

Increased perception of loudness in autism

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Abstract

Clinical reports on autism describe abnormal responses to auditory stimuli such as intolerance to sounds. The present study assessed subjective perception of loudness in subjects with autism compared to healthy controls, using two psychoacoustic tests. First, the auditory dynamic range was evaluated at six different tone frequencies. Secondly, loudness growth as a function of the intensity level of a 1 kHz tone was estimated. Verbal responses from a group of 11 children and adolescents with autism were compared to responses of 11 age- and gender- matched healthy controls. Smaller auditory dynamic ranges were found in the autistic group than in the control group, as well as increased perception of loudness, indicating hyperacusis in subjects with autism.

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1. Introduction

First described by Kanner (1943), autism (or autistic spectrum disorder) is a severe developmental disorder characterized by marked impairment in social interaction and communication, and restricted, repetitive and stereotyped behavioural patterns (American Psychiatric Association, 1994). Atypical reactions to the sensory environment are often reported in people with autism. This is particularly evident with respect to the auditory

modality (Hermelin and O'Connor, 1970; Ornitz, 1974; Dahlgren and Gillberg, 1989; Rosenhall et al., 1999).

People with autism (AUT) are often thought to be deaf due to their unresponsiveness to auditory stimulation e.g. even when they are addressed in a loud voice (Lockyer and Rutter, 1969). However, although the prevalence of severe and profound hearing loss in autism is higher than in the general population (3.5% versus 0.1–0.2%) (Rosenhall et al., 1999), relatively very few children with AUT, even those affected by rubella during foetal development, suffer from a severe hearing loss (Chess, 1977). On the contrary, many children with AUT are very sensitive to noise. They may express panic (flight, crying, etc.), when they hear loud sounds such as a vacuum cleaner or washing machine, and they often avoid acoustic stimulation by covering their ears (Frith and Baron-Cohen, 1987).

Abbreviations: ADR: Auditory Dynamic Range; AUT: autism; CARS: Childhood Rating Scale; CLS: Categorical Loudness Scaling; CONT: Control; IQ: Intellectual Quotient; LDL: Loudness Discomfort Level; QT: Quiet Threshold

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Particular behavioural responses to auditory stimuli such as auditory indifference, and/or hypersensitivity (intolerance to sound) have led researchers to explore hearing function in AUT. Auditory processing has been studied at the cortical level in AUT using electrophysiological methods. Late cortical auditory evoked potentials of normal children mainly show prominent bilateral deflections in temporal regions culminating at around 160 ms; these have been found to be of smaller amplitude in children with AUT compared to controls (CONT) (Bruneau et al., 1999). Such bilateral hyporeactivity could be related to bitemporal hypoperfusion found in associative auditory areas, as reported in recent PET neuroimaging (Zilbovicius et al., 2000; Ohnishi et al., 2000; Boddaert et al., 2001).

Abnormal brain processing of sound intensity could be the origin of such inappropriate reactions to sounds observed in AUT, since intensity (i.e., energy of a sound wave) is strongly associated with the subjective feeling of annoyance and noisiness of an acoustic stimulus (Berglund et al., 1976).

The aim of this study was to verify the hypothesis of abnormal loudness perception, i.e., subjective judgment of intensity, in children and adolescents with and without AUT, and to quantify it by using two psychoacoustic tests.

Previous studies on hyperacusis in tinnitus patients have used measures of loudness discomfort levels (i.e., intensity perceived as very uncomfortable) (LDL), and of auditory dynamic range (ADR) which is the range between absolute or quiet threshold (QT) and LDL. LDL proved to be negatively correlated to hyperacusis (Goldstein and Shulman, 1991). This test thus appeared appropriate to assess loudness in AUT. However, the lower LDL and ADR of hyperacusis patients is sometimes also observed in non-hyperacusis subjects.

Given the lack of sensitivity of this test, we therefore chose to complete our psychoacoustic evaluation of the subjects with a second test, categorical loudness scaling (CLS), which is usually used to assess loudness growth in normal and hearing-impaired individuals (Allen et al., 1990). This is a measure of the subjective intensity of a sound. In this test, which can be performed both in adults and in children (Ellis and Wynne, 1999), pure tones are presented at a range of frequencies and rated for loudness by category. The categorical rating of loudness allows assessment of how the person with AUT perceives the loudness of auditory tones in his ADR.

If indeed the children with AUT are hyperacusis, as suggested by their behavioural reactions to environmental sounds, we might expect that not only would their ADRs be lower than those of normal subjects, but that their CLS would reflect an enhanced perception of sound intensity.

2. Materials and methods

2.1. Subjects

About 11 children and adolescents with primary autistic spectrum disorder (nine males) were included in this study. They were recruited from patients attending a child psychiatry day-care unit of a University Hospital and diagnosed according to DSM-IV criteria (American Psychiatric Association, 1994) by an expert clinician. Autistic behaviour was evaluated by the Childhood Autistic Rating Scale (CARS) (Schopler et al., 1980). The diagnosis of autism was confirmed by the CARS score which was over 30 for all subjects. Their verbal and non-verbal intellectual abilities were assessed using WISC-R (Wechsler, 1981a) and WAIS-R (Wechsler, 1981b). One had no mental retardation (IQ = 117), five had mild mental retardation (IQ between 50 and 70), and five had severe mental retardation (IQ from 41 to 48). According to parents' and clinicians' reports, all the young patients were intolerant of noise when younger.

All subjects had normal hearing and were free of psychotropic medication for at least one month before the study. Children with metabolic or chromosomal disease and/or history of significant neurological disorders were excluded. The inclusion criterion was the subjects' ability to use the set of verbal responses. A few schematic examples were presented before the experimental session to verify that subjects with AUT and the CONT subjects understood the instructions and were able to provide relevant verbal responses. Moreover, the children with AUT were pre-selected by the clinicians in charge of them according to the cognitive demand of the experimental tasks. In addition, subjects were required to demonstrate stable pure tone absolute thresholds (i.e., not more than five dB between the two absolute thresholds measured for each frequency).

In order to ensure the validity of CLS, subjects who did not give reproducible evaluations (i.e., varying in no more than 5 dB) for each set tone intensity were eliminated. About 11 (nine males and two females) of 31 patients tested, met all our criteria and had good test-retest reliability for ADR and 10 (eight males and two females) for CLS. The CLS group was included in the ADR group. Their age range was from 9 to 17 years. The 11 subjects with AUT of the ADR test (mean age: 14.6 ± 0.6) had mean verbal IQ of 56.3 ± 6.6 and non verbal IQ of 64.0 ± 7.0 . The children with AUT in the CLS test (mean age: 14.9 ± 0.7) had mean verbal IQ of 51.8 ± 6.6 and non verbal IQ of 65.9 ± 8.2 . Means age of the CONT group in the ADR test was 14.5 ± 0.6 and 14.8 ± 0.6 in the CLS test.

Each subject with AUT was paired according to chronological age (± 1 year) and gender with a CONT subject drawn from children and adolescents in the nor-

mal school population. All had normal hearing and no history of psychological or educational problems.

The session involved two non-invasive psychoacoustic tests which were administered in a quiet room, with the same portable device, in three different hospital child psychiatry units. To avoid any discomfort, subjects were asked to stop the experiment if the pure tones became too painful, and the child or parent had the right to terminate the session at any moment.

The Ethics Committee (CCPPRB) of Lyon Berard Hospital approved the protocol which complies with APA ethical standards. The testing session took place after signed informed consent had been given by the parents, and assent by the children or adolescents.

2.2. Procedures and stimuli

Sounds were delivered monaurally by the Diagnostic Audiometer AD 28 through TDH 39 earphones, and the first ear studied was randomly chosen (right or left) for each test. All pure tones were of 500 ms duration with onset/offset ramps of 50 ms. All stimuli presentation were performed in one session, and were initiated by the experimenter; stimuli characteristics were otherwise controlled by audiometer. The verbal responses to the two tests had to be given in the first 30 s or so following the stimuli; otherwise, the stimulus was repeated.

2.2.1. Auditory dynamic range

The ADR is the difference between the absolute pure tone threshold (or QT and the LDL; QT is the minimum detectable level of a sound in the absence of any external sounds and LDL corresponds to the intensity at which the sound was considered as uncomfortable by the listener (Goldstein and Shulman, 1996). Tonal audiometry was performed to measure pure tone thresholds expressed in dB HL at various frequencies in the following order: 0.25, 0.5, 1, 2, 4 and 8 kHz. To measure the quiet auditory thresholds, pure tone intensity was decreased until the subject could no longer perceive it. Subjects said “yes” when they could hear the tone and “no” when they were unable to perceive it. The tone level was initially set at 30 dB HL and decreased in 5-dB steps until the subject reported verbally they could no longer hear the tone; it was then increased in 5-dB steps until the subject started to hear the tone again. The lowest intensity at which the tone is perceived corresponds to the absolute threshold. The procedure was repeated a second time to verify the validity of the measurement. If the second threshold obtained differed from the first one, the procedure was repeated until two consecutive QT were identical. However, in the two populations, it was never necessary to repeat the procedure such as to find a third QT value. Therefore, no bias was introduced by spurious responses. After measuring pure tone thresholds, LDLs were obtained for each ear individually and for each frequency.

The children were instructed to indicate verbally or by raising their hands when the sound level became too loud. LDL judgements were made on the basis of presenting stimuli increasing in 5-dB steps from 45 dB HL.

2.2.2. Categorical Loudness Scaling

The CLS used was derived from the loudness growth per 1/2-octave-band test (Allen et al., 1990). Only four ratings and one pure-tone frequency (1 kHz) were tested in the present study instead of six ratings and six frequencies as in Allen’s study; this simplification was made in order to facilitate the assessment of subjects with AUT and their CONT.

The first phase of the CLS is a replication of the ADR measurement at 1 kHz. Indeed, in this phase, the highest (corresponding to LDL, scored 4) and lowest boundaries (scored 0 and corresponding to the QT) of the intensity used for testing are determined using the same instructions as for ADR.

The second phase consisted of random presentations of the 1 kHz tone at various intensities in multiples of 5-dB, ranging from the QT to the LDL determined for each subject in the first phase. When the subject heard the sound, he or she had to verbally rate the tone loudness using a numeric scale where 1 was ‘low’; 2 was ‘medium’; 3 was ‘loud’; and 4 was ‘too loud’. For the subjects with AUT with the lower IQ (five subjects), the verbal correspondence instead of the numbers could be used. The ability to associate the sound loudness to the number and/or the verbal label was verified in a pre-session test with acoustic examples. Each intensity was presented randomly 3 times in total, except when the subject responded ‘too loud’. There were only two presentations of sounds rated ‘too loud’, each followed by a 20-s silent pause. The corresponding averaged intensity (in dB HL) was calculated for each loudness rating to provide a loudness growth curve. The intensity corresponding to the ‘too loud’ sound was only averaged on two intensities.

3. Results

3.1. QT, LDL and ADR

Pure tone thresholds did not display any significant intergroup differences, regardless of the ear stimulated or the frequency tested (see Fig. 1). Since QTs were all lower than 20 dB HL and comparable to clinical norms, the children tested were shown to have no hearing loss and no auditory suprathresholds.

As illustrated by Fig. 1, LDLs were significantly lower in the AUT than in the CONT group ($F(1,20) = 11.1$; $p < 0.005$). Loudness discomfort thresholds used for ADR calculation were lower than 80 dB HL in 63% of the autistic subjects compared with 27% of the CONT

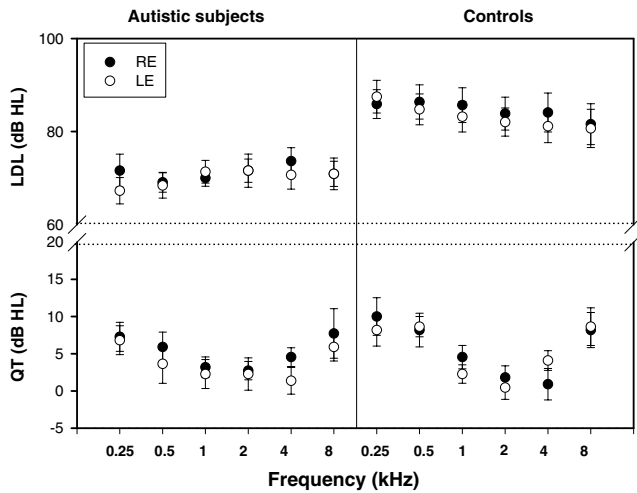


Fig. 1. Means and standard error bars of QT and LDL obtained in both ears of autistic and control subjects for six tones at standard audiometric frequencies.

subjects, regardless of frequency or ear tested. Since the ADR result was obtained for each subject by subtracting the QT value from the LDL value, a significant intergroup difference was therefore found ($F(1,20) = 7.69$, $p < 0.02$) due to lower LDL values in the AUT than in the CONT group (Fig. 1). There was also a main effect of frequency ($F(5,100) = 7.1$, $p < 0.0005$). As shown on Fig. 2, ADR values increased from 0.25 to 4 kHz (significant between 0.5 and 1 kHz, $F(1,20) = 10.2$, $p < 0.004$) and then decreased from 4 to 8 kHz ($F(1,20) = 16.6$, $p < 0.0006$) in both groups. There was no significant effect of whether right or left ear was tested, nor any significant interactions between the different factors

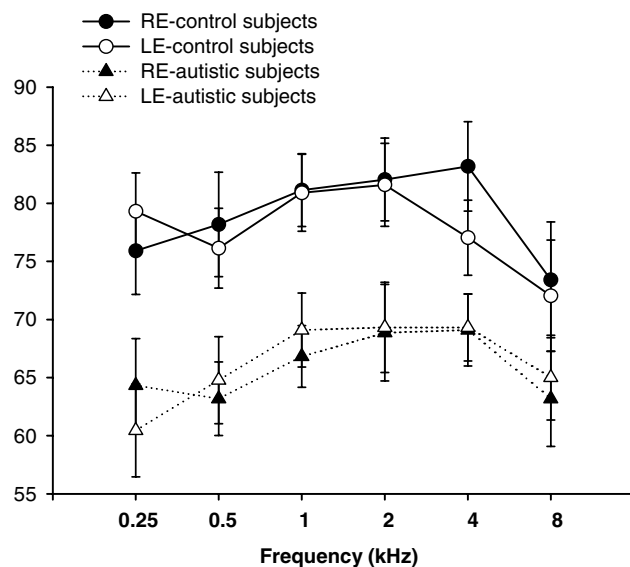


Fig. 2. Means and standard error bars of ADR obtained in both ears of autistic and control subjects for six tones at standard audiometric frequencies.

considered. Tests of homogeneity of variance showed no difference between the two groups.

Furthermore, there was no relationship between ADR results and IQ values in the AUT group as there was a lack of significant Spearman correlation between IQ (verbal, non-verbal or overall) and ADRs for the six frequencies and the 2 ears (all $r^2 < 0.4$, all $p > 0.12$, $N = 11$). In addition, when five autistic subjects with $IQ \geq 50$ were compared with six children with AUT with $IQ \geq 50$, no difference in ADR was observed according to IQ value for ear or frequency with the Mann–Whitney test (all $U \geq 5$, all $p > 0.09$).

3.2. CLS

Loudness growth curves are presented in Fig. 3 for the two groups of subjects and each ear separately. Except for the threshold, which was at the same intensity level for all subjects and both ears, the other four loudness categories were assigned to lower intensity levels in children with AUT compared to CONT children, regardless of the ear stimulated: 21 ± 1 versus 28 ± 1 dB (mean values \pm SEM for left and right ear) for ‘low’ ($F(1,18) = 7.84$; $p < 0.02$), 43 ± 2 versus 61 ± 2 dB for ‘medium’ ($F(1,18) = 20.9$; $p < 0.0005$), 58 ± 2 versus 76 ± 2 dB for ‘loud’ ($F(1,18) = 32.3$; $p < 0.0001$) and 68 ± 2 versus 85 ± 2 dB for ‘too loud’ ($F(1,18) = 19.3$; $p < 0.0005$). Tests for homogeneity of variance did not demonstrate any difference between the two groups.

As above, no significant Spearman correlation was found between IQ values and intensities for the five loudness categories and the two ears in children with AUT (all $r^2 < 0.1$, all $p > 0.9$, $N = 10$). Moreover, when two groups of five autistic subjects with $IQ < 50$ and $IQ \geq 50$ were compared, no difference in CLS values

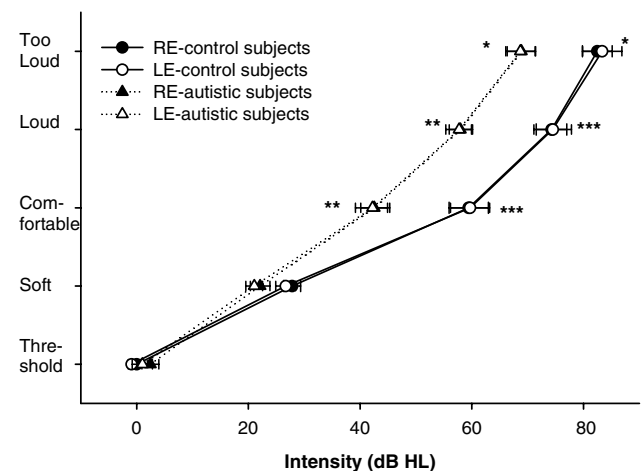


Fig. 3. Means and standard error bars of the 1-kHz pure tone intensity (dB HL) for each loudness category in both right (RE) and left (LE) ears of autistic and control subjects. These average values constitute the loudness growth curves. * $p < 0.05$; ** $p < 0.0005$; *** $p < 0.0001$.

was observed according to IQ for each ear or rating with the Mann–Whitney test (all $U \geq 7$, all $p > 0.3$, $N = 5$).

4. Discussion

4.1. Loudness enhancement in autism

The two psychoacoustic tests used confirmed the hypothesis of an enhanced perception and reduced tolerance of loudness in children with AUT. The ADR results showed that the children with AUT had a restricted dynamic range of perception, although absolute thresholds were normal (less than 20 dB HL). This reduced ADR was due to lower LDLs in the AUT group than in the CONT group. These findings were in accordance with the results of the CLS test since children with AUT rated the 1-kHz pure tone as “loud” at much lower intensities than did CONT children.

Because loudness is a fundamental factor in sound annoyance (Berglund et al., 1976), and because the subjects with AUT in our study perceived pure tones as being relatively louder compared to CONTs, patients may be much more prone than healthy subjects to be irritated by everyday sounds. Furthermore, this auditory hypersensitivity (i.e., higher rating) occurred not only with loud sounds but also with sounds considered to be of moderate intensity. The shift in the loudness growth curves (see Fig. 3) of the AUT group compared to CONT group was about 20 dB for intensities greater than 40 dB HL. This indicates a considerable difference in loudness perception between the two groups. To give a concrete idea of the meaning of such intensities, quiet conversation corresponds to an intensity of about 50 dB SPL (sound pressure level), and a soft whisper to 30 dB SPL (Moore, 1997). Note that for weak sounds (softer than 40 dB HL), the loudness ratings were similar. QTs for all the six frequencies tested were normal and did not differ according to the group.

These results may have therapeutic implications. The auditory integration therapy developed by (Berard, 1993) aims to modify QTs at frequencies showing the lowest thresholds on the audiogram; this has been used in AUT but the results have been inconclusive (Dawson and Watling, 2000). Our experiment suggests that attempts to improve auditory perception in AUT should rather focus on LDL.

Hyperacusis in AUT has also previously been reported by Rosenhall et al. (1999). They showed that 18% of their autistic children and adolescents did not tolerate a click level of 80 dB HL compared to 0% in the CONT group. In the present study, pure tones were used instead of clicks, and 63% of the children with AUT had a LDL below 80 dB HL compared to 27% of CONTs. Differences in patient selection and methods complicate comparisons between the present study and

Rosenhall et al. (1999). However, in both cases, there is evidence of hyperacusis for people with AUT. It is not clear whether hyperacusis in general originates from a central or peripheral (middle ear or cochlea) mechanism (Brandy and Lynn, 1995). Abnormalities in auditory processing have been demonstrated at the cortical level in autism, thus suggesting that hyperacusis could be of central origin.

The ADR results obtained in this study differ somewhat from those reported by Goldstein and Shulman (1996) for hyperacusis adults with tinnitus. They found an average dynamic range of 55 dB or less at any frequency in the majority of hyperacusis patients, whereas in our children with AUT, this was 65–70 dB. It may be that the mechanisms underlying hyperacusis in autism are different from those involved in tinnitus, or that hyperacusis is more pronounced in patients with tinnitus than with AUT, and more pronounced in adults than in children or adolescents. However, hyperacusis in autism may have more detrimental consequences, for example in psychosocial functioning, than in patients with tinnitus.

Despite the concordance of our results with those reported in the literature, they must however be considered with caution. One potential confounding factor is that instead of having an auditory abnormality, the patients with AUT might also demonstrate psychological bias towards responding in a particular way as compared to CONTs in the tests.

4.2. Reliability of results

From a methodological point of view, we might question whether the children with AUT were able to perform the psychoacoustic tests correctly. However, because of our strict inclusion criteria and our observation of the children’s behaviour during the tests, it is clear that these children did not have difficulties in giving a categorical loudness rating, and that their responses were consistent (i.e., reproducible). Moreover, although loudness growth curves of the AUT group (Fig. 3) were skewed to the left, the curves were the same as for the CONT group, with the same inter-individual variability, and the results for both ears were almost identical. The homogeneity of variance between the two groups of children also supports reliable response by the AUT group.

Another important point is the lack of relationship between the psychoacoustic measurements and IQ, suggesting the independence between loudness perception and IQ values. Within the AUT group, the same ADR results were observed for subjects regardless of their IQ values i.e., no correlation was observed between IQ values and psychoacoustic results, and no difference was observed between these measurements according to IQ. However, lower IQ of children with AUT as compared to CONT may contribute to the differences observed between results of the two groups.

Finally, given that only some of the children with AUT were able to perform the two psychoacoustical tests, caution must be exercised in generalising our findings to all patients with AUT, even if this seems likely to be a true result.

5. Conclusions

The CLS and ADR tests enabled us to demonstrate and quantify for the first time an increased loudness perception of pure tone intensity in children and adolescents with AUT, demonstrating the existence of hyperacusis in autism. This disordered loudness processing could be related both to the inappropriate behaviour and electrophysiological abnormalities in response to acoustic stimuli previously observed in autism. Such subjective measurements could be used in parallel with objective measurements of auditory functioning in order to provide better understanding of the underlying mechanisms of hyperacusis in autism.

Our study suggests that loudness perception should be particularly taken into account in autism, perhaps by adapting the acoustic environment where appropriate, to potentially improve quality of life and learning.

Future work in this area could shed further light on the importance of this phenomenon in the wider group of autistic spectrum disorder and understanding of the physiological mechanisms involved, as well as exploring the potential therapeutic applications of this finding.

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