Critical Review:

For individuals with Parkinson's disease whose speech characteristics include both hypophonia and palilalia, does use of a pacing board facilitate speech intelligibility?

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A tool used in a variety of speech disorders involving rate and repetitive speech phenomena is the pacing board. Though therapeutic use of the pacing board has been described since the late 1970s, only one quantitative case report and a few brief case reports examine outcomes of pacing board use with individuals with Parkinson's disease (PD) who exhibit speech characteristics of hypophonia and palilalia (PD+HpPa). This paper critically reviews peer-reviewed accounts of pacing board use with this population, and presents a new case report of a client with PD+HpPa who received Lee Silverman Voice Treatment (LSVT) and rate control therapy using a pacing board. Results were inconclusive due to methodological and data reporting inconsistencies among reports. Some recommendations are made for future research that would establish a higher level of evidence.

Introduction

Of the seven million individuals worldwide with PD, up to 89% experience disordered communication (Ramig, Fox, & 2008). Surveys of Parkinsonian Sapir, speech characteristics suggest that about 20-28% of this group exhibit rate control deficits and/or repetitive speech phenomena (Logemann, Fisher, Boshes, & Blonsky, 1978), and that 42-49% experience a reduction in speech intensity known as hypophonia (Ludlow, Bassich, McNeil, Rosenbek, & Aronson, 1984; Gamboa et al., 1997). A frequently observed type of repetitive speech in PD is palilalia: the compulsive repetition of context-appropriate words and syllables which may be accompanied by progressively decreasing vocal loudness, increasing rate (Benke, Hohenstein, Poewe, & Butterworth, 2000), amelodic prosody (Fleet & Heilman, 1985), and difficulty initiating speech (Van Borsel, Schelpe, Santens, De Vos & De Vos, 2001). Some individuals who seek treatment for PD-related speech disturbance will present with both hypophonia and palilalia (PD+HpPa).

Treatment for PD typically includes Carbidopa-Levodopa drugs to manage dyskinesia, though these drugs have been linked to decreased speech intelligibility and increased palilalia (Ackermann, Ziegler, & Oertel, 1989). Ackermann et al. (1989) have proposed that palilalic speech seen in some individuals with PD may be attributed to impairment of inhibitory motor pathways that terminate speech motor plans, though Van Borsel, Bontinck, Coryn, Paemeleire, & Vandemaele, (2007) conclude that the precise pathogenesis of palilalia remains unknown.

The pacing board is the instrumental basis of a widelypracticed method of pacing therapy which was first documented by Helm (1979). Helm's pacing board consisted of a sectioned board used to control rate by instructing the client to mark utterances at the syllable level with finger tapping. It has been proposed that purposeful pacing motions could act to scaffold impaired inhibitory motor circuits which normally terminate utterances (Luria, 1967). As such, use of a pacing board may be beneficial in therapy addressing repetitive speech in patients with PD, a question considered in the present paper.

LSVT has been subject to extensive study as an intervention method for PD-related speech disturbance (Mahler, Ramig, & Fox, 2015; Sapir, Spielman, Ramig, Story, & Fox, 2007; Ramig, Fox, & Sapir, 2008). Advocates of LSVT identify the systematic manipulation of speech intensity as the best target for improving speech intelligibility in people with PD (Ramig et al., 2008), whereas others consider rate control to be the single most modifiable variable (Yorkston, Dowden, & Beukelman, 1992). Positive outcomes with the LSVT protocol in increasing speech intensity in the majority of people with PD are well documented in the literature (Mahler et al., 2015; Sapir, et al., 2007; Ramig et al., 2008), though Sapir et al. (2007) found that LSVT is most effective for people with mild-moderate speech disturbance. Furthermore, Benke et al. (2000) found that individuals with palilalia are far more likely to be among the advanced PD group, and have severe speech disturbance. It would follow from this that those with both hypophonia and palilalia may benefit from a therapeutic approach combining speech intensity and rate control treatments such as LSVT and a pacing board. The present study addressed this proposal in both a critical review of the literature (Study 1) and case report (Study 2).

Objectives

This critical review examined published case reports involving the use of a pacing board in speech therapy for individuals with PD+HpPa (Study 1). A second objective was to examine the effectiveness of LSVT and pacing therapy using a pacing board with an individual with PD+HpPa in a case report (Study 2).

Study 1

Search Strategy

Computerized databases including PubMed and CINAHL as well as ASHA publications were searched using the following search strategy: [(palilalia) OR (palilalic) AND (Parkinson's) AND ("pacing board") OR (pacing)]. Reference lists of previously searched articles were also used to obtain other relevant studies.

Selection Criteria

Articles were chosen that included reports of clinical treatment and outcomes of individuals with PD+HpPa where the treatment included use of a pacing board. Studies describing therapy techniques in the absence of a specific case description or study were excluded. Anecdotal reports from non-peer reviewed sources such as blogs and best-practice guidelines, though copious, were also excluded

Study 1 Results

The majority of studies included in this review are case reports (Helm 1979; Van Borsel et al., 2007; Lang & Fishbein, 1983). By definition, a case report provides retrospective information about treatment of a client which does not normally include comparisons with a control group, or any experimental manipulation. Case reports are thought to have relatively low internal validity and are rated evidence level 4 according to *The Oxford Centre for Evidence-Based Medicine*, since numerous confounding variables could contribute to the outcomes observed. (Phillips, Ball, Badenoch, Straus, Haynes, & Dawes, 2011). A clinician might use a case report to present an account of a technique or tool that was effective for a single client, which could act as a catalyst for further investigation.

Case reports

Helm (1979): This case report concerns a 54-year-old male described as having slow-progressing Parkinson's disease. His speech was characterized by severe palilalia, such that he was non-communicative, however the client was not observed to be palilalic when performing categorical naming tasks of single-syllable items. Helm trialed metronome pacing and hand tapping, which proved ineffective. A pacing board, described as an 8-segment 13x2 inch apparatus with coloured segments separated by wooden dividers, allowed the client to speak syllable-bysyllable, mirroring the incremental movement of his finger along the board, "without exhibiting palilalia". The client was reportedly able to communicate effectively in the clinic, but needed prompting to use the pacing board when having conversations outside the clinic room. By the end of treatment, the client learned to communicate functionally using syllabic speech facilitated by the pacing board, but needed continuous prompting to use the device.

This case report provides qualitative testimony of a technique for controlling palilalic utterances through syllabic speech. The author included a detailed description of the pacing board she designed, but did not provide any quantitative analysis of the client's baseline speech or progress, and few specifics about the therapy administered. Overall, this report provides suggestive evidence that the external cueing provided by the pacing board could help to reduce repetitive speech.

Lang & Fishbein (1983): This brief case report concerns a 53-year-old male with PD whose speech characteristics included rushed speech, severe palilalia, and frequent hesitations averaging six seconds in duration. In addition, he was found to exhibit hypophonia, particularly during Levodopa "off" phase. These characteristics resulted in low intelligibility, with rate of speech being "30% of normal values". When training using a pacing board was introduced, speech intelligibility improved by 63%.

Though an effort is made at reporting pre-post speech characteristics, the author does not report which speech parameters were measured, or how they were measured. As a result, this case report provides equivocal evidence concerning the utility of pacing boards in reducing repetitive speech.

Van Borsel et al. (2007): This case report, contained within a larger study of palilalic speech characteristics, concerns a 60-year-old male with a diagnosis of Idiopathic Parkinson's disease who exhibited severe palilalia and reduced speech intensity. Over a period of two years, the client received speech therapy (three sessions of 45 minutes per week) that aimed to decreasing overall speech rate and reduce the number of repetitions during palilalic utterances. Active pacing was trailed using a pacing board, and passive pacing was trailed using a metronome. The pacing board had an "immediate and marked" positive effect on speech intelligibility and reduction of repetitions; the metronome was judged to be ineffective. Despite intensive training with the pacing board, the client was not able to perform syllabification consistently without maximal clinician cueing, and eventually abandoned oral communication in favor of a speech generating device.

This report includes a qualitative description of pacing board therapy with a client presenting with PD+HpPa. The report lacks detail regarding methodology used in therapy, and any use of quantitative pre/post measurements. This case provides somewhat suggestive evidence that pacing boards may be effective with this population, at least in the short-term. Suzuki et al. (2013): This single-subject study involves a 61-year-old male Japanese speaker with a diagnosis of PD, who developed palilalia subsequent to implantation of a deep brain stimulator. It uses perceptual measures of intelligibility as an indicator of progress. Sixteen features¹ of the client's speech were rated on a 0-4 scale ("0" being normal; "4" being severely abnormal) by three independent speech-language pathologists, and the average ratings were reported for each feature. Treatment included the LSVT protocol and pacing board training. Perceptual ratings were carried out under three conditions: before treatment, after 16 sessions of LSVT, and after treatment with a pacing board. Prior to treatment, the most disordered features were impaired loudness (rated as 1), rough hoarseness (rated as 1.5) abnormality of utterance speed (rated as 2), changes in utterance speed (rated as 1.5) and repetitive speech phenomena (rated as 3). After 16 sessions of LSVT, all of these features had improved somewhat, however rough hoarseness, abnormality of utterance speed, and changes in utterance speech continued to be rated mildly abnormal (both rated as 1), and repetitive speech phenomena was rated moderately abnormal (with a rating of 2). After introducing the pacing board, and training the client to mark rhythmic units, perceptual ratings of all features approached "0". The authors report that gains in speech production were contingent upon continued use of the pacing board, and could not be reproduced without its use.

This study used a multiple-baseline pre-post design. The use of pre-post perceptual ratings is an ecologically valid way to measure change, since improved intelligibility is the desired outcome of most speech therapy. Attempts are made at maximizing internal and external validity of the data collected by giving detailed explanations of methodology and rating systems used. Nonetheless, some aspects of the study could have been improved. Firstly, use of a visual analogue scale may have resulted in greater degree of accuracy than the rigid 0-4 point scale. Additionally, researchers did not control for the cumulative effect of LSVT and pacing, since the two conditions were carried out consecutively. Nevertheless, this study provides highly suggestive evidence of the facilitative benefits of the pacing board and the advantages of combining it with LSVT for patients with PD.

Group study

Van Nuffelen et al. (2009): This non-randomized, controlled pre-post study investigated the outcomes of seven rate

control methods (RCMs), including pacing board use, on commonly employed measures of speaking rate and articulation rate (SR; AR), and intelligibility in 19 individuals with dysarthria, six of whom presented with PD-related hypokinetic dysarthria. Outcome measures were compared to an unaffected age-matched control group. For each RCM condition, participants were provided with a brief orientation to the RCM and each participant received the same reading materials. The order in which the RCMs were trialed was randomized. Intelligibility was rated by five SLPs using a visual analogue scale. Strong inter-rater reliability was reported (.85). Appropriate statistical analyses included ANOVA with adjustments for multiple comparisons. Results indicated that pacing board use resulted in significantly slower speech and articulation rates in comparison to the habitual rates. Strikingly, however, the rate control methods did not result in significant increases in intelligibility across participants. In individual analyses, only 2/6 PD participants met the cutoff determined for a significant change in intelligibility.

Overall, the results provide suggestive evidence that pacing boards are among the RCMs that are effective in controlling rate. However, addressing rate alone may not yield significant gains in intelligibility, which suggests that targeting more than one parameter (ex: both speech intensity and pacing) might result in more functional speech. However, the extent to which these results specifically apply to individuals with PD+HpPa is uncertain given the lack of detail regarding participants.

Discussion

It is clear that the available evidence regarding combining treatments targeting speech intensity and rate in individuals with PD+HpPa is limited. Nonetheless, most of the reviewed studies provide suggestive evidence of potential benefits, which may guide clinicians in providing clinical services as was the case in the following case report.

Study 2

The following case study reports a therapeutic approach combining LSVT and pacing board use.

Participant

The participant was a 64-year-old male with an approximately 10-year history of speech disturbance related to PD and demonstrating both palilalia and hypophonia (PD+HpPa). Speech intelligibility was severely impaired, and the participant reported being unable to carry out basic daily communication tasks. The participant reported using Carbidopa-Levodopa medication to manage dyskinesia, and indicated that his ability to speak was significantly reduced in the "off" phase. No co-morbidities were reported.

¹ The characteristics rated were: short rushes, impaired loudness, rough hoarseness, breathy hoarseness, asthenic hoarseness, strained hoarseness, abnormality of pitch, vocal tremor, hypernasality, distorted vowels, abnormality of utterance speed, change of utterance speed, repetitive speech phenomena, monoloudness, monopitch, excessive change of loudness.

Procedures

The participant completed a total of 22 therapy sessions, which included eight 60-minute LSVT-only sessions, eight 60-minute LSVT+pacing therapy sessions, and six 60-minute sessions of exclusively pacing therapy.

Objective measures

Repetitive speech: A spontaneous monologue sample was recorded pre- and post- intervention, and the number of partial and whole word repetitions was counted (Rep/M). Rep/M was also determined for each intervention session based on a set of 10 client-generated utterances pertaining to daily life. For the counting of Rep/M, the number of repetitions were recorded by two independent listeners, and the results averaged.

Speech intensity: Average speech intensity was determined in decibels (db SPL) using LSVT Companion software for connected speech tasks (client-generated functional phrases, speech hierarchy, spontaneous speech) throughout the LSVT portion of the intervention.

Perceptual ratings

Three unfamiliar listeners, all of whom were speechlanguage pathology graduate students, were provided with a recording of the participant's spontaneous monologue (pre and post intervention) and instructed to rate the following parameters on a visual analogue scale (0-1): overall intelligibly (poor to good), repetitive speech (very distracting to normal), and speech intensity (highly abnormal-normal). The raters were instructed that that a rating of 1 would indicate perceptually normal speech, whereas 0 would indicate severely disordered speech.

Results

Figure 1 shows the speech fluency and intensity results from pre-assessment, and throughout the 22 intervention sessions. Initial improvements in speech intensity were not sustained across the initial eight sessions, and the pacing board was introduced during session nine to control repetitions. The pacing board had an immediate positive effect on Rep/M, and a decrease of 16 Rep/M from baseline was observed. Overall speech intensity, however, declined to 68.4 dB SPL, which is below baseline. Additionally, the client reported feeling very fatigued. In consideration of the high level of effort required to attend to both loudness and pacing cues, it was decided to restrict cueing mainly to pacing.

During sessions 10 to 16, speech intensity increased to an average of 74.25dB SPL, 5.25dB SPL from baseline, which marginally surpassed gains achieved during the LSVT-only condition. Rep/M, however, dramatically decreased, with mean reduction of 25 Rep/M as compared to baseline.

Upon completion of the 16-session LSVT protocol, an additional 6 sessions focusing exclusively on pacing were carried out. With minimal verbal feedback and modeling, repetitions were reduced to 9 Rep/M during the FP task by end of treatment. A transient spike in repetitions was observed in session 21, which is thought to correspond to client reports of irregularities in timing of Carbidova-Levodopa administration.

Perceptual ratings at pre-assessment were: overall intelligibility, .32; repetitive speech, .23; speech intensity, .55. At post-treatment, they were: overall intelligibility, .68; repetitive speech, .80; speech intensity, .65.

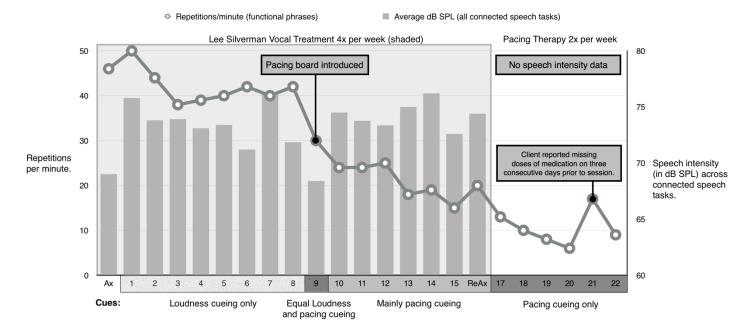


Figure 1. Results of a 22-session block of speech therapy utilizing LSVT and pacing board therapy.

Discussion

During this course of treatment, the client received a combination of LSVT and pacing therapy. In the LSVT-only condition, modest improvements were observed in both dB SPL and Rep/M, and a pacing board was introduced when palilalic repetitions severely hindered the client's ability to produce functional speech. The pacing board resulted in immediate reduction of Rep/M, however speech intensity initially dropped below pre-treatment baseline. During the LSVT+pacing condition, intensity levels recovered, and Rep/M decreased sharply. Finally, the pacing-only condition resulted in even greater reductions in Rep/M. Though the relative contributions of intensity and Rep/M to intelligibility are unclear, pre-post perceptual intelligibility ratings suggest a marked improvement in overall intelligibility.

The data presented here is subject to a number of flaws. Firstly, Rep/M was measured using the same set of ten phrases, repeated every session. Therefore, a practice effect could have contributed to the reduction in repetitions. Additionally, since the pacing board was introduced concurrently with another treatment (LSVT), it cannot be concluded that improvements in intelligibility were due to the pacing board alone. Carbidova-Levodopa drugs that the client self-administered daily have been associated with reduced speech intelligibility and increased palilalia (Ackermann et al., 1989). On one occasion, the client reported having missed doses of his medication for three consecutive days, which could have impacted performance.

General Discussion

The case reports discussed, including Study 2, are subject to a variety of methodological omissions and inconsistencies, making it untenable to either accept or fail to reject the null hypothesis. However, converging evidence, both qualitative and quantitative, seem to suggest that use of a pacing board with individuals presenting with PD+HpPa could have a positive effect on speech intelligibility. In particular, congruencies between the data presented in Study 2, and the findings of Suzuki et a. (2013) suggest a facilitating effect of a pacing board when combined with LSVT in treatment of individuals with PD+HpPa. In both cases, clients experienced markedly greater reductions in palilalic utterances after treatment with LSVT+pacing board than they did in the LSVT-only condition. The only evidence for use of a pacing board as the primary intervention method, however, is found in a series of subjective case reports. Helm (1979), Lang & Fishbein (1983), Van Borsel et al. (2007) all describe using a pacing board with a degree of success, but lack a description of therapy conditions used to achieve the reported improvements. Furthermore, Van Nuffelen et al. (2009) found that the improvements in rate control do not necessarily translate in to increased intelligibility.

Researchers affiliated with LSVT Global have frequently cited the Helm (1979) pacing board case report to support the claim that therapy techniques addressing rate have "modest and short-term results" (Ramig et al. 2004; Sapir et al., 2011; Trail et al., 2005), however a webinar available on the LSVT Global website² demonstrates a type of pacing board used in therapy with an individual with PD+HpPa. Similarly, Guidelines for Speech-Language Therapy in Parkinson's Disease issued by the Dutch Association of Logopedics and Phoniatrics states "When...LSVT does not sufficiently help to counteract accelerated speech, the use of a pacing board...can be considered"3. These expert opinions, combined with an overwhelming volume of other non-peer-reviewed accounts available online, suggest that the pacing board continues to be a widely recommended form of intervention with this population despite a dearth of high quality evidence.

Features of future studies providing a higher level of evidence might include: larger sample sizes, randomized controls, reporting of both objective and perceptual measures, detailed documentation of measurement techniques, and post-treatment follow-up to determine the long term benefits of pacing therapy.

Clinical Implications

- A combined approach including LSVT and a pacing board may result in better speech intelligibility outcomes for individuals with PD+HpPa, though further research is needed.
- Only subjective and anecdotal evidence exists to support pacing board use as a primary intervention method for individuals with PD+HpPa. Addressing rate in isolation does not necessarily result in improved intelligibility.
- Clinicians could consider trialing a pacing board with individuals presenting with PD+HpPa in conjunction with LSVT, particularly if initial progress is slow.

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² The e-seminar entitled "How LSVT LOUD and LSVT BIG Benefit Persons with Advanced Parkinson Disease" can be found at <u>https://www.lsvtglobal.com/patient-resources/free-webinars</u>. Supporting studies cited in the webinar do not address the pacing technique demonstrated, and thus did not meet criteria for this review

³ Retrieved from

 $http://www.parkinsonnet.de/media/11927204/guidelines_for_speech-language_therapy_in_parkinson_s_disease.pdf.$

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