Listening effort and downstream effects due to hearing loss in children and young people: an online quantitative questionnaire-based observational study

Callum Andrew Shields, Mark Sladen, Azita Rajai, Hannah Guest, Iain Bruce, Karolina Kluk, Jaya Nichani

INTRODUCTION
Listening effort (LE) remains an elusive concept and, as such, is yet to be integrated as an outcome within clinical audiology. Perhaps the most contemporary and inclusive definition of LE concept stems from the Framework of Understanding Effortful Listening.

The deliberate allocation of mental resources to overcome obstacles in goal pursuit when carrying out a listening-based task.

The importance of deepening our insight into LE can be illustrated by considering the possible unmet burden experienced within hearing-impaired populations. The impact conditions, such as mild–moderate and unilateral hearing loss, have on individuals has consistently been under-reported. However, recent studies have documented the negative educational, professional and behavioural ramifications of mild hearing loss, especially among children and young people (CYP). This observation is particularly concerning given that hearing loss affects around 11.3% of the general school population. This lack of awareness may have contributed to a paradox whereby, in certain circumstances, children with severe/profound hearing loss perform better educationally than those with mild hearing loss. Moreover, a clinically acceptable measure of LE may also allow us to discern the real-life benefit of particular

STRENGTHS AND LIMITATIONS OF THIS STUDY
⇒ Records data at two time points to allow within-subject comparison.
⇒ Relatively large sample size (n=100).
⇒ Entirely remote to avoid unnecessary expense/time commitments for the participants.
⇒ Technology bias—this study is limited to the recruitment of participants with access to an internet-enabled device.
⇒ Those with severe/profound hearing loss may not be able to complete the Digits in Noise test unaided.
interventions, for example, bilateral cochlear implantation, which may be missed through over-reliance on traditional audiological outcomes.6

One of the significant challenges to the clinical application of LE is the lack of consensus on measuring the abstract notion. Many instruments have been postulated, ranging from self-reported measures to physiological markers.7 Nevertheless, despite the myriad of proposed tools, the evidence to support one optimum measure is nominal.1

This issue is further complicated because correlational analysis between different measures often shows conditional and weak relationships supporting the idea that each tool captures a different component of LE.9 This finding was corroborated in a recent systematic review within our department, which is in press with Trends in Hearing.

An additional challenge pertains to our understanding of cognitive processes involved with effort perception. The interplay between working memory, processing speed, attention, motivation, task difficulty, arousal and cognitive capacity is yet to be fully mapped.1 Although these theories have been developed since Kahneman first posited effort=attention, these mechanistic deficiencies hinder our ability to explain the inconsistencies noted within the literature.10 11

With the potential benefit from broadening our application of LE to clinical practice harshly stifled by these barriers, it may be more prudent to consider a different approach to LE. One such way would be to focus on its corollaries rather than the intrinsic concept. The association between fatigue and LE has been well documented in the literature.8 12–14 However, research remains in its infancy regarding other possible consequences of prolonged exposure to effortful listening. Manifestations in the form of stress, low self-esteem, low motivation and low mood are yet to be fully explored. If an association can be shown, then there is a potential to use pre-existing and prevalidated instruments for these downstream consequences as a clinically informative marker for LE.

Study aim
This study explores the use of existing questionnaire-based outcome measurement instruments in evaluating LE and its associated consequences in CYP with different hearing abilities.

Primary objective
The primary objective of this study is to explore the relationship between LE and measures of fatigue, stress and confidence in CYP and the impact of hearing loss using existing questionnaires.

Secondary objective
The secondary objective of this study is to explore the relationship between LE and fatigue, stress and confidence measures at different time points (working day vs rest day) in CYP with normal hearing and hearing loss using existing questionnaires.

METHODS AND ANALYSIS
This is an exploratory study of the relationship between LE and its downstream effects on fatigue, stress and confidence in a range of CYP with normal hearing and varying degrees of hearing loss; therefore, there will be no initial sampling. This study will take the form of a quantitative questionnaire-based design. The proposed timeline for data collection is 3 January 2023–31 March 2023. The questionnaires will be delivered entirely online using the secure platform REDCap.15 16 CYP with normal hearing and a range hearing loss level will be recruited (n=min 100). Once 50 responses have been collected, the distribution of hearing loss, age and gender of the participants will be evaluated. If there is a bias towards either normal hearing or hearing loss or a cluster of ages and gender, efforts will be made to purposively sample to optimise recruitment with an equal distribution of normal hearing and hearing loss, age and gender.

Participants will be recruited through three avenues:

- Subjects will be selected from our database (Audit base) of paediatric patients who have undergone cochlear implant and/or hearing aid fitting at the Royal Manchester Children’s Hospital (RMCH). This will be done by a clinical audiologist who is normally involved with the care of this subgroup of patients. Potential participants will be sent an email invitation to sign up for the study. This will be via a link to the eligibility questionnaire on REDCap. This will be the source for the hearing-impaired participants.

- An invitation link will also be sent to hearing charities/social media groups for them to share. This will link to the eligibility questionnaire on REDCap. This will rely on the participants self-referring to the study as they will not be directly contacted via this avenue.

- Recruitment will also take place in the Fracture clinic within the outpatient department (OP) of RMCH with posters. Fracture clinic has been specifically chosen as this population is less likely to have chronic health conditions, which may confound the questionnaire results. The participants will self-refer by following the link on the posters to the eligibility questionnaire on REDCap. Periodically, study team members will be in the waiting room of the fracture clinic to highlight the chance to participate in the study to patients before they are seen in the clinic. We will not use any database to target patients within this cohort specifically.

Once an individual has declared interest in participating in the study, they will follow a link to the eligibility questionnaire on REDCap. This process will ensure they meet the criteria set out in table 1. This may be completed by the participants themselves or with help from their parents if required. If they are not eligible for the study, REDCap will inform them, and no details from their questionnaire will be recorded.
The relevant participant information sheet (PIS) will be available via the eligibility link before confirming interest. Those eligible for the study who register their interest via the initial link will then be emailed a copy of the PIS immediately after registration. The participant will then receive a link to the consent process. Participants under the age of 16 will require parental consent in order to take part in the study. This will be documented via an informed consent form which must be electronically signed by the parent. The participant must also complete an assent form. Participants over the age of 16, will provide consent for themselves. This will be documented via an informed consent form which must be electronically signed by the participant. A copy of the signed consent/assent form will be stored on the secure RedCAP repository. Additionally, a copy will be emailed to the participant.

Following eligibility screening and consent gathering, participants will be invited to complete an online battery of questionnaires. This battery consists of four separate sessions, with a total completion time of approximately 1 hour 30 min over 2 weeks. Participants will automatically be sent the links to access each session via email. Once the participant has completed one session, they will be emailed the link to the next session appropriately. The participant can complete the questionnaire any time after receiving the link; it does not need to be completed on the day they receive the link. If the participant does not complete the session, reminder emails will be automatically sent. The sessions are outlined as follows:

- **Participant demographic information.** Basic information relating to age, sex and hearing device use will be collected (see table 2 for the full list of questions). Perceived hearing ability will also be measured using the Speech, Spatial and Qualities of Hearing Scale (SSQ) questionnaire, first created by Gatehouse and Noble and then later validated in children 11 years and over by Galvin and Noble.17 18 This will be sent to the participant immediately following the completion of the informed consent form. This session will take approximately 10 min to complete.

- **Digits in Noise (DIN).** Next, participants will be asked to complete a DIN hearing test (see further for full details). This test will be done using the University of Manchester Software MOSS. The DIN may be done on the same day as the demographic questionnaire or on a different day if the participant wishes. The DIN will allow us to establish an objective spectrum of hearing abilities across the participants. This will be sent to the participant immediately following the completion of the participant demographics questionnaire. The test will take approximately 20 min to complete.

- **Rest Day Questionnaire session.** The participant will be asked to complete a series of four validated questionnaires (see further for full details). These questionnaires aim to measure LE, fatigue, stress, and confidence. The participant will be instructed to complete this session at the end of a rest day when they have not been to school/college/work. The data and time of when the participant completed this session will be recorded. This will be sent to the participant at 18:00 on the next weekend following completion of the participant demographics questionnaire. This session will take approximately 30 min to complete.

- **Workday Questionnaire session.** Finally, the participant will be asked to complete the aforementioned outcome questionnaires again. The participant will be instructed to complete this session at the end of a day when they have been in school/college/work. The data and time of when the participant completed this session will be recorded. This will be sent to the participant at 18:00 on the next weekday following completion of the rest day session. This session will take approximately 30 min to complete.

### Table 1: Inclusion and Exclusion Screening Criteria

<table>
<thead>
<tr>
<th>Inclusion criteria</th>
<th>Exclusion criteria</th>
</tr>
</thead>
<tbody>
<tr>
<td>▶ Aged 12–17 years old.</td>
<td>▶ Aged younger than 12 years old or older than 17 years old.</td>
</tr>
<tr>
<td>▶ Normal hearing or hearing loss of any aetiology.</td>
<td>▶ Presence of any other comorbidity or chronic health condition associated with fatigue.</td>
</tr>
<tr>
<td>▶ No other comorbidity or chronic health condition associated with fatigue.</td>
<td>▶ Dual sensory impairment.</td>
</tr>
<tr>
<td>▶ No dual sensory impairment (uncorrected vision loss).</td>
<td>▶ Inadequate English language understanding.</td>
</tr>
<tr>
<td>▶ Adequate English language understanding.</td>
<td>▶ No access to the internet.</td>
</tr>
<tr>
<td>▶ Access to the internet.</td>
<td></td>
</tr>
</tbody>
</table>

### Table 2: Demographic Information Form

<table>
<thead>
<tr>
<th>Initial question</th>
<th>Follow-up question(s)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Age of participant</td>
<td>NA</td>
</tr>
<tr>
<td>Gender of participant</td>
<td>NA</td>
</tr>
<tr>
<td>Does the participant use a hearing device?</td>
<td>▶ What device do they use?</td>
</tr>
<tr>
<td></td>
<td>▶ Which side do they wear?</td>
</tr>
<tr>
<td></td>
<td>▶ How long have they used the device?</td>
</tr>
<tr>
<td>Does the participant use any other hearing equipment?</td>
<td>What equipment do they use?</td>
</tr>
<tr>
<td>Has the participant been diagnosed with any medical conditions?</td>
<td>Please indicate the conditions.</td>
</tr>
<tr>
<td>SSQ sections A (speech) and B (spatial), 22 items to assess for perceived hearing ability</td>
<td>NA</td>
</tr>
<tr>
<td>NA, not applicable; SSQ, Speech, Spatial and Qualities of Hearing Scale.</td>
<td></td>
</tr>
</tbody>
</table>
Audiological assessment
Each participant will complete an online hearing test, DIN test, with instructions on how to complete it. The test will be completed at home in quiet using the participant’s device (must be a computer or laptop and not a phone or tablet, due to software compatibility) and head/earphones. If a participant uses a hearing device such as a hearing aid or cochlear implant, they should remove this prior to completing the test. The DIN test will take 10–20 min.

The DIN is an online speech in noise test. The participant is presented with a series of three randomised digits within a steady-state speech-shaped background noise. The participant must select (from a number pad within the platform) the digits they heard. They are then presented with further digit triplets until the end of the test. They are given a decibel signal-to-noise ratio, defined as the difference between target digits and background noise, in dB, that is required to correctly recognise 50% of the digit-triplets. This measure was chosen to provide a relatively quick and better approximate hearing function in a ‘real-life’ environment that can be completed at home. More information about the hearing test can be found in online supplemental appendix 1.

LE, fatigue, confidence and stress questionnaires
For sessions 3 and 4, each participant will be presented with four questionnaires, each measuring a different domain (see further). Parents/guardians can ask the participant the items provided in the questionnaire and encourage the participant to answer the item as detailed as possible, or the participant can complete it by themselves. The questionnaires will take approximately 30 min to complete.

The participant will be asked to complete the questionnaires twice. The two time points are defined as part 1: the evening of a rest day (rest day is defined as not attending school/college/work, and part 2: the evening of a workday (workday is defined as attending school/college/work). Each standardised questionnaire will be checked and validated by a research team member. If a participant does not complete a questionnaire session fully and there are missing data, we will send an email requesting the complete the entire session again. The questionnaires are as follows.

Listening effort
SSQ for children, section C, qualities of hearing
This questionnaire was first created by Gatehouse and Noble, then later validated in children 11 years and over by Galvin and Noble. Several studies have used a subscale of the qualities section of the SSQ, which contains three items on a sliding scale, to subjectively measure LE. The sliding scale corresponds to a numerical value (1–10). The sum of each scale is determined, with lower scores representing higher effort levels.

Fatigue
Vanderbilt Fatigue Scale–Child (VFS-C)
This was first created by Hornsby et al for use in adults and recently validated for use in children aged 6–17 years. The Vanderbilt Fatigue Scale is specifically designed to measure fatigue related to hearing. VFS-C is a 10-item, 5-point Likert scale with numerical weighting (0–4). The sum of the 10 items equates to the level of fatigue, with higher scores representing higher fatigue levels.

Stress
Perceived Stress Scale (PSS)
This was first created by Cohen and later shown to apply to children aged 5–18 by White. The PSS is a 10-item, 5-point Likert scale with a numerical weighting (0–4). The sum of the 10 items equates to the stress level, with higher scores representing higher stress levels. Some questions are reversed scored.

Confidence
Rosenberg Self-Esteem Scale (RSES)
This was first created by Rosenberg and has shown to apply to children aged 12–17 by Bagley and Mallick. The RSES is a 10-item, 4-point Likert scale with a numerical weighting (0–4). The sum of the 10 items equates to the confidence level, with higher scores representing higher confidence levels. Some questions are reversed scored.

A flow diagram outlining the process a participant takes through this study is represented in figure 1.

Figure 1 Timeline of participants’ journey through this study. LE, listening effort; PSS, Perceived Stress Scale; RSES, Rosenberg Self-Esteem Scale; SSQ, Speech, Spatial and Qualities of Hearing Scale; VFS-C, Vanderbilt Fatigue Scale–Child.
Statistical analysis plan
Without previous information to power the study, the sample size was determined pragmatically. Tests are exploratory, and for hypothesis-generating purposes, the standard criterion of p<0.05 highlights areas with potential significance.

An appropriate descriptive analysis of participants’ demographics and baselines will be provided. The number (%) of participants who completed each part of the tests will be reported. Depending on the distribution, numerical outcome measures will be summarised using mean/median (SD/IQR) for each test condition. Binary outcomes will be summarised by count (%).

LE will be correlated to other outcome measures using appropriate methods, depending on the distribution of outcome measures. A sample of 100 will have 80% power to detect a Pearson correlation of 0.3 or larger. Univariable regression models may be used to explore the effect of different factors such as day of assessment, age and gender on the correlations. The relationship between outcome measures with demographics will be explored.

Missing data
If there are missing data or clarification is needed, the participant will be contacted to try and ascertain missing data. If they are not able to do this, the quantity of missing data on each variable per individual will be assessed. For individuals with less than 20% missing items per questionnaire, the missing data will be predicted using an appropriate multiple imputation method, such as delta adjusted. This has been chosen to reduce the risk of selection bias associated with complete case sampling (de Goeij et al, 2013; Mackinnon, 2010). A complete case analysis will be conducted alongside multiple imputation.

The data generated by the study will be analysed at Manchester University NHS Foundation Trust, and the analysis will be performed by researchers employed by Manchester University NHS Foundation Trust.

Patient and public involvement
Neither patients nor the public were consulted during the creation of this exploratory study. If correlations between hearing loss and the downstream consequences of LE can be established, then the authors would seek to involve stakeholders at this point.

Data statement
Data collected as part of this study will be pseudonymised and labelled with a unique personal study participant code rather than names. The study ID is assigned to the participant when they sign the Informed Consent Formed (ICF). The contact email address and study ID are downloaded from REDCap and stored in a password-protected file within the MFT secure server drive.

There is also an intention to share an anonymous final data set with external researchers; however, this will only occur if the correct data-sharing agreement is in place.

Ethics and dissemination
This study has been reviewed within the funding organisation (Cochlear Research and Development Limited) by an independent and relevant peer reviewer/committee. Favourable comments and approval of the protocol were given during the peer review process. A favourable opinion has also been granted by the Health Research Authority (HRA) and NHS Research Ethics Committee (REC) for the study and all the supporting documents, including this protocol, information sheets, informed consent forms and other relevant documents (22/WS/0134).

Study results will be disseminated via the Cochlear Implant Children’s Support Group, for example, newsletters and social media, as deemed appropriate by the charity and mailing lists. A summary of this study’s results will be provided to all participants who have consented to be contacted. The aim includes publication of the final study results in medical literature and presentation at medical conferences.

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Contributors
IB, JN, KK, CAS, AR and MS conceived the study concept and design. MS and CAS conducted screening and data collection. AR performed the analysis. HG coordinated the Digits in Noise hearing test. IB, JN, CAS and MS prepared the first draft of the manuscript. All authors provided edits and critiqued the manuscript for intellectual content.

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Competing interests
None declared.

Patient and public involvement
Patients and/or the public were not involved in the design, conduct, reporting or dissemination plans of this research.

Patient consent for publication
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Provenance and peer review
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Supplemental material
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