

Pre-stroke undiagnosed dysphagia lusoria as a rare cause of aspiration pneumonia with respiratory failure in a stroke patient

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Abstract

Introduction: Dysphagia is a risk factor for aspiration pneumonia and acute respiratory failure in acute stroke patients. Dysphagia lusoria is caused by compression on the esophagus from artery lusoria, when the aberrant right subclavian artery arises from the descending aortic arch. We present a rare case report of pre-stroke undiagnosed dysphagia lusoria as a cause of aspiration pneumonia with acute respiratory failure in a 67-year-old female patient admitted with a minor left intracerebral hemorrhage in the left basal ganglia. On admission to the stroke unit, she had Glasgow Coma Scale of 15, National Institutes of Health Stroke Scale of 8, and a negative screening test for dysphagia, dysphasia, and right-sided hemiparesis. After 16 h of admission, dyspnea suddenly occurred with a decrease in SpO₂ (72%). X-ray of the lungs showed less ventilated areas of the lung due to aspiration pneumonia and a broad disfigured shadow of the anterior mediastinum on the base of the lusoria artery. Dysphagia lusoria was confirmed by spiral computed tomography angiography.

Conclusion: One aim of neurocritical care is the prevention of pneumonia from dysphagia due to risk of acute respiratory failure and secondary brain damage. Pre-stroke undiagnosed dysphagia lusoria could be one very rare cause. A broad disfigured shadow of the anterior mediastinum in X-ray of the lungs gives rise to the first suspicion of the possibility of dysphagia lusoria.

Keywords

Neurocritical care, dysphagia, stroke, dysphagia lusoria

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Introduction

Post-stroke dysphagia is a common complication in neurocritical care.¹ It is associated with increased mortality and morbidity due to the risk of aspiration, pneumonia, and respiratory failure. Diagnosing dysphagia is therefore a priority in order to prevent these negative consequences causing secondary brain damage.

Dysphagia is not only caused by neurological illnesses, but it also occurs due to disorders of the esophagus or conditions affecting structures adjacent to the esophagus, such as tumors or infections in the area of the mouth, pharynx, or esophagus.

One very rare cause of dysphagia is artery lusoria, a congenital anomaly occurring when the aberrant right subclavian artery arises from the descending aortic arch, compressing the esophagus.²

Artery lusoria is usually asymptomatic, but can cause dyspnea and asthma by compressing the trachea or Kommerell's aortic diverticulum,^{3,4} which is a diverticular outpouching at the origin of artery lusoria.

The aim of our presentation is to draw attention to the existence of dysphagia lusoria as a very rare cause of dysphagia. Even in this case, the acute stroke patient with an intermittent dysphagia had never been examined or diagnosed by any physician before her admission to neurocritical care.

Case report

This case is reported with the consent of the family. We present a rare case report of pre-stroke undiagnosed dysphagia

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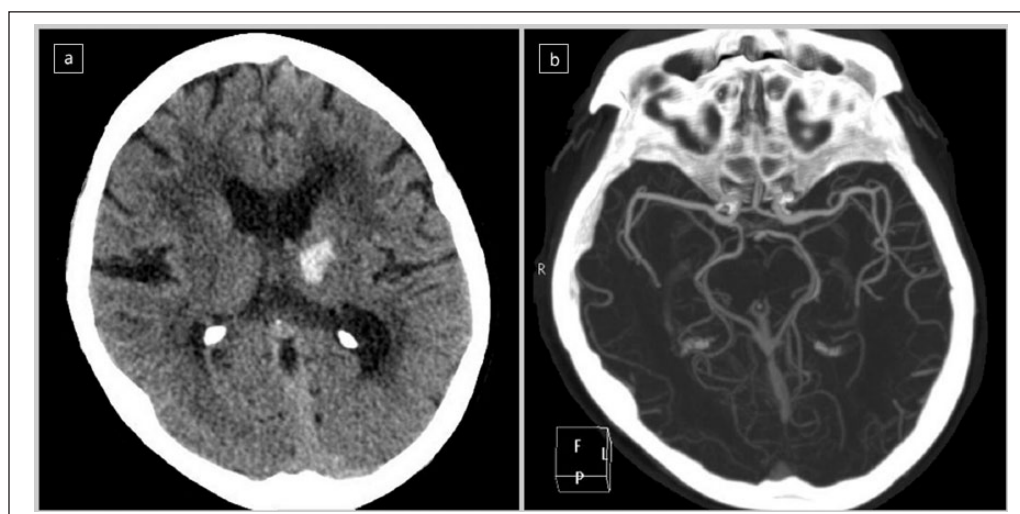


Figure 1. (a) Brain computed tomography and (b) brain computed tomography angiography of intracerebral hemorrhage.

Table 1. Admission screening protocol consisting of eight yes/no questions for dysphagia of the presented patient.

Question	Answer Yes/No	Answer Normal/Abnormal
Is dysarthria present?	Yes	Abnormal
Does patient cough while swallowing thickened fluids?	No	Normal
Is aphasia present?	No	Normal
Is facial nerve palsy present?	No	Normal
Can both shoulders be raised equally?	Yes	Normal
Can the tongue be protruded forwards and to the sides?	Yes	Normal
Can both masticatory muscles be clenched equally?	Yes	Normal
Can the patient cough on demand?	Yes	Normal
Conclusions	No dysphagia	

Two abnormal responses: suspected dysphagia, three or more abnormal responses: dysphagia.

lusoria as a cause of aspiration pneumonia with acute respiratory failure (ARF) in a stroke patient. A 67-year-old female patient with a history of hypertension was admitted to the neurointensive care unit (NICU) with intracerebral hemorrhage (ICH; Figure 1), causing a dysphasia and right-sided hemiparesis. On admission, she had Glasgow Coma Scale of 15, NIHSS (National Institutes of Health Stroke Scale) of 8, and a negative physician-evaluated local NICU screening protocol for dysphagia based on eight yes/no questions (Table 1). Brain computed tomography angiography (CTA) did not show vessel pathology (Figure 1).

After 16 h of admission, dyspnea suddenly occurred with a decrease in SpO₂ (72%). X-ray of the lungs showed less ventilated areas of the lung due to aspiration pneumonia. During this acute complication, the neurological status of the patient had not changed since admission. However, we performed acute brain computed tomography (CT) by portable scanner in the NICU, which showed the same size of ICH. The patient was intubated and kept on mechanical ventilation. X-ray of the lungs (Figure 2) showed a broad disfigured

shadow of the anterior mediastinum on the base of the lusoria artery and signs of aspiration pneumonia. We immediately initiated the intravenous administration of antibiotics (*amoxicillin and clavulanate*). After the acute respiratory complication, the patient was subfebrile, C-reactive protein (CRP) rose from 15.3 g/L on admission to 49 g/L, and leucocytes were $9.9 \times 10^9/L$.

Retro-esophageal dysphagia lusoria was confirmed by spiral CTA, where the aberrant artery lusoria was compressing the esophagus (Figure 2). Following the diagnosis of dysphagia lusoria, we consulted the family, who confirmed intermittent dysphagia in the targeted questionnaire, but it had never been examined or diagnosed by any physician.

For lasting dysphagia, a tracheostomy was performed and the patient was successfully weaned on day 12, and then transferred to the standard neurology department. During the stay in our NICU, we performed an otorhinolaryngological examination with pharyngoscopy and laryngoscopy, which recommended consulting a vascular surgeon at the university hospital. In the standard neurology department, the tracheostomy tube could not

Table 2. Key factors of dysphagia lusoria.

Dysphagia lusoria is a very rare cause of dysphagia.

This dysphagia is caused by artery lusoria compressing the esophagus.

The first suspicion in the diagnosis is a broad disfigured shadow of the anterior mediastinum in X-ray of the lungs.

Dysphagia lusoria is confirmed by vascular imaging.

Contrast enhanced CT or MR angiography can be used to confirm this diagnosis, as well as illustrating the relationship between arteria lusoria and surrounding structures, in particular the esophagus and trachea.

CT: computed tomography; MR: magnetic resonance.

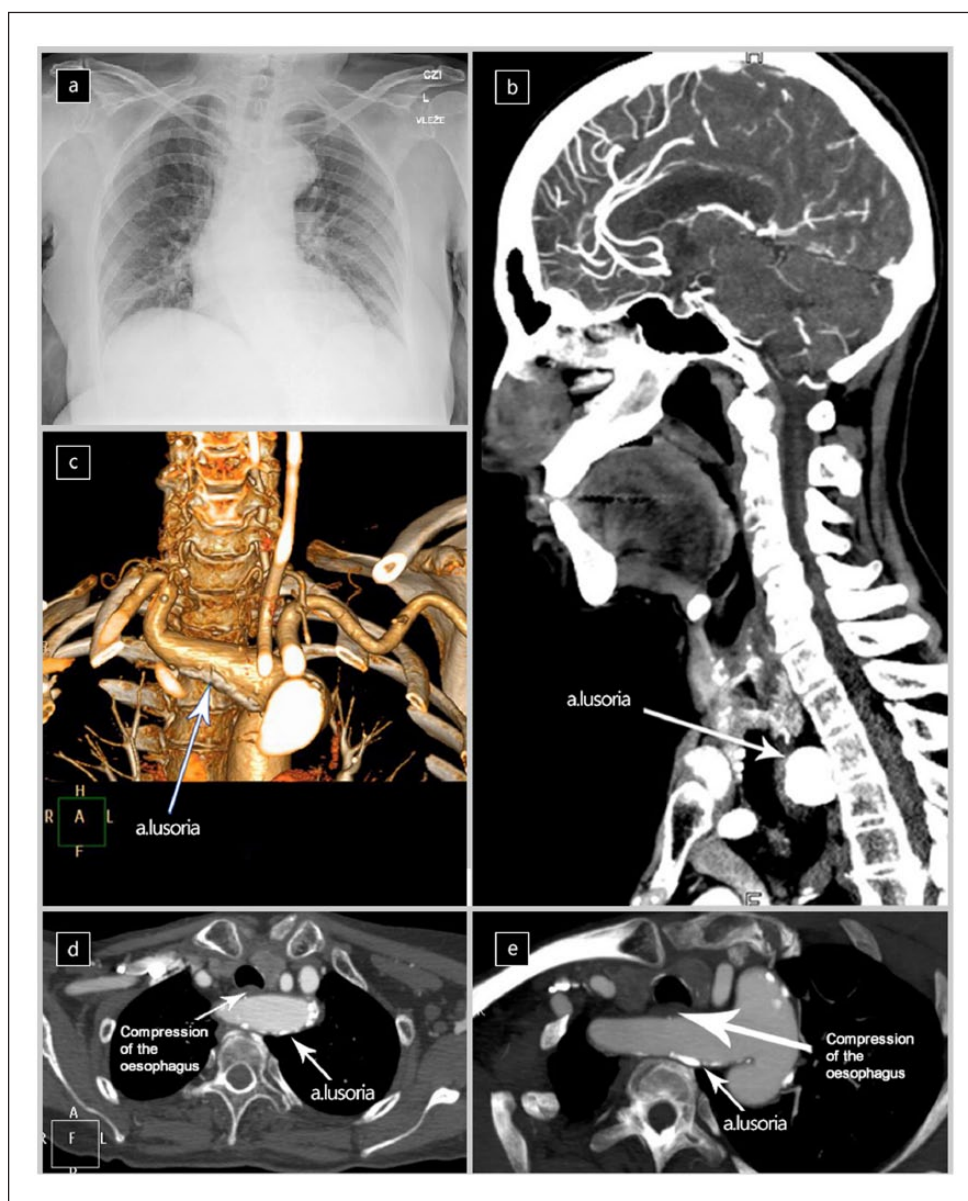


Figure 2. (a) X-ray of the lungs and (b–e) spiral computed tomography angiography of artery lusoria. The compression of esophagus artery lusoria is indicated by arrow.

be removed due to continuing dysphagia, and so a percutaneous endoscopic gastrostomy was performed. The patient suffered depression syndrome, which did not improve despite

anti-depressive therapy. This affected her overall state, although her dysphasia and hemiparesis improved. She was later transferred to aftercare, rehabilitation, and logopedic therapy. No

vascular surgeon was consulted either at the standard neurology department or at the aftercare department due to the patient's overall status.

Discussion

Dysphagia is a common and serious complication in post-stroke critical care because of the risk of aspiration, which can lead to pneumonia and respiratory failure and can cause secondary brain damage.¹ The aim of neurocritical care is to minimize all secondary brain damage in order to reduce morbidity and mortality in post-stroke neurocritically ill patients. For this reason, attention must be paid to every possibility which can increase these risks. Dysphagia lusoria² is a very rare cause of dysphagia, most seriously in cases when it is undiagnosed pre-stroke, as we show in our case report. Of the symptoms associated with artery lusoria, our patient had only intermittent dysphagia from a compression of the esophagus. This syndrome was first described by David Bayford⁵ in London in 1794. A congenital anomaly of the aberrant right subclavian artery arising from descending the aortic arch may have caused this compression impinging on the esophagus, possibly affected by pressure in the artery or the content of the esophagus. One potential risk of embolism from arteria lusoria could lie in a very rare dissecting aberrant right subclavian artery associated with Stanford type B acute aortic dissection.³

In our case, however, dysphagia lusoria was not diagnosed either in the patient's previous medical history or during the admission screening protocol, which was established as our local physician-evaluated screening protocol 3 years prior to this case (Table 1). This protocol consists of eight yes/no questions and was drawn up in cooperation with a logopedic therapist. Of the eight questions, the presence of dysarthria was the only abnormality, which led to the conclusion that the patient did not have dysphagia.

Possible explanations for her negative admission protocol are that she had only had an intermittent form in her history, that she had not eaten beforehand, or that at the time of the test she did not have the arterial hypertension which was also present in her history. We do not suppose that the test was carried out inadequately because it was performed by an experienced stroke physician.

The suspicion of dysphagia lusoria occurred to us due to a broad disfigured shadow of the anterior mediastinum in the X-ray of the lungs (Figure 2). This was confirmed by CT angiography (Figure 2).

In acute stroke critical care, a combination of multiple factors can cause aspiration, as we see in our case report, where pre-disposition dysphagia lusoria and neurological changes from the acute stroke caused serious aspiration pneumonia with ARF, which required intubation and the implementation of artificial ventilation.

In the end, further brain damage was avoided and the patient was successfully weaned, although with a tracheostomy for remaining dysphagia lusoria to prevent further aspiration.

In our presented case report, we want to demonstrate the difficulty of differential diagnosis of dysphagia in a post-stroke critically ill patient and bring to attention the existence of dysphagia lusoria (Table 2), even in a case where it had never been examined or diagnosed by any physician until her admission to neurocritical care.

Conclusion

One aim of neurocritical care is the prevention of pneumonia from dysphagia due to the risk of ARF and secondary brain damage. Pre-stroke undiagnosed dysphagia lusoria could be one very rare cause. Dysphagia lusoria is caused by a compression of the esophagus by artery lusoria, when the aberrant right subclavian artery arises from the aortic arch. A broad disfigured shadow of the anterior mediastinum in X-ray of the lungs could give rise to the first suspicion of the idea of dysphagia lusoria.

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Ethical approval

Our institution does not require ethical approval for reporting individual cases or case series.

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Informed consent

Written informed consent was obtained from a legally authorized representative(s) for anonymized patient information to be published in this article.

References

1. Cohen DL, Roffe C, Beavan J, et al. Post-stroke dysphagia: a review and design considerations for future trials. *Int J Stroke* 2016; 11: 399–411.
2. Kommerell B. Verlagerung des Oesophagus durch eine abnorm verlaufende Arteria subclavia dextra (Arteria lusoria). *Fortschr Geb Roentgenstrahlen* 1936; 54: 590–595.
3. Singh S, Grewal PD, Symons J, et al. Adult-onset dysphagia lusoria secondary to a dissecting aberrant right subclavian artery associated with type B acute aortic dissection. *Can J Cardiol* 2008; 24: 63–65.
4. Morel V, Corbinau H, Lecoz A, et al. Two cases of “asthma” revealing a diverticulum of Kommerell. *Respiration* 2002; 69: 456–460.
5. Bayford D. An account of a singular case of obstructed deglutition. *Memoirs Med Soc London* 1794; 2: 275–286.