



CASE STUDY

DISCREPANT RESULTS OF FLEXIBLE ENDOSCOPIC EVALUATION OF SWALLOWING (FEES) AND VIDEOFLUOROSCOPY SWALLOWING STUDY (VFSS) IN MYASTHENIA GRAVIS PATIENT

Susyana Tamin¹, Devianty Octavia^{1*}

¹Departemen Ilmu Kesehatan Telinga Hidung Tenggorok Bedah Kepala dan Leher, Fakultas Kedokteran, Universitas Indonesia, Jakarta, Indonesia

*Correspondence email : devyoct@gmail.com

ABSTRACT

Myasthenia gravis (MG) is an autoimmune neuromuscular disorder that causes weakness in skeletal muscles. Dysphagia is an early symptom that is often found in 15-40% of patients with myasthenia gravis. Flexible Endoscopic Evaluation of Swallowing (FEES) and Video Fluoroscopic Swallowing Study (VFSS) examinations are routinely used to diagnose dysphagia. This paper reports a case of Male, 51 years old who presented to ENT Outpatient Clinic with swallowing difficulties for 3 months. The patient then underwent initial FEES examination and consulted to Neurology Department with suspicion of MG. A diagnosis of neurogenic oropharyngeal dysphagia with silent aspiration due to MG was supported by clinical and objective swallowing examination findings. Swallowing evaluation post-therapy and rehabilitation programs were carried out. Interestingly both FEES and VFSS had a significant discrepant result. However, FEES and VFSS are complementary tests for assessing dysphagia, each with its own limitations and advantages, enabling a comprehensive evaluation when used together. In cases of dysphagia due to MG, when interpreting objective swallowing function examination, confounding factors such as fatigue, timing of examinations, and time of taking medication specifically anticholinesterase (Pyridostigmine), should be taken into consideration.

Keywords: Dysphagia; FEES; Myasthenia gravis; VFSS

АБСТРАКТ

Миастения гравис (МГ) - это аутоиммунное нервно-мышечное заболевание, вызывающее слабость скелетных мышц. Дисфагия является ранним симптомом, который часто встречается у 15-40% пациентов с миастенией гравис. Для диагностики дисфагии обычно используются гибкая эндоскопическая оценка глотания (FEES) и видеофлюороскопическое исследование глотания (VFSS). В данной статье приводится случай мужчины, 51 года, который обратился в ЛОР-клинику с затрудненным глотанием в течение 3 месяцев. Затем пациент прошел первичное обследование по методике FEES и был направлен в неврологическое отделение с подозрением на МГ. Диагноз нейрогенной орофарингеальной дисфагии с молчаливой аспирацией вследствие МГ был подтвержден клиническими и объективными результатами обследования глотания. Проведена оценка глотания после терапии и реабилитационные программы. Интересно, что результаты FEES и VFSS значительно отличались друг от друга. Тем не менее, FEES и VFSS являются взаимодополняющими тестами для оценки дисфагии, каждый из которых имеет свои ограничения и преимущества, что позволяет проводить комплексную оценку при их совместном использовании. В случаях дисфагии, обусловленной МГ, при интерпретации результатов объективного исследования функции глотания следует учитывать такие факторы, как усталость, время проведения обследования и время приема лекарственных препаратов, в частности антихолинэстеразных средств (пиридостигмина).

Ключевые слова: Дисфагия; FEES; миастения гравис; VFSS

INTRODUCTION

Dysphagia is a symptom or collection of symptoms that broadly describes swallowing difficulty, often found in myasthenia gravis (MG). Myasthenia gravis is an autoimmune disorder, caused fluctuating muscle weakness worsened by exertion. This results from disrupted synaptic transmission at neuromuscular junction. The prevalence of dysphagia in generalized MG is 15-40%, in which the severity is directly proportional to the disease. About 50% of myasthenic crisis are preceded by symptoms of dysphagia.^{1,2}

Flexible Endoscopic Evaluation of Swallowing (FEES) and Videofluoroscopic Swallowing Study (VFSS) are commonly utilized in diagnosing dysphagia, aimed to assess the efficiency and safety of the swallow. VFSS provides information on all swallowing phases, while FEES allows direct visualization of mucosa, management of secretion, and laryngeal sensitivity. Fatigable swallowing test (FST) is proposed as a standardized examination for precisely grading dysphagia MG patients and can be added to standard FEES procedures. Diagnosis and evaluation of dysphagia should be of concern in MG patients, given the severe complications and myasthenic crisis that can be precipitated. Thus, a comprehensive management strategy including pharmacotherapy of the underlying disease, rehabilitation strategy, strength training, and diet modification plays an important role in MG patients experiencing dysphagia.^{3,4,5}

It is essential to remember that MG is a medical condition distinguished by weakness in skeletal muscles, a symptom that intensifies with physical activity and diminishes during periods of rest. This report describes a case of dysphagia in MG patients, who had a significant discrepancy in FEES & VFSS results. Potential causes of those discrepancies include fatigue, different timing of the examinations, and different periods from the last pyridostigmine (cholinesterase inhibitor) consumption.

MATERIAL AND METHODS

A 51-year-old male came to Bronchoesophagology-ENT Outpatient clinic with a chief complaint difficulty of swallowing for 3 months. Accompanied by exhaustion during eating, talking, and daily activities. He felt his voice become hoarse and slurred during speech, sensation of food stuck in the throat after the third swallow, and choking. The patient lost 23 Kg of body weight in the last 3 months. Initial FEES showed severe neurogenic oropharyngeal dysphagia with silent aspiration and paresis of unilateral vocal cord. The patient was then consulted to the neurology department with MG suspicion. Myasthenia gravis composite score (MGCS) was 10, physical examinations showed cranial nerve palsies (IX, X). Brain MRI was normal, so this excluded vascular lesion. Repetitive nerve stimulation was done and there was a decrease of CMAP amplitude > 10% pre-post exercise of orbicularis oculi muscle, this leads to post-synaptic neuromuscular junction lesion (MG). The initial management strategy consisted of Pyridostigmine 2x60mg, enteral feeding (NGT), nutrition counseling, swallowing rehabilitation & NMES.

After finishing the rehabilitation program, VFSS was carried out and showed improvement in swallowing function (Figure 1). VFSS examination was done in the morning, 30 minutes after pyridostigmine consumption. Later, he was sent back to ENT for FEES re-evaluation, and whether or not the NGT can be removed. FEES evaluation was done in the afternoon, 21 hours after last pyridostigmine consumption, it showed the symmetrical movement of vocal fold, pre-swallowing leakage, delayed swallowing reflex initiation, delayed epiglottis retroflexion, residue in vallecula and pyriform sinus (Yale III-IV), and silent aspiration on thin liquid consistency (Figure 2). NGT was maintained, and patient was scheduled for another FEES examination, with addition of fatigable swallowing test (FST), 30 minutes after pyridostigmine consumption (similar to previous VFSS).

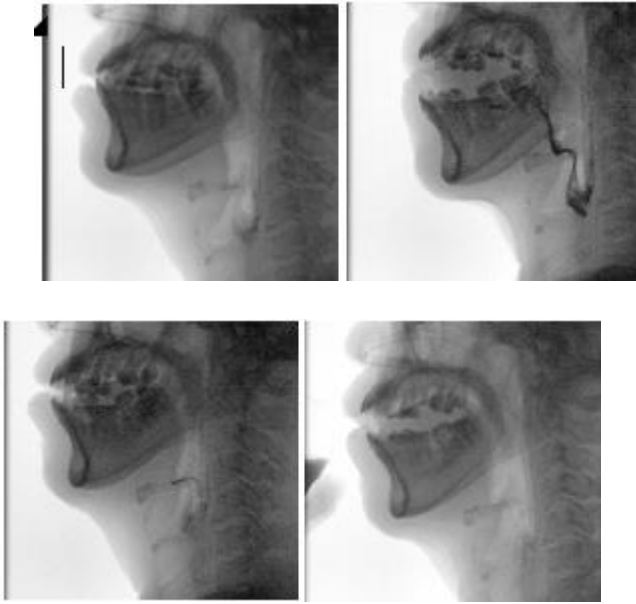


Figure 1. VFSS performed at 7AM, pyridostigmine was taken 30 minutes prior to FEES. Result showed minimal post swallowing residue, PAS Score 1 in all consistencies.



Figure 2. FEES were performed at 2 PM, pyridostigmine was taken 21 hours before FEES. Results showed; A. Pooling secretion in vallecula & pyriform sinus; B. Post swallowing residue 25-50% (Yale IV); C. The opening of UES at rest.

FEES re-evaluation was carried out at 2 PM, 30 minutes after taking pyridostigmine, and the results were different from the previous one. There were no pooling secretions (MSS 0), post swallowing residue was only found in the vallecula (25%). Followed by FST, the patient was able to swallow all 30 pieces of white bread, with minimum residue (25%) and, PAS score of 1. However, the opening of the UES was observed after swallowing the last piece of bread, which may be due to exhaustion (Figure 3). Based on these findings, the NGT was removed and patient was allowed to have an oral diet, good oral hygiene should be

maintained, and if he ever experienced any swallowing difficulty or exhaustion, or decreased body weight in the next 2 weeks, the NGT will be re-inserted for nutritional fulfilment purpose.

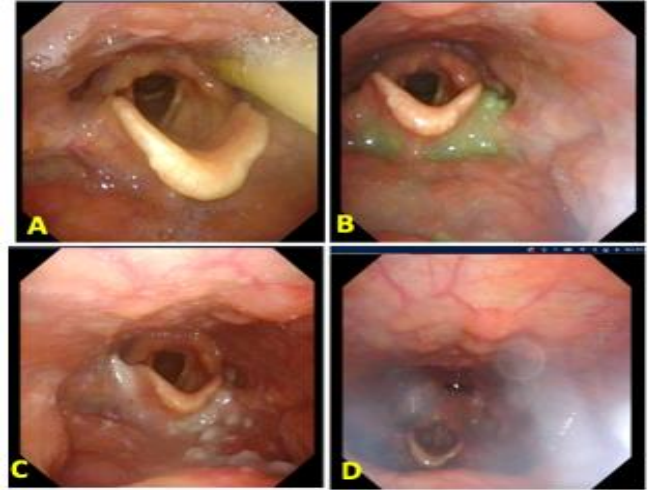


Figure 3. FEES were performed at 2 PM, pyridostigmine was taken 30 minutes before FEES. Results showed; A. No pooling secretion; B. Post swallowing residue 25% in the vallecula (Yale III); C. FST: Post swallowing residue 5-25% (Yale II) in vallecula from 1st-30th bread, PAS Score 1; D. The opening of UES observed after completing FST.

RESULT

Muscle weakness in MG patients arises due to the presence of autoantibodies that target the acetylcholine receptor (AChR) located at the neuromuscular junction. This weakness in the muscles involved in swallowing leads to difficulty in swallowing, which remains a significant cause of health issues in individuals with MG. Assessing and diagnosing dysphagia in MG patients is crucial, considering the potential for serious complications and the risk of myasthenic crisis. This assessment often involves supportive examinations such as Fiberoptic Endoscopic Evaluation of Swallowing (FEES) and Videofluoroscopic Swallow Study (VFSS).^{1,2,6}

DISCUSSION

Flexible Endoscopic Evaluation of Swallowing (FEES) evaluates the anatomical and physiological aspects of swallowing, from the oral cavity to the vocal folds, checking for

silent aspiration. It comprises three stages: pre-swallowing assessment, swallowing assessment, and therapeutic assessment. The clinician evaluates anatomy and condition, then observes swallowing using a flexible endoscope passed through the nasal passage. Various food consistencies are given, and therapeutic interventions, like postural adjustments, are applied as needed.^{7,8,9,10}

Videofluoroscopic Swallow Study (VFSS) is a diagnostic procedure used to evaluate swallowing function by capturing real-time X-ray images of the swallowing mechanism. Throughout the examination, the patient ingests food and liquid mixed with barium, which shows up clearly on X-ray images. These images allow clinicians to evaluate the coordination, timing, and efficiency of swallowing. These images also aid in identifying any abnormalities or difficulties, such as recognizing when food bolus enters the airway, leading to aspiration or penetration. The examination offers crucial information for diagnosing swallowing disorders and guiding treatment interventions.^{8,9,10,11}

FEES and VFSS are commonly used for diagnosing dysphagia due to their complementary capabilities in assessing swallowing function. FEES offers portability, lower cost, use of actual food and liquid, direct laryngeal assessment, and suitability for longer therapy sessions, yet has limitations in observing the larynx and lacks esophageal phase information. VFSS provides comprehensive assessment of mouth, pharynx, esophagus, and swallowing phases, evaluates swallowing phase durations and bolus effects, and produces reviewable images, yet involves ionizing radiation, lacks typical food and liquid use, may not fully represent normal swallowing, focuses on motor function, faces accessibility challenges, and incurs high costs. Together, they offer a comprehensive evaluation, enabling clinicians to identify structural abnormalities, functional deficits, and aspiration events, thus guiding appropriate treatment strategies tailored to individual patient needs.¹¹⁻¹³

This case report discusses a male patient, 51 years old, with a diagnosis of neurogenic oropharyngeal dysphagia with silent aspiration due to myasthenia gravis, supported by clinical and objective swallowing examination findings. The patient had significant discrepancies in the results of the FEES and VFSS examinations (after treatment and rehabilitation program). VFSS examination showed minimal residue in the vallecula and piriform sinus and PAS 1 in all consistencies, while FEES results showed standing secretion in the bilateral piriform sinus, vallecula, post cricoid (MSS 1), moderate residue in the vallecula (The Yale Pharyngeal Residue Severity Rating Scale IV) on gastric rice and oatmeal consistencies, mild residue in bilateral piriform sinuses (The Yale Pharyngeal Residue Severity Rating Scale III) on puree, thin and thick liquid consistencies, and silent aspiration (PAS 8) on thin and thick consistencies liquid, which occurred at the end of the FEES observation, and thought to be an indication of fatigue in the patient. Apart from the findings above, residue was also found in the post-cricoid and the upper esophageal sphincter was observed open at rest, which could increase the risk of post-swallowing aspiration.

According to existing research, there is still no agreement on a gold standard examination for dysphagia. However, both FEES and VFSS offer relevant information in diagnosing oropharyngeal dysphagia. Giraldo-Cadavid et al⁹, stated that FEES had higher sensitivity compared to VFSS in assessing aspiration (0.88 vs. 0.77, $p=0.03$), penetration (0.97 vs. 0.83, $p=0.0002$), and laryngopharyngeal residue (0.97 vs. 0.80, $p=0.0001$). Fattori et al¹⁰, stated that FEES has high sensitivity compared to VFSS in semi-solid and liquid consistencies (85.2%; 80.4%) with overall validity of 83.3% and 80%. Wu et al.¹⁴ highlighted the key differences between FEES and VFSS in assessing swallowing safety. Their study revealed disparities in detecting various parameters such as premature oral leakage (39.3%), pharyngeal stasis (10.7%), laryngeal penetration (14.3%), aspiration (14.3%),

effective cough reflex (39.3%), and velopharyngeal incompetence (67.9%). Notably, FEES demonstrated higher sensitivity in detecting certain aspects like aspiration and cough reflex compared to VFSS. Until now, no studies have been found in the literature comparing FEES and VFSS for diagnosing dysphagia in a specific patient population with myasthenia gravis.

Despite the preference of many experts for VFSS due to its direct visualization of the upper aerodigestive tract during all swallowing phases, it also provides significant and crucial insights into detecting aspiration and assessing the amount of residue in the pharynx compared to FEES. FEES and VFSS have their limitation and advantages. It is best not to compare those two modalities, but to see them as a complementary test to each other, and we can collect all the relevant information to assess dysphagia.^{15,16}

Several things have the potential to cause differences in the findings of the FEES and VFSS examinations in the case above, including (1) Fatigue factors that are typical of myasthenia gravis, (2) Differences in the time of the FEES and VFSS examinations, (3) Time difference between examinations and last consumption of anticholinesterase drugs, considering the pharmacokinetics of pyridostigmine which has an onset of 15-30 minutes, time to peak plasma concentration of 1,5 to 2 hours, and a duration of 6-8 hours.^{3,17,18}

The study's limitation lies in its single-case design, limiting its generalizability. Future research could benefit from larger sample sizes and longitudinal designs to further investigate the effectiveness of rehabilitation programs. Objective measures, like VFSS and FEES, alongside patient-reported outcomes, would provide a more comprehensive understanding of treatment outcomes. Furthermore, it is crucial to investigate the lasting effectiveness of these interventions in enhancing swallowing function and quality of life for individuals with myasthenia gravis. Moreover, there is a lack of literature comparing FEES and VFSS for diagnosing dysphagia in this specific patient group.

CONCLUSION

Both FEES and VFSS play pivotal roles as complementary diagnostic tools in evaluating oropharyngeal dysphagia in MG patients. In the interpretation of objective swallowing function examinations, it is crucial to consider potential confounding factors such as fatigue, the timing of examinations, and the administration of medications, particularly anticholinesterase drugs like Pyridostigmine. This article is expected to provide information on the tests anticipated for evaluating swallowing difficulties in myasthenia gravis patients in Indonesia.

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DECLARATIONS

Conflict of interest. The authors declare that they have no competing interests to disclose.

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