Airway Clearance Management with Vaküm Technology in Subjects with Ineffective Cough: A Pilot Study on the Efficacy, Acceptability Evaluation, and Perception in Children with Cerebral Palsy

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Background: In the medical complexity of a children with cerebral palsy, impaired airway clearance represents a major problem, leading to significative respiratory morbidity and mortality. Its management is difficult because of limited cooperation and poor tolerance to invasive treatments. Free Aspire[®] (FA) is a device designed to remove bronchial secretions noninvasively, without generating cough.

Methods: The aim of our pilot prospective study is to assess the efficacy and acceptability of FA in removing airway obstruction in subjects with cerebral palsy and ineffective cough. We enrolled 11 subjects. At enrollment and after 3, 6, and 12 months, we collected data regarding health care resources use for respiratory exacerbations, perceived efficacy, and acceptability of treatment.

Results: We observed a reduction in emergency room (ER) accesses, home pharmacological treatment, hospitalizations number, and hospital stay length. In particular after 12 months we observed a reduction of 74% in ER accesses and home pharmacological treatment, 38% in hospitalizations number, and 17% in hospital stay length for respiratory exacerbations. The 100% of caregivers considered the treatment effective and simple to use and noted an improvement in subjects' general condition. They also reported good treatment tolerance of subjects, with an overall good compliance.

Conclusion: The study demonstrates that FA is an effective device for the removal of bronchial secretions, with a positive caregivers' perception, that favored a good long-term compliance.

Keywords: cerebral palsy, ineffective cough, Free Aspire

Introduction

AIRWAY CLEARANCE (AC) is the physiological process that promotes the removal of endobronchial secretions.¹ Proper AC management is critical to maintain a good health state.^{2,3} A lot of methods exists for this purpose, each one with not only well-recognized efficacy but also critical issues that could make it difficult to be used.^{5–8} A new device that could be a help in the AC is Free Aspire[®] (FA). FA (MPR, Medical Products Research S.r.l, Italy) is an electromedical device for removing bronchoalveolar secretions. The machine is based on Vaküm technology, which accelerates expiratory flow through the Venturi effect from a special connector. The process happens only during the expiratory phase and is proportional to airflow on spontaneous breathing, according to the natural rhythm of the

subject's respiratory function and without needing any subject's collaborations. The secretions slide along the layer of liquid lining the bronchial epithelium until they reach the glottis from where they are swallowed; therefore, no respiratory effort is required for the elimination of the secretions. No negative pressure is generated inside the lungs; therefore, there is no risk of airways collapse. FA does not require an efficient cough and offers a gentle secretion removal solution (Fig. 1). To date, little is known about pediatric population and neurological conditions or neurological impairment⁹ and, to our knowledge, any studies have ever investigated the perception of efficacy and the acceptability of treatment by caregivers. The aim of our pilot prospective observational study is to assess the efficacy and acceptability of FA in removing airway obstruction in subjects with cerebral palsy followed at the

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FIG. 1. Mucus clearance by 2-phase gas–liquid flow mechanism: The movement of air above a layer of mucus develops a shearing force over the surface of this liquid layer. When the shearing force exceeds the surface tension in the mucous layer, the mucus starts to move in the direction of the air flow.

pediatric pulmonology service of the S. Orsola-Malpighi University Hospital of Bologna.

Materials and Methods

The Institutional Review Board (IRB) of our hospital approved the study and all parents or legal tutors signed an informed consent to the treatment protocol. Inclusion criteria were the following: diagnosis of cerebral palsy by a qualified medical specialist, ineffective cough, defined as at least 1 hospitalization for pneumonia or 2 lung infections per year treated with oral or intravenous antibiotics,¹⁰ age at the beginning of treatment <14 years old, and follow-up of at least 12 months. Exclusion criteria were intolerance to the treatment manifested as desaturation and/or dyspnea at the test session. We prescribed the treatment at home, asking caregivers to perform at least 3 sessions per day lasting 20 min each. Baseline data were collected at enrollment (T0) from the parents of subjects and using our hospital database about clinical history of each subjects, especially the use, in the year before, of health care resources for respiratory exacerbations: number of emergency room (ER) visits, number and length of hospitalizations, and number of respiratory exacerbations treated at home, defined as episodes that did not require hospitalization, but where it was necessary a change of the usual therapy and a prescription of systemic corticosteroids and/or antibiotics by the pediatrician¹¹. Follow-up visits were scheduled at 3 (T1), 6 (T2), and 12 (T3) months after the beginning of treatment. At each visit we evaluated the perception of efficacy and the acceptability of treatment, proposing 2 closed-ended questionnaires to caregivers. We used a validated questionnaire, previously utilized in other published studies,^{12–13} about the perception of efficacy was based on a visual analog scale (VAS). It was divided into 4 parts, each one with a score from 0 (best opinion) to 10 (worst opinion): usefulness of treatment, improvement of symptoms, secretion reduction, and subject's general conditions. The 4 parts together composed a score from 0 (best opinion) to 40 (worst opinion). The acceptability questionnaire was based on a Comfort Scale.¹⁴ It was divided into 3 parts. The first part was composed by 3 claims: “the device is effective in removing secretions from the airways,” “the device is simple to use,” “the treatment is well tolerated,” each one with 6 response options: extremely agree, agree, undecided, disagree, extremely disagree, and abstained. The second part was composed by 2 questions: “how often was the treatment discontinued in the previous period?” and “how often was the treatment not performed in the previous period?” each with 5 response options: never, 1 time, 2–4 times, 5–7 times,

and >7 times. In the third part, we asked parents if they had noticed a change in the subject clinical condition for the previous period, with 7 response options: marked improvement, moderate improvement, slight improvement, no improvement, slight worsening, moderate worsening, and marked worsening. To evaluate the efficacy of treatment, at the last follow-up visit, we collected data about health care resources use in 12 months of treatment (T12), considering the clinical indicators listed earlier, comparing with those at enrollment. The health care use variables were examined with the Shapiro–Wilk test, which revealed a non-normal distribution. Therefore, Wilcoxon signed-rank test for paired pre–post comparisons was adopted. *P* values <0.05 were considered statistically significant. The questionnaire results were expressed descriptively as frequencies and mean values.

Results

Eleven subjects (5 males) met the inclusion criteria. The mean age at enrollment was 5.5 ± 4.3 standard deviations (SD) years. The mean weight at enrollment was 20.1 ± 6.5 kg. Gestational age (GA) average was 35.7 ± 3.8 weeks. Five subjects (4 males) were premature and 2 of them (1 male) were very preterm (GA <32 weeks). All subjects had severe cognitive and behavioral retardation with dependence for all activities of daily living and spastic tetraparesis resulting from hypoxic-ischemic encephalopathy in the perinatal period. All subjects had a history of aspiration and were receiving medical treatment for gastroesophageal reflux. At enrollment 4 subjects were fed by gastrostomy tube, 2 of them had also undergone fundoplication. Two subjects were receiving a chronic therapy with antiasthmatic drugs. One subject had undergone tracheostomy. Clinical characteristics and demographic features of subjects are summarized in Table 1. In the 12 months preceding the beginning of treatment, all subjects had suffered from respiratory exacerbations: 9 subjects had accessed the ER, 6 had been hospitalized, 9 had been treated at home by the general practitioner. All subjects completed a 12-month study period, during which the treatment was performed at home with a mean of 2.5 sessions per day lasting 20 min each. During the entire study period, no adverse events were reported, even when it was performed close to mealtime. Health care resources use, as reported at enrollment and

TABLE 1. DEMOGRAPHIC AND CLINICAL CHARACTERISTICS OF THE STUDY POPULATION (N=11)

Demographic characteristics	
Gender: M/F	5/6
Age: mean $\pm$ SD (range)	5.5 ± 4.3 (1–13)
GA: mean $\pm$ SD (range)	35.7 ± 3.77 (30–40)
Preterm (M/F)	5 (4/1)
Caucasian/other ethnic group	8/3
Clinical characteristics	
PEG	4
Nissen fundoplication	2
Tracheostomy	1
ER admission for respiratory exacerbations	9
Hospitalizations	6
Home treatments	9

M, males; F, females; SD, standard deviation; GA, gestational age; PEG, percutaneous endoscopy gastrostomy; ER, emergency room.

TABLE 2. HEALTH CARE RESOURCES USE IN THE 12 MONTHS BEFORE ENROLLMENT AND IN THE 12 MONTHS OF TREATMENT WITH FREE ASPIRE

	12 months before enrollment			12 months of treatment			Change %	P value
	Mean $\pm$ SD	Maximum	Total	Mean $\pm$ SD	Maximum	Total		
ER accesses number of hospitalizations	2.1 $\pm$ 2.8	10	23	0.5 $\pm$ 0.7	2	5	-74	0.03*
	0.7 $\pm$ 0.8	2	8	0.5 $\pm$ 0.7	2	5	-38	0.18
Length of hospitalizations	8.8 $\pm$ 18.6	61	88	7.3 $\pm$ 20.3	65	73	-17	0.18
Home pharmacological treatment for respiratory exacerbations	4.5 $\pm$ 4.6	12	50	1.2 $\pm$ 1.5	5	13	-74	0.04*

* $P < 0.05$. The mean and maximum are per subject.

after 12 months of treatment, is shown in Table 2. The perception of treatment efficacy, assessed by VAS, is shown in Table 3. In all the 4 questionnaire parts and, therefore, globally, mean scores were below the intermediate value (Fig. 2). In the study period, 100% of the caregivers agreed with the statements “the device is simple to use” and “the device is effective in removing secretions from the airways”. The percentage of parents who expressed agreement with the statement “the treatment is well tolerated” showed a reduction from 81.9% at T1 and T2 to 45.5% at T3. Answering the question about how often the treatment was discontinued in the previous period, 54.5% of parents said “never,” at each visit. Answering the question about how often the treatment was not performed in the previous period, the percentage of caregivers stating “never” decreased during follow-up from 63.6% at T1 to 45.5% at T2 and 36.4% at T3. At each follow-up visit, 100% of caregivers noted an improvement in the subject’s general condition compared with before study period.

Discussion

Cerebral palsy is a neurological disease with neurological impairment and it is part of a wide group of disorders associated with an important respiratory morbidity and mortality consequent to impaired AC, due mostly to ineffective cough. There is evidence that nonpharmacological intervention for AC improvement is useful without major adverse events reported¹⁵. Several techniques, either manual or mechanical, are available for this purpose, each one with not only proven efficacy but also some critical features, such as expensiveness, difficulty of use, need for caregiver

training end/or poor subject collaboration/tolerance that can lead to poor compliance and, therefore, to low long-term efficacy. To date, only 1 published study, by Garuti et al., evaluated the use of FA in pediatric subjects with ineffective cough. In this study, focused on children with cerebral palsy, authors found a reduction in home visits by primary care pediatrician, days spent in hospital, and days of antibiotic treatment for respiratory problems comparing the 6 months before the treatment with 6, 12, and 18 months of treatment. They concluded that this method is feasible and effective in reducing respiratory complications and health care resources need, with importance of regular use to maintain these results in time. In our study, we observed a reduction of 74% in the number of respiratory exacerbations that required ER access ($P = 0.03$) or a home pharmacological therapy prescribed by the pediatrician ($P = 0.04$), during the 12 months of treatment, compared with the 12 months before. Moreover, comparing the same periods, we observed a reduction of 38% in the number of hospital admissions and of 17% in the length of hospitalization, even if without statistical significance ($P = 0.18$ in both cases). These results, although not all statistically significant due to the small sample, are consistent with those obtained from Garuti et al. although considering different temporal periods. We choose to analyze data at 3, 6, and 12 months, to eliminate bias due to the seasonality of the most common forms of airway inflammations. Our results are also consistent with those of other studies regarding the effectiveness of other nonpharmacological interventions for the management of AC in pediatric subjects. Plioplys et al.¹⁶ found a 50% reduction in the number of pneumonia and antibiotic therapy and a 66% reduction in the number of hospitalizations for pneumonia in

TABLE 3. RESULTS OBTAINED IN THE QUESTIONNAIRE, BASED ON VISUAL ANALOG SCALE, ON FREE ASPIRE EFFICACY PERCEPTION BY THE CAREGIVERS

Usefulness of treatment		Symptoms relieve		Secretion reduction		General conditions		Global score	
Mean $\pm$ SD	Range	Mean $\pm$ SD	Range	Mean $\pm$ SD	Range	Mean $\pm$ SD	Range	Mean $\pm$ SD	Range
T1									
0.8 $\pm$ 1.2	0–3	1.5 $\pm$ 1.0	0–3	1.9 $\pm$ 2.9	0–10	2.0 $\pm$ 1.2	0–4	6.2 $\pm$ 4.1	1–13
T2									
0.7 $\pm$ 1.0	0–2	0.6 $\pm$ 0.9	0–2	1.6 $\pm$ 1.4	0–5	1.7 $\pm$ 1.5	0–5	4.8 $\pm$ 2.9	0–8
T3									
1.1 $\pm$ 1.8	0–6	1.4 $\pm$ 1.3	0–4	2.6 $\pm$ 1.5	0–5	1.0 $\pm$ 1.2	0–3	5.8 $\pm$ 3.8	2–14

The questionnaire was divided into 4 parts: usefulness of treatment, symptoms relieve, secretion reduction, and patient’s general conditions. Each one had a score ranging from 0 (best opinion) to 10 (worst opinion). A global score, ranging from 0 (best opinion) to 40 (worst opinion), resulting from the 4 parts, is also shown. T1: 3 months, T2: 6 months, and T3: 12 months of treatment.

FIG. 2. Improvement in the subject's general condition compared with the free-of-treatment period.

the year of VEST therapy for the previous year was found in 7 children with quadriplegic cerebral palsy. Yuan et al. compared high-frequency chest wall oscillation with chest physiotherapy and found no significant reduction in the need for hospitalization and home antibiotic treatment in the follow-up period for both group in a pediatric population composed of children with a neuromuscular disease or cerebral palsy. These results show the lack of a significant inferiority of the treat-

ment with FA than high-frequency chest wall oscillation. It is not possible to compare the effectiveness of other AC methods as the parameters used to assess their effectiveness are different from ours. When we started prescribing this treatment, we decided to monitor not only health care resources need, but also the perception of efficacy and the acceptability of treatment by caregivers, which is an important feature in maintaining a good long-term compliance. In their study, Yuan

et al. found a greater adherence to high-frequency chest wall oscillation therapy, with some caregivers reporting that manual chest physiotherapy was harder to perform especially in subjects with spine deformity. In the 12 months of treatment with FA, the caregivers of our subjects expressed a positive overall judgment, as evidenced by the scores assigned to the questionnaires. In addition, all parents felt the treatment simple to use and effective in removing endobronchial secretions. Subjects showed well tolerance to the treatment, especially in the first 6 months of use, whereas at 12 months 3 families reported a low tolerance. Compliance was overall good, although a reduction of tolerability and compliance was reported, as no more than 1 family reported the need to interrupt the treatment or its nonexecution for >7 times in a month before each visit. No family had to stop the treatment completely, the reduction of tolerance and compliance could be explained, more for the patient's preference, than for the technical use and characteristics of the device. In particular the reduction of use is reported in the summer months where the feeling "of the necessity of use" of the families was lower. At every visit, all parents noticed an improvement in the general conditions (reduction of prescription of corticosteroids and/or antibiotics, reduction of infections, improvement of food tolerance, and "more interacting children") compared with the previous period. The principal limits of our study are the small sample size and the lack of comparison with a group of subjects using other methods of AC management.

Conclusions

Our experience demonstrates a positive effect of home FA treatment in children with cerebral palsy, improving general health state and decreasing health care resources utilization, with a positive caregivers' perception, that favored a good compliance even several months after the prescription. This treatment is noninvasive, it does not require caregiver training, nor subject cooperation, and it can be performed close to mealtime. FA offers an effective method for lower airway secretions clearance for children with cerebral palsy.

Author Disclosure Statement

The authors declare no conflicts of interest.

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