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Diagnostic value of "dysphagia limit" for neurogenic dysphagia: 17 years of experience in 1278 adults

Ibrahim Aydogdu *, Nefati Kiylioglu, Sultan Tarlaci, Zeynep Tanriverdi, Sezin Alpaydin, Ahmet Acarer, Leyla Baysal, Esra Arpaci, Nur Yuceyar, Yaprak Secil, Tolga Ozdemirkiran, Cumhur Ertekin [1]

Ege University Medical School Hospital, Departments of Neurology and Clinical Neurophysiology, Bornova, Izmir, Turkey

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HIGHLIGHTS

- Dysphagia limit is a simple electrophysiologic test for the diagnosis and evaluation of neurogenic dysphagia.
- Dysphagia limit is a reliable, noninvasive, quantitative test with a high sensitivity (92%) and specificity (91%) to detect and follow both clinical and subclinical neurogenic dysphagia.
- Dysphagia limit is easy to use, safe, and can be readily utilized by a neurologist within an EMG laboratory.

ABSTRACT

Objective: Neurogenic dysphagia (ND) is a prevalent condition that accounts for significant mortality and morbidity worldwide. Screening and follow-up are critical for early diagnosis and management which can mitigate its complications and be cost-saving. The aims of this study are to provide a comprehensive investigation of the dysphagia limit (DL) in a large diverse cohort and to provide a longitudinal assessment of dysphagia in a subset of subjects.
Methods: We developed a quantitative and noninvasive method for objective assessment of dysphagia by using laryngeal sensor and submental electromyography. DL is the volume at which second or more swallows become necessary to swallow the whole amount of bolus. This study represents 17 years experience with the DL approach in assessing ND in a cohort of 1278 adult subjects consisting of 292 healthy controls, 784 patients with dysphagia, and 202 patients without dysphagia. A total of 192 of all patients were also reevaluated longitudinally over a period of 1–19 months.
Results: DL has 92% sensitivity, 91% specificity, 94% positive predictive value, and 88% negative predictive value with an accuracy of 0.92. Patients with ALS, stroke, and movement disorders have the highest sensitivity (85–97%) and positive predictive value (90–99%). The clinical severity of dysphagia has significant negative correlation with DL ($r = -0.67$, $p < 0.0001$).
Conclusions: We propose the DL as a reliable, quick, noninvasive, quantitative test to detect and follow both clinical and subclinical dysphagia and it can be performed in an EMG laboratory.
Significance: Our study provides specific quantitative features of DL test that can be readily utilized by the neurologic community and nominates DL as an objective and robust method to evaluate dysphagia in a wide range of neurologic conditions.

1. Introduction

Swallowing is a complex sensory-motor event that consists of both voluntary and involuntary movements involving the oropharyngeal structures, esophagus, and laryngeal and respiratory muscles. Dysphagia can result from lesions anywhere along

* Corresponding author. Tel.: +90 232 3903851; fax: +90 232 3880980.
E-mail addresses: ibrahimxaydogdu@gmail.com, ibrahim.aydogdu@ege.edu.tr (I. Aydogdu).
[1] Member of the Science Academy of Turkey.

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the neuromuscular pathway from the cerebral cortex to the swallowing muscles (Hughes and Wiles, 1998; Bakheit, 2001). Neurological disorders account for about 70–80% of all patients with swallowing problems especially for the oropharyngeal region (Tally et al., 1991; Groher, 1997; Logemann, 1998). Neurogenic dysphagia can lead to failure of bolus transport and misdirection of bolus into the nasal cavity, larynx, and/or trachea (Logemann, 1998; Ertekin and Aydoğdu, 2003). Although neurogenic dysphagia accounts for three-quarters of the etiology of all dysphagias, it is often not considered among the differential diagnoses by neurologists (Humbert and Robbins, 2007). Despite being a major source of morbidity and mortality for a myriad of neurologic conditions, the interest of neurologists in neurogenic dysphagia remains low. There is a great need to conduct research in individuals with dysphagia that aims to diagnose the underlying neurologic causes. The neurogenic dysphagia should be first differentiated from the oropharyngeal focal/structural disease. The mechanism causing neurogenic dysphagia may differ in patients. However, neurologists have an important role to play to help elucidate the mechanism of the dysphagia and to give advice in a multidisciplinary context about dysphagia management such as the patient's diagnosis and prognosis (Hughes and Wiles, 1998).

In the United States, it is estimated that about 300,000–600,000 persons each year experience dysphagia, as a result of stroke or other neurological disorders (Mann et al., 2000; Paciaroni et al., 2004) and that up to 6 million adults are at risk for it (Sura et al., 2012). Dysphagia affects up to 68% of elderly nursing home residents (Steele et al., 1997), 30% of the elderly admitted to the hospital (Lee et al., 1999), 64% of stroke patients (Mann et al., 2000), and 13–38% of elderly who live independently (Kawashima et al., 2004; Roy et al., 2007).

The main consequences of neurogenic dysphagia include aspirations often resulting in pneumonia, malnutrition, and dehydration that are potentially fatal complications for patients with stroke, dementia, and other progressive neurological disorders. Further, the financial burden of chronic dysphagia is very high. Thus, screening and early diagnosis of dysphagia in high-risk populations can be significantly cost-saving (Cichero and Altman, 2012). Early diagnosis of neurogenic dysphagia is important for the quality of life and survival of affected patients. In some acute-onset neurologic conditions, such as stroke, dysphagia will often improve spontaneously over a few weeks and therefore does not need to be investigated by invasive diagnostic tests such as videofluoroscopy (VFS) or endoscopy. These cases of neurogenic dysphagia may be followed up with noninvasive screening tests such as water swallowing, water timed-test, or dysphagia limit (DL) (Hughes and Wiles, 1996; Ertekin et al., 1996, 1998a; Suiter and Leder, 2008).

On the other hand, neurogenic dysphagia could be insidious and slowly progressive in conditions such as amyotrophic lateral sclerosis (ALS), Parkinson's disease, dementia, multiple sclerosis, and some neuromuscular disorders. There may be a subclinical period before the onset of clinically overt dysphagia in these chronic neurologic disorders. The diagnosis of dysphagia in the early subclinical period may be critical for the management of its complications. Early diagnosis of this potentially devastating condition can enable early establishment of rehabilitative, medical, and surgical management approaches (Ertekin et al., 1998a, 2000a; Stambler et al., 1998; Kawai et al., 2003; Terzi et al., 2007; Bautmans et al., 2008; Chio et al., 2009; Pieterse et al., 2009; Spataro et al., 2011; Pena et al., 2012). Often, patients with chronic, progressive neurologic conditions are submitted to invasive swallowing tests. We propose a noninvasive and quantitative method for the diagnosis of subclinical and the follow-up monitoring of clinical dysphagia, which can be applied in a wide variety of conditions with neurogenic dysphagia. We and others suggest that the best-practice diagnostic method for neurogenic dysphagia should fit the following criteria: (Bautmans et al., 2008; Clave et al., 2008; Bours et al., 2009):

(1) It should be easy to apply with a low risk for adverse effects.
(2) It must be well tolerated even by patients with advanced neurologic disease.
(3) It must be objective and quantifiable.
(4) It should be rapidly performed and repeatable even at short intervals.
(5) It should be reproducible across different laboratories and clinics.
(6) It should be of low cost.

There are three types of water swallowing screening tests that are noninvasive: the 3-ounce water test (Gordon et al., 1987; De Pippo et al., 1992; Nilsson et al., 1996), the swallowing speed/timed test (Nathadwarawala et al., 1992; Hughes and Wiles, 1996), and the DL method developed by our group (Ertekin et al., 1996, 1998a). The 3-ounce water and swallowing speed/timed tests fulfill some, but not all, of the above criteria as they are not capable of demonstrating airway aspirations in patients with severe dysphagia. Further, they are not able to exclude focal structural lesions (Ertekin et al., 1998a; Hughes and Wiles, 1998; Bakheit, 2001). Although there are reports supporting the use of the 3-ounce water swallowing test to detect aspiration during swallowing (De Pippo et al., 1992; Mari et al., 1997; McCullough et al., 2000; Rosenbek et al., 2004), there is no clear consensus on this issue (Suiter and Leder, 2008).

This study is a detailed investigation of the method of DL in a large cohort of 1278 subjects consisting of 292 healthy controls, 784 neurologic patients with overt or suspected dysphagia, and 202 patients without dysphagia. The first aim of this study is to provide a comprehensive investigation of the DL in this diverse cohort. The second aim is to provide a longitudinal assessment of dysphagia in a subgroup of subjects using the DL. Given the findings from our study spanning a 17-year period, we propose that DL is a useful and an objective method that can be applied in a wide variety of neurologic conditions and severity of neurogenic dysphagia. It is easy to use, of low cost, and is a quantitative approach, thus representing an optimal swallowing method, in contrast to the traditional invasive swallowing methods with far more restricted utility.

2. Materials and methods

2.1. Subjects

A total of 292 normal healthy adult subjects (157 female, 135 male) participated in the study. These subjects were without a history of neurological and swallowing disorders and were recruited from among the hospital staff, volunteers, and relatives of patients. These control subjects ranged in age from 17 to 83 years (mean age: 45).

As many as 784 patients with overt or suspected dysphagia (those without overt symptoms but with swallowing complaints) were investigated both clinically and electrophysiologically. Another group of patients with neurological disorders (202 cases) but without overt or suspected dysphagia were also separately examined. Thus, 986 patients in total were examined (388 females, 598 males). Their ages ranged from 19 to 88 years (mean age: 51.3). They were selected from the inpatient and outpatient clinics at the Department of Neurology, Ege University Medical School, from 1995 to 2012. A further 80 patients were excluded from the study because their clinical diagnosis could not be classified among those mentioned in Table 1, which summarizes the neurological