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Impact of tracheostomy on swallowing performance in Duchenne muscular dystrophy

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ABSTRACT

Mechanical ventilation has improved survival in patients with Duchenne muscular dystrophy (DMD). Over time, these patients experience upper airway dysfunction, swallowing impairments, and dependency on the ventilator that may require invasive mechanical ventilation via a tracheostomy. Tracheostomy is traditionally believed to further impair swallowing. We assessed swallowing performance and breathing-swallowing interactions before and after tracheostomy in 7 consecutive wheelchair-bound DMD patients, aged 25 ± 4 years, over a 4-year period. Chin electromyography, laryngeal motion, and inductive respiratory plethysmography recordings were obtained during swallowing of three water-bolus sizes in random order. Piecemeal deglutition occurred in all patients over several breathing cycles. Half the swallows were followed by inspiration before tracheostomy. Total bolus swallowing time was significantly shorter ($P = 0.009$), and the number of swallows per bolus significantly smaller ($P = 0.01$), after than before tracheostomy. Invasive ventilation via a tracheostomy may improve swallowing.

1. Introduction

The introduction of mechanical ventilation (MV) has improved survival in patients with Duchenne muscular dystrophy (DMD), from about 15 years in the 1960s to more than 30 years now [1–4]. However, the patients experience increasing physical disability and dependence on care with advancing age [3].

Invasive or noninvasive mechanical ventilation can be used for long-term ventilatory assistance [5,6]. Noninvasive ventilation (NIV) is initially offered at night to treat sleep-related breathing disorders and hypoventilation, which are common in patients with DMD [7]. As the disease progresses, hypoventilation starts to occur during the day, requiring daytime ventilatory assistance, generally as intermittent positive-pressure ventilation via a mouthpiece or nasal mask [8]. Dependence on the ventilator increases over time, and many patients become unable to use a mouthpiece and to tolerate prolonged nasal ventilation during the day. Eventually, severe upper airway dysfunction develops, impairing NIV efficiency. At this stage, tracheostomy may be considered [6,8].

The severe upper airway dysfunction seen in DMD is often associated with swallowing difficulties. These difficulties affect 18% of patients above the age of 18 years and increase with advancing age [9]. Other factors contribute to cause feeding difficulties, including weakness of the masticatory muscles, malocclusion, and inability to self-feed [9]. The feeding difficulties often develop insidiously, being frequently missed by family members and healthcare professionals, [10] and may lead to inadequate food intake. Thus, malnutrition develops in up to 44% of DMD patients [8,11]. Aspiration is another severe consequence of impaired swallowing that becomes increasingly common as the disease progresses [8]. The ATS recommends gastrostomy tube placement when adequate nutrition cannot be safely achieved via oral feeding [8]. However, we have noted that some patients who underwent gastrostomy before tracheostomy became capable of oral feeding during invasive mechanical ventilation via the tracheostomy. This observation suggests that tracheostomy may provide improvements in quality of life by obviating the need for gastrostomy.

The objective of this study was to assess the impact of tracheostomy combined with positive-pressure ventilation on breathing/swallowing interactions and swallowing performance in DMD patients in whom permanent NIV was either not sufficiently effective or not well tolerated.

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2. Methods

2.1. Study population

We conducted a prospective observational study from February 2006 to February 2009. The relevant ethics committee (Hôpital A. Paré, Paris, France) approved the study, and written informed consent was obtained from all patients before study inclusion.

Patients with DMD were recruited during routine follow-up visits at the ventilation unit of the Intensive Care Department of the Raymond Poincaré Teaching hospital (Garches, France) if they had been in stable clinical condition for at least the past month, were receiving NIV, and had consented to undergo tracheostomy. Tracheostomy was decided because NIV was inadequately effective or poorly tolerated. Informed consent for tracheostomy was obtained after a detailed discussion of social factors and living arrangements [6].

2.2. Study procedures

Thoracic and abdominal movements were monitored using respiratory inductive plethysmography. Swallowing was monitored noninvasively, using electromyography to detect submental muscle activity via skin-surface electrodes on the chin and a piezoelectric sensor placed between the cricoid and thyroid cartilages, as described elsewhere [12–14]. All signals were digitized and recorded directly on a personal computer.

2.2.1. Experimental protocol

The patient was seated comfortably with the head and neck maintained in the preferred position. All patients were assessed just before tracheostomy and 3 months after tracheostomy. After tracheostomy, mechanical ventilation was used during the assessment [6]. Before tracheostomy, patients were allowed to choose whether to use NIV during the assessment, as no recommendations existed for this situation [15]. Water boluses were placed in the mouth using a syringe. Three bolus sizes were used, (5, 10, and 15 ml), in random order. Patients were blinded to bolus size. As previously published [12,14,16–18], four sets of three boluses were studied, without using the same bolus size twice consecutively. Each bolus was separated by 2 min. Patients were asked to swallow normally and as efficiently as possible. The recording was checked and swallows were counted until the patient was able to open his mouth in order to demonstrate mouth emptying. At completion of each measurement session, patients used the modified 10-point Borg scale to evaluate their worst sensation of respiratory difficulty during the session [19].

2.3. Data analysis

The investigator who read the recordings was blinded to the experimental condition. Blinding was maintained by analyzing swallowing independently from thoracic and abdominal movements. Swallowing onset was defined as the onset of phasic submental electromyographic activity and swallowing termination as the onset of downward laryngeal movement detected by the piezoelectric sensor [12,14]. For each bolus size, we recorded swallowing duration, number of swallows, and number of ventilatory cycles required to swallow the entire bolus. The percentages of swallows followed by expiration were computed. In normal individuals, swallowing is nearly always followed by expiration [20–22].

2.4. Statistical analysis

All results are reported as mean ± standard deviation in the text and as mean ± standard error of the mean in the figures. Statistical tests were run using the Stat View 5 package (SAS Institute, Grenoble, France).

We used two-way analysis of variance (ANOVA) for repeated measurements, with tracheostomy and bolus size as the two factors. In patients who were unable to swallow the largest bolus, [12,14] a paired *t*-test was used to compare results obtained with the smallest bolus. A paired *t*-test was also used to compare Borg scale results. *P* values less than 0.05 were considered statistically significant.

3. Results

3.1. Study population

We included 7 consecutive DMD patients. Table 1 reports their main anthropometric characteristics and respiratory parameters. All patients had a Hauser Ambulatory Index [23] of 9/9, indicating confinement to the wheelchair and inability to self-transfer. In addition, none of the patients was able to self-feed. However, all patients were able to swallow fluids and blended food; thus, none required a gastrostomy. The reason for tracheostomy was inadequate effectiveness of NIV in 1 patient (#5) and poor tolerance of NIV during the long periods of use required in the other 6 patients. The duration of noninvasive ventilation before tracheostomy was 60 ± 32 months (Table 1).

Assist-control mechanical ventilation was used, with an uncuffed tracheostomy tube after tracheostomy. Initially, tidal volume was set between 10 and 12 ml/kg and the backup respiratory rate was set two to three breaths per minute below

Table 1
Patient characteristics.

#	Age	Height	VC Sitting	MEP	MIP	SB $PaCO_2$	Duration of NIV before tracheostomy (months)	$PaCO_2$ before/after tracheostomy under MV	Weight before/after tracheostomy
1	24	167	10	15	5	6.2	53	4.7/5.4	23.1/23.2
2	19	163	6	<5	<5	6.1	26	5.0/5.0	28.0/NA
3	23	164	8	10	15	6.6	27	4.1/3.0	24.0/27.5
4	29	163	12	10	8	5.7	51	3.6/4.5	33.0/36.0
5	25	168	12	11	11	6.8	61	6.6/5.4	49.0/52.0
6	28	175	10	13	5	NA	98	3.0/3.8	38.0/41.0
7	28	158	7	7	6	9.1	107	3.7/6.1	40.0/43.0
Mean	25	165	9	10	8	6.8	60	4.4/4.7	33.6/37.1
SD	4	5	2	3	4	1.2	32	1.2/1.0	9.4/10.5

Abbreviations: age in years; height in cm; weight in kg; VC, vital capacity expressed as the % of the predicted value; MEP, maximal expiratory pressure in cmH_2O; MIP, maximal inspiratory pressure in cmH_2O; $PaCO_2$, partial pressure of CO_2 in arterial blood in kPa; NA, not available; SB, spontaneous breathing; NIV, noninvasive ventilation; MV, mechanical ventilation via tracheostomy.