well as severity scale of the FSSM (Gensthaler et al., 2020b), elevated levels of trait social anxiety and BI in both children with SM and SAD indicated by the SPAI-C score (Muris & Ollendick, 2015) and RIBI-score (Gensthaler et al., 2016a) respectively. Additionally, there were no significant correlations between age, gender and the dependent variables (Table 2). Regarding the exploratory sum score of the three freezing items of the FSSM, we found group differences ($p < 0.001$,

SM > SAD > TD) with the highest score was in children with SM (range: 0–3, $M = 1.96$, $SD = 1.10$), followed by children with SAD ($M = 0.96$, $SD = 1.07$) and the lowest score was seen in children with TD ($M = 0.11$, $SD = 0.57$).

The correlation analysis revealed a negative relation between the symptom severity score of SM and fixation time on the eye-region across all video sequences in the whole sample ($N = 84$). We did not find any significant relation between SAD-symptoms or BI-scores and gaze behavior during complete video sequences (Table 2).

Attentional Bias

Regarding (a) the *fixation time on the eye-region* ($F(2,81) = 1.030$, $p = 0.362$, $\eta^2 = 0.025$) for *complete video sequences* we did not find group differences.

Early Attentional Bias

Regarding (b) the *fixation time on eye-region during the first 500 ms of the whole video sequences* ($F(2,81) = 0.746$, $p = 0.477$, $\eta^2 = 0.018$), we did not find group differences. Further, we did not find any main effect for condition ($F(2,162) = 0.018$, $p = 0.982$, $\eta^2 = 0.001$), group ($F(2,80) = 0.379$, $p = 0.686$, $\eta^2 = 0.009$) or interaction ($F(4,162) = 0.352$, $p = 0.842$, $\eta^2 = 0.009$) for (c) the *fixation time on eye-region during the first 500 ms of the 4 s-free-viewing task* in response to conditions.

Delayed Disengagement and Avoidance

Regarding analysis of a potential *delayed disengagement or avoidance,* we did not find any main effect for condition ($F(2,162) = 0.340$, $p = 0.712$, $\eta^2 = 0.004$), group ($F(2,160) = 1.013$, $p = 0.368$, $\eta^2 = 0.024$) or interaction ($F(4,162) = 0.446$, $p = 0.775$, $\eta^2 = 0.011$) for the second half of the 4 s—free-viewing task.

Time Course of Gaze Behavior During the 4 s Free Viewing Task

We displayed the time course of attention for each condition in Figs. 1, 2 and 3. There was a main effect for time interval ($F(181.779, 4.477) = 6.139$, $p < 0.001$, $\eta^2 = 0.071$). As contrasts revealed a significant linear decline of fixation time on the eye-region over the course of attention ($p < 0.001$, $\eta^2 = 0.167$), individuals looked increasingly less into the eyes of the social counterpart as the duration of the stimulus presentation progressed. We neither found a main effect for group ($F(626.602, 2) = 0.215$, $p = 0.807$, $\eta^2 = 0.005$) nor any interactions for condition x group ($F(7263.199, 3.537) = 0.215$, $p = 0.807$, $\eta^2 = 0.007$), condition x time ($F(2168.349, 9.226) = 0.495$, $p < 0.882$, $\eta^2 = 0.006$) or time x group ($F(7984.392, 8.953) = 1.348$, $p = 0.174$, $\eta^2 = 0.033$). Given that the interaction of group x time did not reach a significant level, a similar decline of fixation time on the eye-region for each group is suggested.

Extent of Visual Exploration

Main Analysis

Regarding (a) the *length of the scanpath* ($F(2,81) = 0.174$, $p = 0.840$, $\eta^2 = 0.004$) for *complete video sequences* we did not find group differences. Groups differed regarding the *length of scanpath* during the 4 s free-viewing task ($F(2,81) = 5.839$, $p = 0.004$, $\eta^2 = 0.126$). We neither found a main effect of condition ($F(2,162) = 0.211$, $p = 0.810$, $\eta^2 = 0.003$) nor an interaction of condition and group ($F(4,162) = 0.771$, $p = 0.535$, $\eta^2 = 0.019$), indicating that group differences did not depend on condition.

Table 2 Correlations between dependent variables of gaze behavior analyzed for whole video sequences and age, gender, and questionnaire sum scores based on the whole sample ($N = 84$)

	SPL (pixel)		FTeye (ms)		FTeye f500 (ms)	
	r	p	r	p	r	p
Age	− .114	.304	.080	.467	.128	.247
Gender	.109	.325	− .094	.394	− .146	.184
FSSM-SS	− .063	.568	**− .275**	**.011**	− .177	.108
FSSM-Freeze	− .208	.057	− .176	.110	− .109	.326
SPAI-C	.110	.320	− .112	.312	− .129	.241
RIBI	.002	.987	− .060	.616	− .048	.691

Significant correlations (p < .05) are marked in bold

SPL mean scanpath length across all videos in pixel, *FTeye* mean fixation time on eye-region across all videos in ms, *FTeye f500* mean fixation time on eye-region during the first 500 ms of each video in ms, *FSSM-SS* Frankfurter Scale of Selective Mutism—Severity Scale, *FSSM-Freeze* Freezing-related Items 3, 4 and 5 of Frankfurter Scale of Selective Mutism, *SPAI-C* Social Phobia and Anxiety Inventory for Children, *RIBI* Retrocspective Infant Behavioral Inhibition questionnaire

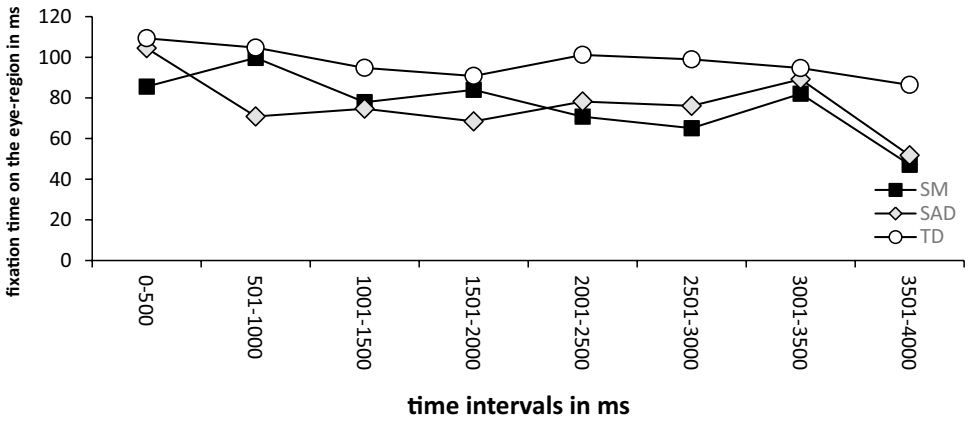

Fig. 1 Fixation time in ms on the AOI of eye-region during the 4 s free-viewing task averaged per group across all videos of condition *question*. *SM* Selective Mutism, *SAD* Social Anxiety Disorder, *TD* Typical Development

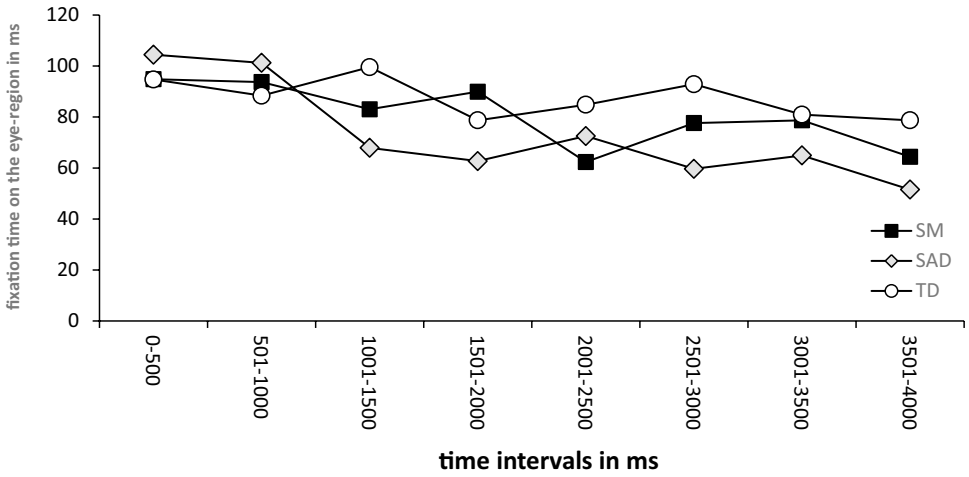

Fig. 2 Fixation time in ms on the AOI of eye-region during the 4 s free-viewing task averaged per group across all videos of condition *evaluation*. *SM* Selective Mutism, *SAD* Social Anxiety Disorder, *TD* Typical Development

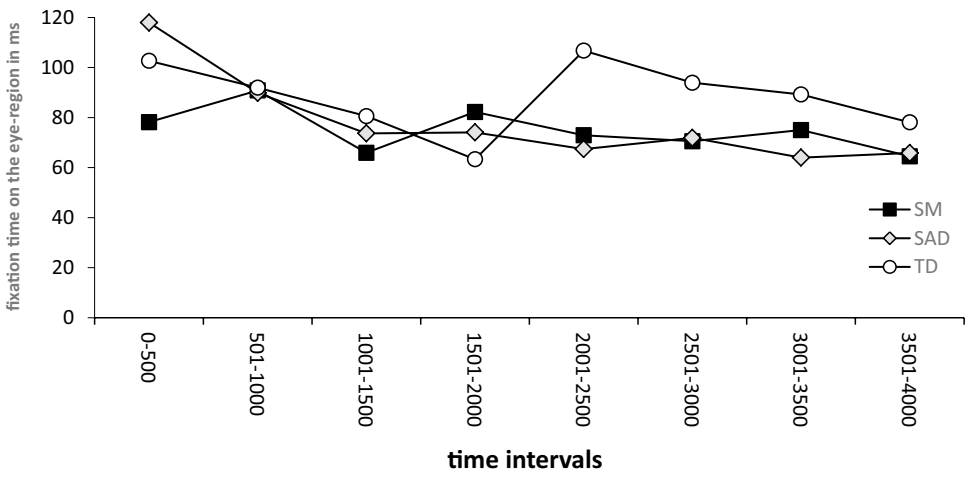

Fig. 3 Fixation time in ms on the AOI of eye-region during the 4 s free-viewing task averaged per group across all videos of condition *neutral*. *SM* Selective Mutism, *SAD* Social Anxiety Disorder, *TD* Typical Development

Bonferroni-corrected post-hoc tests revealed a significantly lower length of scanpath in the SM-group compared to the TD-group ($p = 0.003$; SM < TD). There were no differences between groups of SM and SAD ($p = 0.542$) and groups of SAD and TD ($p = 0.133$). Because we only found a significant main effect for group, but not a significant interaction for group x condition, we could not perform

group comparisons per condition. The extent of visual exploration of groups per condition are displayed in Fig. 4.

Further Analysis

In order to additionally test whether group differences in the scanpath are due to a fundamentally lower level of visual exploration or a longer attentional focus towards another area, we performed two further analyses with the Bonferroni-corrected alpha level ($\alpha = 0.025$). Groups did not differ regarding the length of the scanpath during the presentation of the fixation crosses (as a kind of baseline condition without a threatening stimulus) based on an ANOVA ($F(161.632, 2) = 0.424$, $p = 0.656$), indicating a similar level of visual exploration across groups. Furthermore, groups did not differ on the duration of all detected fixations on the screen (independent of AOI) during the 4 s free-viewing task based on an ANOVA ($F(115.530, 2) = 1.405$, $p = 0.251$), indicating on average a similar duration of fixation time on any area across groups. Fixation detection parameters were a minimum focus duration of 80 ms with a maximal dispersion of 2°.

Correlational analysis regarding 4 s free-viewing task in response to conditions revealed a negative relation between the exploratory calculated score of freezing-items of FSSM-DS and the length of the scanpath for all conditions (Table 3) in the whole sample ($N = 84$). As the extent of freezing increases, the extent of visual exploration decreases. Because of the exploratory nature of these correlations, we adjusted for multiple testing according to the conservative Bonferroni correction. The adjusted alpha level for this is $\alpha = 0.001$.

To further investigate which variable is able to predict the extent of visual exploration while statistically controlling for the remaining variables, we also performed a multiple regression with SPAI-C, FSSM-SS, and FSSM-Freezing items. In the significant model ($R^2 = 0.142$, $F(3, 83) = 4.407$, $p = 0.006$) only the freezing items ($\beta = -0.412$, $p = 0.004$) predicted the length of the scanpath, while SPAI-C ($\beta = -0.001$, $p = 0.991$) and FSSM-SS ($\beta = 0.059$, $p = 0.401$) did not have an influence on the extent of visual exploration. Due to the exploratory nature of the regression, we again applied a Bonferroni alpha level correction per each included predictor. The adjusted significance level is $\alpha = 0.016$.

Sensitivity Analysis

To check whether we had sufficient statistical power for the analyses we performed, we conducted a sensitivity analysis as recommended in the literature (Perugini et al., 2018). The mixed ANOVAs comparing the dependent variables between groups for the three conditions (question, evaluation, neutral) with conservatively expected small effect size of a group x condition interaction and correlations found between within-variables of average $r = 0.80$ between the three conditions, had a power of 0.81. The mixed ANOVAs based on the eight time intervals and correlations of an average of $r = 0.65$ between the eight time intervals within conditions, had a power of 0.81. Dimensional analyses

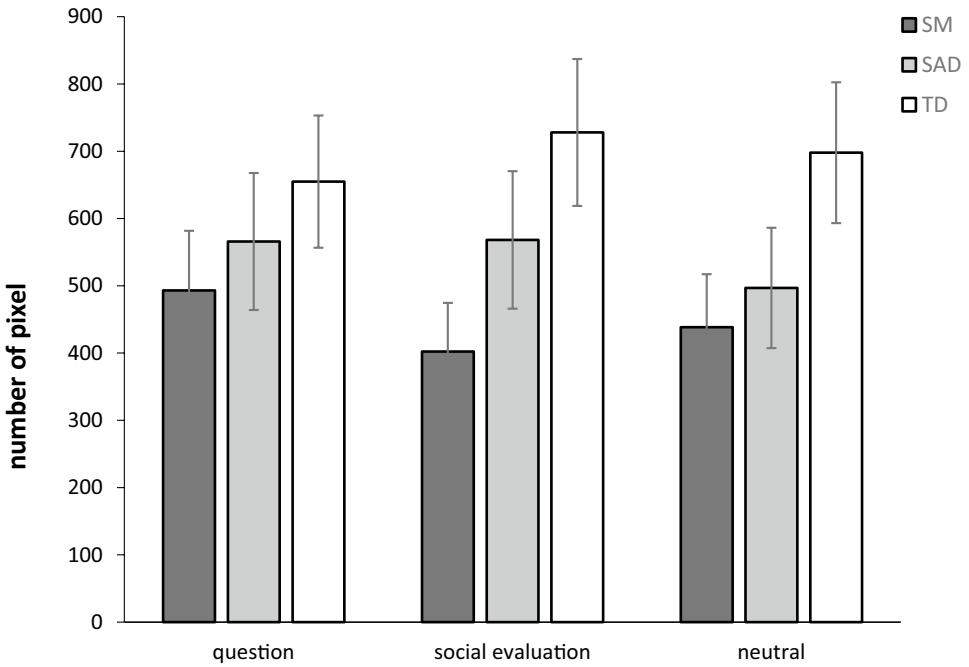

Fig. 4 Total length of Scanpath in number of pixel during the 4 s free-viewing task averaged per group across all videos of each condition. *SM* Selective Mutism, *SAD* Social Anxiety Disorder, *TD* Typical Development

Table 3 Correlations between dependent variables of gaze behavior during 4 s-free viewing task and age, gender, and questionnaire sum scores based on the whole sample ($N = 84$)

	SPL-Q (pixel)		SPL-E (pixel)		SPL-N (pixel)		SPLfvt (pixel)		FT-Q (ms)		FT-E (ms)		FT-N (ms)	
	r	p	r	p	r	p	r	p	r	p	r	p	r	p
Age	.046	.675	.104	.094	.094	.394	.106	.339	.045	.686	.074	.503	.091	.412
Gender	.009	.933	.014	− .034	− .034	.758	− .003	.981	− .134	.224	− .104	.347	− .047	.670
FSSM-SS	− .097	.380	− .205	.061	− .192	.080	− .212	.052	− .162	.141	− .147	.183	− .107	.332
FSSM-Freeze	− .252	.021	**− .358**	**.001**	− .257	.018	**− .374**	**.001**	− .151	.171	− .101	.360	− .094	.396
SPAI-C	− .140	.204	.005	.967	− .086	.436	− .088	.426	.089	.421	.003	.979	.044	.691
RIBI	.148	.215	− .031	.026	.026	.830	.055	.646	− .054	.625	− .026	.826	.016	.893

Significant correlations are marked in bold

We corrected for multiple testing using the conservative Bonferroni correction. The adjusted alpha level is = .001

SPL-Q mean scanpath length during 4 s free-viewing task after videos of condition question in pixel, *SPL-E* mean scanpath length during 4 s free-viewing task after videos of evaluation in pixel, *SPL-N* mean scanpath length during 4 s free-viewing task after videos of condition neutral in pixel, *SPL-fvt* mean scanpath length during 4 s free-viewing task after all videos, *FT-Q* mean fixation time on eye-region during the last 2 s of the free viewing task after condition question in ms, *FT-E* mean fixation time on eye-region during the last 2 s of the free viewing task after condition evaluation in ms, *FT-N* mean fixation time on eye-region during the last 2 s of the free viewing task after condition neutral in ms, *FSSM-SS* Frankfurter Scale of Selective Mutism—Severity Scale, *FSSM-Freeze* = Freezing-related Items 3, 4 and 5 of Frankfurter Scale of Selective Mutism, *SPAI-C* Social Phobia and Anxiety Inventory for Children, *RIBI* Retrocspective Infant Behavioral Inhibition questionnaire

(correlations and regression) based on the entire sample of the current study also had a sufficient power (0.84–0.88) assuming medium effect sizes as reported in the literature (Seefeldt et al., 2014). Group comparisons on the dependent variables and conducted MANOVA were slightly underpowered with a power of 0.64 and 0.65, assuming medium effect sizes between socially anxious and healthy children of the same age group as reported in the literature (Seefeldt et al., 2014).

Discussion

The purpose of the present study was to examine different components of attentional bias as well as the extent of visual exploration in the presence of threat in children with SM compared to children with SAD and children with TD. We measured attentional processing using eye-tracking during videos displaying a social counterpart directly looking at the child as well as during a free-viewing task that followed these video sequences of threatening disorder-congruent situations.

Early Attentional Bias Towards Threat

Results contradict our assumption that children with SAD and children with SM show an early attentional bias towards threat, which was indicated by previous studies in socially anxious children (Keil et al., 2018; Schmidtendorf et al., 2018; Seefeldt et al., 2014; Waters et al., 2010). Contradictory findings might be explained by differences between paradigms, as our study is the first to investigate attentional bias in children with SAD during the course of a dynamic social situation. Most previous studies in children with SAD have measured attention bias by contrasting threatening and neutral static stimuli. Additionally, so far only Keil et al. (2018) used the eye region of the counterpart as threatening stimulus in children with SAD, whereas the other disorder-specific studies in children with SAD each used the entire face (Schmidtendorf et al., 2018; Seefeldt et al., 2014; Waters et al., 2010). In the study done by Keil et al. (2018), children with SAD showed an early attentional bias toward the eye area in only one of three conditions. This raises the question regarding the robustness of the early bias towards threatening stimuli in children with SAD when a differentiated analysis of gaze behavior with respect to the eye area is performed, which is, according to previous research, of high importance (Weeks et al., 2013). An additional explanation for our finding of no attentional bias, especially in children with SM, is that attentional focus may depend on the amount of visual exploration, masking possible effects. Given the finding that children with SM show reduced visual exploration, it could be that their eye movement is frozen, so that they show less eye-movement towards or away from the eye-region and thus demonstrate no attentional bias. Beyond these methodological and theoretical considerations, it is important to emphasize that our group comparisons had somewhat too little statistical power and thus we might have failed to detect possible effects in children with SAD or SM.

Avoidance

Our results do not indicate a stronger avoidance or delayed disengagement in children with SM and children with SAD compared to children with TD. However, we found a negative correlation between fixation time on the eye-region across conditions and symptom severity of SM across the entire sample. Taking into account that our group comparisons were underpowered, it stands to reason that we could not detect this potential effect in the contrast between groups and thus captured it only based on the dimensional analyses. In addition to other possibilities that might explain why a child gazes at a certain stimulus for less time (for example, loss of interest in the stimulus or a generally reduced attention span), avoiding a threatening stimulus seems to be a reasonable explanation in the context of SM. This would be in line with the conceptualization of SM as an anxiety disorder and findings that avoidance of threatening stimuli is a component of attention processing in the context of anxiety (Cisler & Koster, 2010). Interestingly, several case reports already exist suggesting that some children with SM avoid eye contact due to fear (Kovac & Furr, 2019; Muris & Ollendick, 2015; Wong, 2010). Given that there are numerous studies showing that socially anxious adult individuals avoid eye contact in dynamic social situations and social anxiety is a core feature of SM (Muris & Ollendick, 2021), it would seem reasonable to assume that this relationship may be driven by social anxiety. However, in our study, we did not find a correlation between social anxiety and avoidance of eye contact. In previous research on children with SAD, the only study that examined gaze behavior with respect to the eye region in socially anxious children also did not find attentional avoidance in children with SAD. Although speculative, this may suggest that social anxiety does not yet play a role in relation to gaze avoidance in children, and gaze avoidance may rather be a mechanism associated with the symptomatology of SM in children.

We also found a gradual decrease of attention on, and thus probably an avoidance of, the social counterpart's eye-region over time in response to the videos for all three groups. This finding could be in line with developmental research (Dudeney et al., 2015; Field & Lester, 2010), which suggests that all individuals, healthy ones included, display an attentional bias. While healthy individuals learn to regulate their attention during development, anxious individuals retain an attentional bias regarding threat. It is suggested that this process is associated with a lack of maturation of top-down regulatory cognitive functions in anxious individuals (Dudeney et al., 2015). Again reasons other than avoidance could also be considered as reason why a decrease in focusing on the eye region with the duration of stimulus presentation have been observed. For example, preliminary research suggests that boredom is also associated with a loss of attentional focus (Kim et al., 2018).

Extent of Visual Exploration

Interestingly, we found that only children with SM had a significantly lower visual exploration compared to children with TD. Although only children with SM differed from children with TD in terms of the scanpath length, there seems to be a gradient in the extent of visual exploration across groups, with children with SM showing the strongest inhibition of visual exploration (see Fig. 4). Because our group comparisons were underpowered, we may not have detected a possible difference between children with SAD (without SM) and children with TD. Therefore, reduced visual exploration, although possibly less pronounced, might also occur in children with SAD. Additionally, given that a large proportion of children with SM in our study also met criteria for SAD, it is questionable whether reduced visual exploration is only a characteristic of children with SM and SAD or also occurs in non-socially anxious children with SM. Future studies should disentangle this by examining visual exploration in subgroups of children with SM with sufficiently large sample sizes.

Various explanations are possible for the finding of reduced visual exploration in our SM-group. For example, it could be that children with SM show a fundamentally lower level of visual exploration than children with TD or focus longer on another area during social interaction and remain there with their attention. However, both explanations are contradicted by the result that the groups did not differ on visual exploration during the presentation of the fixation crosses as well as regarding the average duration of fixations, irrespective of areas of interest. However speculative, the current finding might be an indicator of the involvement of the mechanism of attentive freezing in SM, which is also associated with reduced eye-movement and thus a reduced visual exploration. This would be in line with previous research which indicates an association between reduced visual exploration and the fear response of freezing in healthy adults (Löw et al., 2015; Rösler & Gamer, 2019; Wendt et al., 2017). Furthermore, this consideration is supported by our correlation analysis (see Table 3) as well as the conducted multiple regression, in which reduced visual exploration (indicated by length of the scanpath) was predicted by the freezing-items of the FSSM.

Implications for SM-Symptomatology

Although it can only be speculated based on the current results whether reduced exploration was due to attentive freezing, there is evidence that suggests that this may be

linked with the inability to speak in certain situations in children with SM. Attentive freezing is considered as part of a biologically-driven defense cascade that occurs across species (Kozlowska et al., 2015) and includes a pattern of psychophysiological reactions such as a decline of heart rate as well as reduced motor activity including eye-movement and vocal inhibition (Kozlowska et al., 2015; Roelofs, 2017; Rösler & Gamer, 2019). Findings that children with SM display high levels of BI (Gensthaler et al., 2016a), are described as frozen with fear by clinicians (Anstendig, 1999) and report a paralyzing fear themselves (Vogel et al., 2019) might also support this assumption. Furthermore, the mechanism of freezing would also provide an explanation as to why in the current study children with SM showed reduced visual exploration in general, regardless of whether the situation contained a question or not. Because of the assumed biological foundation of the mechanism, freezing might be compared to attentional biases that depend on disorder-congruency of stimuli (Pergamin-Hight et al., 2015), less dependent on learning experiences and on the content of the social situation (i.e., whether it has a speech component or not). Consistently, evidence from recent research indicates that eye contact and the presence of strangers per se, induces fear in children with SM (Schwenck et al., 2021) and that children with SM exhibit longer latency to movement, even in social situations where they do not need to speak (Milic et al., 2020).

In order to draw a valid conclusion regarding whether the reduced visual exploration found in children with SM can be explained by freezing and in order to determine whether freezing is involved in SM-symptomatology, it would be important for future studies to also assess other features of freezing (e.g., physiological responses) during a task that requires speech-production in children with SM. Although we did not find an association between visual exploration and retrospectively recorded BI in our study, due to the conceptual proximity of freezing and BI on the one hand and BI and SM on the other, it would be important to further investigate the interaction of these variables. In this context, research in very young children with SM or high BI that is not dependent on retrospective data would be important.

Clinical Implications

Our findings might also have important clinical implications. Our results suggest that a frozen motor activity might be involved in SM-symptomatology. Furthermore, they indicate that this inhibition occurs in various types of social situations, including neutral situations that do not involve an expectation to speak or a social evaluation. For therapy, this means that even in the absence of an expectation to speak, and despite the use of techniques such as defocused communication, increased inhibition in children with SM might be expected. Thus, interventions that counteract this state of freezing during social situations might be promising. Although this is the first finding that suggests that it may be important to address the state of freezing in therapy, this approach is already found in the therapeutic literature on SM. Here, for example, it is described that activation exercises can be applied as a supportive element of an exposure (McHolm et al., 2005). Another clinical implication may arise from the finding that the symptom severity of SM is associated with avoidance of eye contact, which has already been described in single-case studies. This might suggest that eye contact is experienced as aversive by children with SM and is consequently avoided. Consequently, clinicians should be aware that direct eye contact may be counterproductive when interacting with children with SM during defocused communication. However, over time, learning to maintain direct eye contact could be a valuable target for exposure therapy.

Limitations

The current study has some limitations to acknowledge. First, comparability between our study and previously conducted studies on attentional bias in socially anxious children is limited due to differences in the applied paradigm as well as a descriptively lower symptom level of SAD in our sample compared to samples of previous studies (Schmidtendorf et al., 2018; Seefeldt et al., 2014). Second, due to a rather narrow age range of 8–12 years for our sample, applying these results to the typical onset of pre-school age or to early phases of SM might be limited. Third, three of the children assigned to the SM group in our study, consistent with findings on comorbidity rates between SM and SAD, did not have comorbid SAD. In contrast to the regression analyses based on SM- and SAD-symptomatology, the group comparison in our study does not allow an entirely accurate conclusion about which findings are specific to children with SM and whether SM subgroups (e.g. SM with and without SAD) would differ with respect to mechanisms. Fourth, individuals did not actually have to answer during the speech-demanding condition, so that this condition might not have had the expected fear-inducing effect. Although we did not assess state anxiety with respect to the stimuli, a previous study indicates a fear-inducing effect of the chosen conditions based on a subjective anxiety level (Schwenck et al., 2019). Fifth, four of the thirteen neutral statements included statements that pertained to the person depicted in the video (e.g., "I feel pretty good today."), which could lead to attention being focused on that person. Given that a comparison of fixation time in response to these four neutral items with fixation time in response to the remaining neutral items did not reveal a difference, this does not seem to have resulted in

a bias in the measures of attention. Sixth, the actors shown in the videos were adults, as is the case in previous research of gaze behavior in socially anxious children. Given that some children with SM are more likely to show symptoms in the presence of adults, while others are more likely to have difficulty in speaking in the presence of peers, it would be useful to investigate gaze behavior towards peers as well. Seventh, the group comparisons show a power that is slightly too low to detect the effects between socially anxious and healthy children assumed in the literature. However, the remaining analyses conducted in this study had sufficient statistical power.

Conclusion

In conclusion, this is the first study that has investigated attention processing in children with SM. We did not find evidence of the presence of any of the components of an attention bias in dynamic social situations in children with SM nor in children with SAD. However, we found a lower attentional focus on the eye-region to be associated with a higher level of SM-symptoms, probably indicating a relation between avoidance of eye-contact and the presence of SM. Given that there is already evidence of an early bias in children with SAD from studies with static threatening stimuli, the different findings could be due to differences in the applied paradigm. We also found that children with SM showed reduced visual exploration regardless of the video condition. This suggests that reduced visual exploration in children with SM generally occurs in social situations and does not depend on the context of the social situation. Reduced visual exploration might be explained by attentive freezing (inhibition of the visual motor system). The literature suggests that attentive freezing is part of a more fundamental psychophysiological response that may also affect speech production. Thus, this mechanism may be used to explore the occurrence of the inability to speak in certain situations in children with SM. Additional experimental research is needed to address the assumption that freezing is involved in the symptomatology of children with SM.

Supplementary Information The online version contains supplementary material available at https://doi.org/10.1007/s10608-021-10289-3.

Author Contributions All authors contributed to the study conception and design. The first draft of the manuscript was written by Felix Vogel and all authors commented on previous versions of the manuscript. All authors read and approved the final manuscript.

Funding Open Access funding enabled and organized by Projekt DEAL.

Declarations

Conflict of interest The authors declare that they have no conflict of interest.

Ethical Approval The study was approved by the local ethics committee of the Department of Psychology of the University of Giessen and therefore has been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments. All persons who were included in this study gave their informed consent prior to the start of their participation.

Informed Consent Informed consent was obtained from all individual participants included in the study and written informed consent was obtained from the parents.

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