Prosthodontic management of hypernasality: Two very different cases

Gestion de l'hypernasalité à l'aide de prothèses vélopalatines: deux cas très différents

Tim Bressmann[1, 2]
James D. Anderson[2, 3]
Robert P. Carmichael[2, 4]
Christina Mellies[5]

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[1]Department of Speech-Language Pathology, University of Toronto, Toronto, Ontario

[2]Faculty of Dentistry, University of Toronto, Toronto, Ontario

[3]Craniofacial Prosthetic Unit, Odette Cancer Centre, Sunnybrook Health Sciences Centre, Toronto, Ontario

[4]Department of Dental Services, Holland Bloorview Kids Rehabilitation Hospital, Toronto, Ontario

[5]Hamilton Health Sciences Centre, Hamilton, Ontario

Abstract

This study describes two cases of structurally related hypernasality that were treated with speech bulb prostheses. The first case, a young woman with hypernasality of unknown etiology, was treated with a combined palatal lift and speech bulb in order to improve velopharyngeal closure and oral-nasal balance in speech. In order to help her maximize the benefit of the prosthesis, the patient practiced with online nasalance and pitch biofeedback. However, it was only when she resorted to a vocal play manoeuvre that she was able to consistently improve her velopharyngeal closure with the speech bulb. The second case had undergone radiation therapy for a brain stem tumour, resulting in a velar paralysis. The patient was first treated with a standard acrylic speech bulb prosthesis, which lead to only moderate improvement of her speech. An experimental prosthesis with a flexible silicone end piece was created in order to achieve a greater occlusion of the velopharyngeal opening. The two cases illustrate that speech bulbs are currently not being used to their full potential and that more research is needed to improve both the behavioural interventions and the prosthesis design to achieve consistent success for all patients.

Abrégé

Cette étude décrit deux cas d'hypernasalité à base structurelle traitée à l'aide d'une prothèse vélopalatine obturatrice. Le premier cas, une jeune femme avec une hypernasalité dont l'étiologie est inconnue, a été traitée à l'aide d'une combinaison d'une prothèse vélopalatine élévatrice avec obturateur pour améliorer sa fermeture vélopharyngée et son équilibre oral-nasal pendant la parole. Afin de l'aider à maximiser les avantages de sa prothèse, la patiente a effectué des exercices de rétroaction biologique en ligne de la nasalance et de la fréquence. Cependant, ce n'est que quand elle a eu recours à une manoeuvre de jeu vocal qu'elle a été en mesure d'améliorer de façon consistante sa fermeture vélopharyngée avec sa prothèse obturatrice. Le deuxième cas est une patiente qui avait reçu de la radiothérapie pour une tumeur au tronc cérébral et qui avait par conséquent une paralysie du vélum. La patiente a initialement été traitée à l'aide d'une prothèse obturatrice standard en acrylique, mais cet appareillage n'a mené qu'à une amélioration modérée de sa parole. Une prothèse expérimentale avec un embout flexible en silicone a été créée pour l'aider à produire une meilleure occlusion de l'ouverture vélopharyngée. Ces deux cas démontrent que les prothèses obturatrices ne sont pas actuellement utilisées à leur plein potentiel et qu'il existe un besoin de recherche pour améliorer tant les interventions comportementales que les conceptions de prothèses de façon à atteindre un succès répété avec tous les patients.

INTRODUCTION

The velopharyngeal mechanism is responsible for the regulation of the oral-nasal flow of air and acoustic energy in speech. Successful oral-nasal balance is an important prerequisite for intelligible and socially acceptable speech. Patients with hypernasal resonance disorders may experience social stigmatization (Kummer, 2008). Hypernasal resonance can be a consequence of various structural, neurological and functional etiologies.

The velopharyngeal closure mechanism is complex and relies on a number of muscles for its function. According to the seminal description by Fritzell (1969), the levator veli palatini elevates the velum and pulls it in a cranio-dorsal direction towards the posterior pharyngeal wall. Activity of the palatopharyngeus medializes the lateral pharyngeal walls, which narrows the pharyngeal space (Moon, Smith, Folkins, Lemke, & Gartlan, 1994). Hypernasal patients, most notably those with cleft palate, also sometimes exhibit muscle bulges in the posterior pharyngeal wall. This phenomenon is called 'Passavant's ridge' and is attributed to the pharyngeal constrictor muscles (Glaser, Skolnick, & Shprintzen, 1979). The uvular muscle contracts the velum along its length and contributes to the elevation of the posterior velum towards the posterior pharyngeal wall (Azzam & Kuehn, 1977; Huang, Lee, & Rajendran, 1997).

Clinical research using transnasal endoscopy has found four typical patterns of velopharyngeal closure. Croft, Shprintzen and Rakoff (1981) described a coronal closure pattern (characterized by mainly velar elevation), a sagittal pattern (characterized by approximation of the lateral pharyngeal walls), and a circular pattern (characterized by joint movement of the velum and the lateral pharyngeal walls). The authors also found a variation that was observed in patients with cleft palate. This fourth pattern was characterized by circular closure with a Passavant's ridge.

The effective treatment of hypernasal resonance disorders poses significant challenges. Many patients, such as head and neck cancer patients with velar resections or neurological patients with paralyses, will not be able to control their velopharyngeal closure mechanism at all. Other patients, such as hypernasal patients with a repaired cleft palate, will have better potential for velopharyngeal closure. However, while the structures contributing to velopharyngeal closure are highly sensitive to touch (which may trigger a gag reflex), they offer almost no proprioceptive information. This means that the speaker has virtually no information about the position and the movement of the velopharyngeal sphincter. As a result, teaching velopharyngeal closure to a patient with a hypernasal resonance disorder is difficult and may not always lead to a successful outcome (Ruscello, 1997). A major focus of the treatment of hypernasality has been to change the geometry of the velopharynx, with either surgical or prosthodontic interventions. The goal of these procedures is to reduce the size of the velopharyngeal gap, with the hope that the client will then find it easier to achieve velopharyngeal closure (Kummer, 2008; Peterson-Falzone, Hardin-Jones, & Karnell, 2001).

The surgical approaches for the treatment of hypernasal resonance disorders are primarily used for patients with cleft palate and comprise velopharyngeal operations such as pharyngeal flaps or pharyngoplasties (Peterson-Falzone et al., 2001). However, surgical therapy may not be feasible or desirable for all patients. Another, less commonly used approach to treat patients with hypernasality of different aetiologies is prosthodontic treatment. There are two types of prosthodontic appliances to support velopharyngeal closure. Palatal lifts are shoehorn-shaped appliances that serve to elevate a velum that is sufficiently long but does not elevate sufficiently. Speech bulbs (also sometimes called pharyngeal obturators) are used to partially fill the velopharyngeal space when there is insufficient velar tissue. The patient can use the lateral and posterior pharyngeal walls to make contact with the speech bulb to close off the nasopharynx. The two designs may be combined to maximize the effectiveness of the appliance (Esposito, Mitsumoto, & Shanks, 2000). Compared to surgical interventions, prosthodontic interventions are less invasive, less expensive and easily reversible.

Neither palatal lifts nor speech bulbs can be made so big that they would fill the velopharynx completely because this would interfere with the nasal air passage, make speech hyponasal, and cause discomfort during swallowing (Esposito et al., 2000). Therefore, in many cases, speech therapy has a vital role following the fitting of a palatal appliance, when the patient has to learn to actively use the appliance. However, behavioural modification of the velopharyngeal closure mechanism is notoriously difficult. Velopharyngeal closure is a complex, non-volitional movement. While one can clearly determine the position of the tongue tip in one's own mouth, the state of the velopharyngeal sphincter is inaccessible to introspection and voluntary control.

Despite the prosthodontist's and the speech-language pathologist's best efforts, many patients will only make moderate improvement after a palatal appliance has been fitted. The appliance will passively take up velopharyngeal space and mildly reduce the hypernasality. Patients who can actively use the appliance by closing their velopharyngeal sphincter around it will often be able to achieve a near-normal oral-nasal balance in speech. However, it is often difficult to optimize the device for both speech and swallowing.

Speech-Language Pathologists struggle to find ways to facilitate velopharyngeal learning for patients with hypernasal resonance disorders. Kummer (2008) stresses the role of biofeedback in the therapy of hypernasality. The techniques she discusses range from simple paper air paddles and listening tubes to visual biofeedback using a nasometer. The biofeedback allows the patient to anchor him- or herself in a different perceptual modality.

This paper describes two case studies of hypernasal patients who were fitted with speech bulb/ palatal lift appliances. The first case illustrates how a palatal lift appliance and different biofeedback techniques can be used successfully to significantly reduce a patient's hypernasality. The second case describes a scenario in which a re-design of the palatal lift appliance was used to help a hypernasal patient who could not be expected to make active use of the appliance.

CASE DESCRIPTION 1: JASMINE

Jasmine (name changed), was a 19-year-old female patient. She spoke fluent English with the accent that is typical for Southern Ontario. The patient was also fluent in two other languages. Jasmine was referred to the fourth author because of persistent hypernasality of unknown etiology. The patient reported that she had been hypernasal all her life. Her velum looked normal but did not elevate sufficiently on oral inspection. In order to evaluate the velopharyngeal sphincter in greater detail, a multi-view videofluoroscopic exam with barium contrast was conducted. The anterior-posterior view demonstrated active lateral pharyngeal walls that approximated moderately well during speech. In the lateral view (Figure 1), the velum appeared sufficiently long. The anterior portion of the velum elevated during speech production but the posterior aspect (around the uvula) sagged on elevation. Velopharyngeal closure was complete during swallowing. While the imaging data appeared consistent with occult submucous cleft palate, no definite diagnosis was reached and a genetic test for 22q11 microdeletion syndrome with the fluorescence in situ hybridization technique (FISH; Driscoll et al., 1993) was negative. Jasmine reported that at least one other member of her extended family presented with a similar form of hypernasality but no additional family members were evaluated.

Since Jasmine did not wish to undergo surgery, she was referred to the second author for prosthodontic treatment. The second author decided to construct a combined speech bulb/ palatal lift prosthesis that elevated the velum along its whole length (Figure 2). The bulb extension at the end of the lift appliance was designed to provide additional mass in the centre of the velopharyngeal tract. This prosthesis was fabricated and adjusted over a series of appointments using nasoendoscopic assessment. When a satisfactory shape had been reached, Jasmine was seen by the first author for an acoustic and perceptual appraisal at the Voice and Resonance Laboratory at the University of Toronto.

Figure 1. Lateral videofluoroscopic view of Jasmine's vocal tract. This frame demonstrates maximum velar elevation during speech. Despite the maximum elevation, there is a considerable gap between the velum and the posterior pharyngeal wall. The velum also does not show the knee-shaped posterior eminence that often accompanies velar elevation in normal speakers.

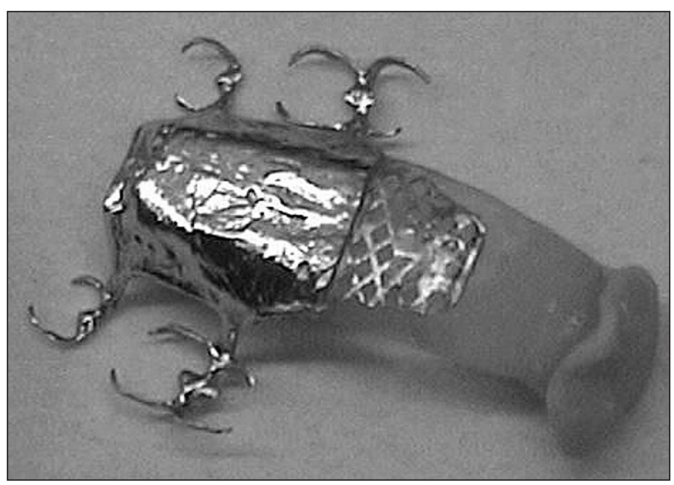

Figure 2. Jasmine's combined palatal lift/speech bulb appliance. The anterior part of the appliance was designed to minimize its long-term effects on the gums around the teeth. The uvula fit into the 'notch' on the anterior side of the bulb, which also gives an indication of the size of the gap to the posterior wall of the pharynx.

Jasmine's nasal resonance was evaluated using the NasalView nasometer (Tiger DRS, Seattle, WA 98125). The NasalView (Awan, 2001) allows the clinician to evaluate the oral-nasal balance of a speaker. The measurements are made with a sound separation plate with separate microphones for the nose and the mouth. The separate sound signals from the mouth and the nose are recorded to a computer and analyzed. The NasalView software allows the experimenter to display the sound signals and measure the 'nasalance' of the speaker (i.e., the proportion of nasal to oral resonance). Research has demonstrated that nasalance values are closely related to a speaker's perceived nasal resonance (Hardin, Van Denmark, Morris, & Payne, 1992).

First, Jasmine's nasal resonance without the prosthesis was determined. It was found that her nasalance score for the Zoo Passage (Fletcher, 1978) was 52%. The normal scores for this passage when measured with the NasalView have been reported to be around 21-23% with a standard deviation of 5% (Awan, 2001; Bressmann, 2005). The Zoo Passage is a standard text passage without nasal consonants, which is used for the assessment of hypernasal resonance disorders. Jasmine's scores were more than double the expected, which confirmed the perceptual impression of severe hypernasality. The recording was repeated with the prosthesis in place. Her nasalance values improved to 47%. Perceptually, a slight improvement was noted but Jasmine was still severely hypernasal.

While minor improvement was noted with the prosthesis, the overall result was not satisfactory. To help the patient further reduce the hypernasality, the real-time biofeedback feature of the NasalView was used. In this mode, a nasalance trace is displayed on the computer screen (Figure 3). Jasmine was instructed to 'move the line towards the bottom of the screen when you speak'. She wore the prosthesis during these exercises. Jasmine achieved a little improvement on some of the sustained vowels but was not able to consistently transfer this improvement to words or sentences.

During a second session of nasalance feedback, Jasmine was instructed to try different forms of vocal play. If patients can imitate different voice qualities or get into a vocal character, this can sometimes facilitate the modification of the voice or the nasal resonance. Jasmine struggled with this exercise. She found it difficult to raise or lower her habitual pitch and loudness for speaking, let alone change her resonance. However, the exercise prompted her to demonstrate a vocal quality, which she referred to as her 'baby voice'. Jasmine reported that she sometimes used this vocal character with family and friends (mainly for comic relief). The 'baby voice' was characterized by a high pitch (264 Hz, as opposed to

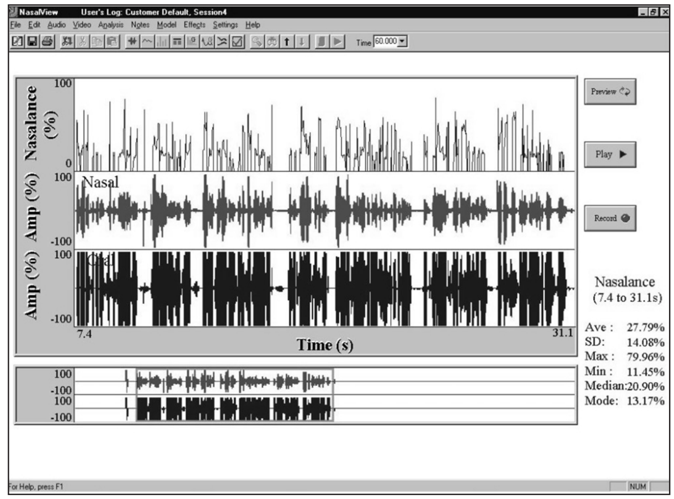

Figure 3. Screenshot from the NasalView software, showing Jasmine's production of the Zoo Passage during the final session. The patient is wearing the speech bulb/palatal lift appliance and constricting her pharyngeal walls. The nasalance trace is displayed together with the oral and nasal sound signals.

her regular fundamental frequency of 220 Hz), a rough voice quality, and pronounced pharyngeal constriction. Perceptually, this resulted in a child-like voice quality. When Jasmine used her 'baby voice' without the prosthesis in place, her nasalance values dropped immediately to 39%. Initially, she had a difficult time producing the 'baby voice' with the prosthesis in place. Jasmine experienced gagging and discomfort, probably because the pharyngeal constriction associated with the 'baby voice' brought the pharyngeal walls in contact with the acrylic speech bulb.

By the third session, Jasmine's pharynx was desensitized and she was able to produce the 'baby voice' with the prosthesis in place. However, her voice was still rough and her fundamental frequency was unacceptably high. When a sound recording was played back to her, Jasmine agreed that the quality and pitch of the voice were undesirable. However, she was initially unable to lower her voice while still maintaining the pharyngeal constriction. The Real Time Pitch module of the MultiSpeech software (MultiSpeech 3700, KayPentax, NJ 07035-1488) was used to provide her with online feedback about the fundamental frequency of her voice. The Real Time Pitch software displays the fundamental frequency of the speaker as a line on the computer screen. With this visual feedback, Jasmine was quickly able to lower her fundamental frequency to her habitual pitch.

For the rest of the third session, Jasmine practiced wearing the prosthesis while constricting her pharynx without raising her fundamental frequency. She was provided with online visual feedback from the NasalView. A nasalance recording demonstrated a value of 28% nasalance for the Zoo Passage. Perceptually, Jasmine was as close to normal as could reasonably be expected after only three sessions of training. She was able to transfer the new vocal quality to phonetically balanced reading passages and spontaneous speech towards the end of the session. However, she continued to experience difficulties with quick nasal to oral transitions where it often took her a short moment to constrict her pharynx again. Given Jasmine's rapid progress, it was likely that further biofeedback sessions could have helped to further improve and consolidate her oral-nasal balance. Unfortunately, she was scheduled to return to her native country and therapy could not be continued.

Jasmine was seen again at the Voice and Resonance Laboratory during a visit to Toronto six months later. At that time, new nasalance recordings with the NasalView were made to check her progress. The recordings showed a score of 23% nasalance for the Zoo Passage with the prosthesis in place. This indicated that Jasmine had not lost the skill of constricting her pharynx and lowering her pitch with the prosthesis in place. However, she had not improved her nasal-oral transitions in phonetically balanced reading passages and spontaneous speech. Jasmine had not sought the help of a Speech-Language Pathologist in her home country and admitted to not practicing regularly. Nevertheless, she reported that people were able to understand her more easily with the prosthesis in place and that she felt more confident about her speech. Overall, she expressed satisfaction with the treatment.

CASE DESCRIPTION 2: LISA

Lisa (name changed), a 17-year-old female patient, underwent a resection of a brain stem tumour, which, as a side effect of the radiation therapy, resulted in a flaccid paralysis of the velum and pharynx. The patient was hypernasal and her velum did not elevate sufficiently on oral inspection. Because the nature and the extent of the velar paralysis were readily apparent, videofluoroscopic and endoscopic exams were not undertaken.

Owing to the extent and nature of velar paralysis, Lisa was not a good candidate for pharyngeal flap surgery. Therefore, she was referred to the third author for prosthodontic treatment. The third author constructed a combined speech bulb/ palatal lift prosthesis that elevated the velum along its whole length (Figure 4), similar to the prosthesis made for Jasmine.

Lisa's nasal resonance was evaluated using the NasalView. Her nasal resonance when reading the Zoo Passage without the prosthesis was 58%. The recording was repeated with the prosthesis in place. Her nasalance values improved to 55%. Perceptually, a slight improvement was noted but Lisa was still severely hypernasal.

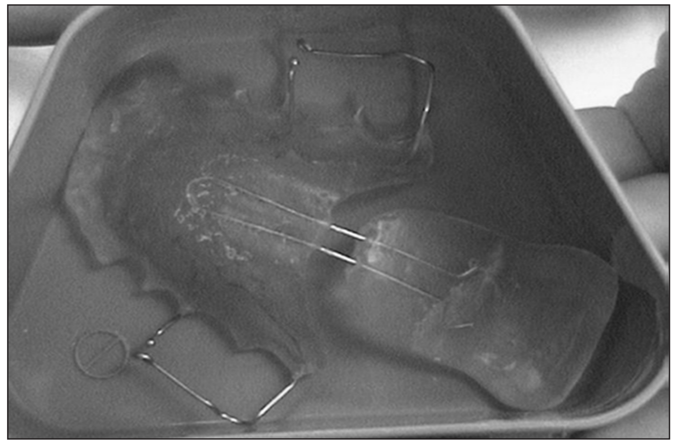

Figure 4. Lisa's combined palatal lift/ speech bulb appliance.

Because the patient experienced persistent hypernasality with the prosthesis, it was decided to redo the prosthesis with a different material to achieve a better occlusion of the velopharyngeal sphincter. Vogel, Sauermann & Ziegler (1996) first described a redesigned palatal lift appliance with a soft silicone end piece. Vogel (2001) demonstrated the usefulness of the Palatoflex appliance with a group of 23 dysarthric patients with different etiologies. The prosthesis consists of a removable dental appliance with a posterior rod with retention hooks. The metal rod supports the palatal lift extension. The lift is made from a flat sheet of silicone rubber which is tapered to paper-thickness. The thin edges of the prosthesis increase patient comfort with the appliance. Because the prosthesis is relatively thin and pliable, it is also possible to make the device larger than an acrylic prosthesis. Vogel (2001) argues that the pliability of the extension reduces patient discomfort during swallowing. One note of caution that should be extended to readers: a retention rod may be prone to mechanical failure if it is supporting an extensive lift. This could potentially result in the extension coming loose, with subsequent occlusion of the airway. If any signs of stress appear on the prosthesis, the patient should contact their prosthodontist immediately.

The third author made an appliance for Lisa that was based on the Palatoflex design. The soft silicone rubber extension was made from Implantech silicone block. This material has a durometer grade of 40 (Implantech Associates, Ventura, CA 93003-7602). Durometry determines the hardness and resistance of a material to indentation or deformation. The appliance is shown in Figure 5. Since the patient had already learned to tolerate a hard acrylic speech bulb in her pharynx, she was able to tolerate the large soft silicone attachment very well. Because of the experimental nature of the prosthesis, the patient was instructed to check the device carefully for signs of wear and tear on a daily basis. The patient was also instructed not to wear the appliance while eating and drinking, during sleep, or during physical exercise.

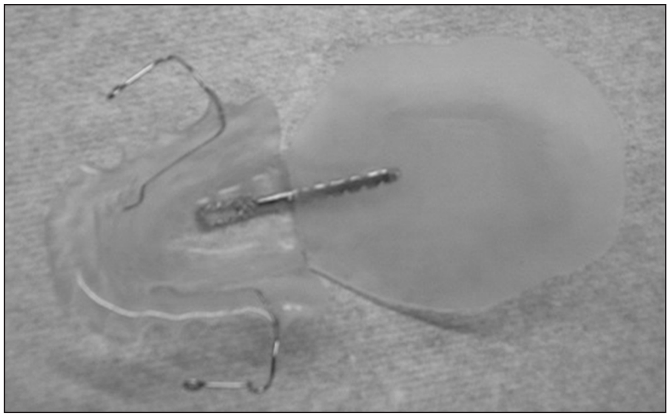

Figure 5. Lisa's Palatoflex-style appliance. The posterior extension of the appliance is made from flexible silicone. The silicone is held in place with a retention rod. The silicone is tapered so that the posterior and lateral edges of the attachment are paper-thin.

With the silicone extension, a NasalView recording of the Zoo Passage indicated a mean nasalance score of 38%, which was closer to the range of normal speakers of 21-23% (Awan, 2001; Bressmann, 2005). Lisa's voice quality with the appliance had a hyponasal quality, owing to the extensive obturation of her velopharyngeal space. This was further underscored by the nasalance scores for the Nasal Sentences (Fletcher, 1978). Lisa obtained a nasalance value of 33% with the appliance. For normal speakers, the scores for the Nasal Sentences have been reported to be around 56% with a standard deviation of 5% (Bressmann, 2005). Hyponasal speech is not normal but it is more intelligible and socially acceptable than hypernasal speech (Shprintzen, Lewin, & Croft, 1979; Sullivan, Marriman, & Mulliken, 2010).

DISCUSSION

The generalizability of any case study is necessarily limited, and the findings may not necessarily be repeatable with different patients. Nevertheless, the present case studies afford us interesting insights into the prosthodontic management of hypernasal resonance disorders.

The first case, Jasmine, illustrates the importance of both biofeedback and vocal play in the therapy of a hypernasal resonance disorder. The successful remediation of hypernasal resonance continues to be a challenging conundrum in Speech-Language Pathology. One of the main recommendations for treatment is to use biofeedback to externalize the effects of velopharyngeal movement for the patient (Kummer, 2008). This treatment approach assumes that the patient will be able to recognize his or her own successful attempts based on the biofeedback, develop a new motor strategy, and then incrementally improve the outcomes using further biofeedback.

On the other hand, Stemple (2000) recommends exploring the patient's abilities for vocal play. According to his recommendations, a hypernasal patient should be instructed to try to 'speak like you have a cold' (i.e., try to find a hyponasal voice quality). For most hypernasal patients, such an instruction would be unrealistic and would only serve to confuse the patient. However, for an individual who can get into a different vocal character, such vocal play can greatly facilitate the learning process and the transfer to everyday speech.

In the present case, it was the combination of biofeedback with a vocal play manoeuvre (Jasmine's 'baby voice') that lead to a successful therapy outcome. It was only when the patient was encouraged to experiment with a different way of producing voice that her nasalance values and the perceptual impression of hypernasality changed markedly. The 'baby voice' apparently enabled Jasmine to achieve pharyngeal constriction and reduce the diameter of her velopharyngeal opening and to contact the palatal lift/ speech prosthesis with her lateral pharyngeal walls. Once this crucial step had been achieved, the biofeedback became more useful to the patient and she was able to improve her oral-nasal balance. With the additional pitch feedback, Jasmine was able to also drop her pitch and to achieve an acceptable conversational voice. This underlines the observation by Peterson-Falzone et al. (2001) that "if biofeedback is to be useful, it must be used with techniques capable of establishing skilled patterns of opening and closing the velopharyngeal port during connected speech" (p. 310).

While vocal play turned out to be a central building block for the therapy of the present patient, it cannot be stressed enough that the therapist must exercise utmost caution when encouraging a patient to make radical changes to their voice quality or to go to extremes in their vocal range. Since the velopharyngeal sphincter is not easily available to kinaesthetic or proprioceptive feedback, a patient attempting to achieve velopharyngeal closure will often build up substantial muscle tension in the neck area. Consequently, patients who try to improve their velopharyngeal closure may inadvertently lift their larynx and squeeze their vocal folds, thus putting themselves at risk for developing a voice disorder (Peterson-Falzone et al., 2001). The patient Jasmine in the present case study exhibited vocal roughness and described a feeling of physical strain when first employing the 'baby voice' in the second session. The experimenters were aware of the risks to her voice and focused the third therapy session on modification of the fundamental frequency in order to achieve a voice quality that was comfortable and sustainable for the patient.

The second case, Lisa, presented a very different scenario. Because of the velopharyngeal paralysis, Lisa did not have the same movement capacities and potential for behavioural therapy as Jasmine. The treatment approach for this patient therefore focused on maximizing her oral resonance with an appliance that would reduce her velopharyngeal gap during speech. Lisa's acrylic speech bulb prosthesis afforded her only a marginal improvement of her nasalance values. The new prosthesis based on the Palatoflex design (Vogel et al., 1996; Vogel, 2001) allowed the fabrication of a very large end piece. The tapered design of the speech bulb extension and the pliability of the material made the appliance more tolerable for the patient.

In the case of Lisa, the improvement of the oral-nasal balance came at the price of increased hyponasality. While a hyponasal voice will be more intelligible and socially more acceptable than hypernasality (Shprintzen et al., 1979), the large and obturating silicone attachment may interfere with nasal breathing. This trade-off must be carefully considered when designing the appliance so that the impact on nasal breathing will be as minimal as possible.

A curious feature of Lisa's speech was that her nasalance scores for the non-nasal text were higher than her values for the nasal sentences. This was an unusual finding because even severely hypernasal speakers will normally have higher nasalance scores on the nasal sentences, although the distance between their nasal and non-nasal productions may diminish (Bressmann et al., 2000). In the case of Lisa, possibly owing to the severity of the velar paralysis, there was no clear differentiation of the nasal and non-nasal productions. This pattern persisted with the Palatoflex prosthesis in place.

Despite a considerable body of literature published on the topic, speech bulbs and palatal lift prostheses are not often utilized in the care of patients with hypernasal resonance disorders (Peterson-Falzone et al., 2001). The two cases described in this paper illustrate two continuing challenges for the practical use of in this type of appliance.

Jasmine's case illustrated the importance of active use of the prosthesis by the patient. Unfortunately, Speech-Language Pathologists have not established effective and reliable methods of stimulating velopharyngeal movement in speech. This is an area that merits further investigation because it is still unclear to what extent speakers can voluntarily modify their velopharyngeal closure pattern (Siegel-Sadewitz & Shprintzen, 1982).

The current designs of speech bulb and palatal lift appliances have been established over 50 years ago, with only minor variations. Lisa's case underlines that it can sometimes be necessary to stray from the original, time-honoured ways of creating speech bulb or palatal lift prostheses. The Palatoflex design (Vogel et al., 1996; Vogel, 2001) enabled the treatment team to drastically improve Lisa's oral-nasal balance in speech, beyond what could have been achieved with a conventional prosthesis. This area, too, merits further research and innovation so that more patients will be able to benefit from prosthodontic treatment for hypernasal resonance disorders in the future.

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AUTHORS' NOTE

Correspondence should be sent to Tim Bressmann, Ph.D., Associate Professor, Department of Speech-Language Pathology, University of Toronto, 160-500 University Avenue, Toronto, ON, M5G 1V7. E-mail: tim.bressmann@utoronto.ca

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