Does use of the Cough Assist Machine reduce respiratory morbidity for children with neuromuscular disease?

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ABSTRACT

The role of the Cough Assist Machine (CAM) in the long term respiratory management of children with neuromuscular disease (NMD) is unclear. This study examined the impact of regular, home use of the CAM on the respiratory status of six children with NMD and significant respiratory morbidity. Individualised CAM programmes were devised to be undertaken regularly. Retrospective review of hospital records was undertaken to obtain admission data, lung function data, community antibiotic prescriptions and chest radiology reports for the two years prior to CAM initiation. These data were compared to data collected for two years following CAM initiation. Fewer days hospitalised for respiratory infections following machine initiation were evident for all participants. Qualitative feedback indicated high treatment compliance and satisfaction. Four of the five participants, with persistent or recurrent chest radiology abnormalities on enrolment, achieved resolution. Half of the participants had a reduction in community antibiotic prescriptions. No adverse events were reported. Acknowledging the small sample size, domiciliary use of the CAM appears a safe and effective form of airway clearance for some children with NMD. In addition, CAM's may potentially reduce respiratory admission time for some children with severe respiratory morbidity as a result of their NMD. Furthermore an impact on radiological abnormalities and community antibiotic prescriptions may also be possible.

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INTRODUCTION

Neuromuscular diseases (NMD's) are disorders caused by an abnormality of any component of the lower motor neuron system. This can include anterior horn cell, peripheral nerve, neuromuscular junction, or muscle (Rossi et al 2004). The most common NMD's in children are genetic and include a range of dystrophies, spinal muscular atrophy (SMA), hereditary motor sensory neuropathy, and congenital myasthenia gravis (Rossi et al 2004). Early symptoms include infant floppiness, poor feeding, and failure or delay in achieving motor milestones. In more severe neuromuscular disorders, symptoms can progress to include loss of ambulation, scoliosis, and respiratory impairment (Rossi et al 2004). Despite significant advances in respiratory management, respiratory complications remain the most common cause of hospital admission and often, the eventual cause of death in this severe group (Chatwin and Simonds 2009).

Children with NMD have normal chest wall, lung, and upper airway structure at birth. In those with severe respiratory involvement prolonged low lung volumes can result in chronic microatelectasis, muscular fibrosis, articular contracture, and subsequent chest wall deformity. Furthermore, reduced muscle activity can contribute to chronic hypoventilation, reduced cough peak flows (CPF), and subsequent impaired chest clearance (Chatwin et al 2003). Long term this impairment in chest clearance can result in pneumonia, atelectasis, and altered gas exchange, with subsequent supplemental oxygen dependency and respiratory acidosis (Miske et al 2004).

Consensus statements for the management of Duchenne's Muscular Dystrophy (DMD), SMA and Congenital Muscular Dystrophy (CMD) all highlight regular pulmonary function testing, polysomnography, Non-Invasive Ventilation (NIV), and appropriate airway clearance techniques as imperative (Finder 2009, Wang et al 2007, Wang et al 2010), if the inevitable spiral to respiratory failure is to be delayed (Homnick 2007). Airway clearance strategies involve techniques to:

- Loosen secretions (manual percussion vibrations and High Frequency Chest Wall Oscillation),
- Assist insufflation (glossopharyngeal (frog) breathing, breath stacking with an ambu bag, insufflation with cough assist machine),
- Augment cough (manually assisted cough and mechanically assisted cough (CAM)) (Wang et al 2010).

Given children with NMD's tend to retain mucociliary clearance while losing cough clearance, at least in the early stages, it is the latter two categories that tend to be of greatest effect (Finder 2009). Despite an absence of randomised controlled trial data suggesting an advantage, the use of CAM's has increased over the last two decades (Hull et al 2012). However, for many

children and young people in New Zealand access to devices is precluded by the relatively high cost and limited expertise to support optimal use.

The use of CAM's to augment cough peak flow (CPF) in NMD was championed by Bach in the early nineties (Bach 1993). Observational studies followed, suggesting that in combination with non-invasive ventilation (NIV), CAM's may reduce pulmonary morbidity in both adults and children with a range of NMD's (Bach et al 1997, Simonds 2006, Tzeng and Bach 2000). In an acute paediatric setting, CAM's have been shown to reduce chest physiotherapy treatment time (Chatwin and Simonds 2009) and reduce the incidence of treatment failure leading to tracheostomy (Vianello et al 2005). Further studies have concluded that CAM's may be a useful tool post operatively for children with NMD to avoid prolonged intubation, particularly following scoliosis surgery (Marchant and Fox 2002).

In an out-patient setting CAM's have been shown to result in significant increases in CPF both after (Fauroux et al 2008) and during use (Chatwin et al 2003). This increased CPF is particularly evident when used in combination with manually assisted cough manoeuvres (Chatwin et al 2003). Three previous studies have examined the safety and efficacy of long term routine use of the CAM at home in the paediatric population (Chatwin et al 2011, Miske et al 2004, Moran et al 2013). Only one has previously considered hospitalisation as an outcome measure (Moran et al 2013). The primary focus of Moran et al's study was to assess the impact of home use of the CAM on hospital admissions in children with NMD. Though only considering a small sample of ten children, they concluded that CAM's were an effective and well-received means of managing inter-current infections at home (Moran et al 2013). The current study builds on Moran et al's findings by considering the impact of the CAM on both hospital admissions and the wider respiratory health of participants.

BACKGROUND

In 2006, monies donated by the local Muscular Dystrophy Association (MDA) funded a CAM for acute use at the Starship Children's Hospital (SCH) in Auckland, New Zealand. Initial outcomes from its use were overwhelmingly positive. Seven years on, SCH now has fourteen machines in total, three for acute in-patient use and eleven for use in the community. Funding for these was sought annually through the hospital budget and Starship Charitable Foundation. Due to the significant cost of the machine ($12,000 NZD) evidence of treatment effect was requested by hospital management, hence implementation of this study. We hypothesised that regular domiciliary use of the CAM would be safe and improve the respiratory morbidity of participants over a two year period.

METHODS

Participant recruitment

Any child with a diagnosed neuromuscular disorder (excluding SMA I) admitted between January 2007 and May 2010 fitting the criteria below were approached to participate. Children with severe respiratory morbidity were identified following acute admissions for respiratory infection. If these children presented with severely impaired cough and failure to maintain respiratory health despite optimal conventional management then they were invited into the study. Conventional management was defined as consisting of parenteral or enteral antibiotics, nutritional supplementation, non-invasive ventilation, and age appropriate traditional chest clearance techniques as indicated. Failure to maintain respiratory health was defined as experiencing:

- One severe respiratory infection resulting in Paediatric Intensive Care Unit admission or a significant deterioration in respiratory status from their baseline which is not significantly improved by discharge.

  or

- Two or more admissions with significant respiratory infections in a six month period requiring antibiotics and a prolonged hospital stay.

  or

- Three or more prolonged courses of oral antibiotics at home for repeated chest infections over a nine month period.

Children with SMA I were excluded from the study due to their young age and difficulties in identifying optimal conventional respiratory management (Chatwin et al 2011).

Children were identified by their treating therapist or respiratory consultant as an in-patient. Both families and children were provided with age appropriate written and/or pictorial information regarding the study. Written consent was obtained from each child's legal guardian. Children provided informed verbal or written, assent or consent appropriate to their age and developmental stage. Over the enrolment period seven young people were identified as fitting the inclusion criteria. One young person refused consent to participate, the further six families consented. Once enrolled, all programme prescription, education, and machine set-up was undertaken by the researchers.

Investigations / observations undertaken

Once identified and consented, management was identical to the optimal conventional management outlined above with the addition only of the CAM as part of an airway clearance programme. Follow-up was as per standard best practice care. This consisted of annual reviews with both the respiratory physician and the physiotherapist, with routine chest x-ray (CXR) and lung function performed at these times. Respiratory physiologists performing lung function tests and radiologists reading and reporting on CXR's had no knowledge of the study and as such were fully blinded. Participants were individually assessed by an experienced respiratory physiotherapist and two tailored CAM programmes were developed in partnership with the patient and family. One programme was for use when 'well' and one when 'unwell' with respiratory symptoms. Participants and their care-giver were fully trained on the implementation of the two programmes. The lowest effective pressure that enabled thoracic expansion on inspiration and effectively cleared secretions on expiration was prescribed for each participant (range ±25cmH20 to ± 45 cmH20). This was increased slightly (by 5cmH20) for some participants when they became unwell. Adaptations to programmes were made based on patient need and presentation throughout the two year study period. All were asked to use the CAM at least once a day.

Over a two year period from initiation, the number of inpatient days for respiratory infections was measured. Their data were compared to that collected in the two years prior to CAM initiation by retrospective review of hospital records. Secondary outcomes considered were the number of oral antibiotic prescriptions, pulmonary function tests (PFT) (forced vital capacity (FVC), forced expiratory volume in the first second ($FEV_{1)}$, and cough peak flow (CPF) where available) and radiological changes. Qualitative feedback about use of CAM and compliance with its use was obtained from families and children where appropriate. In the absence of any suitable published questionnaires, a treatment satisfaction survey and a treatment impact questionnaire, specifically developed for this study, were utilised. The survey consisted of ten statements to be scored on a Likert scale and three open ended questions regarding participant's views of the positive and negative elements of the CAM (Appendix 1, see website). The treatment impact questionnaire consisted of six open ended questions regarding compliance and treatment impact on both the child and the wider family (Appendix 2, see website). The questionnaire was not validated or checked for reliability but was based on past literature and our previous research experience. The New Zealand Health and Disability Ethics Committee approved the study.

RESULTS

Demographics

Six children with NMD were prospectively followed for two years following initiation of a regular, at-home CAM programme (Table 1). All were wheelchair bound and had scoliosis either on study initiation or developed one during the study. Four underwent spinal rodding surgery to correct their spinal deformity prior to or during the study. In case 3, the parents declined operative intervention, and in case 2 the surgery was completed after the study finished. One child was gastrostomy fed due to failure to thrive. None had significant symptoms of or were prescribed therapy for gastro-oesophageal reflux which has been highlighted as a precaution of use of the CAM (Miske et al 2004).

Table 1: Patient characteristics

Case	Diagnosis	Sex	Age at initiation of CAM (years)	Age at initiation of home NIV (years)
1	SMAII	F	14.9	12.5
2	SMAII	F	2.8	2.8
3	CMD	M	12.0	6.8
4	SMAII	F	5.6	-
5	SMAIII	F	7.5	-
6	SMAII	F	12.8	6.5

CAM: Cough Assist Machine, NIV: Non-Invasive Ventilation, SMA: Spinal Muscular Atrophy, CMD: Congenital muscular dystrophy; F: female; M: male

Outcomes

Admission numbers and days hospitalised for respiratory infections for each child are summarised in Table 2. Statistical analysis of the admission and community antibiotic data is summarised in Table 3. Data from two years prior to CAM initiation were compared with those collected during the two years following initiation using a paired t-test. Secondary outcome measures are summarised in Table 4.

Table 2: Summary of days hospitalised (admission numbers) 2 years before and after initiation of CAM

Case	-2 years	-1 year	CAM initiated	+1 year	+2 years
1	60 (2)	85 (5)	26/01/07	37 (2)	0
2	4 (2)	39 (3)	23/10/07	36 (4)	10 (3)
3	6 (1)	5 (1)	09/11/07	0	0
4	3 (1)	13 (1)	30/08/08	0	0
5	0	29 (1)	22/01/10	4 (4)	6 (1)
6	0	10 (3)	01/04/10	1 (1)	4 (1)
Total	73 (6)	181 (14)		78 (11)	20 (5)

CAM: Cough Assist Machine

All participants had fewer days hospitalised for respiratory infections per year following CAM initiation. Though p-values are not significant, mean values demonstrated a clear trend to lower admission days and to a lesser extent of admission numbers post CAM initiation.

In cases 1 and 3, a reduction in community antibiotic use per year was marked, with no antibiotics prescribed in the second year of study. In case 2, antibiotic use reduced slightly per year. In cases 4-6, community antibiotic use remained static or slowly escalated despite the introduction of the CAM. Of note, Case 6 was prescribed Cotrimoxazole once daily as prophylaxis for her recurrent chest symptoms prior to study initiation and this was continued throughout the study period.

Five of the six participants had persistent or recurrent CXR abnormalities prior to the study. Of these five, four demonstrated radiological resolution after initiation of the CAM. Overall, there were no trends or changes in PFT's during the course of the study.

Qualitative information

Patient and family satisfaction with the CAM was high with mean treatment satisfaction scores of 92% (range 85%-97.5%). Qualitative feedback was overwhelmingly positive. Ease of use, prophylactic benefits, reduced treatment time, greater treatment comfort and efficiency were highlighted as the most common positive aspects. Negative aspects identified the limited portability of the device, noise and age appropriate resistance to use initially in the two youngest participants.

Compliance

The recommended daily usage was undertaken in all cases for the first six months of the study period, and for the entire study period in three cases. All participants reported regular usage (ranging from twice daily to twice weekly) when well for the entire two year study period. This was increased to multiple times a day when unwell in all cases to aid secretion clearance. One patient (case 4) complained of sore ears and another (case 6) experienced initial musculoskeletal chest pain. Inspiratory and expiratory pressure was subsequently reduced by ±5cmH2O with acceptable effect in both cases. No other complications were reported.

Table 3: Statistical analysis of outcome measures at 1 and 2 years post CAM initiation

Outcome measure	Mean (SD) 1 yr Pre CAM	Mean (SD) 1 yr Post CAM	Mean difference (95% CI)	p-value	Mean (SD) 2 yrs post CAM	Mean difference (95% CI)	p-value
Days hospitalised	30.2 (29.7)	13.0 (18.3)	-17.2 (-35.0 to 0.7)	0.06	3.3 (4.1)	-26.8 (-58.3 to 4.7)	0.08
Number of hospital admissions	2.3 (1.6)	1.3 (1.5)	-1.0 (-2.5 to 0.5)	0.14	0.8 (1.2)	-1.5 (-3.5 to 0.5)	0.11
Number of community antibiotic prescriptions	3.7 (2.4)	3.5 (2.1)	-0.2 (-1.7 to 1.4)	0.79	3.0 (2.7)	-0.7 (-3.9 to 2.6)	0.62

CAM: Cough Assist Machine

Table 4: Summary of secondary outcome measures

	Pre CAM Initiation					Post CAM initiation				
Case	PFT 2 years prior to enrolment		Chronic radiological abnormalities	Community Antibiotic courses		PFT 2 years following study enrolment		Chronic radiological abnormalities	Community Antibiotic courses	
	FVC	FEV_1		-2 years	-1 year	FVC	FEV_1		+1 yrs	+2 yrs
1	0.92 (29%)	0.83 (30%)	Y	7	4	0.79 (20%)	0.69 (18%)	N	3	0
2	Too Young		Y	2	8	Too Young		N	7	6
3	1.48 (62%)	1.18 (58%)	Y	6	4	1.73 (59%)	1.16 (43%)	N	2	0
4	0.65 (50%)	0.50 (41%),	N	1	1	0.72 (40%)	0.68 (42%)	N	1	2
5	1.31 (96%)	0.91 (74%)	Y	1	2	1.58 (90%)	1.42 (89%)	N	4	5
6	0.59 (19%)	0.46 (15%),	Y	2	3	0.55 (19%)	0.57 (19%)	Y	4	5

CAM: Cough Assist Machine; PFT: pulmonary function tests; FVC: forced vital capacity; FEV_1: forced expiratory volume in the first second; Y: yes; N: no;

DISCUSSION

CAM's are an expensive and therefore scarce resource in New Zealand. Furthermore, there is limited information regarding the benefits of long term use in children with NMD. This study supports previous work highlighting the safety, tolerance, and acceptability of CAM's in children (Fauroux et al 2008, Gauld 2009, Miske et al 2004, Moran et al 2013). While the cohort of this study was small and heterogeneous, it demonstrates the ability of CAMs to reduce hospitalisation time despite the inexorable decline in pulmonary function documented in children with severe NMD's. Furthermore, in some cases, resolution of radiological abnormalities, and reduced community antibiotic use was also seen.

Two previous studies have examined the effects of long-term use of CAM's in children. In a similar study, Moran et al (2013) considered hospitalisation time for children with NMD using CAM at home. Our study supports the findings of Moran et al demonstrating days hospitalised as the primary improvement for all participants over the study period. Though Moran was able to demonstrate a slightly larger magnitude of change, the trend is clear in both studies. The limiting factor for both studies is seen in the small sample size and subsequent limited statistical power.

Neither study was able to demonstrate a statistically significant effect on the number of hospital admissions, though a reducing trend is evident in our data. This may suggest a reduction in both severity of respiratory infections or greater empowerment of families to manage inter-current infections out of hospital. Feedback from our families suggests elements of both are true, along with improved overall respiratory health.

In addition to reduced hospitalisation time and radiological resolution, a high level of treatment satisfaction was observed. A similar level of treatment satisfaction was seen in both an on-demand CAM programme for adults with ALS (Vitacca et al 2010) and a patient survey regarding the impact of CAM on life style in paediatrics (Moran et al 2013).

The positive qualitative findings reported from parental surveys in Moran et al (2013) are supported by the feedback we received from the families involved in our study. Key elements in both studies were the ability to remain at home during respiratory infections, prevention of infections through regular effective clearance, and increased well-being. Both study groups reported the size of the CAM and its reliance on mains power as negative elements of the machine but none felt these outweighed the positive aspects. In addition to these elements, our study families identified reduced treatment time and greater treatment practicality as major bonuses. These elements have been highlighted previously as benefits in the acute paediatric setting (Chatwin and Simonds 2009); though these have not been considered before in the community context.

Although we had intended to measure quality of life using the PEDS-QOL, our participants and their families found this questionnaire inappropriate to their situations and as such they were not completed. The questionnaires that we designed for this study were not validated, but based on domains found to be important in past research of parent satisfaction for paediatric nursing and hospital care i.e. information, outcome, harm from treatment, staff professionalism and overall satisfaction (Bitzer et al 2012, Erden et al 2006, Thomas et al 1995, Thompson and Sunol 1995).

In our questionnaires we also included device effectiveness, reliability convenience, ease of use and portability, because these had been identified as important aspects of parental satisfaction with medical technology (McNamara et al 2009). In future research we suggest a specific quality of life questionnaire needs to be constructed and validated for use in this population.

Miske et al (2004) described a cohort of 62 young people (median age 11.3 years) using CAM's for mean duration of 13.4 months (Miske et al 2004). Only five of the 62 had reduced lower respiratory tract infections (LRTI's) post-treatment and demonstration of this was reported to be difficult due to the small number of LRTI's experienced pre-treatment (Miske et al 2004). In our series, the primary criterion for allocation of CAM's was increasing LRTI's, and subsequent hospitalisation. As such, an effect on this variable was more easily demonstrated.

In the study by Miske et al (2004), resolution of chronic atelectasis over the study period was reported in only four of the 64 participants. In contrast, in our study, four of the five cases with chronic radiological changes demonstrated complete resolution over the study period. This suggests that an impact on radiology is possible and the difference in findings for both LRTI's and radiological resolution may simply reflect the stage in disease at which the CAM was introduced. This may be similar in regards to the findings for community antibiotic use. In our first three cases, both hospitalisation and community antibiotic use declined, suggesting an overall improvement in respiratory management. Many children with respiratory complications of NMD present with persistent atelectasis and likely increased bacterial load. As such, they are at greater risk of bacterial super-infection. Though CAM's are unable to prevent infections, we suggest that, through optimising airway clearance, the severity and complications of respiratory infections can be reduced. As identified previously, reduced hospital admissions may reflect a greater capacity and confidence of families to manage increased respiratory symptoms at home. This may explain why for some participants, community antibiotic prescriptions increased as hospitalisation time reduced. Given the small cohort of this study, however, these suggestions can only be speculative. Further research is required in this area to determine the optimal point of initiation of the device to balance burden of care and cost with maintenance of optimal respiratory health.

Increased LRTI, and failure to maintain respiratory status with standard ACT's have been suggested as clinical indicators for the initiation of the CAM (Chatwin and Simonds 2009, Miske et al 2004). Pulmonary function tests have been proposed as the main means of guiding ACT initiation in some types of NMD. In Duchenne's Muscular Dystrophy (DMD) a CPF of <160L/min has been proposed as the level at which a CAM is likely to be required (Bach et al 1997). In children, initiation of domiciliary CAM has been proposed when Peak Expiratory (PE) Max dropped below 60cmH20 (Miske et al 2004). However, clinically these thresholds have their limitations due to difficulties gaining reproducible figures in those with severe restrictive patterns and children under the age of six (Miske et al 2004).

Though an increase in FVC and Peak Inspiratory (PI) Max has been hypothesised with CAM use (Bach 2002), no evidence of effect was seen on PFT's of participants in our group. This may reflect both the longevity of disease prior to CAM initiation and disease severity of participants. It is also noted that while five of the six children had established kyphoscoliosis at initiation of CAM. Case 2, the youngest child, developed kyphoscoliosis regardless of regular CAM use from two years nine months of age. It has been proposed that CAM may assist in maintaining lung compliance and chest wall range of motion (Chatwin et al 2011), which should theoretically prevent development of kyposcoliosis. The patients and caregivers in this series were encouraged to utilise the CAM a minimum of once daily when well and increase use as required to clear secretions when unwell. This decision was made because, although limited evidence exists, daily prophylactic use has been proposed to maintain expansion (Bach 2002), clear secretions in those with chronic sputum retention, and ensure familiarity (and thus treatment effectiveness) is maintained (Miske et al 2004).

A limitation of the CAM model used in this study was the inability to log usage data to provide an objective measure of treatment compliance. The machines were reported to be used from twice daily to twice weekly when participants were clinically well and multiple times a day when unwell to clear secretions. This varied usage is similar to that reported in other studies (Miske et al 2004, Moran et al 2013). As newer digital CAM's with compliance downloadable data become more available, further research opportunities in the area of compliance can be explored.

Pressure settings were individualised in a comparable way to that described in other studies (Chatwin et al 2011, Miske et al 2004). Despite a relatively homogenous group, pressures prescribed ranged from ±25 to 45cmH2O (median ±35cmH2O). Model based research suggests a minimally effective pressure of ±30cmH2O (Go´mez-Merino et al 2002). Clinical papers have described pressure use in paediatrics from ±15cmH2O (Miske et al 2004), to ±50cmH2O (Bach 2002), and this reflects the need to develop individualised programmes based on clinical assessment rather than on age or disease type (Miske et al 2004). Inspiratory and expiratory times for the participants in this study were not set, as all caregivers were taught to utilise the device on manual mode. This was done to enable caregivers to synchronise the device to the child, depending on individual needs and clinical presentation, and to provide the children with a level of control. To date, criteria to determine when manual or automatic modalities should be used in children are lacking (Panitch 2009).

A limitation to this study was that our sample comprised of a small and heterogeneous group. However, given the small prevalence of children with such significant neuromuscular weakness and subsequent respiratory morbidity to fulfil our criteria, an extended multi-centre international trial would

be required to address this. Though a randomised controlled trial would be considered optimal, growing research support for CAM's, along with their gradual increased use in clinical practice, means withholding this treatment from study participants may be difficult to justify ethically.

In addition, although community antibiotic prescription was recorded for each case, this does not necessarily equate with antibiotic use, due to the potential of non-compliance with medication use.

Lastly, specific cost benefit analysis has not been undertaken for this study. However with a day's admission to the medical subspecialty ward at Starship costing $1600 NZD and one day in the Paediatric Intensive Care Unit costing $6000 NZD the approximately $12,000 NZD outlay for a CAM presents a fiscally as well as clinically attractive option.

CONCLUSION

This report demonstrates the potential effectiveness of CAM's in reducing hospitalisation for respiratory infections and improving chest radiology and community antibiotic prescriptions in children with severe NMD. We hope this study and its report will assist both those considering utilising the device clinically and in the development of a protocol for larger prospective studies. Furthermore, the findings may encourage paediatric services to consider trial of CAM's as an effective means of supporting those children who are experiencing significant respiratory morbidity despite optimal traditional management. Even if this intervention and investment only off sets the healthcare costs from paediatric to adult years, the feedback from children, young people and their families suggests the investment is justified.

KEY POINTS

- Home use of the Cough Assist Machine can safely reduce hospitalisation for respiratory infections in children with neuromuscular disease within the New Zealand context.
- A positive impact may also be seen on chronic chest radiological changes with home use of the Cough Assist Machine.
- Qualitative feedback from children and families supports a high level of treatment acceptability and satisfaction with home Cough Assist Machine use.
- Prophylactic home treatment with Cough Assist Machines could be considered a safe, effective and fiscally responsible means of managing severe respiratory morbidity in children with neuromuscular disease.

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Permissions

Ethics

The study received ethical approval from the Northern X branch of the Health and Disability Ethics Committee (Ref #NTX/09/03/018) In addition approval was obtained from the Auckland District Health Board Research Review Committee (A+4351) and the Maori Research Review Committee All caregivers gave written informed consent for inclusion in the study. All children provided informed verbal or written, assent or consent appropriate to their age and developmental stage.

DISCLOSURES

This study received no funding from any source. None of the authors identify any conflicting or competing interests

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