

Is High-Intensity Speech Intervention Better? A Comparison of High-Intensity Intervention Versus Low-Intensity Intervention in Children With a Cleft Palate

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Abstract

Purpose: The purpose of this study was to compare the effect of speech intervention provided with a low intensity with speech intervention provided with a high intensity on the speech and health-related quality of life (HRQoL) in Dutch-speaking children with a cleft palate with or without a cleft lip (CP ± L) between 4 and 12 years.

Method: A longitudinal, prospective, randomized controlled trial with a multiple baseline design was used. Twelve children with a CP ± L ($M_{age} = 8.0$ years, $SD = 1.54$) were divided into two groups using block randomization stratified by age and gender: One group received low-intensity speech intervention (LISI; $n = 6$) and one group received high-intensity speech intervention (HISI; $n = 6$). Children in the LISI group received intervention with a session duration of 1 hr, a dose frequency of 1 session per week, and a total intervention duration of 10 weeks. Children in the HISI group received intervention with a session duration of 1 hr, a dose frequency of 5 sessions per week, and a total intervention duration of 2 weeks. The cumulative intervention intensity was kept constant. Both groups received identical therapy programs provided by the same experienced speech therapist. Perceptual speech assessments were performed on baseline and posttreatment data points. Changes in HRQoL were assessed using the Velopharyngeal Insufficiency Effects on Life Outcomes (VELO) questionnaire. Both groups were compared over time using (generalized) linear mixed models.

Result: No significant Time × Group interactions were observed for the percentage of correctly produced consonants at the word and sentence levels, indicating no differences in evolution over time among the two groups. The variables speech understandability, speech acceptability, and the total VELO scores significantly improved following HISI, but not following LISI.

Conclusions: Children in the HISI group made equal and, for some variables, even superior progress in only 2 weeks of therapy compared to children in the LISI group who received 10

weeks of therapy. HISI is a promising strategy to improve speech outcomes and HRQoL in a shorter time period.

The presence of active or compensatory speech errors severely impacts the speech understandability and speech acceptability of children with a cleft of the palate with or without a cleft of the lip (CP ± L; Harding & Grunwell, 1998). To eliminate these active or compensatory speech errors, behavioral intervention in terms of the provision of speech therapy is required (American Cleft Palate-Craniofacial Association, 2016). The intensity of speech therapy is a variable that may be key to optimizing intervention effects (Warren et al., 2007; Zeng et al., 2012). In literature, there is no consensus on the precise definition of the term *intensity*. *Intervention intensity* is, for example, defined as the quality and quantity of services delivered in a given time (Barnett & Escobar, 1990) or the number of hours of intervention over a specific time period (Lovaas, 1987). Some authors, on the contrary, define intervention intensity as the number of specific teaching episodes per unit of time (Guralnick, 1997). Determination of the intervention intensity, however, involves more than counting the hours spent practicing a skill (Kaipa & Peterson, 2016; Togher, 2012). With the eye on the application of a uniform definition, Warren et al. (2007) proposed a framework for defining intervention intensity. This framework includes six different parameters: (a) dose form, (b) dose, (c) session duration, (d) dose frequency, (e) total intervention duration, and (f) cumulative intervention intensity. “Dose form” includes the type of task or activity in which teaching episodes are delivered, for example, playing or drill exercises. *Dose* is defined as the number of properly administrated teaching episodes during a single intervention session, for example, the amount of trials within a therapy session. “Session duration” and “dose frequency,” respectively, involve the length of a session in time (e.g., 1 hr) and the number of sessions per unit of time (e.g., 1 or 2 times per week). “Total intervention duration” refers to the time period over which a specified intervention is presented, for example, 10 weeks. “Cumulative intervention intensity” is the product of dose, dose frequency, and total intervention duration.

To date, the optimal intensity for speech intervention in children with a CP ± L is unknown (Bessell et al., 2013). The decision for selecting a particular intervention intensity has been dependent on the clinician's own experience and the countries' health regulations and insurance providers (Bessell et al., 2013; Howard & Lohmander, 2011; Mullen & Schooling, 2010; Sweeney et al., 2020). In European countries, speech therapy sessions of 30–60 min are traditionally provided once or twice per week during several years (Baker & McLeod, 2011; Maas et al., 2008; Mullen & Schooling, 2010). Speech therapy provided with an inaccurate intervention intensity (i.e., too high or too low) can result in ineffective intervention outcomes (Baker, 2012). While providing too much intervention may not result in additional gains and may be a waste of resources, providing too little intervention may not be beneficial at all (Kaipa & Peterson, 2016).

In the past few years, the intensity of speech intervention in children with speech sound disorders (SSD) but without a CP ± L has received more attention. Allen (2013) performed a randomized controlled trial in 54 children (age range: 3–5 years) with SSD without a CP ± L. The authors compared the effect of high-intensity speech intervention (HISI) with the effect of low-intensity speech intervention (LISI). The children in the HISI group received three 30-min sessions per week for 8 weeks, whereas the children in the LISI group received one 30-

min session per week for 24 weeks. Interestingly, the findings demonstrated that children receiving high-intensity intervention made significantly more progress following therapy when compared to children who received low-intensity intervention. More recently, Cummings et al. (2021) investigated how dose frequency affected phonological acquisition in eight children with SSD without a CP \pm L. The children received twenty 50-min sessions that were either provided twice a week ($n = 4$ children) for 10 weeks or 4 times a week for 5 weeks ($n = 4$ children). Regardless of whether speech intervention was provided 2 times per week or 4 times per week, the children demonstrated similar phonological gains. However, children who received speech intervention 4 times per week made their phonological gains in approximately half the time compared to children who received speech intervention 2 times per week. The findings of this study hence suggest that more intensive weekly speech intervention sessions could be more efficient in teaching phonological information compared to less intensive speech intervention sessions. Considering the limited amount of evidence, several authors argued that research on the effect of different speech intervention intensities is urgently required (Allen, 2013; Bessell et al., 2013; Cummings et al., 2021; Maas et al., 2008). If more intensive speech intervention is recommended compared to the current traditional intervention intensity, a systematic change of speech therapy-related health care should be considered.

The traditional intervention intensity applied in children with a CP \pm L in European countries (i.e., therapy sessions of 30–60 min once or twice per week provided during several years) can be considered a form of LISI (Mullen & Schooling, 2010). To date, it is unknown whether this intervention intensity is evidence based considering that there are few studies that have yet investigated the effect of (the intensity of) speech intervention in children with a CP \pm L (Bessell et al., 2013). These traditional low-intensity interventions that are provided during several years may increase the burden for patients and their families and can hypothetically result in treatment fatigue with higher cancellation rates and dropouts (Pamplona et al., 2005). Given the lack of financial resources and available speech-language pathologists (SLPs), the delivery of years-long speech intervention is particularly challenging in developing countries (Sell et al., 2011). Within these contexts, other types of speech therapy models have been described (Sell et al., 2011). Speech therapy delivery in terms of “camp models” has received much attention. Camp models include the provision of intensive speech intervention programs and can be considered an example of HISI (Sell et al., 2011). Such intensive speech camps are usually provided for 1–2 weeks, with patients receiving multiple hours of treatment per day (Sell et al., 2011). The effectiveness of high-intensity speech therapy camps has been demonstrated in Mexico (i.e., 4 hr of intervention per day for a period of 3 weeks; Pamplona et al., 2005, 2009), Uganda (i.e., 2 hr of intervention per day for a period of three consecutive days; Alighieri, Bettens, Bruneel, Sseremba, et al., 2020; Luyten et al., 2016), and Thailand (i.e., 6 hr of intervention per day for a period of four consecutive days; Makarabhirom et al., 2015). These intensive speech intervention models do not only increase patient adherence but are also described to be considerably cheaper than the current European provision models (Pamplona et al., 2005). Unfortunately, the unique character of developing countries hampers the generalization of study findings to European countries. Moreover, the interpretation of the results should be performed carefully since these studies were subject to several sources of bias (e.g., trainer and observer bias). The application of high-intensity intervention models in children with a CP \pm L is, however, not only described in developing countries. In a recently performed randomized controlled trial that compared the effect of a motor-phonetic versus a linguistic-phonological intervention in 14 children with a CP \pm L, speech intervention was provided with a session duration of 1 hr, a dose frequency of 5 sessions per week, and a total intervention duration of 2 weeks

(Alighieri, Bettens, Bruneel, D'haeseleer, et al., 2020). The findings demonstrated that both high-intensive motor-phonetic and linguistic-phonological speech interventions can have a positive impact on the occurrence of cleft speech characteristics (CSCs) and consonant proficiency in children with a CP \pm L. A high-intensive linguistic-phonological approach, however, was observed to be more effective in terms of improving these speech outcomes compared with a high-intensive motor-phonetic approach. The authors questioned whether this high-intensity linguistic-phonological intervention model would lead to equal or superior results when compared with more traditional (lower intensity) intervention models.

To the best of our knowledge, only two studies actually compared the effect of HISI with the effect of LISI in patients with a CP \pm L (Albery & Enderby, 1984; Pamplona et al., 2005). Albery and Enderby (1984) performed a randomized controlled trial that compared two speech intervention intensities. Forty-six children with a CP \pm L were randomly assigned to two groups. One group received three speech therapy sessions per day (two individual 30-min sessions and one 30-min group session) during 6 weeks (i.e., group receiving HISI) and one group received one 1-hr speech therapy session per week with an undefined total intervention duration (i.e., group receiving LISI). The findings demonstrated that, regardless of the treatment intensity, the children exhibited a significant reduction in articulation errors immediately post-intervention. Interestingly, the children who received HISI showed more immediate and larger improvements in terms of the correct production of targeted consonants compared with children who received LISI. Pamplona et al. (2005) compared the effect of an intensive speech therapy camp (i.e., 4 hr of therapy per day during 5 days for a period of 3 weeks) with conventional speech therapy (i.e., 1-hr treatment sessions, twice a week for a period of 12 months) in 90 children with a CP \pm L. The results showed no significant difference in evolution between the two groups. Based on these two studies, one can conclude that evidence supporting HISI in children with a CP \pm L is conflicting (Howard & Lohmander, 2011). More recently, Sweeney et al. (2020) compared the effectiveness of a parent-led, therapist-supervised articulation therapy (PLAT) with the effectiveness of a routine speech intervention delivered by a specialized SLP in 44 children with a CP \pm L. Children in the PLAT group received therapy with a session duration of 10–15 min, a dose frequency of 5 times per week, and a total intervention duration of 12 weeks. Children in the routine intervention group (routine group) received therapy with a session duration of 1 hr, a dose frequency of 1 time per week, and a total intervention duration of 12 weeks. Thereby, the intervention in the PLAT group can be considered high intensive, whereas the intervention in the routine group can be considered low intensive. Despite that investigating intervention intensity was not the purpose of this study, the results suggested that parent-led, therapist-supervised intervention (which was provided with a higher intervention intensity) could lead to equal results when compared with routine intervention (which was provided with a lower intervention intensity) in children with a CP \pm L. In summary, the effect of intervention intensity in children with a CP \pm L is inconclusive and questionable due to heterogeneity of previous studies.

Intervention approaches used to investigate the effects of intervention intensity should be able to provide descriptions of the six intensity parameters proposed by Warren et al. (2007) (Allen, 2013). The Metaphon approach, which is a metaphonological intervention approach that is often used by SLPs to treat speech sound errors, seems to be eligible for this matter (Dean et al., 1995). The approach consists of two separate phases. During Phase 1, the child's metaphonological awareness is stimulated by exploring the sound system using child-friendly games and vocabulary. During Phase 2, this acquired metaphonological knowledge is used in communicative situations using a structured treatment task. Thereby, the dose form for the

first phase is reflected by a child-directed, play-based approach, whereas the dose form for the second phase is more consistent with structured (drill) play (Dean et al., 1995, 1996; Grundy, 1995; Howell & Dean, 1991). An intervention study can thus control for the variable “dose form” while using the Metaphon approach. In addition, documentation on session duration (i.e., 1 hr), dose frequency (i.e., 5 sessions per week), and total intervention duration (i.e., 2 weeks) was provided by the randomized controlled trial performed by Alighieri, Bettens, Bruneel, D'haeseleer, et al. (2020). Since the latter study demonstrated the effect of the Metaphon approach in children with a CP ± L delivered in accordance with a high-intensity intervention model, this information on the different intervention intensity parameters can be used as a basis for further research.

At present, there is a lack of evidence on the effect of intervention intensity in children with a CP ± L (Bessell et al., 2013). The lack of evidence-based practice forces us to ask the question: “Are we applying the available resources in the most effective way?” To respond to this gap in literature, the purpose of this study was to compare the effect of speech intervention provided with a low intensity (i.e., a session duration of 1 hr, a dose frequency of 1 session per week, and a total intervention duration of 10 weeks) with the effect of speech intervention provided with a high intensity (i.e., a session duration of 1 hour, a dose frequency of 5 sessions per week, and a total intervention duration of 2 weeks) on the speech and health-related quality of life (HRQoL) in Dutch-speaking children with a CP ± L using perceptual and psychosocial outcome measures. Regardless of the intervention intensity, the children received the same intervention approach (i.e., Metaphon approach). Based on the literature review, it was hypothesized that HISI may be equally effective or even more effective in reducing active or compensatory speech errors and in increasing the child's consonant proficiency and HRQoL compared with LISI.

Method

This study was approved by the Ethics Committee of the Ghent University Hospital (2018/1218). The trial registration number is B670201837572.

Participants

Dutch-speaking children with a CP ± L and aged between 4 and 12 years were recruited at the Ghent University Hospital between January 2020 and December 2020 using a purposive sampling method. Children who presented with at least one active or compensatory speech error, based on the perceptual assessment of one SLP (C. A.), could be included in this study. Children were excluded if they presented with (a) cognitive and related learning disabilities or syndromes based on the patients' files and questioning the parents, (b) an oronasal fistula based on oral examination performed by an SLP, (c) velopharyngeal insufficiency based on videofluoroscopic examination if this was performed as a part of the clinical evaluation, and (d) hearing disabilities based on pure-tone audiometry (> 25 dB HL) and five otologic health screening questions (the use of hearing aids, frequency and chronicity of otitis media, past placement of tympanostomy tubes, and development of complications such as cholesteatoma and mastoiditis; Allori et al., 2017). Included patients were not allowed to receive any other type of speech intervention nor any type of surgical intervention during the study period. Figure 1 presents the inclusion and exclusion of possible participants.

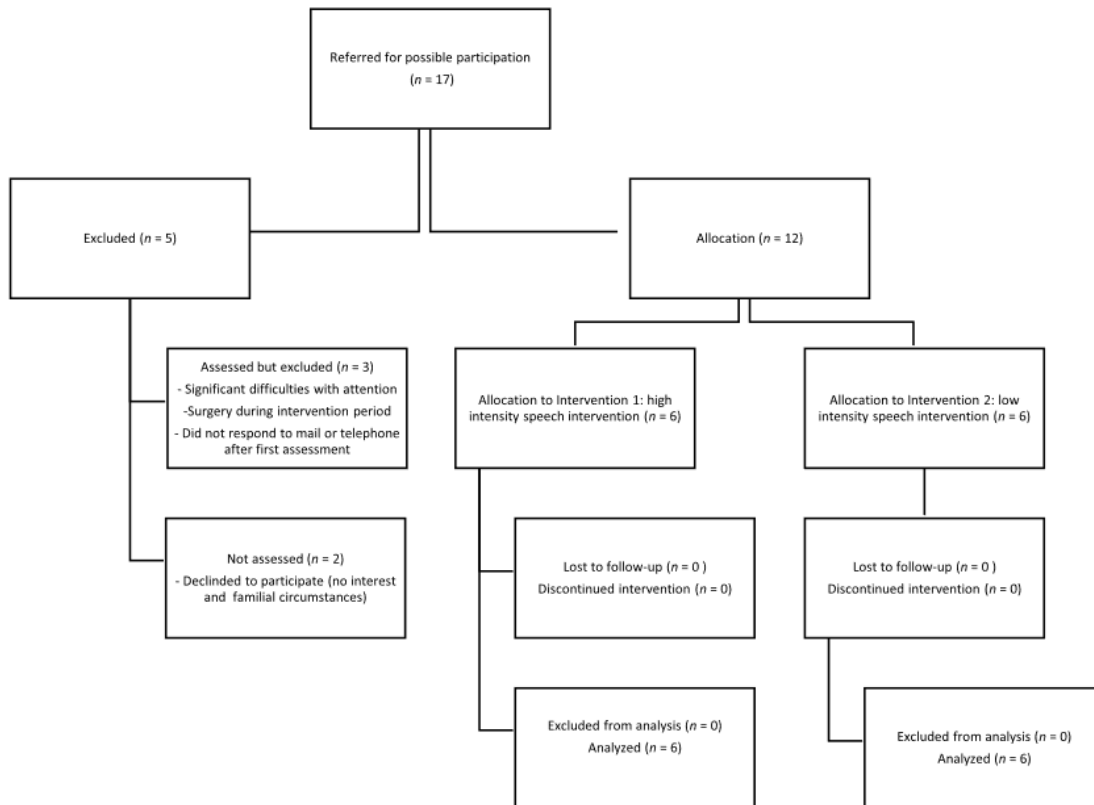


Figure 1. Flowchart for inclusion or exclusion of the participants.

The socioeconomic status (SES) of each child was measured based on Hollingshead's four-factor index of social status (Hollingshead, 1975). The four factors measured in this index are marital status, retired/employed status, education, and occupational prestige. The minimal SES score is 8 (lowest SES); the maximal SES score is 66 (highest SES).

Study Design

A longitudinal, prospective, randomized controlled trial with a multiple-baseline design was used with the LISI group serving as the control group. Participants were randomly assigned to two groups using block randomization stratified by age and gender. One group received speech intervention with a low intensity (i.e., a session duration of 1 hr, a dose frequency of 1 session per week, and a total intervention duration of 10 weeks) and the other group received speech intervention with a high intensity (i.e., a session duration of 1 hr, a dose frequency of 5 sessions per week, and a total intervention duration of 2 weeks). The cumulative intervention intensity was kept constant (see Table 1). Each child received 10 hr of speech intervention regardless of the group allocation. A summary of the intervention intensity parameters is presented in Table 1.

Table 1. Intervention intensity parameters according to the framework proposed by Warren et al. (2007).

Group	Dose form	Dose	Session duration	Dose frequency	Total intervention duration	Cumulative intervention intensity
LISI group	Phase 1: child-directed, play based Phase 2: structured (drill) play	80–100 responses per session ^a	1 hr	1 time per week	10 weeks	800–1,000
HISI group	Phase 1: child-directed, play based Phase 2: structured (drill) play	80–100 responses per session ^a	1 hr	5 times per week	2 weeks	800–1,000

Note. LISI = low-intensity speech intervention; HISI = high-intensity speech intervention.

^aDose recommendations in accordance with Williams (2003, 2005).

Every child went through three study phases: a baseline phase, an intervention phase, and a follow-up phase (see Table 2). Two identical speech samples were collected once a week before the start of the intervention (T1 and T2). During the intervention phase, speech samples were collected after 1 hr of therapy (i.e., after 1 day in the HISI group and after 1 week in the LISI group: T3) and after 5 hr of therapy (i.e., after 1 week in the HISI group and after 5 weeks in the LISI group: T4). After the intervention phase, data were collected on three occasions: immediately after the intervention (T5), 3 weeks postintervention (T6), and 3 months post-intervention (T7). Data were collected by two different SLPs (A. D. C./C. A.).

Table 2. Overview of the different study phases.

Group	Baseline phase		Intervention phase		Follow-up phase		
	T1	T2	T3	T4	T5	T6	T7
LISI	1 week pre-intervention	Immediately pre-intervention	After 1 hr of intervention (i.e., after 1 week)	After 5 hr of intervention (i.e., after 5 weeks)	Immediately post-intervention (i.e., after 10 weeks)	3 weeks post-intervention	3 months post-intervention
HISI	1 week pre-intervention	Immediately pre-intervention	After 1 hr of intervention (i.e., after 1 day)	After 5 hr of intervention (i.e., after 5 days)	Immediately post-intervention (i.e., after 2 weeks)	3 weeks post-intervention	3 months post-intervention

Note. LISI = low-intensity speech intervention; HISI = high-intensity speech intervention.

Intervention

The children received individual HISI or LISI provided by the same Dutch-speaking SLP (C. A.) with 4 years of experience with cleft palate speech disorders. Target consonants differed between the patients and were determined based on the data collected during the baseline phase. Table 3 provides an overview of the affected target consonants and error types per patient. Speech sounds that affected speech understandability the most were treated first (Kummer, 2011). This was determined on the perceptual assessment of the treating SLP (C. A.) and on the judgment of the child and their parents. If several speech sounds affected the child's speech understandability similarly, consonants that are normally acquired first during normal speech development were addressed first. Every therapy session was provided in a quiet room at Ghent University Hospital or at the child's home environment.

Table 3. Overview of the affected target consonants and error type per patient.

Patient	Group	Affected target consonant	Error type	Therapy session	Phonological features
1	LISI	[s] [z] [t]	Palatalization Backing to a velar place of articulation	Sessions 1–5: [s] [z] Sessions 6–9: [t] Session 10: [s] [z] [t]	Front–back
2	LISI	[s] [z] [v] [f] (inconsistent)	Active nasal fricatives	Sessions 1–5: [s] [z] Sessions 5–9: [v] [f] Session 10: [s] [z] [v] [f]	Nose–mouth
3	LISI	[p] [b] [k] [t] [d]	Glottal articulation Double articulation with a glottal stop (i.e., glottal reinforcement)	Sessions 1–5: [p] [b] [k] Sessions 5–9: [t] [d] Session 10: [p] [b] [k] [t] [d]	Throat – mouth
4	LISI	[s] [z] [v] [f]	Active nasal fricatives	Sessions 1–5: [s] [z] Sessions 5–9: [v] [f] Session 10: [s] [z] [v] [f]	Nose–mouth
5	LISI	[s] [z] [t] (inconsistent)	Palatalization	Sessions 1–5: [s] [z] Sessions 5–9: [t] (and [s] [z]) Session 10: [s] [z] [t]	Front–back
6	LISI	[b] [p] [t] [d] [k] [ɣ] [z] [s]	Glottal articulation Active nasal fricatives	Sessions 1–5: [b] [p] [t] [d] [k] [ɣ] Sessions 5–9: [z] [s] (and [k] [ɣ]) Session 10: [b] [p] [t] [d] [k] [ɣ] [z] [s]	Throat–mouth Nose–mouth
7	HISI	[t] [d] [k] [ɣ]	Glottal articulation	Sessions 1–5: [t] [d] Sessions 6–9: [k] [ɣ] Session 10: [t] [d] [k] [ɣ]	Throat–mouth
8	HISI	[s] [z] [ʃ] [t]	Palatalization	Sessions 1–5: [s] [z] Sessions 6–9: [ʃ] [t] Session 10: [s] [z] [ʃ] [t]	Front–back
9	HISI	[b] [p] [t] [d] [k] [ɣ]	Glottal articulation	Sessions 1–5: [b] [p] [t] [d] Sessions 5–9: [k] [ɣ] Session 10: [b] [p] [t] [d] [k] [ɣ]	Throat–mouth
10	HISI	[s] [z] [ʃ] [t] [d]	Palatalization	Sessions 1–5: [s] [z] [ʃ] Sessions 6–9: [t] [d] Session 10: [s] [z] [ʃ] [t] [d]	Front–back
11	HISI	[s] [z] [t]	Active nasal fricatives Backing to a velar place of articulation	Sessions 1–5: [s] [z] Sessions 6–9: [t] Session 10: [s] [z]	Nose–mouth Front–back
12	HISI	[s] [z] [v] [f]	Active nasal fricatives	Sessions 1–5: [s] [z] Sessions 5–9: [v] [f] Session 10: [s] [z] [v] [f]	Nose–mouth

Note. LISI = low-intensity speech intervention; HISI = high-intensity speech intervention.

The HISI and LISI groups received speech intervention using a linguistic-phonological approach following the principles of the Metaphon treatment with modifications for children with a CP ± L (Alighieri, Bettens, Bruneel, D'haeseleer, et al., 2020; Dean et al., 1995, 1996). The Metaphon approach was found to be effective in reducing active or compensatory speech errors and in increasing consonant proficiency in children with a CP ± L (Alighieri, Bettens, Bruneel, D'haeseleer, et al., 2020). Multiple active or compensatory speech sound errors were treated simultaneously (Dean et al., 1995, 1996). The Metaphon approach consists of two phases. Phase 1 is divided into four levels (i.e., the concept level, the sound level, the phoneme level, and the word level). During the first level, the contrastive features of the speech sound system were practiced, which were relevant to the particular error that was targeted. Terms describing these features were introduced using child-friendly and age-appropriate words (e.g., for backing, the terms *front* and *back* were used, and for active nasal fricatives, the terms *nose* and *mouth* were used). Child-friendly games were introduced to practice and illustrate the contrast between the concepts (e.g., “give a cookie to mister nose/mister mouth”). This vocabulary was transferred to the level of nonspeech sounds (e.g., pictures of animals making sounds, sounds made by toys). During the phoneme level, the SLP produced sounds that varied along the dimension in question (e.g., the SLP produced “nose” sounds). At the end of Phase 1, minimal word pairs (e.g., *tea/key*), produced by the SLP, were used to facilitate the patient's awareness of sounds in words. During the second phase, child-friendly minimal word pair games were used to emphasize phonological awareness, in which the speaker named a picture and the listener chose from the two pictures on the table according to the word they heard. Once the child became proficient at making distinctions in the production of minimal pairs, the words were put into sentences. These sentences were either imitated or self-generated by the child.

Home Practice

At the end of every therapy session, parents were provided with information about the used techniques. They were also encouraged to practice 5–10 min per day with their child. This information was provided orally and written in a notebook. Parents were also provided the possibility to contact the SLP by phone or by e-mail if they had any further questions.

To verify compliance with home practice, the notebook contained a daily schedule (i.e., a log book). In this schedule, parents could indicate (a) if they practiced with their child (yes/no), (b) how long they practiced (in minutes), (c) which exercises they did, and (d) what the results were.

Outcome Measures

Speech Sample

A Dutch speech sample was audio- and video-recorded at each data point using a unidirectional condenser microphone (Samson CO1U) and a Sony Handycam HDR-CQ280E with a high-quality built-in microphone. The speech sample, which was constructed as part of a Dutch outcome tool for cleft palate speech in accordance with the guidelines described by Henningsson et al. (2008), consisted of spontaneous speech, automatic rote speech, sentences, and words (Bruneel et al., 2020). Spontaneous speech was elicited by asking questions about the holidays, free time, and so forth. For the automatic rote speech, patients younger than 7 years old were instructed to count from 1 to 10 and to recite the days of the week. When patients were older than 7 years, they were asked to count from 1 to 20 as well as from 60 to 70 and to recite the days of the week. The sample included 13 sentences and 31 words targeting all the pressure consonants of the Dutch language in initial, medial, and final positions. To allow for a more structured and standardized speech sample, the children were asked to repeat the sentences produced by the SLP. At the word level, children were asked to name pictures. If the child was unable to produce the word spontaneously, a semantic or phonological cue was provided. If semantic and phonological cueing was found to be insufficient, the child was asked to repeat the word after modeling of the SLP. The speech samples were anonymized and randomized for further analysis.

Assessment Categories

A Dutch outcome tool that was constructed based on the Cleft Audit Protocol for Speech-Augmented (CAPS-A protocol) was used for the perceptual evaluation of the speech samples (Bruneel et al., 2020). Different categories were assessed, including speech understandability, which is defined as “the degree to which the speaker's message can be understood by the listener,” and speech acceptability, which is referred to as “the degree to which speech calls attention to itself apart from the content of the spoken message” (Henningsson et al., 2008). For these variables, an ordinal rating scale was used: 0 = *within normal limits*, 1 = *mildly disturbed*, 2 = *moderately disturbed*, and 3 = *severely disturbed*. In addition, consonant production was assessed in terms of CSCs (i.e., anterior oral CSCs, posterior oral CSCs, and nonoral CSCs). For the CSCs, a score of “0” corresponded with no affected target consonants in the speech sample, whereas a score of “1” corresponded with one or two affected consonants. A score of “2” was provided when the sample included three or more affected target consonants. Finally, the need for speech-language therapy (SLT) intervention for CSCs (0 = *no*, 1 = *yes*) was assessed.

Beside these categorical speech-related outcome measures, the children's consonant proficiency was assessed in terms of the percentage of correct consonants–revised (PCC-R; Shriberg et al., 1997), the percentage of correct places (PCP), and the percentage of correct manners (PCM; Klintö et al., 2011). For the PCC-R, consonants produced with a correct place, manner, and voice but with an (inter)dental quality or weak realization or passive nasal emission were considered to be correct (Sell & Sweeney, 2020; Shriberg et al., 1997).

Assessment Procedure

Two SLPs (K. B. and A. D. C.), with respectively 8 and 2 years of experience in assessing cleft-related speech disorders, perceptually assessed the speech samples. The first rater (K. B.) was officially trained in using the original CAPS-A tool and was involved in the construction of the Dutch outcome tool (John et al., 2006; Sell et al., 2009). The second rater (A. D. C.) was an SLP who had obtained a bachelor degree in speech-language pathology. She participated in this project in the context of her master's thesis on the effect of speech therapy in children with a CP ± L. The raters did not provide speech therapy to any of the included patients and were blinded to the treatment allocation of the patients.

A structured listening protocol was used for the perceptual analysis of the speech samples (Bruneel et al., 2020). First, the raters listened to the audio recordings of the spontaneous speech sample to assess speech understandability. Subsequently, the SLPs listened to the audio recordings of the automatic rote speech to perform an initial evaluation of the consonant productions. The audio recordings of the sentences were used to evaluate the consonant production in terms of active CSCs. Based on the video-recorded spontaneous speech samples and automatic rote speech samples, a revision of the visual aspects of the consonant productions was performed. Consequently, the SLPs listened to the video-recorded speech samples at the sentence level to revise the consonant productions in terms of the CSCs. Finally, speech acceptability and need for SLT intervention for CSCs were assessed based on the raters' overall judgment of the audio and video recordings.

The two assessors transcribed the speech samples at the word and sentence levels using the International Phonetic Association (IPA, 1999) and the IPA extensions as well as additional symbols to describe specific cleft-related articulation errors (Peterson-Falzone et al., 2016). The speech sample at the word level included the 13 target high-pressure consonants of the Dutch language in all possible positions (i.e., /p/, /b/, /t/, /d/, /s/, /z/, /ʃ/, /ʒ/, /f/, /v/, /k/, /x/, /ɣ/). No voiced pressure consonants were targeted in the word-final position since voiced high-pressure consonants are devoiced in the word-final position in the Dutch language (Grijzenhout & Krämer, 2000). The sample at the sentence level included the same target consonants supplemented by sentences with s-clusters (i.e., /st/, /sp./, /sk/; Bruneel et al., 2020). Based on these transcriptions, PCC-R, PCP, and PCM at the word and sentence levels were calculated (Klintö et al., 2011; Shriberg et al., 1997). The PCC-R was calculated by dividing the amount of correctly produced target consonants (numerator) by the total amount of target consonants elicited (denominator), multiplied by 100. The PCPs and PCMs were calculated similarly to the PCC-R following the guidelines described by Klintö et al. (2011). Thereby, if a child omitted a sound as part of developmental speech immaturity (e.g., consonant cluster deletion or deletion of [final] consonants), this consonant was not counted within the “total amount of target consonants elicited” (Sell & Sweeney, 2020).

The first rater (K. B.) analyzed 100% of the speech samples ($n = 84$). This SLP reassessed 20% ($n = 17$) of the speech samples in order to calculate intrarater reliability. To calculate

interrater reliability, the second rater (A. D. C.) also analyzed 100% of the speech samples ($n = 84$). The ratings of the first SLP were used for further analysis.

HRQoL Assessment

To assess the parents' and children's HRQoL, the Dutch version of the Velopharyngeal Insufficiency Effects on Life Outcomes (VELO) questionnaire was used (Bruneel, Bettens, & Van Lierde, 2019; Bruneel et al., 2017). The VELO questionnaire was administered on three different occasions: (a) immediately pre-intervention (i.e., T2), (b) immediately post-intervention (i.e., T5), and (c) 3 months post-intervention (i.e., T7). The VELO questionnaire consists of a parent and child report (for children older than 8 years) addressing different domains: speech limitation (seven items), swallowing problems (three items), situational difficulty (five items), emotional impact (four items), and perception by others (four items). In the parent report, an additional domain “caregiver impact” (three items) is included (Skirko et al., 2012, 2013). Each item was scored on a Likert scale ranging from 0 (*never*) to 4 (*almost always*). The responses on the items were recoded for further analyses using a mathematical formula: total score = 100 – (mean of all items \times 25). The scores on the reports ranged from 0 (the lowest HRQoL) to 100 (the highest HRQoL).

Statistical Analyses

SPSS Version 26 (SPSS Corporation) was used for the statistical analysis of the data. Analyses were conducted at $\alpha = .05$.

To assess inter- and intrarater reliability for the perceptual speech assessment, two-way mixed single-measure intraclass correlation coefficients (ICCs) were calculated. These ICCs were interpreted following the classification of Altman (1990; ICC < .20, poor; ICC = .21–.40, fair; ICC = .41–.60, moderate; ICC = .61–.80, good; ICC = .81–1.00, very good).

Linear mixed models (LMMs) were fitted to compare the HISI and LISI groups over time on each continuous outcome measure using the restricted maximum likelihood estimation and the scaled identity covariance structure. This covariance structure was chosen based on comparison of the Akaike's information criterion values. Time, group, and Time \times Group effects were specified as fixed factors. Likewise, generalized LMMs were fitted for the categorical outcome measures. If a significant main effect (time, group, or Time \times Group) was found, a comparison of time within the groups was determined using pairwise comparisons with Bonferroni corrections at $\alpha < .017$ (.05/3). The pre-intervention values were compared with immediate post-intervention values (T5). To investigate the short-term effect, T5 was compared with T6 (3 weeks follow-up). To investigate the long-term effect, T5 was compared with T7 (3 months follow-up).

Both unstandardized and standardized effect sizes were reported. Unstandardized effect sizes were measured by providing the estimated mean differences and 95% confidence intervals for the outcome variables (Baguley, 2009). In addition, standardized effect sizes were calculated for the Time \times Group interactions using Cohen's *d*s dividing the estimated mean difference by the standard deviation of a linear null model on the baseline data (Feingold, 2013).

Results

Participants

Five boys and seven girls with a CP ± L were included in this study. The LISI group consisted of six children (three boys and three girls) with a median age of 7.00 years (range: 6.00–10.00 years). The HISI group also consisted of six children (two boys and four girls) with a median age of 9.00 years (range: 6.00–10.00 years). The Mann–Whitney *U* test revealed no statistically significant differences in age between the two groups ($U = 9.50, p = .180$). In addition, a chi-square test revealed that there were no statistically significant differences in gender between the two groups, $\chi^2(1) = 0.343, p = .500$.

Each included child had a history of speech therapy. Median amount of speech therapy was 2.75 years (interquartile range [IQR] = 2.00–5.00 years) and 4.33 years (IQR = 3.00–5.00 years) in the LISI and HISI groups, respectively. No statistically significant differences in the total amount of previous speech therapy were found when comparing the LISI and HISI groups (Mann–Whitney $U = 7.00, p = .093$). Table 4 presents a detailed comparison of the cleft characteristics as well as demographic and otologic information between the LISI and HISI groups.

Table 4. Comparison of cleft characteristics as well as demographic and otologic information between the low-intensity speech intervention (LISI) group and the high-intensity speech intervention (HISI) group.

Variable	LISI (n = 6)	HISI (n = 6)	U, p ^a
Cleft characteristics			
Cleft type	CP: n = 0 CLP: n = 4 SC: n = 2	CP: n = 2 CLP: n = 4 SC: n = 0	—
Palatal closure, Mdn (IQR)	11.50 m (10.00–14.75 m)	11.50 m (9.75–14.00 m)	U = 19.50, p = .818
Lip closure, Mdn (IQR)	3.00 m (3.00–3.00 m)	3.00 m (3.00–3.00 m)	U = 4.00, p = 1.000
Secondary surgery	Secondary lip repair: n = 1	Sphincter pharyngoplasty: n = 1	—
Demographic information			
SES, Mdn (IQR)	33.42 (20.50–39.63)	34.25 (18.63–57.25)	U = 15.50, p = .699
Otologic information			
Use of hearing aids	n = 0	n = 0	—
Frequency of otitis media during the child's lifetime, Mdn (IQR)	1.00 (0.00–3.00)	1.00 (0.00–2.00)	U = 16.50, p = .818
Past placement of tympanostomy tubes, Mdn (IQR)	2.00 (0.00–4.00)	1.00 (0.00–2.00)	U = 12.50, p = .394
Development of otologic complications	n = 0	n = 0	—

Note. CP = cleft palate; CLP = cleft lip and palate; SC = submucosal cleft; IQR = interquartile range; m = months; SES = socioeconomic status.
^aStatistical analyses based on the Mann–Whitney *U* test ($\alpha = .05$).

Inter- and Intrarater Reliability

Results for the inter- and intrarater reliability are presented in Table 5. Interrater reliability was good to very good for all the assessment categories, except for “anterior oral CSCs,” “PCM at the word level,” and “targets correct at the word level in terms of manner of articulation.” For these variables, interrater reliability was fair to moderate. Intrarater was very good for the majority of the assessment categories. For the variables “posterior oral CSCs,” “PCM at the word level,” and “targets correct at the word level in terms of manner of articulation,” intrarater reliability was moderate.

Table 5. Inter- and intrarater reliability by means of a two-way mixed single intraclass correlation coefficient (ICC; consistency).

Variable	Interrater reliability			Intrarater reliability		
	Single ICC consistency	95% CI single ICC consistency	Interpretation of single ICC ^a	Single ICC consistency	95% CI single ICC consistency	Interpretation of single ICC ^a
Speech understandability	.71	[.48, .84]	Good	.95	[.89, .97]	Very good
Speech acceptability	.84	[.69, .92]	Very good	.90	[.75, .96]	Very good
Anterior oral CSCs	.53	[.24, .74]	Moderate	.84	[.60, .94]	Very good
Posterior oral CSCs	.64	[.28, .81]	Good	.48	[.02, .78]	Moderate
Nonoral CSCs	.78	[.61, .88]	Good	.84	[.62, .94]	Very good
PCC-R word level	.83	[.67, .91]	Very good	.94	[.89, .98]	Very good
Targets elicited word level	— ^b	— ^b	— ^b	.86	[.66, .95]	Very good
Targets correct word level	.98	[.73, .99]	Very good	.98	[.90, .99]	Very good
PCC-R sentence level	.76	[.56, .88]	Good	.87	[.73, .94]	Very good
Targets elicited sentence level	.73	[.34, .97]	Good	.87	[.67, .96]	Very good
Targets correct sentence level	.92	[.56, .99]	Very good	1.00	[1.00, 1.00]	Very good
PCP word level	.75	[.55, .87]	Good	.89	[.73, .96]	Very good
Targets correct in terms of place of articulation (word level)	.86	[.67, .96]	Very good	.88	[.69, .96]	Very good
PCP sentence level	.61	[.32, .78]	Good	.91	[.72, .97]	Very good
Targets correct in terms of place of articulation (sentence level)	.63	[.24, .93]	Good	.97	[.91, .99]	Very good
PCM word level	.56	[.26, .75]	Moderate	.48	[-.02, .80]	Moderate
Targets correct in terms of manner of articulation (word level)	.40	[.11, .69]	Fair	.43	[.02, .72]	Moderate
PCM sentence level	.68	[.43, .83]	Good	.90	[.72, .97]	Very good
Targets correct in terms of manner of articulation (sentence level)	.63	[.45, .86]	Good	— ^b	— ^b	— ^b
Need for SLT intervention	.87	[.76, .94]	Very good	1.00	[1.00, 1.00]	Very good

Note. CI = confidence interval; CSCs = cleft speech characteristics; PCC-R = percentage of correct consonants-revised; PCP = percentage of correct places; PCM = percentage of correct manners; SLT = speech-language therapy.

^aBased on Altman (1990): ICC < .20, poor; ICC = .21–.40, fair; ICC = .41–.60, moderate; ICC = .61–.80, good; ICC = .81–1.00, very good.

^bICC was impossible to calculate due to zero variance.

Baseline Outcome Measures

Comparison of the two data points in the baseline phase (i.e., T1 and T2) within and between the LISI and HISI groups revealed no statistically significant differences for the outcome variables ($p > .025$). Within each group, there was a stable baseline. In addition, there were no baseline differences between the two groups. The mean value of the two baseline measures was calculated for each outcome variable and used for further analyses.

Evolution of the Outcome Measures

Table 6 and 7 and Figures 2 and 3 present the evolution of the speech-related outcome variables and consonant proficiency. Generalized LMMs revealed a significant time effect for the variables speech understandability, $F(6, 18) = 5.384, p < .001$; speech acceptability, $F(6, 14) = 4.549, p = .001$; anterior oral CSCs, $F(6, 16) = 8.061, p < .001$; posterior oral CSCs, $F(5, 6) = 5.365, p < .001$; nonoral CSCs, $F(6, 10) = 5.155, p < .001$; and need for SLT intervention, $F(2, 6) = 3.436, p = .005$. The significant time effects indicated an improvement of these outcome measures irrespective of the group assignment. Post hoc pairwise comparison of time within the groups revealed that the variables speech understandability ($p = .002$), speech acceptability ($p = .003$), anterior oral CSCs ($p \leq .001$), and nonoral CSCs significantly improved immediately after the intervention in the HISI group but not in the LISI group. These immediate improvements remained present in the short term (3 weeks post-intervention, $p = .672$) and in the long term (3 months post-intervention, $p = .672$). In contrast, the variable “posterior oral CSCs” improved immediately after the LISI intervention but not after the HISI intervention ($p = .016$).

Table 6. Comparison of the categorical speech-related outcome variables between the high-intensity speech intervention (HISI) and low-intensity speech intervention (LISI) groups.

Variable	Group	Pre Mdn (IQR)	Post T5 (immediately) Mdn (IQR)	Post T6 (3 weeks) Mdn (IQR)	Post T7 (3 months) Mdn (IQR)	Time x Group <i>p</i>	Group <i>p</i>	Time <i>p</i>	Comparison time within groups		
									Evolution Pre-T5 <i>p</i>	Evolution T5-T6 <i>p</i>	Evolution T5-T7 <i>p</i>
Speech understandability	LISI	2 (1.5-3)	1 (0.5-2)	2 (0.5-2)	1 (0.5-2)	.654	< .001 ^a	< .001 ^a	.069	.266	1.000
	HISI	1.5 (1-2)	0 (0-1)	0 (0-1)	0 (0-1)				.002 ^a	.672	.672
Speech acceptability	LISI	2.5 (2-3)	2 (1-2.5)	2 (1.5-3)	1 (0.5-2)	.716	< .001 ^a	.001 ^a	.122	1.000	.242
	HISI	2 (1-2.5)	0.5 (0-1)	0.5 (0-1)	0 (0-1)				.003 ^a	1.000	.671
Anterior oral CSCs	LISI	1.5 (1-2)	1 (1-1.5)	0.5 (0-1)	0.5 (0-1)	.757	.083	< .001 ^a	.116	.289	1.000
	HISI	1.5 (1-2)	0 (0-1)	0 (0-0.5)	0 (0-0.5)				< .001 ^a	.618	.618
Posterior oral CSCs	LISI	1 (0.5-1)	0 (0-0.5)	0 (0-0.5)	0 (0-0.5)	.356	.018	< .001 ^a	.016 ^a	1.000	1.000
	HISI	0.5 (0-1)	0 (0-0.5)	0 (0-0.5)	0 (0-0.5)				.076	1.000	1.000
Nonoral CSCs	LISI	1.5 (1-2)	1 (0-1.5)	1.0 (0.5-1.5)	1 (0-1)	.795	< .001 ^a	< .001 ^a	.043	.601	.601
	HISI	1 (1-2)	0 (0-0.5)	0 (0-1.5)	0 (0-0.5)				.002 ^a	1.000	1.000
Need for SLT intervention	LISI	1 (1-1.5)	1 (1-1.5)	1 (1-1.5)	1 (1-1.5)	.226	.001 ^a	.005 ^a	1.000	1.000	1.000
	HISI	1 (1-1.5)	0.5 (0.5-1)	0 (0-1)	0 (0-1)				.049	.502	.502

Note. Mdn = median; IQR = interquartile range; CSCs = cleft speech characteristics; SLT = speech-language therapy.

^aIndicates a significant effect based on generalized linear mixed models ($\alpha < .017$).

Table 7. Comparison of consonant proficiency between the high-intensity speech intervention (HISI) and low-intensity speech intervention (LISI) groups.

Variable	Group	Pre	Post T5 (immediately)	Post T6 (3 weeks)	Post T7 (3 months)	Time x Group
		EM [95% CI]	EM [95% CI]	EM [95% CI]	EM [95% CI]	<i>p</i>
PCC-R WL	LISI	64.72 [57.57, 71.87]	87.10 [79.94, 94.25]	89.03 [81.87, 96.18]	87.88 [80.73, 95.03]	.490
	HISI	69.31 [63.08, 76.14]	97.31 [90.78, 103.84]	100.00 [93.47, 106.53]	100.00 [93.47, 106.53]	
PCC-R SL	LISI	66.92 [59.15, 74.70]	84.77 [76.99, 92.55]	82.70 [74.92, 90.48]	86.50 [78.72, 94.28]	.183
	HISI	65.71 [58.61, 72.82]	96.40 [89.29, 103.50]	100.00 [92.90, 107.10]	100.00 [92.90, 107.10]	
PCP WL	LISI	66.17 [58.69, 73.67]	83.03 [81.54, 96.53]	90.96 [83.47, 98.46]	88.52 [81.03, 96.02]	.682
	HISI	68.28 [61.43, 75.12]	97.85 [91.01, 104.69]	100.00 [93.16, 106.84]	100.00 [93.16, 106.84]	
PCP SL	LISI	71.82 [64.17, 79.48]	86.39 [78.34, 94.05]	84.32 [76.67, 91.98]	86.50 [78.84, 94.16]	.070
	HISI	67.07 [60.08, 74.06]	96.85 [89.96, 103.83]	100.00 [93.010, 106.99]	100.00 [93.010, 106.99]	
PCM WL	LISI	93.71 [81.41, 106.02]	98.71 [86.41, 111.01]	98.06 [85.76, 110.37]	100.00 [87.70, 112.30]	.448
	HISI	87.37 [76.14, 98.60]	99.46 [88.23, 110.69]	100.00 [88.77, 111.23]	100.00 [88.77, 111.23]	
PCM SL	LISI	95.67 [92.46, 98.89]	98.38 [95.16, 101.60]	98.37 [95.16, 101.60]	100.00 [96.78, 103.22]	.864
	HISI	98.65 [95.71, 101.58]	99.55 [96.61, 102.48]	100.00 [97.06, 102.94]	100.00 [97.06, 102.94]	

Table 7. (continued).

Variable	Group	Time	Comparison time within groups									
			Evolution Pre-T5				Evolution T5-T6				Evolution T5-T7	
			<i>p</i>	<i>p</i>	EM difference [95% CI]	<i>p</i>	Time x Group	EM difference [95% CI]	<i>p</i>	Time x Group	<i>p</i>	Time x Group
PCC-R WL	< .001 ^a	< .001 ^a	+22.38 [+10.67, +34.09]	.001 ^a	.115	+1.93 [-9.78, +13.65]	.738	.093	.892	.066		
			+28.00 [+19.42, +35.98]	< .001 ^a		+2.69 [-5.60, +10.97]	.514		.514			
PCC-R SL	< .001 ^a	< .001 ^a	+17.85 [+3.67, +32.02]	.015 ^a	.185	-2.07 [-16.25, +12.10]	.767	.040	.805	.116		
			+30.69 [+23.21, +38.15]	< .001 ^a		+3.60 [-3.87, +11.08]	.334		.334			
PCP WL	.002 ^a	< .001 ^a	+22.86 [+11.36, +34.35]	< .001 ^a	.220	+1.93 [-9.56, +13.43]	.733	.210	.929	.113		
			+29.57 [+20.23, +38.92]	< .001 ^a		+2.15 [-7.20, +11.50]	.643		.643			
PCP SL	.001	< .001 ^a	+14.57 [-0.25, +29.40]	.054	.009 ^a	-2.07 [-16.90, +12.75]	.766	.123	.988	.052		
			+29.77 [+23.64, +35.91]	< .001 ^a		+3.16 [-2.98, +9.29]	.303		.303			
PCM WL	.811	.085	+4.99 [-1.67, +11.66]	.136	.048	-0.65 [-7.31, +6.02]	.845	.485	.694	.592		
			+12.06 [-8.87, +33.06]	.249		+0.54 [-20.42, +21.50]	.956		.956			
PCM SL	.030	.306	+2.71 [-3.34, +8.74]	.367	.310	-0.01 [-6.04, +6.04]	1.000	.384	.587	.165		
			+1.35 [-1.50, +4.21]	.342		+0.45 [-2.40, +3.30]	.750		.750			

Note. EM = estimated mean; CI = confidence interval; PCC-R = percentage of correct consonants-revised; WL = word level; SL = sentence level; PCP = percentage of correct places; PCM = percentage of correct manners.

^aIndicates a post hoc significant effect ($\alpha < .017$).

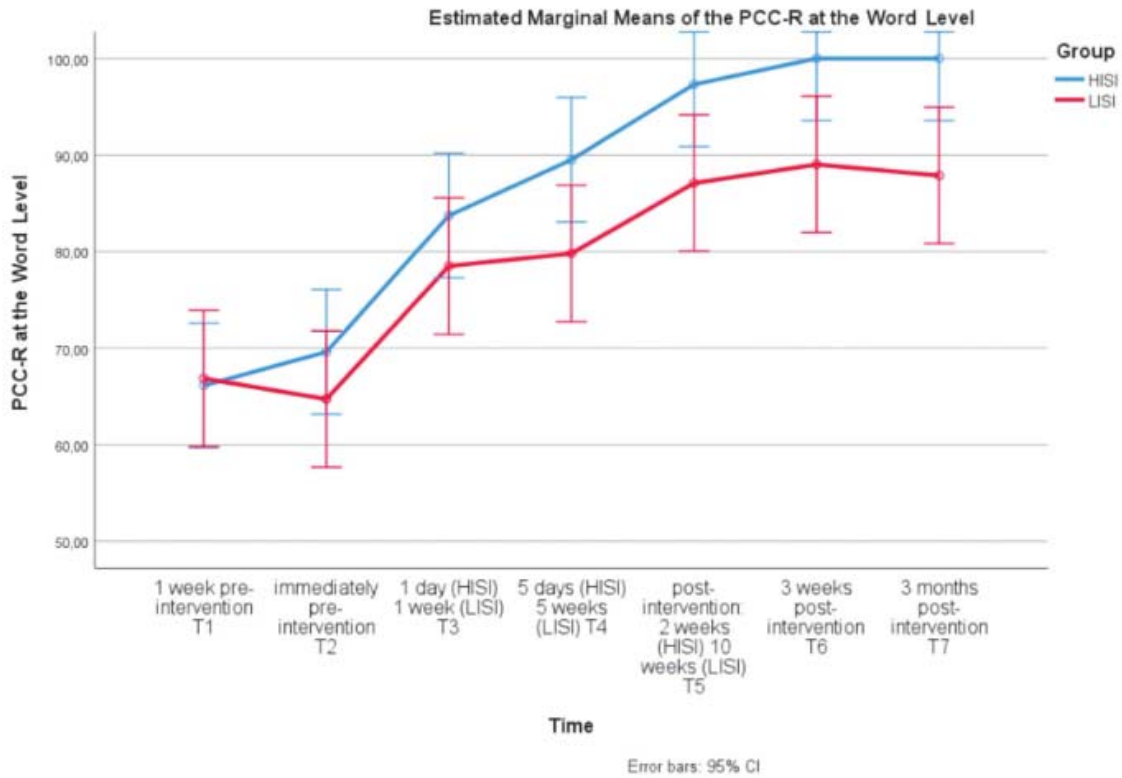


Figure 2. Evolution of the percentage of consonants correct–revised (PCC-R) at the word level. HISI = high-intensity speech intervention; LISI = low-intensity speech intervention; CI = confidence interval.

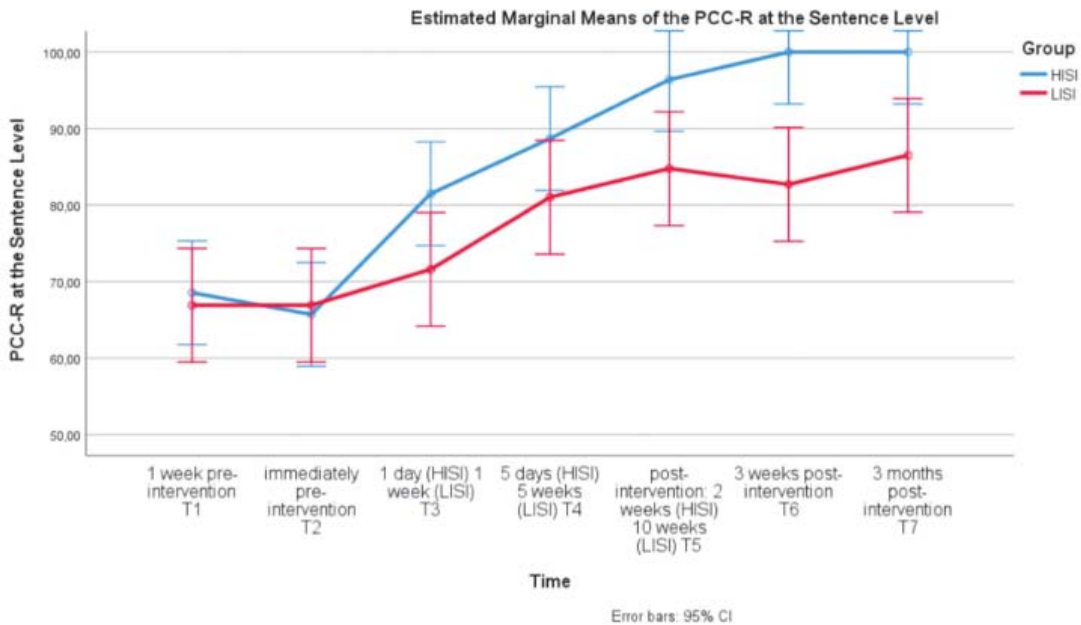


Figure 3. Evolution of the percentage of consonants correct–revised (PCC-R) at the sentence level. HISI = high-intensity speech intervention; LISI = low-intensity speech intervention; CI = confidence interval.

LMMs revealed a significant time effect for the variables “PCC-R at the word level and at the sentence level” (word level: $F(6, 63) = 26.333, p < .001$; sentence level: $F(6, 63) = 19.758, p$

< .001) and “PCP at the word level and at the sentence level” (word level: $F(6, 63) = 25.316$, $p < .001$; sentence level: $F(6, 63) = 16.556$, $p < .001$). The significant time effects indicated an improvement of these outcome measures irrespective of the group assignment. Post hoc analyses of the evolution between the pre-intervention values and the immediate post-intervention values revealed no significant Time \times Group effects for the variables “PCC-R at the word and sentence levels” (word level, $p = .115$; sentence level, $p = .185$) and “PCP at the word level” ($p = .220$). The lack of Time \times Group effects indicated that there were no significant differences in evolution between the HISI and LISI groups for these variables. For the variable “PCP at the sentence level,” a significant Time \times Group interaction with a large effect size was found for the evolution between the pre-intervention values and the immediate post-intervention values ($p = .009$, $d = 1.34$). This significant Time \times Group effect revealed that the PCP score at the sentence level significantly improved immediately following the intervention in the HISI group, but not in the LISI group ($p < .001$). This improvement remained in the short term (3 weeks post-intervention, $p = .303$) and in the long term (3 months post-intervention, $p = .303$).

Evolution of the HRQoL

A comparison of the HRQoL between the HISI and LISI groups is provided in Table 8. LMM revealed significant time effects for the total VELO scores of the parent report, $F(2, 30) = 4.906$, $p = .014$, and the subscales Speech Limitation, $F(2, 30) = 6.971$, $p = .003$, and Situational Difficulties, $F(2, 13) = 21.713$, $p < .001$. These effects indicate a positive evolution of these HRQoL measures over time.

Significant Time \times Group interactions with large effect sizes were observed for the pre–post evolution of the total VELO scores of the parents ($p = .010$, $d = 0.932$) and for the subscales Speech Limitations ($p = .002$, $d = 0.846$), Situational Difficulties ($p = .003$, $d = 0.963$), and Caregiver Impact ($p = .008$, $d = 0.868$). These scales only significantly improved following speech intervention in the HISI group (see Table 8).

Table 8. Comparison of the health-related quality of life between the low-intensity speech intervention (LISI) and high-intensity speech intervention (HISI) groups.

Variable	Group	Between-groups analyses					
		Pre (T2)	Post (T5) (immediately)	Follow-up (T7) (3 months)	Time \times Group	Group	Time
		EM [95% CI]	EM [95% CI]	EM [95% CI]	<i>p</i>	<i>p</i>	<i>p</i>
Parent–total	LISI	71.37 [60.57, 82.18]	81.20 [70.40, 92.01]	79.23 [68.43, 90.04]	.488	.353	.014 ^a
	HISI	68.14 [57.33, 78.94]	87.92 [77.11, 98.73]	87.99 [77.18, 98.79]			
Speech	LISI	56.55 [43.38, 69.72]	71.03 [57.86, 84.19]	74.41 [61.24, 87.57]	.671	.362	.003 ^a
	HISI	55.22 [42.05, 68.38]	81.18 [68.01, 94.35]	80.21 [67.05, 93.38]			
Swallowing	LISI	93.07 [82.51, 103.64]	93.78 [83.22, 104.35]	90.28 [79.71, 100.85]	.584	.878	.776
	HISI	88.51 [77.94, 99.07]	92.44 [81.87, 103.004]	93.40 [82.83, 103.96]			
Situational Difficulties	LISI	58.33 [42.81, 73.86]	76.67 [61.14, 92.19]	72.50 [56.97, 88.03]	.163	.270	< .001 ^a
	HISI	57.39 [41.86, 72.91]	91.27 [75.74, 106.80]	90.28 [74.76, 105.81]			
Emotional Impact	LISI	70.83 [53.37, 88.30]	77.54 [60.08, 95.00]	81.25 [63.79, 98.71]	.568	.331	.066
	HISI	78.08 [60.62, 95.55]	91.40 [73.94, 108.86]	91.46 [74.00, 108.92]			
Perception by Others	LISI	84.38 [71.37, 97.39]	88.75 [75.74, 101.76]	88.54 [75.53, 101.55]	.942	.704	.509
	HISI	88.50 [75.50, 101.51]	91.40 [78.40, 104.41]	90.63 [77.62, 103.64]			
Caregiver Impact	LISI	81.94 [66.87, 97.02]	83.89 [68.82, 98.97]	81.94 [66.87, 97.02]	.188	.748	.102
	HISI	74.62 [59.54, 89.70]	91.41 [76.32, 106.48]	90.42 [75.34, 105.50]			
Child–total	LISI	80.95 [58.90, 102.99]	— ^b	— ^b	—	—	—
	HISI	82.15 [66.56, 97.74]	93.04 [77.45, 108.63]	93.16 [77.57, 108.74]			

Table 8. (continued).

Variable	Comparison time within groups							
	Evolution pre-post		Time x Group	Evolution pre-follow-up		Evolution post-follow-up		Time x Group
	EM difference [95% CI]	p		EM difference [95% CI]	p	EM difference [95% CI]	p	
Parent-total	+9.83 [-7.63, +27.30]	.249	0.010 ^a	+7.86 [-9.60, +25.32]	.352	-1.97 [-19.43, +15.49]	.813	681
	+19.78 [+5.50, +34.06]	.010 ^a		+19.85 [+5.57, +34.13]	.010 ^a	+0.07 [-14.21, +14.35]	.992	
Speech	+14.48 [-5.22, +34.17]	.138	0.002 ^a	+17.86 [-1.84, +37.55]	.072	+3.38 [-16.32, +23.08]	.720	0.885
	+25.96 [+6.79, +45.13]	.011 ^a		+24.99 [+5.83, +44.17]	.014 ^a	-0.97 [-20.13, +18.20]	.916	
Swallowing	+0.71 [-13.16, +14.59]	.909	0.630	-2.79 [-18.31, +12.72]	.578	-3.51 [-17.38, +10.37]	.578	0.598
	+3.93 [-1.93, +9.79]	.163		+4.89 [-2.76, +12.54]	.173	+0.96 [-4.90, +6.82]	.719	
Situational Difficulties	+18.33 [+5.95, +30.72]	.008	0.003 ^a	+14.17 [-4.34, +32.67]	.115	-4.17 [-16.55, +8.22]	.468	0.928
	+33.88 [+20.98, +46.79]	<.001 ^a		+32.90 [+16.83, +48.96]	.002 ^a	-0.99 [-13.89, +11.92]	.866	
Emotional Impact	+6.71 [-8.90, +22.31]	.360	0.066	+10.42 [-14.70, +35.53]	.368	+3.71 [-11.90, +19.31]	.608	0.629
	+13.32 [+6.23, +20.41]	.002 ^a		+13.38 [+2.76, +23.99]	.002 ^a	+0.06 [-7.03, +7.14]	.987	
Perception by Others	+4.38 [-15.59, +24.34]	.609	0.861	+4.17 [-7.60, +15.93]	.419	-0.21 [-20.18, +19.76]	.980	0.642
	+2.90 [-1.75, +7.56]	.194		+2.13 [-6.04, +10.29]	.548	-0.78 [-5.43, +3.88]	.716	
Caregiver Impact	+1.95 [-12.05, +15.95]	.758	0.008 ^a	+0.00 [-16.94, +16.94]	1.000	-1.95 [-15.95, +12.05]	.758	0.477
	+16.79 [+4.57, +29.01]	.013 ^a		+15.80 [+0.09, +31.51]	.049	-0.99 [-13.21, +11.24]	.859	
Child-total	+10.83 [+2.82, +18.94]	.018		+10.95 [-5.01, +26.91]	.110	+0.07 [-9.03, +9.17]	.986	

Note. EM = estimated mean; CI = confidence interval.

^aStatistically significant effect based on linear mixed models ($\alpha < .017$). ^bSample size was too small to calculate the estimate mean.

In the HISI group, five children were 8 years old or older and completed the VELO child report. In the LISI group, only two children were older than 8 years of age and completed the VELO child report. Because of this small sample size, no time, group, and Time × Group effects could be calculated.

Home Practice

Based on the reports in the log books, the mean time of home practice was 35.00 min per week in the HISI group ($SD = 27.386$ min, range: 0–60 min) and 45.83 min per week in the LISI group ($SD = 33.229$, range: 0–100 min). LMM revealed no significant group effects, $F(1, 63) = 0.297, p = .588$, nor any Time × Group effects, $F(6, 63) = 0.901, p = .500$, for the variable “home practice.”

Discussion

At present, there is a lack of evidence on the effect of intervention intensity in children with a CP ± L (Bessell et al., 2013). The lack of evidence-based practice forces us to ask the question: “Are we applying the available resources in the most effective way?” To respond to this issue, this study compared the effect of speech intervention provided with a low intensity and speech intervention provided with a high intensity on the speech and HRQoL in Dutch-speaking children with a CP ± L. Based on previous studies, it was hypothesized that HISI may be equally effective or even more effective in reducing active or compensatory speech errors and in increasing the child's consonant proficiency and HRQoL compared with LISI.

The hypothesis was confirmed by the results of this study. Our findings indicated that consonant proficiency (in terms of PCC-R at the word level and at the sentence level) significantly improved in the HISI group and in the LISI group (see Table 7). Strikingly, children in the HISI group made greater progress in a shorter amount of time: Children in the HISI group received 2 weeks of speech intervention compared with the children in the LISI group who received 10 weeks of speech intervention (see Tables 6 and 7 and Figures 2 and 3). This greater progress was not only reflected by the PCC-R measures but also by the

significant decrease in the occurrence of anterior oral and nonoral CSCs. These improvements were maintained in the short term and also in the long term. This finding might seem surprising since the children in the HISI group only received 2 weeks of actual treatment. Considering the positive long-term results in the HISI group, it seems that the learning effect of the short (but high-intensive) intervention remained present. Interestingly, this finding is in line with previous research in the area of voice therapy. Wenke et al. (2014) compared the effect of high-intensity voice therapy (four 1-hr treatment sessions per week during 2 weeks) with the effect of low-intensity voice therapy (i.e., one 1-hr treatment session per week during 8 weeks). Positive long-term results were observed in the group who received high-intensity voice intervention. The authors assumed that this positive follow-up might indicate that the patients were capable of progressive self-management of their voice in the absence of any treatment. In this study, these positive long-term results might perhaps be related to the emphasis that was placed on metaphonological knowledge and self-correction during the intervention period. Two weeks of intensive metaphonological intervention seems enough to stimulate the phonological systems to maintain the learning effect. In general, the findings of this study confirm earlier reports, which suggested that more intensive intervention results in better speech outcomes in the short term and in the long term (Albery & Enderby, 1984; Allen, 2013; Cummings et al., 2021). HISI minimizes the amount of rest between therapy sessions so there is more time on task than there is time spent on rest (Magill & Anderson, 2010; Muratori et al., 2013). Thereby, HISI is hypothesized to increase learning opportunities (Magill & Anderson, 2010; Muratori et al., 2013). It should also be noted that the variable “posterior oral CSCs” significantly improved following LISI, but not following HISI (see Table 6). Elimination of the error “backing to a velar place of articulation,” which is a posterior oral CSC, was a treatment goal in two children ($n = 1$ in the LISI group and $n = 1$ in the HISI group). The results for this speech outcome might have been different if more children with posterior oral CSCs were included in the sample.

In the literature, there is growing consensus that speech outcomes should not only include individual measures of speech (e.g., articulation, which was reflected by the measures of consonant proficiency and CSCs in this study) but should also be extended to global speech outcome measures (e.g., speech understandability and speech acceptability; Lohmander et al., 2009). Interestingly, the variables “speech understandability” and “speech acceptability” significantly improved in the HISI group, but not in the LISI group (see Table 6). This study demonstrated that more global (holistic) outcome measures can improve following 2 weeks of intensive speech intervention. One of the primary goals of speech intervention in children with a CP ± L is to ensure that their speech is understandable and acceptable to others (Howard & Lohmander, 2011; Nagarajan et al., 2009). Since HISI improves these important speech outcomes, it can be considered a promising intervention model.

Beside improving speech outcomes, speech intervention aims to improve quality of life (Howard & Lohmander, 2011). Therefore, it is important to determine to what extent the better speech outcomes are also meaningful to our patients. To assess changes in the patient's HRQoL, the VELO questionnaire was included in this study (Bruneel, Alighieri, et al., 2019; Bruneel et al., 2017; Skirko et al., 2012, 2013). The analyses were based on the parent reports because the sample size was too small to perform analyses on the child reports ($n = 5$ children in the HISI group and $n = 2$ children in the LISI group). Bruneel et al. (2017) demonstrated that the parent proxy assignment was a valid and reliable tool for the assessment of the child's HRQoL. The findings revealed that the total VELO scores of the parents and the scores for the subscales Speech Limitations, Situational Difficulties, and Caregiver Impact only significantly improved in the HISI group (see Table 8). The different subscales of the VELO

questionnaire can be considered in light of the International Classification of Functioning, Disability and Health (ICF; Neumann & Romonath, 2012; World Health Organization, 2001). The subscale Speech Limitations is related to the ICF domain “body functions” and maps specific speech difficulties that can be experienced. The subscale Situational Difficulties is related to the ICF domains “activities and participation” and “environmental factors,” questioning the patient's speech understandability as experienced by different listeners (family, friends, and strangers) in different situations (in the car and on the phone). The subscale Caregiver Impact is associated with the ICF domain “environmental factors” and identifies the impact of the speech disorders on the caregiver. According to the parent reports, the effect of HISI was twofold: (a) HISI decreased the occurrence of speech difficulties and their impact on the caregivers, and (b) HISI increased the child's participation in society. It should be acknowledged that the VELO scores also improved in the LISI group, but these improvements were not as pronounced as the improvements in the HISI group (see Table 8). The finding that LISI does not result in a significant improvement of VELO scores was in line with previous research. Bruneel, Alighieri, et al. (2019) compared changes in parent VELO scores between parents whose children received 1 year of LISI (with a session duration and dose frequency of two 30-min sessions per week or one 1-hr session per week) and parents whose children did not receive speech intervention. Remarkably, no significant difference in the evolution of the VELO scores was observed between the two groups. This finding forces us to reconsider the LISI models. It may be questioned whether parents' familiarity with the VELO questionnaire (especially in the HISI group where parents completed the questionnaire twice in a short time: before the intervention and following the 2-week intervention) may have impacted the results. Since Bruneel, Alighieri, et al. (2019) demonstrated the 2-week test–retest reliability of this instrument, these results may be considered reliable. Parents were blinded to the specific study purposes (i.e., the comparison of HISI with LISI), but they were not blinded to the treatment intensity of their own child. They obviously knew how much therapy their child received. Parents' perceptions on the treatment intensity may possibly have influenced the VELO scores.

In summary, the results of this study suggest that HISI is more effective in reducing active or compensatory speech errors and in increasing the child's consonant proficiency and HRQoL compared with LISI. In literature, several benefits of HISI have been described. HISI may not only result in a faster speech improvement, but other studies argued that HISI may also reduce treatment fatigue and the financial costs of speech intervention for the patient, the health insurances, and the society (Makarabhirom et al., 2015; Pamplona et al., 2005). The implementation of HISI in clinical practice will thus have several consequences for the child and their family, for the SLP, and for the health care regulations. For health care to be truly evidence based, there needs to be an integration and harmonization of (a) the health practitioner's expertise and opinions; (b) the patient's needs, values, and preferences; and (c) the best available research evidence (Steglitz et al., 2015). A certain intervention intensity will only be successful if the child and their family are willing to participate and are able to tolerate the prescribed intervention intensity level. Beside information on the patients' experiences with the different intervention intensities, we must also have insight in the SLPs' opinions and how they make decisions regarding intervention intensity. An understanding of this decision-making process is essential before HISI can really be applied in clinical practice. Several authors report that SLPs seem to rely on practical factors to make decisions on intervention intensities, for example, caseload size and scheduling constraints (Brandel & Loeb, 2011; Brumbaugh & Smit, 2013; Farquharson et al., 2020). For some clinicians, it might be difficult to implement HISI in their busy schedule. Therefore, we must carefully consider how to support a cultural shift away from the traditional “one or two therapy

sessions per week for many years” models, which are currently predominant (Hegarty et al., 2018). As suggested before in other domains of speech therapy, for example, in voice therapy, the golden mean between HISI and LISI might be an achievable and effective solution for clinical practice (Meerschman et al., 2019).

This innovative study was one of the first to compare the effect of HISI with the effect of LISI in children with a CP ± L. This study responded to the shortcomings of earlier investigations. An important strength of this study is that we accounted for the different intervention intensity parameters as recommended by Warren et al. (2007). In addition, a condition-specific patient-reported outcome measure (i.e., the VELO questionnaire) was included to assess the value placed on the results by the parents. The design of this study was a longitudinal, prospective, randomized controlled trial with multiple baselines and post-intervention data points. The three post-intervention data points provided the opportunity to assess the short- and long-term effects of the HISI and LISI interventions. Previous studies that have investigated treatment intensity in children with SSD or in children with a CP ± L often did not report if there was long-term maintenance of the observed effects (Bessell et al., 2013; Kaipa & Peterson, 2016).

Despite the strengths and the innovative character of this study, there were some limitations that should be acknowledged. This study included a small sample, which may hamper generalization of the results. Because of the unavailability of other SLPs, some data were collected by the same SLP who provided speech intervention to the children in the LISI and HISI groups. By including two blinded raters who did not provide speech intervention to the included children and who were unaware of the treatment allocation, we responded to this limitation. The reliability results demonstrated good to very good inter- and intrarater reliability for the majority of the outcome variables (see Table 5). Another limitation was the variability in the intervention intensity parameter “dose” (i.e., the number of properly administered teaching episode or trials during an intervention session; Warren et al., 2007). In accordance with the recommendations of Williams (2003, 2005), the dose ranged from 80 to 100 responses per session. Children who received a higher dose might have had extra time on task. Future studies should therefore control for this variable. Despite that this study controlled for the amount of home practice by including a log book, parents did not go through a training session with the SLP. They only received information about the used treatment techniques. Hence, some parents may have reinforced incorrect behaviors, and home practice could potentially be counterproductive in these cases. Future studies can include a parent training program to control for this possible source of bias. An example of such parent training program is provided by Sweeney et al. (2020). Because an individualized treatment plan was developed for each child, the type and the amount of target consonants differed between the children (see Table 3). In some children, only three consonants were targeted, which might have resulted in deeper learning compared to children in whom more consonants were targeted. Replication in larger samples, accounting for the type and amount of CSCs, is necessary in order to confirm our results. Controlling for the type of CSCs will also deepen our understanding of the most effective intervention for a given patient (i.e., “what works for whom”).

Conclusions

This study was the first to compare the effect of HISI with the effect of LISI in children with a CP ± L using a longitudinal, prospective, randomized controlled trial with a multiple-baseline design. The study responded to the shortcomings of earlier investigations by

including patient-reported outcome measures and taking into account the intervention intensity framework. The findings revealed that children in the HISI group made superior progress in only 2 weeks of therapy compared to children in the LISI group who needed 10 weeks of therapy. HISI is thus a promising strategy to improve speech outcomes in a shorter time period. The implementation of HISI in clinical practice will have some consequences for the different stakeholders. Before HISI can be applied in everyday clinical practice, we must carefully consider how to support a cultural shift away from the traditional “one or two therapy sessions per week for many years” models, which are currently predominant. In the future, attention should be paid on how the patients, their families, and the SLPs perceive and experience HISI.

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