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Treatment of unilateral vocal fold paralysis with ansa cervicalis to recurrent nerve anastomosis in a young adolescent: European case report

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Abstract

Background. Laryngeal re-innervation in paediatric unilateral vocal fold paralysis is a relatively new treatment option, of which there has been little reported experience in Europe.
Methods. In this European case report of a 13-year-old boy with dysphonia secondary to left-sided unilateral vocal fold paralysis after cardiac surgery, the patient underwent re-innervation using an ansa cervicalis to recurrent laryngeal nerve transfer, in combination with fat augmentation, after 12 years of nerve denervation. Perceptual analysis data, and acoustic and laryngoscopy recordings were acquired pre-operatively, and at one and two years post-operatively.
Results. The patient's perceptual voice quality was improved. He experienced subjective improvement and is very satisfied with the result. As expected, laryngoscopy at one and two years after surgery showed no physiological mobility of the vocal fold concerned, but improved closure during phonation was achieved. Electromyography showed evidence of re-innervation.
Conclusion. Laryngeal re-innervation could be considered as a treatment option for unilateral vocal fold paralysis in children and adolescents, even after a long-term delay.

Introduction

Unilateral vocal fold paralysis is a challenging pathology for clinicians when it occurs in children and adolescents. About 50 per cent of the patients recover function naturally over time, with a wide range of recovery time depending on the cause of the paralysis. The majority of children with persistent paralysis are able to compensate for the glottic defect.[1–3] When dysphonia persists, an observation time of 8–12 months is common before treatment options other than speech therapy are considered. Alternative treatment options for children and adolescents include vocal fold injection augmentation, thyroplasty or re-innervation. Laryngeal re-innervation has been described in children and adolescents as a relatively new treatment option. Based on existing clinical experience from the USA, it is considered safe.[4–10] Crumley was the first to describe ansa cervicalis to recurrent laryngeal nerve (RLN) anastomosis as a treatment option in children, during the 1980s.[11]

In the Netherlands, and possibly in other European countries, there is little experience with laryngeal re-innervation in children, as there are no European publications on this subject to date. We describe our experience with ansa cervicalis to RLN anastomosis in the case of a 13-year-old boy with unilateral vocal fold paralysis.

Case report

A 13-year-old boy presented with unilateral vocal fold paralysis, which had become apparent after cardiac surgery performed in his first year of life. He was treated with voice therapy at the age of six years, which had little effect. He was about to start secondary school when the persisting hoarseness became an issue again. He had become more aware of the dysphonia and was more conscious of his presentation. His parents noted that he had shown signs of voice mutation at the age of 12 years; the voice was unstable.

Different treatment options were discussed with the patient and his parents, including surgical treatment. The parents were not in favour of repetitive treatments, and had personal objections to the implantation or injection of foreign body materials. It was decided that a re-innervation procedure would be performed using an ansa cervicalis to RLN nerve transfer combined with autologous fat injection augmentation.

Ansa cervicalis to RLN nerve transfer surgery was performed on the left side, with a horizontal skin incision of 4 cm at the level of the cricothyroid joint. Strap muscles were lateralised to identify the omohyoid muscle. The ansa cervicalis was identified lateral to the omohyoid muscle. The RLN was found paratracheally, cranial to the thyroid gland.

Table 1. Pre- and post-operative measurements

Assessment	Pre-surgery	1 year post-surgery	2 years post-surgery
Perceptual measurements			
– Therapist 1 GRBAS score	G2R2B1A0S0	G2R1B1A0S0	G1R1B0A0S0
– Therapist 2 GRBAS score	G2R1B1A1S2	G2R1B2A1S1	G2R2B0A0S1
– Therapist 3 GRBAS score	G3R2B1A0S0	G2R1B2A1S0	G1R1B0A1S0
– Therapist 4 GRBAS score	G3R2B1A1S0	G2R1B2A2S0	G2R1B0A0S1
– Therapist 5 GRBAS score	G3R3B1A0S3	G2R2B1A0S2	G2R2B0A0S1
– Therapist 6 GRBAS score	G3R3B0A0S1	G2R1B2A2S0	G2R2B1A1S0
– Mean GRBAS score	G3R2B1A0S1	G2R1B2A1S1	G2R2B0A0S1
– Voice ranking	Worst	Middle	Best
– Voice description	Falsetto, creaky, hoarse, diplophonic	Modal register, hoarse, low, asthenic	Modal register, creaky, bit strained
Acoustic measurements			
– Normal speech volume (dB)	58	60	57
– Normal speech SF0 (Hz)	228	148	93
– Loud speech volume (dB)	62	60	62
– Loud speech SF0 (Hz)	272	133	94
– Shouting volume (dB)	60	72	76
– Shouting SF0 (Hz)	304	166	105
– Maximum phonation time (seconds)	10.34	6.41	8.31
Psychosocial impact			
– Voice Handicap Index	57/120	19/120	16/120
EMG findings	Fibrillations present	Polyphasic action potentials (confirming that re-innervation has taken place)	Not performed

GRBAS = grade, roughness, breathiness, asthenia, strain; SF0 = speaking fundamental frequency; EMG = electromyography

The ansa cervicalis was cut with sufficient length to the RLN to avoid tension; the branch to the omohyoid muscle was spared. The ansa cervicalis was sutured with an end-to-end anastomosis to the distal RLN, with size 9.0 Ethilon micro-sutures, using microsurgical instruments. Limited fat augmentation was performed, as only a very small amount could be harvested from the belly area. There were no peri- or post-operative complications.

We conducted voice assessment according to Cohen *et al.*,[12] who suggested an adaptation of the European Laryngological Society protocol (Dejonckere *et al.*[13]) for paediatric voice. Vocal function parameter assessment, conducted at one and two years after surgery, included: perceptual analysis, acoustic measurements (pitch and speaking volume, using LingWaves software version 3.0 (Wevosys, Baunach, Germany), aerodynamic analysis, subjective measurement of voice disorder symptoms (Voice Handicap Index) and laryngoscopy. Blinded perceptual evaluations (grade, roughness, breathiness, asthenia, strain ('GRBAS') scale and ranking of speech audio files) were carried out by six speech therapists from other hospitals who were not involved in the treatment. Electromyography of the right and left vocalis muscles was performed pre-operatively and at one year post-operatively. The pre- and post-operative measurements are shown in Table 1.

Laryngoscopy before surgery showed an immobile left vocal fold, which was slightly atrophic and in a paramedian position; the arytenoid joint position was rotated inferoanteriorly. During phonation, there was incomplete glottic closure. At one and two years after surgery, laryngoscopy showed an immobile left vocal fold, with improved closure during phonation, as shown in the supplementary video material (Appendix 1).

Discussion

Hoarseness in children can lead to social problems, especially during school years when a child's social network broadens.[12] Surgical intervention is recommended in children and adolescents with unilateral vocal fold paralysis when speech therapy has led to unsatisfactory results.[2] Surgical interventions aim for medialisation of the paralysed, immobile vocal fold, in order to improve glottic closure during phonation.

The range of possibilities in adults may not be equally favourable in children or adolescents. Injection augmentation is temporary; furthermore, autologous fat injection material has unknown long-term effects in children. Thyroplasty in a growing larynx will also prove inadequate in the long term. Re-innervation techniques using an ansa cervicalis to RLN nerve transfer would offer a permanent solution. However, re-innervation is not likely to be successful when there is denervation-induced laryngeal muscle atrophy and fibrosis.[14] Electromyography can be used to make the diagnosis of vocal fold paralysis or ankylosis in cases of immobility. It is also useful to confirm the extent of denervation, to anticipate the results of a new non-selective re-innervation. If there are signs of total denervation, with a lack of fibrillations, there is a good chance that the re-innervation process will fail, and an alternative rehabilitation technique is then preferred.

A considerable period of delay will usually have passed before the choice is made to perform re-innervation surgery, and the length of delay may influence the results. The time between nerve injury and re-innervation in children ranges widely in the available literature. In cases presented by Zur and Carroll, denervation interval times of 25 months to 13 years prior to re-innervation surgery were included.[5] Marcum *et al.* reported two cases, including one of a six-year-old child with six years of denervation with similar

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results.[6] All of these patients experienced a subjective beneficial effect after surgery.

Smith and Houtz published a review of prospectively collected data of 35 children and adolescents (aged 1–21 years) with an average denervation interval time of 4.6 years (range, 1–21.8 years).[7] Seven patients had a denervation period of 5–10 years and 3 had a denervation interval of more than 10 years. There was no correlation found between age at re-innervation and subjective and perceptual voice outcomes. A slight negative correlation was found between the time of RLN injury and time of re-innervation.[7] Follow-up data were available for 28 patients. Based on these data, it was concluded that ideally ansa cervicalis to RLN anastomosis should take place between two to three years of denervation.[7] A systematic review stated that permanent treatment should ideally take place after a few years of observation and before the age of six years.[1] In our case, the age of 13 years with 12 years of denervation was not contraindicated, as experience with a beneficial effect of re-innervation after a long-term denervation period can be found in the literature.[5,7,8]

To evaluate the voice, standardised methods of assessment should be used. In cases of poor voice quality and young age, it can be challenging to assess voice quality objectively. In previous studies on the effect of paediatric ansa cervicalis to RLN re-innervation, perceptual evaluation and objective assessment methods were used, including maximum phonation time and pitch range. The Pediatric Voice Handicap Index and Pediatric Voice-Related Quality of Life survey have been used to evaluate the effects on the child's life and quality of voice. In our case, we used the adult Voice Handicap Index, as the Pediatric Voice Handicap Index was not yet validated in Dutch and no other validated Dutch questionnaires regarding quality of life in dysphonic patients were suitable. Despite this limitation, the Voice Handicap Index showed an improvement in the psychosocial impact of the voice disorder, decreasing to normal values at one and two years after surgery.

The perceptual voice improvement was the most striking outcome in our case report. The blinded ranking of the voice by the independent speech therapists was unanimous: 'worst' before surgery, 'middle' at one year and 'best' at two years after surgery. The main improvements were observed in acoustic measurements of pitch, register, breathiness and stability. Pre-surgery jitter and shimmer measurements failed because of the poor voice quality, and were therefore excluded from evaluation. Before surgery, the voice was unstable in falsetto. The falsetto was probably a compensation for the breathy voice due to the paralysis.

Phonation in the modal register in running speech was accomplished with voice therapy about eight months after surgery; earlier attempts to lower and stabilise the voice before re-innervation surgery had not succeeded. Mutation can interfere with voice quality, especially in boys.[15] As there was a clear voice improvement, with a reduction of breathiness, re-innervation appears to have been effective during the voice mutation phase. Autologous fat augmentation could still be effective two years after surgery,[16,17] but the finding that the voice actually improved between the first and second year after surgery cannot be explained by the fat augmentation and must be ascribed to the effect of re-innervation. A longer follow-up period could confirm this.

The voice that results from a non-selective ansa cervicalis to RLN transfer cannot be expected to be completely normal. The vocal fold is innervated by regenerated ansa cervicalis axons, which simultaneously innervate the abductor and adductor muscles.[18] Re-innervation provides muscle tension and tone that stabilises the arytenoid and prevents inferior displacement of the vocal process.[19] Nevertheless, our case illustrates clear subjective voice improvement. Two years after surgery, the patient is well understood when speaking from the back of the classroom.

- Treating children and adolescents with unilateral vocal fold paralysis is challenging
- Laryngeal re-innervation with ansa cervicalis to recurrent laryngeal nerve anastomosis is a relatively new treatment in children and adolescents
- Research is limited to small series of children and adolescents; there are no European publications to date
- This European case report describes laryngeal re-innervation, with fat augmentation, in a 13-year-old boy
- Re-innervation was effective, with perceptual voice improvement at 1 and 2 years post-surgery, even after 12 years of denervation
- Laryngeal re-innervation is an option for children and adolescents with persistent hoarseness secondary to unilateral vocal fold paralysis

In adolescent cases of unilateral vocal fold paralysis, wherein speech therapy has led to inadequate voice improvement and the expected recovery of the paralysis is poor, re-innervation could be considered in order to achieve effective improvements in voice quality and quality of life.

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Competing interests. None declared

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Appendix 1. Supplementary video material

A short video of laryngoscopy pre- and post-surgery, showing an immobile left vocal fold and improved closure during phonation, is available online at *The Journal of Laryngology & Otology* website, at https://doi.org/10.1017/S0022215118001007

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