

Case Series – General Neurology

Isolated Dysphagia in a Patient with Medial Medullary Infarction – Effects of Evidence-Based Dysphagia Therapy: A Case Report

Samra Hamzic^{a, c} Patrick Schramm^{b, c} Hassan Khilan^d
Tibo Gerriets^{a, c, d} Martin Juenemann^{a, c}

^aDepartment of Neurology, University Hospital Giessen and Marburg, Campus Giessen, Giessen, Germany; ^bDepartment of Anaesthesiology, University Medical Centre of the Johannes Gutenberg-University, Mainz, Germany; ^cHeart and Brain Research Group, Kerckhoff Heart and Thorax Centre, Bad Nauheim, Germany; ^dStroke Unit, Department of Neurology, Gesundheitszentrum Wetterau, Friedberg, Germany

Keywords

Dysphagia · Flexible endoscopic evaluation of swallowing · Medial medullary infarction · Dysphagia therapy · Evidence-based therapy · Case report

Abstract

Medial medullary infarction (MMI) is a vascular occlusion in the medulla oblongata leading to certain constellations of neurological symptoms and seriously affecting the patient. Effective evidence-based treatment of severe dysphagia as sole symptom of MMI has not yet been reported. This case study aims to report successful effects of evidence-based therapy based on findings of dysphagia symptoms and pathophysiology of swallowing by flexible endoscopic evaluation of swallowing (FEES) in severe isolated dysphagia after MMI. FEES was performed to evaluate swallowing pathophysiology and dysphagia symptoms in a 57-year-old male with severe dysphagia after MMI. On the basis of FEES findings, simple and high-frequent evidence-based exercises for improvement of swallowing were implemented: thermal stimulation of faucial arches, Jaw Opening Exercise, and Jaw Opening Against Resistance. After 7 weeks of high-frequent evidence-based therapy and regular FEES evaluation the patient was set on full oral diet with no evidence of aspiration risk. In a first case report of isolated dysphagia in MMI our

case illustrates that high-frequent evidence-based dysphagia therapy in combination with FEES as the method to evaluate and monitor swallowing pathophysiology can lead to successful and quick rehabilitation of severely affected dysphagic patients.

© 2021 The Author(s)
Published by S. Karger AG, Basel

Introduction

Up to 80% of stroke survivors suffer dysphagia as a consequence of stroke [1]. Characteristic impairments of delayed swallowing reflex, decreased laryngeal elevation, and reduced motility of the upper esophageal sphincter (UES), leading to missequence of swallowing events, massive hypopharyngeal residue, and aspiration, are frequently seen in patients with brain stem stroke [2, 3]. Dysphagia occurs in up to 78% of cases in medial medullary infarction (MMI) [4]. Delayed swallowing reflex is described as typical symptom of dysphagia in MMI [4] whereas decreased laryngeal elevation is mostly attributed to patients with lateral medullary infarction [4]. This case report describes a case of successful treatment of dysphagia as a sole symptom of stroke in medial medulla oblongata with delayed swallow reflex, impairment of laryngeal elevation, and reduced motility of UES as leading pathophysiology of swallowing as defined by flexible endoscopic evaluation of swallowing (FEES).

Case Report

We present a case of a 57-year-old German male scientist who was admitted to a municipal stroke unit (SU) in October 2016 with MMI and severe swallowing impairment as a sole neurological symptom.

The patient was first admitted to the department of Internal Medicine with symptoms of severe dysphagia and inability to elicit the swallowing reflex. Thoracic computed tomography and gastroscopy did not reveal any pathological findings. Due to suspected stroke the patient was then admitted to the SU. Modified Rankin Scale (mRS) and National Institutes of Health Stroke Scale (NIHSS) on admission to the SU were both zero (0). The neurological and general physical examination did not discover any neurological impairments. Further clinical findings included a struma multinodosa, insuline-dependent type II diabetes mellitus, hypothyreosis due to Hashimoto thyroiditis, vitiligo, and temporary Addison's disease. The patient had several allergies: hay fever, penicillin allergy, allergy to insect bites, and allergy to contrast agents for magnetic resonance tomography (MRI) and computed tomography.

Diffusion-weighted MRI and ADC map were able to visualize an elongated acute ischemia in the medial medulla oblongata on the left side. There were no indications of further strokes or a pronounced leukoencephalopathy. The brain-supplying arteries were examined using TOF-MRI and color-coded duplex sonography, showing a fetal supply of the middle cerebral artery on the right side, a high-grade stenosis of the basilar artery in the middle third, and an asymptomatic >50% stenosis [5] of the middle cerebral artery on the right side. Further stroke workup included a Holter-ECG, transesophageal echocardiography (TEE), as well as examinations for thrombophilia and vasculitis. TEE revealed hypermobile interatrial septum and patent foramen ovale, but no other sources of cardiac embolism. Blood tests showed neither a predisposition to thrombophilia nor evidence of vasculitis.

On admission to the SU the patient was allocated to a more specific clinical swallowing examination (CSE) by a speech and language therapist (SLT). CSE from the day of admission on the SU showed an impairment of oropharyngeal sensitivity and a dysfunctional gag reflex. The palatal elevation was triggered with an extreme delay. During the CSE no spontaneous elicitation of swallowing reflex was recorded. There was no dysarthria, no facial palsy, no aphasia, no speech apraxia nor buccofacial apraxia. Clinical water swallow test was not conducted, since the swallow reflex was not elicitable. A second CSE 24 h later did not show any improvement in swallowing, not even after stimulation of palatal structures with ice. In order to evaluate the range of swallowing impairment and the risk of aspiration FEES was conducted 2 days after admission. The examination of hypopharyngeal and laryngeal structures at rest and in motion showed no impairment. No food or fluids were administered since no swallowing reflex was elicited. The main symptoms found in the initial FEES were severe saliva residue in piriform sinus and valleculae and the inability to initiate swallowing reflex (Fig. 1a). There were no hints on saliva aspiration since the patient was regularly expectorating saliva residue.

Initial secondary prevention with acetylsalicylic acid was substituted by oral anticoagulation with Edoxaban 60 mg/day following the findings of TEE. Furthermore, high-dose simvastatin was added. In principle, with a high-grade stenosis of the A. basilaris and under the condition of supply variants of the brain stem, a macroangiopathic etiology of cerebral infarction is possible. Nevertheless, initial secondary prevention with acetylsalicylic acid was substituted by oral anticoagulation with Edoxaban 60 mg/day following the findings of TEE. Furthermore, high-dose simvastatin was added.

Since the admission on the SU the patient was nourished via nasogastric tube. Due to dysphagia as sole symptom of stroke the discharge was planned for day 7 after admission and continuation of dysphagia therapy in an outpatient setting was recommended. Nourishment was to be proceeded via nasogastric tube. Since the patient planned on resuming his professional activity immediately after discharge PEG was placed 5 days after admission upon patient's request.

The underlying symptoms of dysphagia found in FEES (non-elicitable swallowing reflex, severe residue of saliva in valleculae and piriform sinus, Fig. 1a) gave hint of swallowing pathophysiology. The swallowing reflex in normal subjects is elicited at the end of the volitional phase of swallowing when bolus contacts the anterior faucal arches (AFA) allowing the glossopharyngeal nerve to elicit swallowing reflex by transmitting afferent signals to the reticular formation (RF) and nucleus tractus solitarii (NTS) in the dorsolateral medulla oblongata. The inability to trigger the swallowing reflex in MMI may be caused by the stroke-related damage caused to RF and NTS (Fig. 2) [2, 6]. Studies conducted on different patient groups with neurogenic dysphagia showed that thermal stimulation of AFA with ice can lead to quicker triggering of swallowing reflex [6]. In our case study stimulation of oropharyngeal structures with ice was conducted 3×/day by rubbing the AFA five times on each side with an ice stick [6]. Swallowing reflex was elicited for the first time after 3 days of ice stimulation.

The symptoms of residue in piriform sinus and valleculae may point out to impairments in hyoid bone elevation, laryngeal elevation, and tongue base retraction as well as impaired opening of the UES due to impaired hyoid bone elevation and laryngeal elevation [2]. We introduced exercises which are proven to amend these mechanisms: Chin tuck against resistance (CTAR) [7] and jaw opening exercise (JOE) [8]. These exercises have a positive influence on strengthening suprahyoidal muscles and can improve hyoid bone and laryngeal elevation and opening width of UES, thus reducing residue in piriform sinus and valleculae [7, 8].

We conducted the following high-frequent treatment (Fig. 3): (1) 3×/day ice stimulation of anterior faucial arches five times on each side with an ice stick; (2) each five sets of five repetitions of CTAR and JOE for 10 s with 10 s break between each repetition. At the time of discharge from hospital (7 days after admission on the SU), we recommended to continue with the abovementioned treatment frequency at home plus an extra SLT therapy treatment five times a week for 7 weeks. Only oral intake of small liquid boli was recommended at the time of discharge from hospital.

Outcome measure for swallow security as diagnosed via FEES were Penetration-Aspiration-Scale (PAS) [9], Yale Pharyngeal Residue Severity Rating Scale for valleculae (YSv) and for piriform sinus (YSps) [10], and the Functional Oral Intake Scale [11, 12].

The patient conducted the recommended swallow exercises at home and five times a week in the outpatient clinic of the SU with the same SLT who conducted all FEES examinations. To control the effects of the therapy the SLT conducted five FEES examinations within the 7 weeks of therapy, two FEES during the hospital stay, and three follow-up FEES after discharge (Fig. 1, 3, 4). At discharge the ice stimulation was not needed anymore, since the swallowing reflex was elicited regularly as confirmed via FEES. Nevertheless, pharyngeal residue scores for saliva were continuously severe (YSv 5/YSps 5) at discharge. However, the patient was able to swallow small liquid boli without aspiration (PAS 2) despite high residue score (YSps 4). Only oral intake of small liquid boli was allowed at discharge, since overall residue and penetration-aspiration scores for puree were severe (YSv 5/YSps 5/PAS 5). Six weeks after discharge all dysphagia scores showed no relevant aspiration risk (Fig. 4). The rapid therapy success allowed for full oral diet and a quick PEG removal a few weeks later.

Discussion

According to previous studies, 78% of patients suffer dysphagia after MMI [4]. In our case study the patient showed symptoms both of an impairment in the timing of swallowing reflex as well as in the range of laryngeal elevation, thus contributing to an insufficient opening of the UES. Whereas direct involvement of the nucleus ambiguus (NA) may be the cause of impairment in the range of laryngeal elevation in lateral medullary infarction, it is suggested that in MMI the impairment in the timing of swallowing reflex may be due to the destruction of the corticobulbar fibers which are innervating the NA. However, damage to other swallow-relevant brainstem structures may also explain the impairment in the timing of swallowing reflex: central pattern generators (CPGs) are situated in RF which encompasses the NA. Impairment in the timing of swallowing reflex could be explained by potential damage to CPGs in MMI [4].

New methods of dysphagia therapy (transcranial direct current stimulation, repetitive transcranial magnetic stimulation, pharyngeal electrical stimulation) are all very effective in the treatment of neurogenic dysphagia on the one side, but not always applicable to every patient. This case study shows that standard, but high-intensity evidence-based dysphagia treatment based on FEES-proven pathophysiology can improve the swallowing of patients with severe dysphagia after MMI within a rather short period of time. In recent years FEES diagnostics has gained more importance in the diagnostics of dysphagic stroke patients. Various studies have confirmed that near-time implementation of FEES after stroke as primary instrument for diagnostics of dysphagia can predict severe dysphagia 3 months post-onset and contributes to better outcome, lower rates of mechanical ventilation and pneumonia, as well as shorter length of hospital stay [13–16]. Works of Braun et al. on the usefulness of FEES

in neurologic, stroke, and intensive care patients have shown that FEES diagnostics is crucial and has an important influence on overall outcome solely on the basis of dietary adjustment after FEES. In an investigation of 241 patients with various neurological diseases only 33.1% had an adequate oral diet prior to FEES [14]. In this study significant lower rates of mortality and aspiration pneumonia were recorded after the change of oral diet based on FEES findings. In only 31% of investigated dysphagic cases on a stroke unit (SU) did the clinical assessment identify a proper oral diet [15]. In this study the change of oral diet based on FEES findings correlated with a better overall outcome at discharge from SU, shorter length of stay, lower rate of aspiration pneumonia, and lower need for mechanical ventilation. Similar results are found in the work of Braun et al. on the usefulness of FEES in the neurological intensive care unit (ICU) [16]. FEES detected dysphagia in 72% of investigated cases and allowed for adjustment of oral diet in 64% of investigated cases after FEES contributing to lower mortality and morbidity. Therefore, our case study emphasizes all the more the relevance of frequent use of FEES during dysphagia treatment in acute and post-acute phase of stroke to determine not only an adequate oral diet but also effective therapy methods and prove its efficacy based on genuine description of pathophysiology as seen in FEES.

Conclusion

This case report describes a case of successful treatment of dysphagia as a sole symptom of stroke in medial medulla with impairments in the timing of swallowing reflex and the range of laryngeal elevation. Simple high-frequent and evidence-based methods of dysphagia therapy combined with FEES as diagnostic and biofeedback method of evaluation of therapy efficiency were applied ending in total oral diet and removal of PEG tube after a short period of time.

Acknowledgement

We would like to thank our patient who gave his written informed consent for this case report.

Statement of Ethics

This study was approved by the ethics committee of the University of Giessen (Az. 208/16). This case study has been carried out in accordance with The World Medical Association's Declaration of Helsinki for experiments involving humans. Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Conflict of Interest Statement

S. Hamzic, P. Schramm, H. Khilan, T. Gerriets declared that they have no conflict of interest. M. Juenemann has received grants from German Heart Research Foundation (Deutsche

Stiftung für Herzforschung) and from Kerckhoff-Foundation (Kerckhoff-Stiftung) with no relevance to the paper.

Funding Sources

No funding has been received for this case report.

Author Contributions

S.H. designed the case study, performed all examinations and therapies. T.G. supervised the project. S.H. wrote the manuscript. S.H. and H.K. provided data for all figures. P.S. and M.J. contributed to the final version of the manuscript. All authors discussed the results and reviewed the final manuscript.

Case Report Guidelines (CARE) Compliance

This case report was written according to the Case Report Guidelines (CARE): <https://www.care-statement.org>.

References

- 1 Barer DH. The natural history and functional consequences of dysphagia after hemispheric stroke. *J Neurol Neurosurg Psychiatry*. 1989 Feb;52(2):236–41.
- 2 Prosiel M, Höling R, Heintze M, Wagner-Sonntag E, Wiseman K. The localization of central pattern generators for swallowing in humans – a clinical-anatomical study on patients with unilateral paresis of the vagal nerve, Avellis' syndrome, Wallenberg's syndrome, posterior fossa tumours and cerebellar hemorrhage. *Acta Neurochir Suppl (Wien)*. 2005;93:85–8.
- 3 Nakao M, Oshima F, Maeno Y, Izumi S. Disruption of the Obligatory Swallowing Sequence in Patients with Wallenberg Syndrome. *Dysphagia*. 2019 Oct;34(5):673–80.
- 4 Kwon M, Lee JH, Kim JS. Dysphagia in unilateral medullary infarction: lateral vs medial lesions. *Neurology*. 2005 Sep;65(5):714–8.
- 5 Baumgartner RW, Mattle HP, Schroth G. Assessment of [GT]/=50% and [LT]50% intracranial stenoses by transcranial color-coded duplex sonography. *Stroke*. 1999 Jan;30(1):87–92.
- 6 Regan J, Walshe M, Tobin WO. Immediate effects of thermal-tactile stimulation on timing of swallow in idiopathic Parkinson's disease. *Dysphagia*. 2010 Sep;25(3):207–15.
- 7 Yoon WL, Khoo JK, Rickard Liow SJ. Chin tuck against resistance (CTAR): new method for enhancing suprahyoid muscle activity using a Shaker-type exercise. *Dysphagia*. 2014 Apr;29(2):243–8.
- 8 Wada S, Tohara H, Iida T, Inoue M, Sato M, Ueda K. Jaw-opening exercise for insufficient opening of upper esophageal sphincter. *Arch Phys Med Rehabil*. 2012 Nov;93(11):1995–9.
- 9 Rosenbek JC, Robbins JA, Roecker EB, Coyle JL, Wood JL. A penetration-aspiration scale. *Dysphagia*. 1996;11(2):93–8.
- 10 Neubauer PD, Rademaker AW, Leder SB. The Yale Pharyngeal Residue Severity Rating Scale: An Anatomically Defined and Image-Based Tool. *Dysphagia*. 2015 Oct;30(5):521–8.
- 11 Crary MA, Mann GD, Groher ME. Initial psychometric assessment of a functional oral intake scale for dysphagia in stroke patients. *Arch Phys Med Rehabil*. 2005 Aug;86(8):1516–20.
- 12 Hamzic S, Braun T, Juenemann M, Butz M, Voswinckel R, Belly M, et al. Validation of the German Version of Functional Oral Intake Scale (FOIS-G) for Flexible Endoscopic Evaluation of Swallowing (FEES). *Dysphagia*. 2021 Feb;36(1):130–9.

Hamzic et al.: Isolated Dysphagia in a Patient with Medial Medullary Infarction – Effects of Evidence-Based Dysphagia Therapy: A Case Report

- 13 Ickenstein GW, Höhlig C, Prosiegel M, Koch H, Dziewas R, Bodechtel U, et al. Prediction of outcome in neurogenic oropharyngeal dysphagia within 72 hours of acute stroke. *J Stroke Cerebrovasc Dis.* 2012 Oct;21(7):569–76.
- 14 Braun T, Juenemann M, Viard M, Meyer M, Fuest S, Reuter I, et al. What is the value of fibre-endoscopic evaluation of swallowing (FEES) in neurological patients? A cross-sectional hospital-based registry study. *BMJ Open.* 2018 Mar;8(3):e019016.
- 15 Braun T, Juenemann M, Viard M, Meyer M, Reuter I, Prosiegel M, et al. Adjustment of oral diet based on flexible endoscopic evaluation of swallowing (FEES) in acute stroke patients: a cross-sectional hospital-based registry study. *BMC Neurol.* 2019 Nov;19(1):282.
- 16 Braun T, Juenemann M, Viard M, Meyer M, Reuter I, Mausbach S, et al. Flexible endoscopic evaluation of swallowing (FEES) to determine neurological intensive care patients' oral diet. *Int J Speech Lang Pathol.* 2020 Apr 3:1–9.

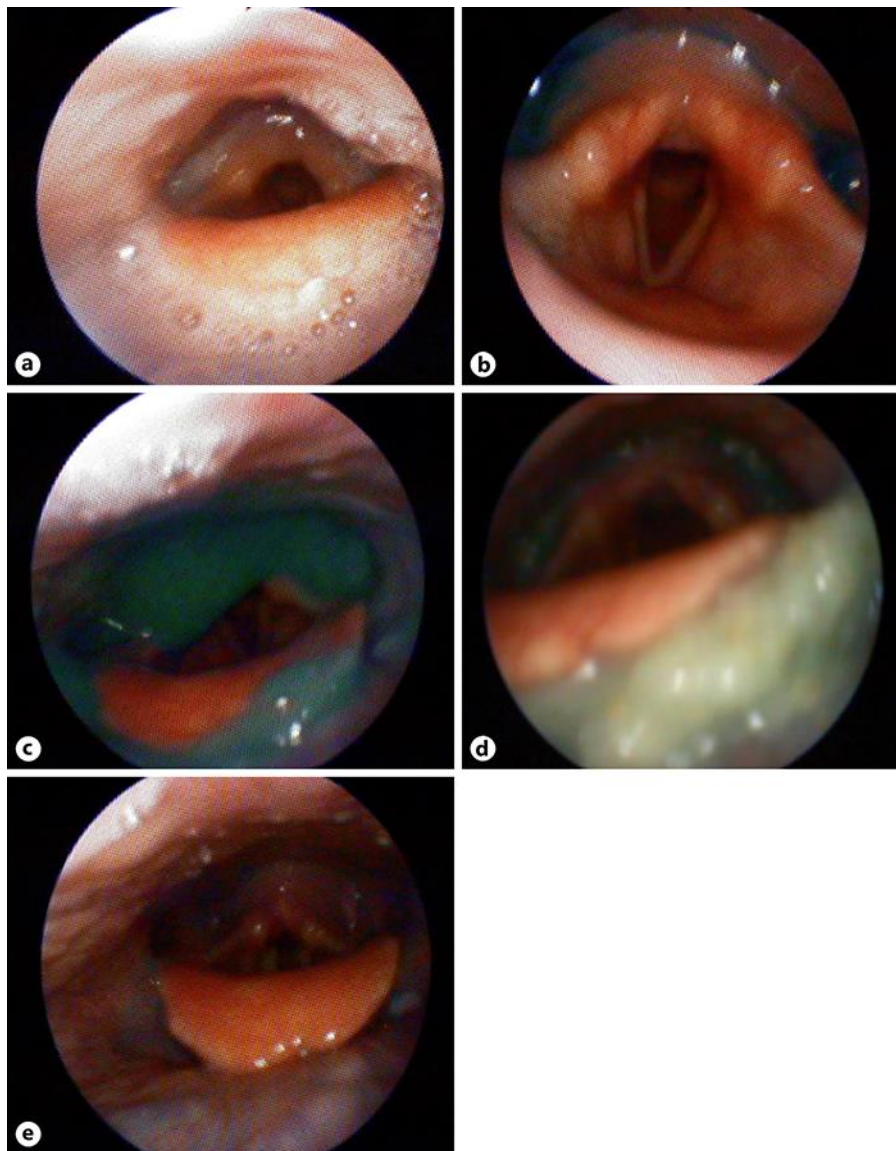


Fig. 1. Symptoms of dysphagia in the course of treatment as seen in flexible endoscopic evaluation of swallowing (FEES). Figure demonstrates the photo documentation of FEES examinations in the course of the treatment with Penetration-Aspiration-Scale (PAS) and Yale Pharyngeal Residue Severity Rating Scale for valleculae (YSv) and piriform sinus (YSps) scores: **a** shows the massive saliva residue in valleculae and piriform sinus in the initial FEES (YSv 5/YSps 5); in **b** penetration of liquid on the right side (PAS 3) and mild residue of liquid bolus (YSps 3) is perceived in piriform sinus 3 days after admission; **c** depicts penetration (PAS 3) and massive residue of puree in valleculae and piriform sinus (YSv 5/YSps 5) 2 weeks after stroke; **d** (24 days post onset) shows massive residue of solid bolus in valleculae and only moderate residue in piriform sinus (YSv 5/YSps 4); in **e** no pathological results were documented.

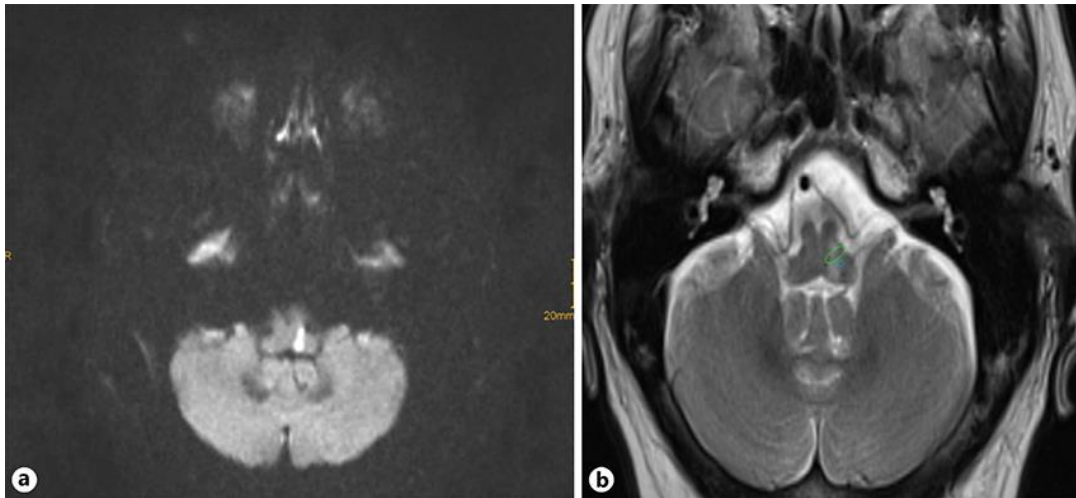


Fig. 2. MRI scan. **a** Diffusion-weighted imaging. **b** T2-weighted MRI. Green circle represents reticular formation. Blue circle represents nucleus ambiguus.

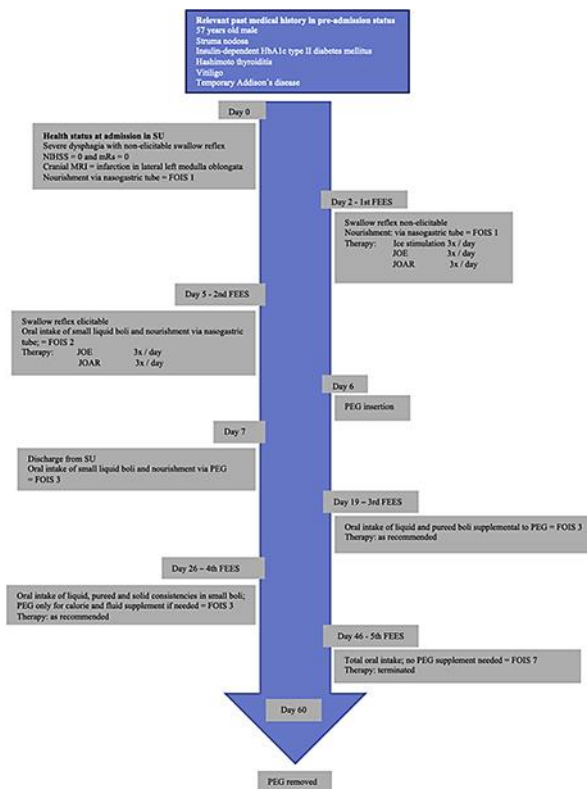


Fig. 3. Timeline. Figure shows the milestones of symptoms, diagnostics and therapy. FEES, flexible endoscopic evaluation of swallowing; FOIS, Functional Oral Intake Scale; JOAR, Jaw Opening Against Resistance; JOE, Jaw Opening Exercise; MRI, magnetic resonance imaging; mRS, Modified Rankin Scale; NIHSS, National Institutes of Health Stroke Scale; PEG, percutaneous endoscopic gastrostomy; SU, stroke unit.

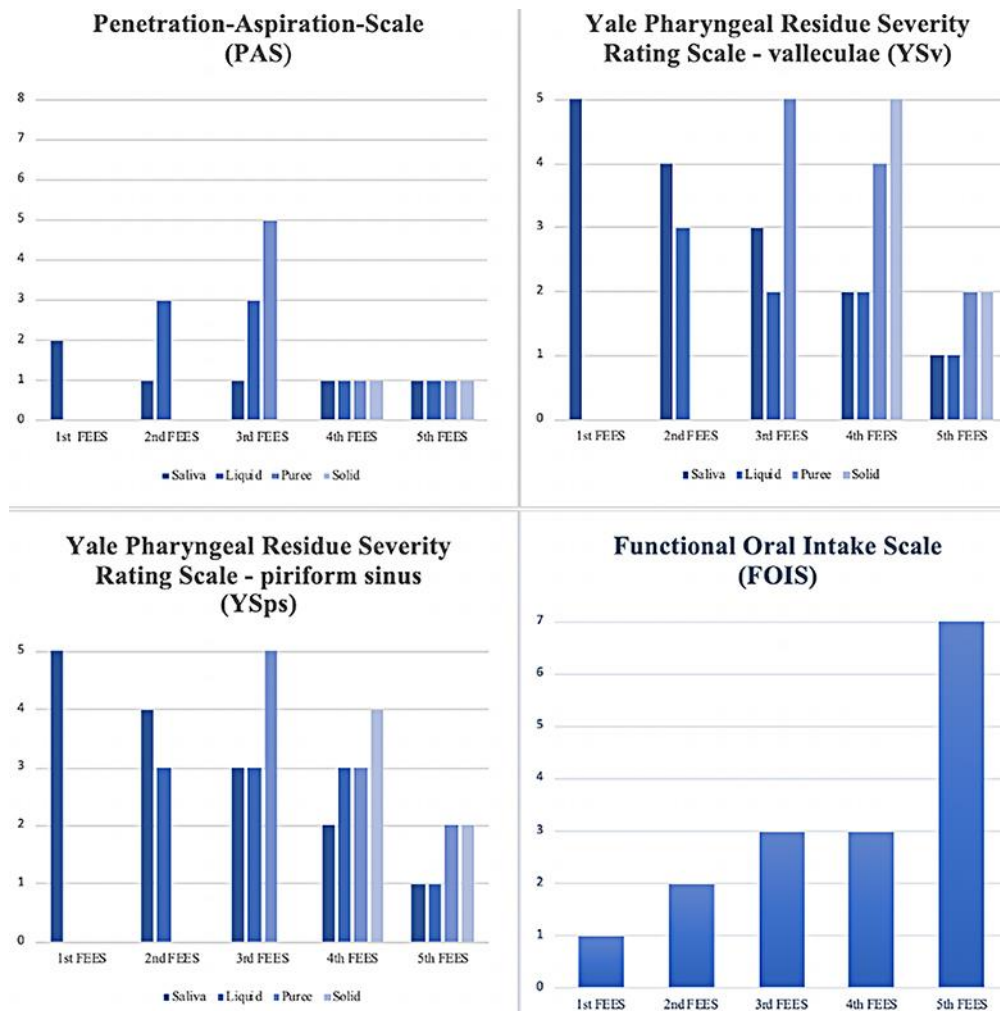


Fig. 4. Dysphagia severity scores as measured by flexible endoscopic evaluation of swallowing (FEES) in the course of 7 weeks of dysphagia therapy with Jaw Opening Exercise (JOE) and Jaw Opening Against Resistance (JOAR). The figure shows the measured dysphagia scores in the consecutive FEES in the course of treatment. Increasing Penetration-Aspiration-Scale (PAS; left upper graph) and Yale Pharyngeal Residue Severity Rating Scale for valleculae (YSv; right upper graph) and piriform sinus (YSps; left lower graph) scores indicate increased dysphagia severity. Increasing Functional Oral Intake Scale (FOIS; right lower graph) scores indicate an improvement in total oral intake.