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ORIGINAL PAPER

Reducing Listening-Related Stress in School-Aged Children with Autism Spectrum Disorder

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Abstract High levels of stress and anxiety are common in children with Autism Spectrum Disorder (ASD). Within this study of school-aged children (20 male, 6 female) we hypothesised that functional hearing deficits (also pervasive in ASD) could be ameliorated by auditory interventions and that, as a consequence, stress levels would be reduced. The use of Ear-Level Remote Microphone devices and Classroom Amplification systems resulted in significantly improved listening, communication and social interaction and a reduction in physiologic stress levels (salivary cortisol) in both one-on-one and group listening situations.

Keywords Autism spectrum disorder · Hearing · Stress · Remote microphone listening system · Classroom amplification

Abbreviations

ASD	Autism Spectrum Disorder
SNR	Signal to noise ratio
HPA	Hypothalamic–pituitary–adrenal
dBSPL	Decibels sound pressure level
dBHL	Decibels hearing level

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Introduction

Autism spectrum disorder (ASD) is a neurodevelopmental condition involving two primary symptom dimensions: social interaction impairment and restricted or repetitive patterns of activity/interest (APA 2013). In addition, affected individuals often present with a range of co-occurring symptoms including stress and anxiety (Morgan 2006). "Stress" is a state in which an individual's homeostasis is disrupted by real or perceived challenges and many of the core features of ASD (impaired social reciprocity, inability to cope with changes to routine, ritualistic patterns of behaviour etc.) may be seen as either causes of, or manifestations of stress (Howlin 1998; Sukhodolsky et al. 2008; Rodgers et al. 2012).

Abnormal stress responses in children with ASD have been demonstrated through physiological measures, most notably cortisol concentration. Cortisol is a hormone released from the adrenal gland following stimulation of the hypothalamic–pituitary–adrenal (HPA) axis (Hennessey and Levine 1979; Herman and Cullinan 1997), with wide-ranging effects on biological functions (Sapolsky et al. 2000). Elevated baseline cortisol concentrations have been demonstrated in children with ASD, particularly in low functioning individuals (Corbett et al. 2008; Kidd et al. 2012; Putnam et al. 2015), and may reflect an inability to adapt to the persistent environmental challenges that are central to the disorder (Romanczyk and Gillis 2006). Furthermore, abnormal cortisol responses have been demonstrated to specific psychosocial stressors. Interestingly, where children with the disorder show a hypo-reactive cortisol response to socio-evaluative tasks (such as public speaking), they typically demonstrate hyper-reactivity to situations requiring interaction with others (Corbett et al. 2012; Schupp et al. 2013).

Abnormal responses to sensory stimuli across multiple modalities are a consistently reported feature of ASD and are now a recognised component of diagnosis (APA 2013). Hearing impairment is relatively common in this population with 2–10% of cases presenting with impaired sound detection thresholds (Gillberg et al. 1983; Rosenhall et al. 1999; Szymanski et al. 2012; Beers et al. 2014). In addition, a high proportion (>50% of paediatric cases) show auditory processing deficits which are thought to be related to a distorted representation of temporal cues in the central auditory pathways (Alcantára et al. 2012; Rance et al. 2014a). The functional consequences of these auditory abnormalities are impaired speech perception and real-world listening deficits severe enough to exacerbate the communication issues central to ASD (Rance et al. 2014a). Listening in background noise (figure/ground perception) is a particular problem (Alcántara et al. 2004; Rance et al. 2014a) and recent findings have, in fact, suggested that for school-aged children with ASD, the ability to identify speech and maintain concentration in the (noisy) classroom is the most significant predictor of academic progress (Ashburner et al. 2008). The mechanisms underlying auditory figure/ground deficit in children with ASD are yet to be fully established. Co-existing intellectual, attention and language deficits are likely to play a part (Dawson et al. 1998; Čeponienė et al. 2003), but fundamental limitations in the neural representation of both monaural and binaural acoustic cues are likely to limit perceptual ability in a high proportion of cases (Alcántara et al. 2004; Groen et al. 2009; Kwakye et al. 2011; Rance et al. 2014a).

The link between impaired hearing ability and stress in children with ASD is yet to be explored, but it has been established that hearing impairment, and it's concomitant communication challenges, can affect the physiologic stress response (Bess et al. 1998, 2016; Bess and Hornsby 2014). Bess and colleagues have recently, compared salivary cortisol concentrations in a group of children with mild-moderate degree hearing loss and a cohort with normal hearing and found significant elevation of the Cortisol Awakening Response (CAR) in the hearing impaired subjects. The CAR is a well-defined phenomenon involving an approximate doubling of cortisol concentration in the first 30–45 min after awakening. It's elevation in the hearing impaired group was thought by the authors to indicate either a need to mobilize energy in preparation for the day or heightened levels of anxiety (Bess et al. 2016).

Listening in a noisy environment can be improved by optimizing the quality of the acoustic signal reaching the listener's ears. One approach is to use "remote-microphone" technologies which record the speaker's voice in close proximity to the mouth (usually via a microphone worn at the lapel) and send the signal directly to the listener. Ear-level remote microphone systems employ this technique, transmitting the acoustic signal via radio waves to an earpiece worn by the child. In the classroom, this can dramatically improve the signal-to-noise ratio, the level of the speech relative to the background noise, replicating an acoustic situation where the teacher is only 30 cm from the student regardless of their relative positions within the room. These devices are readily available and their efficacy has been demonstrated both in listeners with sensorineural hearing loss (Hawkins 1984; Anderson and Goldstein 2004; Toe 2008; Thibodeau 2010) and auditory processing disorder (Johnstone et al. 2009; Schafer et al. 2014). Recent work has also suggested that Ear-Level Remote Microphone systems may improve hearing/communication in children with ASD (Rance et al. 2014a; Schafer et al. 2014, 2016).

"Soundfield Classroom Distribution" is another auditory intervention designed to improve classroom listening. In this case, the teacher's voice (recorded at the lapel), is amplified via strategically placed loudspeakers, improving the signal-to-noise ratio for all students in the listening space. To date there are no published studies exploring the efficacy of Soundfield Distribution for children with ASD, but this approach has been shown to improve classroom listening behaviours in children with other forms of developmental disability (Flexer et al. 1990; Blake et al. 1991).

This study is the first to objectively evaluate the effect of auditory intervention on the stress response in children. In particular we explored the relation between functional hearing ability and stress and evaluated the potential ameliorating effect of auditory interventions in school-aged children with ASD. In Study A we hypothesized that (1) children with the poorest auditory perception would show the greatest physiologic stress response (increase in cortisol concentration) during a challenging one-on-one listening session (2) provision of an Ear Level Remote Microphone listening system would improve speech perception and decrease self-reported perception of hearing disability and (3) that use of an Ear Level Remote Microphone device would reduce the physiologic stress response during a one-on-one listening session. In Study B we hypothesised that amplification of the teacher's voice would reduce physiologic stress levels in a challenging group listening session.

Method

Participants

Twenty-six children (6 girls) with ASD participated. The disorder was identified in each subject via multidisciplinary clinical assessment with input from paediatricians, psychologists and speech pathologists and employing a range of instruments (Table 1). Three children had been diagnosed

Table 1 Participant demographics

Participant	Gender	Age at assessment (years)	Av. hearing level (dBHL)	ASD diagnosis	FSIQ	CBCL/TRF anxiety subscale (percentile)
ASD1	F	12.0	10.0	DSM-IV, CELF-4 WISC-IV, BASC-2	105	54
ASD2	M	8.0	30.0	ABC, BASC-2 CELF-P, CBCL CELF 4, WPPSI-III	112	97
ASD3	M	12.0	7.5	DSM-IV, CELF-4	99	62
ASD4	M	11.0	7.5	DSM-IV, CBCL, WISC-IV, CELF-4	116	79
ASD5	F	6.0	12.5	ADOS, DSM-IV, WISC-IV	92	97
ASD6	M	8.0	11.2	DSM-IV, CELF-4, WISC-IV	91	54
ASD7	M	7.5	27.5	DSM-IV, CELF-4	*	69
ASD8	M	11.3	10.0	DSM-IV, CELF-4	82	97
ASD9	M	11.3	7.5	DSM-IV, CELF-4, TOPL-2	*	93
ASD10	F	9.0	10.0	DSM-IV, WISC-IV, ADOS, CELF-4, TOPL-2	97	97
ASD11	M	9.6	10.0	ADOS, ADI-R DSM-IV, CBCL, CELF-4	52	93
ASD12	M	9.8	25.0	DSM-IV, CELF-4	91	69
ASD13	F	9.2	12.5	DSM-IV, WPPSI-IV, CELF-4	65	81
ASD14	F	8.3	13.7	DSM–IV, ADI-R, CBCL, WISC	103	>97
ASD15	M	9.7	1.2	DSM-IV, CELF-4	*	>97
ASD16	F	8.6	7.5	DSM-V,WISC-IV,DBC,ADOS-2	108	>97
ASD17	M	14.3	11.2	DSM-IV, CBCL	63	93
ASD18	M	16.3	11.2	DSM-IV, WPPSI-R, CELF-4	98	>97
ASD19	M	14.3	13.7	DSM-IV, CBCL, TRF	100	97
ASD20	M	13.3	10.0	ABC, CBCL	83	93
ASD21	M	16.8	12.5	ABC, WISC-IV	87	97
ASD22	M	14.3	12.5	DSM-IV, CELF-4	*	89
ASD23	M	14.1	10.0	ADOS-2	*	73
ASD24	M	14.2	11.2	ASRS, BASC WISC-IV	104	>97
ASD25	M	15.9	12.5	DSM-IV, CELF-4	87	<50
ASD26	M	15.9	10.0	DSM-IV,CELF-4, WISC-IV, BASC	78	97

*Denotes data not available, *ABC* autism behaviour checklist, *ADI-R* autism diagnostic interview – revised, *ADOS* autism diagnostic observation scale, *ASRS* autism spectrum rating scale, *Av.* hearing level, Better ear 4-frequency average hearing level (500 Hz/1 kHz/2 kHz/4 kHz), *BASC* behaviour assessment system for children, *CBCL* childhood behaviour checklist – parent (Achenbach System of Empirically Based Assessment), >70 indicates clinically anxious, *CELF-4* clinical evaluation of language fundamentals (Edition 4), *CELF-P* clinical evaluation of language fundamentals (Pre-School), *DBC* developmental behavioural checklist, *DSM-IV* diagnostic and statistical manual of mental disorders (Edition IV), *dBHL* decibels hearing level, *FSIQ* full scale intelligence quotient, <70 indicates intellectual disability, *TOPL-2* Test of Pragmatic Language (Edition 2), *TRF* teacher's report form (Achenbach System of Empirically Based Assessment), >70 indicates clinically anxious, *WISC-IV* wechsler intelligence scales for children (Version IV), *WPPSI-III* wechsler pre-school and primary scale of intelligence (Version III), *WPPSI-R* wechsler pre-school and primary scale of intelligence (Revised)

with intellectual disability, but all participants were able to communicate verbally and attended a mainstream school. Sixteen children (ASD1-16) participated in Study A, and ten children (ASD17-26) took part in Study B. Children were assigned to each Study based on their age and educational setting. Those in Study A were younger (6.0–12.0 years, M = 9.5 years, SD = 1.7 years) and attended their local primary school where they each had a consistent classroom teacher. Previous work (Rance et al. 2014a) has shown good acceptance of Ear-Level Listening Devices in this context. Participants in Study B were older (13.3–16.8 years, M = 14.9, 1.2 years), and attended local secondary schools where they each had a number of subject teachers. As our previous studies had suggested that personal (ear-level) devices were not well tolerated by adolescents with ASD in mainstream secondary schools (Rance et al.

2014a), a Classroom Amplification strategy was trialled for this group.

Participant anxiety levels were evaluated using the Achenbach System of Empirically Based Assessment. For children in Study A, the parents completed the Child Behaviour Checklist (CBCL) and for Study B, the Teacher Report Form (TRF) was used. Twenty of the 26 participants were clinically anxious (Table 1).

Child Behaviour Checklist (Achenbach et al. 1991a)

The CBCL is a parent report form exploring problem behaviours that occur in childhood across multiple domains including anxiety, stress, attention and social interaction. It is a well validated instrument with high reliability coefficients (≥0.80) reported across the various subscales (Achenbach and Rescorla 2001). The checklist can be used to identify behaviours indicative of psychiatric issues in children aged 6–18 years and has been employed previously in ASD populations, with affected children typically showing higher scores across all domains (Schroeder et al. 2011). The questionnaire requires that the parent rate the frequency of behaviours using a Likert scale ranging from 0 = "not true" to 2 = "very often true".

Teacher Report Form (TRF) (Achenbach et al. 1991b)

The TRF is the parallel form to the CBCL and can be used by teachers to assess problem behaviours in the classroom. As with the CBCL, this instrument has been well validated and shown to have high reliability coefficients (r ≥ 0.80) across the anxiety-related domains (Achenbach and Rescorla 2001). Child behaviour is rated on a Likert scale with response options ranging from 0 = "not true" to 2 = "very true".

Procedures

Study A: Ear-Level Remote Microphone Systems

Each subject participated in three test sessions conducted in a quiet room at the family home. The experimental schedule for this study is summarized in Fig. 1. At-home testing was used for family convenience and to ensure that the child was as relaxed as possible. The ambient noise level was <45 dBSPL in all cases, and as such, would not have affected the perceptual hearing assessments. In the initial session, each participant (with parental assistance) completed a hearing disability survey (Abbreviated Profile of Hearing Aid Benefit) and an audiometric assessment.

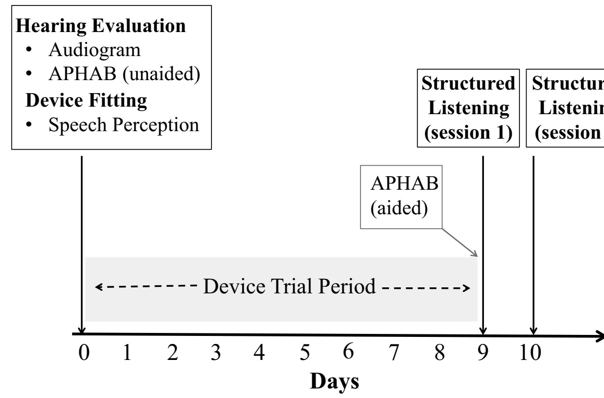

Fig. 1 Experimental schedule for Study A. An average device trial period of 9 days is shown

Abbreviated Profile of Hearing Aid Benefit (APHAB) (Cox and Alexander 1995)

The APHAB is a hearing disability-based inventory that can be used to explore the degree of hearing/communication difficulty experienced in a range of common listening scenarios. The questionnaire authors (Cox and Alexander 1995) report a reliability coefficient in the moderate-to-high range (r = 0.76) and suggest that the instrument can be used to document the effect of auditory intervention. The questionnaire asks the subject to rate their degree of disability in a range of listening situations using a Likert scale ranging from "Always (99% of occasions)" to "Never (1% of occasions)". From these responses a percentage score representing the proportion of everyday situations in which the subject experiences difficulty across a range of domains is calculated. In the present study, responses for the "Ease of Communication", "Listening in Background Noise" and "Aversiveness to Sound" subscales were obtained. This questionnaire has been used previously in school-aged children with ASD and found to provide reliable estimates of real-world listening ability (Rance et al. 2014a).

Sound Detection Threshold Levels

Audiometric testing was carried using pure tone stimuli presented to each ear separately. Sound detection thresholds were obtained at octave frequencies between 250 Hz and 4 kHz and a 4-frequency average hearing level based on hearing levels obtained at the 500 Hz, 1, 2 and 4 kHz test frequencies was calculated for the better hearing ear (Table 1).

After completion of the baseline hearing assessments the child was fitted with an Ear-Level Remote Microphone system. The device was a Phonak Roger Focus receiver fit monaurally and paired with a Roger

Inspiro-FM transmitter. The receiver consists of a small device placed behind the pinna and a small rubber ear-piece which sits in the ear canal. The transmitter is worn by the person communicating most with the child—typically a teacher in the classroom environment. Set-up of the system was as per manufacturer recommendations. The Roger Focus receiver was fit to the ear with better sound detection thresholds, or to the child's preferred side in cases where the average hearing levels were equal. No gain was applied to the signal at the receiver as each of the participants had normal or near-normal sound detection thresholds. The "Bluetooth" connection between transmitter and receiver was checked at the start of each test session and an informal listening test was carried out in which the child was required to answer a series of simple questions posed by the tester (wearing the transmitter) from another room. Speech perception assessment (Consonant-Nucleus-Consonant [CNC] Word test) was then conducted in both unaided and aided listening conditions.

Consonant-Nucleus-Consonant [CNC] Word test (Peterson and Lehiste 1962)

Open set speech perception assessment (Consonant-Nucleus-Consonant [CNC] Word test) was conducted using recorded speech stimuli (50 words per condition). The CNC-word test is widely used in both clinical and experimental contexts and has been shown to have excellent reliability ($r > 0.90$) (Causey et al. 1984). Speech stimuli were presented from a loudspeaker in front of the subject and competing noise (4-Talker Babble) from a loudspeaker behind (Rance et al. 2014a). Target speech and noise were both calibrated to reach the child's head at 65 dBSPL (0dB signal-to-noise ratio [SNR]) replicating listening conditions in a typical school classroom (Crandell and Smaldino 2000). The child verbally repeated the CNC stimulus and responses were phonetically transcribed in real-time. The tester (scorer) was positioned in front of the child (where participant voice-levels were typically 10–15 dB higher than the background noise) to ensure scoring accuracy.

Each participant was then afforded 1–2 weeks of device experience ($M = 8.8$ days, $SD = 2.1$ days) in which the listening system was worn at school and home for 4–6 h per day. At the completion of this trial period each child/parent repeated the APHAB questionnaire estimating the degree of listening/communication difficulty when wearing the device. Parents were also invited to repeat the CBCL anxiety measure evaluating their child's behaviour when wearing the listening device.

Structured Listening/Comprehension Sessions

Following the device acclimatization period, two structured listening sessions were carried out at the same time of day (4.00–4.20 pm) and usually on consecutive days. These 20 min sessions were designed to provide a significant listening challenge and involved a series of auditory perceptual and comprehension tasks presented in competing background noise (4-Talker Babble). The perceptual task was a list CNC-words and the comprehension material was taken from the Clinical Evaluation of Language Fundamentals (CELF-4) test and included the "Understanding Spoken Paragraphs" and "Concepts and Directions" subsections (Semel et al. 2003). Content was matched to the child's speech and language abilities. Different test items were used in each structured session to prevent familiarity effects. For each of the listening/comprehension tasks the child's response was recorded (to maintain concentration), but the findings were not analysed. The listening material was typically presented "live voice", and as such, the participant had access to both auditory and speech-reading (mouth/facial) cues–as per normal communicative interactions. The tester was seated 2.5 m in front of the child and a loudspeaker providing competing noise was placed behind. The direct signal (tester's voice) and noise were both 65 dBSPL at the child's head (Fig. 2a). In both sessions the tester wore the transmitter and the child wore the Ear-Level receiver. The system was turned on in one session and off in the other. Eight children were tested in the unaided condition first and eight in the aided first.

Salivary Cortisol concentrations were obtained from saliva samples collected before and after each structured listening session. The 'baseline' sample was obtained 5 min prior to the session starting. The 'post session' sample was obtained 25 min after the start of the session. Although the cortisol response time course and magnitude depend on the type of stressor, it is generally accepted that the maximum cortisol response occurs within 15 min after the stressor in blood (Schlotz et al. 2008), followed within 2 min by maximum cortisol concentration in saliva (Schlotz et al. 2008; Kirschbaum and Hellhammer 1994, 2000). Hence, the post-session data point was expected to reflect the participant's maximum cortisol response to the structured listening activity. Testing was in the afternoon to minimise circadian rhythm effects. Participants refrained from eating and drinking for 30 min before the session and rinsed their mouth with water 3–5 min before sample collection (Groschl et al. 2001). Each child expectorated at least 3 mL of saliva into a collection tube. Samples were kept for <5 days at room temperature, which should not have led to significant steroid degradation (Groschl et al. 2001), prior to being centrifuged at 3500 rpm for 10 min and the supernatant stored at −20 °C. Melbourne Pathology Inc. conducted

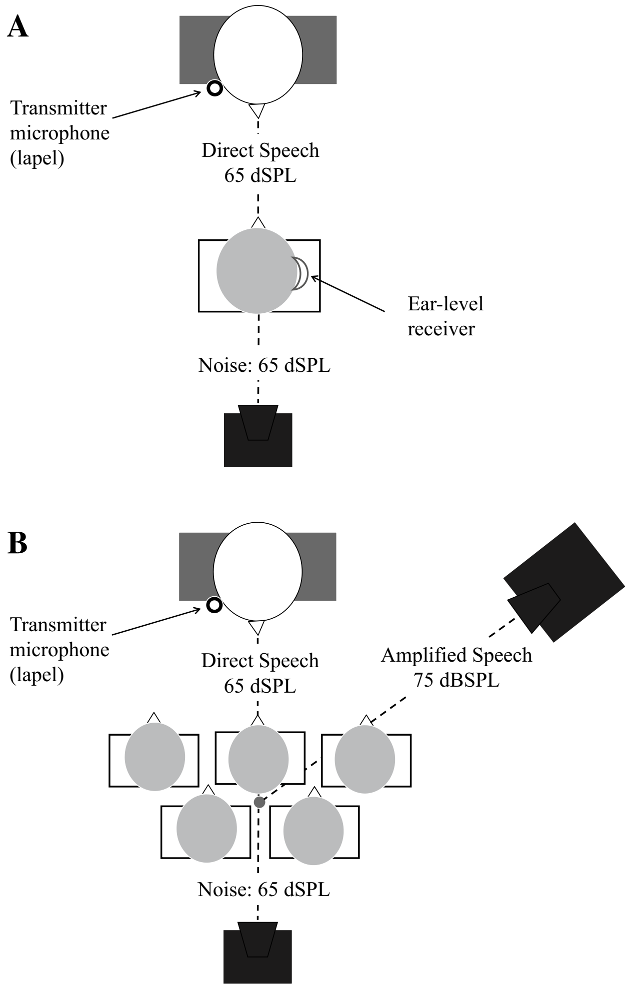

Fig. 2 Test configurations for the structured listening sessions. *Panel A* shows the set-up for the Ear-Level listening device with tester (*top*) seated in front of the single subject and loudspeaker behind. *Panel B* shows the layout for the Soundfield experiment with five participants situated between the tester (*top*) and loudspeaker behind. For the amplified-speech condition, the tester's voice was 10 dB more intense than the noise (+10 dBSNR) at a calibration point in the centre of the group

the cortisol analysis using validated procedures (Vogeser et al. 2006) with a coefficient of variation (CV) value less than 6% on repeated sample analysis. Samples were assayed for concentrations of the free, biologically active fraction of cortisol within 1 month using an electrochemiluminescence immunoassay (Cobas, Roche Diagnostics, Rotkreuz, Switzerland), following the manufacturer's instruction.

Cortisol concentrations for the first 6 participants (ASD1-6) were abnormally high, even for baseline samples (>5 nmol/L). Previous studies involving groups of children assessed mid-afternoon (with no stressor) have shown average concentration levels around 1.5–2.0 nmol/L (Hunt et al. 2007; Bess et al. 2016). Participants ASD1-6 all found the sampling procedure (spitting on-demand) extremely challenging, often requiring >5 min to produce 3 mL of saliva. Cortisol results for these subjects were not included in the analysis. Our test protocol for subsequent testing (participant ASD7-16) was modified to include expectoration training in the initial (device fitting) session. Parents were also encouraged to undertake spitting practice with their child in the week prior to the listening sessions. Adequate saliva samples for each participant who underwent the training were obtained in <3 min.

Study B: Soundfield Classroom Distribution

Each child participated in three test sessions carried out in a standard classroom. The first involved audiometric assessment carried out one-on-one in a quiet (empty) classroom and expectoration training as per Study A. Subjects then underwent two, 20 min listening sessions in a standard (otherwise unoccupied) classroom conducted at 2.00 PM on consecutive days. Ambient noise level in the classroom was <50 dBSPL. Five participants were involved per session. Listening/comprehension tasks were as per Study A and subject attention was maintained by test questions posed at regular intervals throughout the activity. The tester was positioned 2.5 m in front of the group and noise (4-Talker Babble) was presented from behind (Fig. 2b). In one listening session the tester's voice was unamplified and reached the centre of the group at 65 dBSPL. In the other, the voice was amplified by a loudspeaker (Phonak Digimaster 5000) and reached the centre of the group at 75 dBSPL. Tester voice level and the level of the background noise varied by <2 dB across the area in which participants were seated.

Saliva samples were obtained 5 min before and 25 min after the start of each listening/comprehension session. Cortisol analysis was as per Study A. One group was tested in the unamplified condition first, the other in the Soundfield-amplified condition first.

Statistical Analysis

The data were analysed by the first author using the MINTAB 17 statistics package. Paired t-tests with test result (1) speech perception score (2) Hearing Disability Rating (3) salivary cortisol concentration or (4) change in salivary cortisol concentration) as the dependent variable and listening condition (unaided or device-aided) as independent variable. Multiple regression analyses were also undertaken with test result as dependent variable and participant age, average hearing threshold level, Full Scale Intelligence Quotient (FSIQ) and CBCL anxiety score as independent variables.

Results

Study A: Ear-Level Remote Microphone Systems

Speech Perception

Nine of the 16 participants in Study A presented with abnormal speech perception in the presence of background noise. Figure 3a shows CNC-phoneme scores for each individual as a function of age compared with the expected performance range for normally developing children. Provision of the ear-level remote microphone device did, however, provide clear perceptual benefit boosting each individual's speech score to within the normal performance range. (Fig. 3b). Mean CNC-score for the aided listening condition ($M=76.5$, $SD=8.2\%$) was significantly higher than for the unaided ($M=55.5\%$, $SD=13.8\%$; $t(15)=-8.49$, $P<0.001$, $d=-2.12$). Percentage correct scores were arcsine transformed as per Studebaker (1985) to account for unequal variances. Paired T-testing for the transformed values again showed a significant difference between aided ($M=88.0\%$, $SD=13.3\%$) and unaided ($M=59.9\%$, $SD=16.9\%$) scores; $t(15)=-9.32$, $P<0.001$, $d=-2.33$. Multiple regression analysis was carried out with unaided speech score as the dependent variable and participant age, average hearing threshold level, Full Scale Intelligence Quotient (FSIQ) or CBCL anxiety score as independent variables. None of these variables were significantly correlated with unaided speech score ($P>0.05$). Similarly, there were no correlations when the analysis was repeated with aided CNC-score as dependent variable ($P>0.05$).

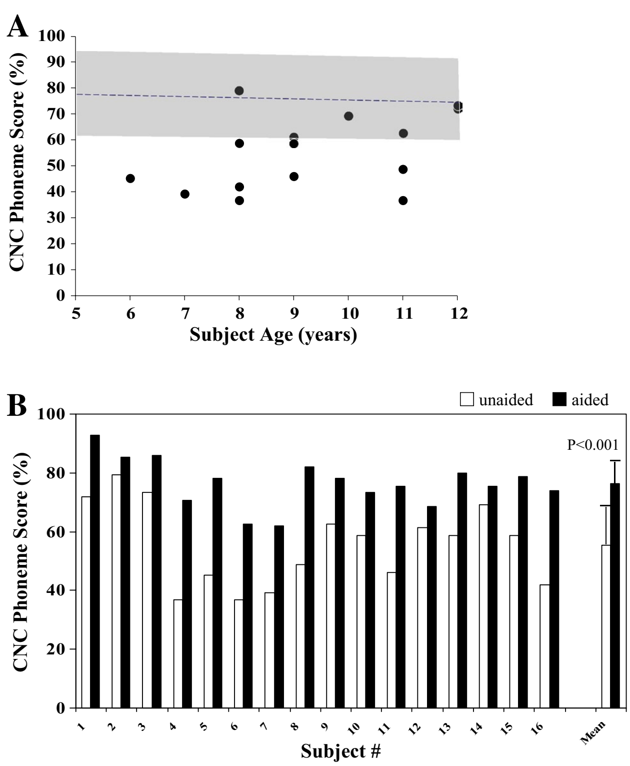

Fig. 3 **a** Open set speech perception in noise (0 dBSNR) score as a function of participant age. CNC-phoneme score is the percentage of phonemes (speech sounds) that the listener could correctly perceive and imitate in a series of monosyllabic words. The *shaded area* represents the 95% performance range for normally developing children based on published findings from Rance et al. 2010; 2012; 2014a. **b** Unaided and device-aided speech perception in noise (0 dBSNR) scores for each participant (Study A). Group scores represent the mean + 1 SD

Hearing Disability

Prior to device trial, most participants reported considerable listening difficulty in everyday listening situations. In 11 of the 16 children, APHAB responses for the "Background Noise" subscale were beyond the 95% range for normally developing children (Fig. 4a). Participants considered that use of the Ear-Level system over the 1–2 week take home trial afforded significant everyday listening and communication benefits and in each case, their perceived difficulty ratings reduced to within the normal range when wearing the device (Fig. 4b). Hearing disability ratings for the Background Noise subscale were significantly lower for the device-aided listening condition ($M=28.7\%$, $SD=11.3\%$) than for the unaided ($M=51.6\%$, $SD=20.0\%$; $t(15)=5.74$, $P<0.001$, $d=1.43$). Similarly, disability ratings for the Ease of Communication subscale were lower for the aided listening condition ($M=15.0\%$, $SD=15.7\%$) than for the unaided ($M=26.5\%$, $SD=21.3\%$; $t(15)=2.50$, $P=0.025$, $d=0.62$).

Sound aversion was common in everyday (unaided) listening with participants reporting discomfort when exposed to loud, and particularly, unexpected sounds in approximately 50% of routine listening situations ($M=52.0\%$, $SD=28.8\%$). Degree of sound aversion was unaffected by the listening device ($M=50.0$, $SD=28.6\%$); $t(15)=0.39$, $P=0.70$, $d=0.10$) reflecting the fact that, while the remote microphone system improved the level of the speaker's voice relative to the background noise, the acoustic signal was not actually amplified by the device. Multiple regression analysis showed no relation between age, hearing level, FSIQ or CBCL anxiety score and any of the APHAB subscale results in either unaided or aided listening conditions ($P>0.05$).

Parent-Reported Anxiety

Six parents completed the CBCL questionnaire after the device-trial. Two of these felt that their child's social anxiety levels were considerably lower (scores decreasing by ≥15 points) when wearing the system. CBCL results

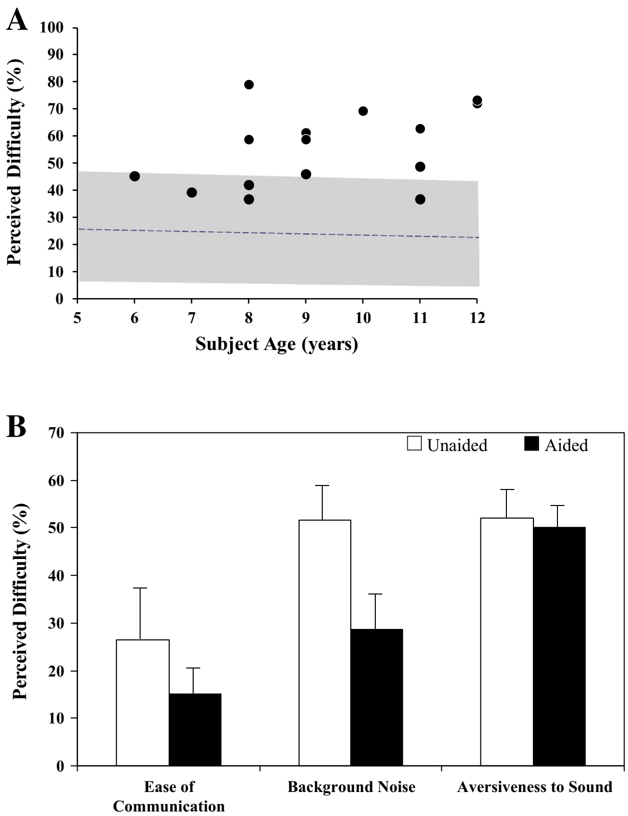

Fig. 4 **a** Hearing Disability ratings (APHAB questionnaire) for the "Listening in Background Noise" subscale. Perceived Difficulty is the proportion of everyday situations in which the participant felt they struggled with their hearing/communication. The shaded area represents the 95% performance range for normally developing children based on published findings from Rance et al. 2010, 2012, 2014a. **b** Abbreviated Profile of Hearing Aid Benefit questionnaire results for unaided and aided listening conditions. Shown are the group means for the Ease of Communication, Listening in Background Noise and Aversiveness to Sound subscales in each listening condition. *Error bars* represent 1 SD

Table 2 Child Behaviour Checklist (CBCL) scores for participants when not wearing (unaided) and wearing (aided) the ear level listening system

Subject	Gender	Age (years)	CBCL unaided (percentile)	CBCL aided (percentile)
ASD6	M	8.0	54	54
ASD11	M	9.6	93	93
ASD12	M	9.8	69	54
ASD13	F	9.2	81	<50
ASD14	F	8.3	>97	>97
ASD15	M	9.7	>97	>97

CBCL: percentile score >70 indicates clinically anxious

for the other children were identical in unaided and aided conditions (Table 2).

Cortisol Concentration

Salivary cortisol concentrations were affected by participation in the structured listening sessions and the changes varied with listening condition (unaided vs. aided). Comparison of cortisol concentration values at the baseline sample point showed no (within-child) difference between unaided and aided conditions suggesting that participant stress levels were comparable at the start of each session (unaided baseline: M = 2.72 nmol/L, SD = 0.93 nmol/L; aided baseline: M = 2.86 nmol/L, SD = 0.77 nmol/L; $t(9) = -0.91$, $P = 0.40$, $d = -0.29$). Over the course of the unaided listening session, cortisol concentration showed a non-significant increase (unaided baseline: M = 2.72 nmol/L, SD = 0.93 nmol/L; unaided post-session: M = 3.02 nmol/L, SD = 0.83 nmol/L; $t(9) = -1.69$, $P = 0.12$, $d = -0.53$). In contrast, cortisol concentration showed a non-significant decrease over the course of the aided listening session (aided baseline: (M = 2.86 nmol/L SD = 0.77 nmol/L; M = 2.71 nmol/L, SD = 0.90 nmol/L); $t(9) = 1.01$, $P = 0.34$, $d = 0.32$). As a result, comparison of within-session cortisol change for each individual revealed a significant difference between unaided (M = 0.30 nmol/L SD = 0.56 nmol/L) and aided (M = −0.18 nmol/L, SD = 0.45 nmol/L) listening conditions ($t(9) = 4.45$, $P = 0.003$, $d = 1.48$). (Table 3).

Multiple regression showed no correlation between change in cortisol concentration across the unaided and aided listening sessions and test order, participant age, hearing level, FSIQ or CBCL anxiety score ($P > 0.05$).

Cortisol Response and Functional Hearing

Cortisol responses for each child were compared with their hearing ability. For the unaided listening condition, change in concentration across the test session was significantly correlated with unaided CNC-word score, with those children showing the poorest speech perception also demonstrating the greatest cortisol increase ($r = -0.67$, $P = 0.03$). Unaided change in cortisol was not significantly correlated with self-reported hearing disability in background noise ($P = 0.88$). For the aided listening condition, cortisol change was correlated with both aided speech score ($r = -0.73$, $P = 0.02$) and aided hearing disability in Background Noise ($r = -0.68$, $P = 0.04$) with those children experiencing the best functional hearing demonstrating the greatest cortisol decrease over the course of the listening session.

Table 3 Mean cortisol concentrations (nmol/L) for children wearing the Ear-Level listening system

Listening condition	Baseline sample	Post-session Sample	Cortisol change
Device off (unaided)	2.72 ± 0.93	3.02 ± 0.83	0.30 ± 0.56
Device on (aided)	2.86 ± 0.77	2.71 ± 0.90	−0.18 ± 0.45
P-value (Paired T-test)	NS (P = 0.40)	NS (P = 0.46)	**P = 0.003**

The bold item highlight the Paired T-test analyses that showed a significant result

Study B: Soundfield Classroom Distribution

Cortisol Concentration

Mean cortisol concentrations for unamplified and Soundfield-amplified conditions were comparable at the baseline data point (unamplified session: M = 3.10 nmol/L, SD = 1.26 nmol/L; amplified session: M = 3.40 nmol/L, SD = 1.04 nmol/L; t(9) = −0.children based on published findings from Rance92, P = 0.38, d = −0.29) (Table 4). Over the course of the unamplified session, mean cortisol concentrations showed a non-significant increase (unamplified baseline: M = 3.10 nmol/L, SD = 1.26 nmol/L; unamplified post-session: M = 3.58 nmol/L, SD = 0.70 nmol/L; t(9) = −1.89, P = 0.085, d = −0.60). For the Soundfield-amplified condition, mean cortisol concentrations showed a significant decrease (amplified baseline: M = 3.40 nmol/L, SD = 1.04 nmol/L; amplified post-session: M = 2.53 nmol/L, SD = 0.66 nmol/L; t(9) = 3.00, P = 0.015, d = 0.95). Comparison of within-session cortisol change for each individual also revealed a significant difference (unamplified change: M = 0.48 nmol/L, SD = 0.88 nmol/L; amplified change: M = −0.87 nmol/L, SD = 0.81 nmol/L); t(9) = 3.64, P = 0.005, d = 1.15) (Table 4).

Multiple regression showed no correlation between cortisol change across the unamplified and amplified listening sessions and test order, age, hearing level, FSIQ or TRF anxiety score (P > 0.05).

Discussion

Stress responses in a group of school-aged children with ASD and varying degrees of auditory dysfunction were evaluated in two experiments. Participants with the poorest functional hearing ability showed the highest physiological stress levels in structured listening/comprehension sessions and also demonstrated the greatest stress reduction with the provision of auditory intervention.

Most children in this study presented with functional hearing impairment, despite not being selected based on hearing history. Eleven of the 16 participants in Study A reported a significantly higher rate of everyday listening difficulty than expected for normally developing children and 9/16 showed abnormal speech perception in noise (Rance et al. 2010, 2012, 2014a). Identification of auditory deficit (and subsequent intervention) is crucial in such cases, as the ability to understand speech and maintain concentration in background noise is a significant predictor of academic outcome in ASD populations (Ashburner et al. 2008). Interestingly, there was no correlation between average hearing levels and auditory capacity in these children suggesting that other factors, such as auditory processing deficits, were likely to have limited functional hearing ability. As such, clinicians should endeavour to assess auditory processing and functional hearing ability (in noise) when evaluating the hearing of children with the disorder.

Individuals with ASD are susceptible to high levels of stress and anxiety, particularly when confronted with social situations. These may be exacerbated by hearing impairment which can, in itself, heighten stress (Bess et al. 1998; Bess and Hornsby 2014). There are a number of ways in which the particular auditory abnormalities associated with ASD might affect stress levels. Firstly, they may increase as a consequence of a "fright" response to sudden or unpleasant sounds. "Phonophobia" or aversion to sound is a widely reported (if poorly understood) feature of ASD (Rosenhall et al. 1999). The effect of auditory hypersensitivity on the

Table 4 Mean cortisol concentrations (nmol/L) for children using Soundfield Classroom Distribution

Listening condition	Baseline sample	Post-session sample	Cortisol change
Unamplified	3.10 ± 1.26	3.58 ± 0.70	0.48 ± 0.88
Soundfield amplified	3.40 ± 1.04	2.53 ± 0.66	−0.87 ± 0.81
P-value (Paired T-test)	NS (P = 0.38)	**P < 0.001**	**P = 0.005**

The bold items highlight the Paired T-test analyses that showed a significant result stress response has not been studied, but there is evidence to suggest that cortisol reactivity to unpleasant stimuli (such as noisy or uncomfortable medical procedures) is elevated in children with ASD (Corbett et al. 2006). Phonophobic reactions were not observed in any of the children in this study, but it is noteworthy that each participant in Study A reported aversion to loud sounds in a high proportion of everyday listening situations. Secondly, exposure to everyday (non-threatening) noise is stressful and may be particularly impactful in ASD listeners with hearing difficulties for whom greater listening effort is required to perceive/understand speech. The effect of noise stressors in individuals with ASD has not been explored, but significant increases in both cortisol (Melamed and Bruhis 1996; Jahncke and Halin 2012) and urinary catecholamine concentration (Evans et al. 1998; Ghotbi et al. 2013) have been reported in normally-hearing individuals exposed to high levels of environmental noise. Thirdly, the functional hearing deficits common in ASD increase the likelihood that affected children will misunderstand what is said to them, increasing the risk of communication breakdown and social anxiety. Again, the relationship between ease of auditory communication and stress in ASD is unclear, but there is preliminary evidence suggesting that hearing deficits in non-ASD populations result in elevated "cortisol awakening responses" (Bess and Hornsby 2014; Bess et al. 2016) which may reflect chronic stress and/or anxiety about the challenges of the upcoming day (Wust et al. 2002).

In the present study, participants with the poorest speech perception in background noise found the auditory comprehension tasks most challenging and demonstrated the greatest cortisol change over the course of the structured listening sessions. This suggests that the cortisol response reflected the amount of listening effort required to complete the listening activities (Hicks and Tharpe 2002). Another possibility is that the exaggerated cortisol response in the worst listeners was associated with increased anxiety about their (poor) performance on the task. Children with ASD are reported to show reduced stress responses to socio-evaluative tasks (Corbett et al. 2012; Schupp et al. 2013), but the participants were all aware that they were being tested, and examination pressures are known to affect cortisol levels (Hellhammer et al. 1985).

The provision of Ear-Level Remote Microphones system improved listening and communication in background noise. Study participants were all afforded significant speech perception benefits on formal assessment and reported fewer hearing/communication difficulties when wearing the device during the "take-home" trial period. These results are consistent with recently published findings (Rance 2014b; Schafer et al. 2014, 2016) which have revealed significant perceptual advantages, social interaction improvements and long-term device acceptance–at least in high-functioning pre-adolescent children with ASD. Remote-microphone listening systems improve figure/ground perception by increasing the level of speaker's voice relative to that of the background noise. Conventional hearing aids, in contrast, amplify the sound (both speech and noise) recorded at the listener's ear. This amplification is beneficial for individuals with sensory hearing loss who can't hear low-level sounds, but may not be helpful for listeners with temporal processing deficit who may simply experience a louder, but equally distorted, auditory percept (Rance et al. 2002). Remote-microphone systems work not by correcting inherent auditory pathway limitations, but by improving the quality of the incoming auditory signal.

Use of Ear-Level Remote Microphone systems also reduced listening-related stress. Participants showed significantly reduced cortisol responses over the course of a challenging listening session when aided. Where children tended to show an increase in cortisol concentration in the unaided condition (reflecting the difficulty they had completing the tasks), they typically showed a decrease when the device afforded better access to the tester's voice. Furthermore, the degree of cortisol reduction in the device condition was correlated with aided hearing ability, suggesting that the children were relatively anxious at the start of the session (as they knew they were being tested) and then relaxed if they found the listening/comprehension tasks easier than expected. It is difficult to know precisely which aspects of the interaction improved with the intervention and were responsible for the cortisol changes. The hearing devices, by enhancing the auditory signal, may also have improved the child's ability to focus, reduced distractibility and/or resulted in better joint attention.

Unfortunately, Ear-Level devices are not a viable management option for all school-aged children with ASD. Maintaining consistent device use in teenagers (with or without ASD) is challenging, as adolescents typically do not wish their hearing difficulties to be conspicuous (Rekkedal 2012). In our previous study for example, where 80% of the 8–12 year old participant group were enthusiastic device users at the end of a 6 week trial, none of the teenagers were prepared to wear the system at school for more than a few days (Rance et al. 2014a). Tactile defensiveness is another potential barrier to device use in some ASD cases. The receiver earpiece, whilst light and unobtrusive, can be intolerable for some children with tactile hypersensitivity (Schafer et al. 2013; Rance et al. 2014a).

Soundfield Classroom Distribution is an option that offers improved listening in the schoolroom without the need for individual students to wear a device. In this study we found that amplification of the teacher's voice could reduce listening stress in children with ASD. Participation in group auditory comprehension activities tended to produce cortisol increases when the teacher's voice was unamplified, whereas cortisol concentrations dropped significantly in the amplified condition, presumably because the children found the listening tasks easy and their initial anxieties abated.

Limitations and Recommendations

This study represents the first attempt to measure the potential ameliorating effects of auditory interventions on stress levels in children with ASD. There are several limitations that can be addressed in future research. First, while effect sizes for our objective stress measure (cortisol concentration) were large in both Study A and Study B, each trial involved a relatively small number of participants (N = 10). Furthermore, while subjective anxiety measures (CBCL/TRF) were obtained prior to the study for each of the participants, only six families in Study A completed the questionnaire post device-trial. All reported either no change, or considerable decreases in social anxiety when wearing the Ear-Level system, but the small response numbers mean that this result should be interpreted with caution. Second, we measured cortisol concentrations in saliva which tends to reflect stressors occurring over a relatively short time period (<20 min). Other sampling techniques, for example extracting cortisol from urine or hair, may be useful in tracking the effects of auditory interventions on chronic stress levels over a longer time course (Wosu et al. 2013). Third, while the participants in this study were all diagnosed with ASD by experienced clinicians, the children were assessed by different clinicians employing a variety of instruments. We think it unlikely that this would have affected participant selection, but a standardized diagnostic approach would have been preferable. Fourth, the psychophysical and subjective test methodologies involved in the study required that only “high functioning” children participate. While it is likely that youngsters with ASD and coexisting disabilities also suffer auditory deficits, the applicability of remote microphone listening systems, particularly those involving a head-worn device in such cases is uncertain.

Conclusions and Implications

Communication difficulties are a core feature of ASD and major source of anxiety for affected individuals. The findings of this study confirm that a high proportion of school-aged children with the disorder suffer functional hearing deficits severe enough to impede communication. Furthermore, the results demonstrate that auditory interventions can improve speech perception in everyday (noisy) listening conditions and, in so doing, can make social interaction easier. The findings also suggest that remote microphone auditory systems can reduce listening-related stress in both one-on-one and group-listening contexts. Minimisation of stress and anxiety is important for the wellbeing of affected children. Although the stress response is adaptive for short-term reaction to environmental stressors, frequent or prolonged activation of the HPA axis can have harmful effects including neuroendocrine dysregulation (McEwan 1998) and immune system suppression (Munck and Guyre 1991; Derijk and Sternberg 1994). As such, auditory interventions that can minimise stress may be important for the health of children with ASD and should be considered for affected youngsters in the school setting.

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Author contributions GR conceptualized and designed the study, drafted the initial manuscript and approved the final manuscript as submitted. DC coordinated and supervised data collection, critically reviewed the manuscript and approved the final manuscript as submitted. KS was involved in study design, subject recruitment/data collection, reviewed/revised the manuscript and approved the final manuscript as submitted. JLR was involved in study design and critically reviewed the manuscript and approved the final manuscript as submitted.

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Compliance with Ethical Standards

Conflict of interest Gary Rance has received research Grants from Phonak Org. Donella Chisari, Kerryn Saunders and Jean-Loup Rault declare that they have no conflict of interest.

Ethical Approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed Consent Informed consent was obtained from all individual participants included in the study. This article does not contain any studies with animals performed by any of the authors.

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