eNeurologicalSci 23 (2021) 100337

Contents lists available at ScienceDirect

eNeurologicalSci

journal homepage: www.elsevier.com/locate/ensci

Case report

Ti.: "High" vagus nerve lesions in varicella Zoster infection☆

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ARTICLE INFO

Keywords:
Cranial nerve
Vagus nerve
"High" vagus nerve lesion
Headache
Swallowing difficulties
Herpes zoster

ABSTRACT

"High" vagus nerve lesions are rare and refer to the region of the nerve from the jugular foramen through the branching of the auricular (Arnold's branch) and the pharyngeal branch. Rapid onset of vagus nerve palsy is observed predominantly in trauma, and rarely in inflammation. An insidious onset points to a neoplastic cause.

The acute "high" vagus nerve lesion is characterized by a unilateral paralysis of the recurrent laryngeal nerve, an incomplete paresis of the soft palate and a transient inability to swallow.

This is a case description of a 79-year-old woman who presented with painful swelling of the left ear and occipital headache, followed by inability to swallow for 3 weeks. A markedly elevated Varicella Zoster titer suggested a herpes virus infection.

1. Introduction

Sudden onset and complete loss of swallowing ability are rare events and require acute medical intervention.

Acute swallowing disorders can be caused by central causes (e.g., stroke, brainstem disease), or neuromuscular diseases such as myasthenia gravis, botulism, and a rare brachiofacial variant of polyradiculitis [1]. More frequent are chronic progressive swallowing disorders, such as in motor neuron disease, pseudobulbar syndromes, and myopathies [2].

Isolated recurrent laryngeal nerve lesions produce hoarseness and some degree of swallowing impairment, but usually no complete loss of swallowing ability.

This case report illustrates the rare event of a "high" vagus nerve lesion, resulting in a transient complete swallowing loss needing parenteral feeding, and followed by complete reversibility. The anatomical distinction between a "high" and low vagus nerve lesion is crucial and refers to peripheral vagus nerve lesions only [3,4].

2. Case report

A 79-year-old previously healthy woman presented with a reddish, erysipelas like swelling of the left ear and cheek. Local pain radiated into the occipital part of the skull. Within 24 h she was unable to swallow. She was admitted to the hospital and a recurrent laryngeal nerve palsy was noted at the first otolaryngologic examination.

On examination, she had a swollen and reddish-colored edematous left ear and also swelling of the left side of the face. (Fig. 1) She spoke with a hoarse voice. She was unable to swallow and parenteral feeding had to be initiated.

The soft palate was lower on the left side, and elevated only minimally upon phonation. The mucous membranes of the oral cavity showed no inflammation or vesicles. There was no numbness in the oral cavity and taste sensation was normal also in the posterior part of the tongue. The rest of the neurological examination was normal, and there were no reflex abnormalities, and no long tract signs or gait abnormalities that pointed to central nervous system involvement.

Neurological investigation excluded a brainstem lesion and no associated cranial nerve lesions were detected. The clinical syndrome correlated with a "high" vagus nerve lesion. The pain distribution was

☆ The Patient consented to the publication. Records are available.

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https://doi.org/10.1016/j.ensci.2021.100337
Received 26 November 2020; Received in revised form 26 March 2021; Accepted 1 April 2021
Available online 3 April 2021

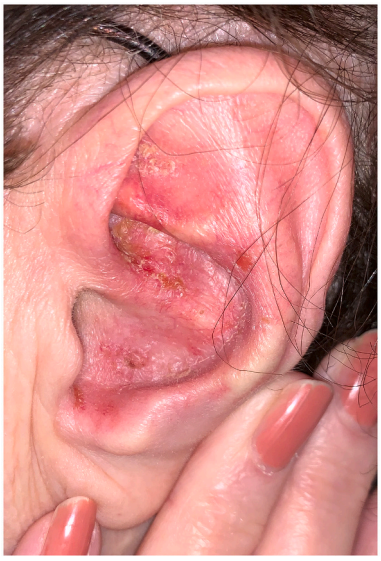

Fig. 1. Acute phase after admission. Painful swelling of the left ear.

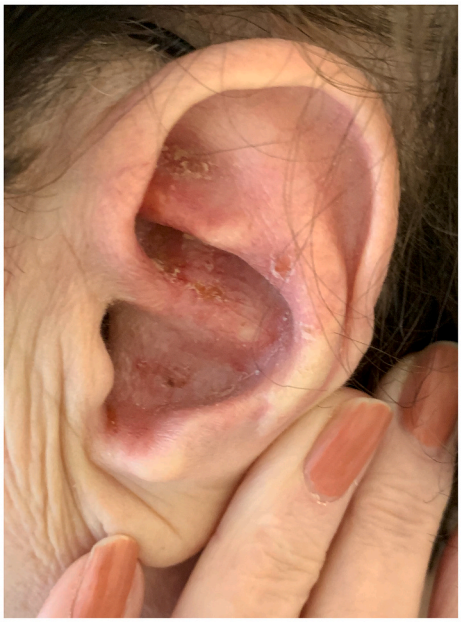

Fig. 2. Ear after 3 weeks. Complete remission of swelling, crusts in the auricle.

attributed to involvement of the meningeal rami and the auricular nerve.

The otolaryngologic examination confirmed a left vocal cord palsy, which was also evident on MRI (Fig. 4). Swallowing was impossible and there was no evidence of an auditory deficit at the time. An endoscopic examination was not performed.

The dermatological exam suspected erysipelas and grouped, small, and eroded papules on an erythematous base on the left ear (Fig. 1) indicating a Herpes infection. Due to the acute and painful development, antimicrobial therapy with Meropenem and Zovirax was initiated and after 5 days steroids were also added.

Under speech therapy treatment, mild improvement of swallowing occurred after 10 days and the patient subsequently completely recovered. After the regression of the ear swelling, crusts were detected in the auricle. (Fig. 2).

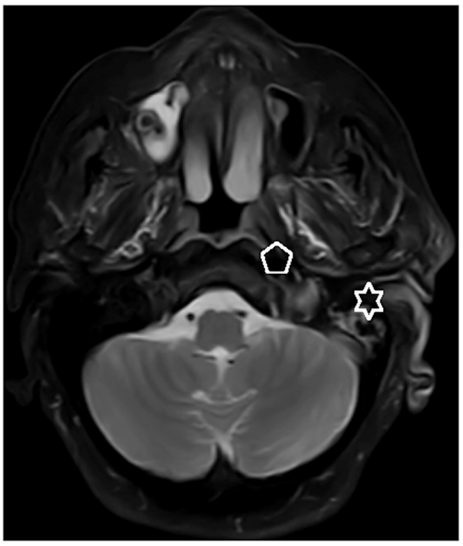

Fig. 3. Axial MRI showing swelling and edema of the left ear (star). Soft tissue edema at the base of the skull is also shown (pentagon).

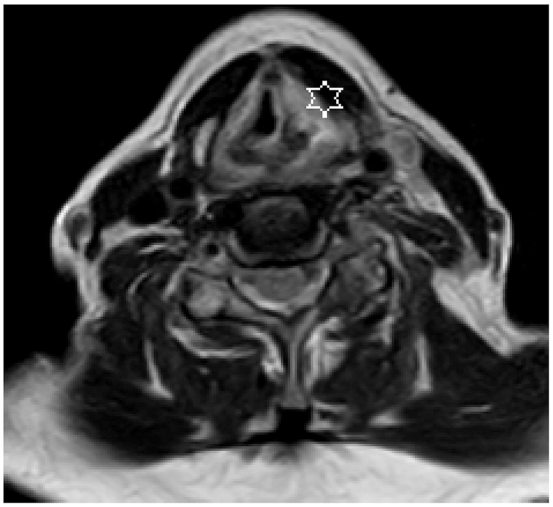

Fig. 4. Axial MRI showing unilateral vocal cord paralysis. Star marks unilateral cord paralysis.

3. Findings

3.1. MRI of the skull and neck

The brain and bony skull were normal, the swelling of the left ear and adjacent tissue were visible, and no abscess formation or tumor was found (Fig. 3). The left pharyngeal fold was paralyzed (Fig. 4).

3.2. Laboratory results

C-reactive protein (CRP) was not elevated. A mild lymphocytopenia (1100/µl) was noted.

Virology was negative for the following: Herpes simplex, tick borne encephalitis (FSME), Coxsackie, Cytomegalovirus, Epstein-Barr, Influenza A, B, measles, adenovirus, ECHO-virus, Mumps,

Borrelia IgM and IgG antibodies.

Varicella Zoster IgG was elevated at >4000 MIU/ml (normal high up to 50), and varicella Zoster IgM was 0.351 (index).

Table 1
Causes of a "High"lesion of the vagus nerve.

Cause	Time course	Details
Trauma [5,18]	Acute	Fracture base of the skull and occipital condyles
Infection [19–21]	Acute	Diphteria, Herpes, Lyme, TBC, viral, HZ
Immune mediated [1,22]	Acute	Brachiofacial variant of of polyradiculitis
Vascular [23]	Acute	Aneurysm, carotid artery dissection
Base of skull tumors	Chronic progressive	Neurilemmomas, 31%, [5,24,25] Neurofibromas, 14%, Paragangliomas, 50%, Plasmacytoma, Metastasis- eg prostate cancer
Radiotherapy [26]	Chronic progressive	Local radiotherapy, radio –Surgery and Gamma knife

4. Discussion

Isolated caudal cranial nerve lesions are rare [2] and symptoms and signs are difficult to discriminate due to overlap in their distribution.

The case history, findings, and symptoms match the description of a rare "high"vagus nerve lesion [3,5]. An additional glossopharyngeal nerve lesion [6,7] seems unlikely, as there was only an incomplete paralysis of the soft palate, no vesicles in the oral cavity were detected, and taste sensations were normal. An isolated herpes zoster (HZ) infection of the vagus nerve was suspected based on skin changes and the high titer of Varicella Zoster IgG. A selective involvement of the vagal nerve ganglia could not be demonstrated in the MR images, probably also due to technical reasons [8].

The swelling and reddening of the left ear was possibly due to a concomitant erysipelas, although the sparing of the pinna [9] is unlikely to be seen in erysipelas and also the CRP level was low. The swelling of the ear subsided within days after initiation of the antibiotic and antiviral therapy, whereas the swallowing difficulties continued. They disappeared only after addition of prednisone. The finding of crusts in the auricle is the only visible and traceable evidence for HZ, in addition to the high serological titer.

As a differential diagnosis, several cases of the so called „red ear syndrome"have been reported, which is a non-infections skin condition [10], possibly also due to autonomic dysfunction [11], but without a swallowing disorder.

The vagus nerve can be damaged at several sites in its peripheral course. The most frequent lesions are recurrent laryngeal nerve palsies, which can occur in a number of conditions [12,13] and must also be considered in all vocal cord dysfunctions [14].

Lesions of the proximal part of the vagus nerve between the jugular foramen and the nerve sheath are termed "high" vagus nerve lesions [3,5]. The clinical hallmark of an acute "high" vagus nerve lesion is abrupt and complete swallowing inability, combined with focal headache radiating into the posterior part of the skull. The paralysis of the vocal fold can be misinterpreted as a recurrent laryngeal nerve lesion, which would point to a more distal anatomical site. Several cases [15,16] and a number of causes, usually characterized as either acute or chronic, have been described (Table 1). According to Fang [5], isolated "high" vagus nerve lesions are rare and represent about 10% of vagus nerve lesions. Conversely, in HZ, vagus nerve lesions represented less than 1% in a series of 330 patients with cranial nerve lesions due to HZ [17].

The mechanism of the "high" vagus nerve lesion is presumably neurapraxia of the nerve due to local swelling and inflammation, with preservation of the axons. This would be compatible with the effectiveness of steroids and the rapid reversibility, which could not be explained by regeneration alone. This remains hypothetical as electrophysiological studies are not available for this part of the nervous system.

The identification of a rare "high" vagus nerve lesion is important to consider in an acute swallowing disorder, and needs to be included in the differential diagnostic considerations. The combination of local headache, abrupt swallowing inability, and local swelling of the ear may be a useful clinical guide.

Table 1 summarizes several causes of "high" vagus nerve lesions. The clinical course is mainly distinguished between acute (trauma, infection and vascular) and insidious onset (usually space occupying lesions).

References

[1] B.R. Wakerley, N. Yuki, Pharyngeal-cervical-brachial variant of Guillain-Barre syndrome, J. Neurol. Neurosurg. Psychiatry 2014 (85) (2014) 339–344.
[2] J. Finsterer, W. Grisold, Disorders of the lower cranial nerves, J. Neurosci. Rural Pract. 6 (2015) 377–391.
[3] M. Rontal, E. Rontal, Lesions of the vagus nerve: diagnosis, treatment and rehabilitation, Laryngoscope. 87 (1977) 72–86.
[4] P.K.M.C.J. Thomas, Diseases of the ninth, tenth, eleventh, and twelfth cranial nerve, Chapter 52, in: P.J. Dyck, P.K. Thomas (Eds.), Peripheral Neuropathy, Vol. 2, fourth edition, Elsevier Saunders, 2005.
[5] T.J. Fang, Y.Y. Tam, M.S. Courey, H.Y. Li, H.C. Chiang, Unilateral high vagal paralysis: relationship of the severity of swallowing disturbance and types of injuries, Laryngoscope 121 (2011) 245–249.
[6] H. Nakagawa, M. Nagasao, T. Kusuyama, H. Fukuda, K. Ogawa, A case of glossopharyngeal zoster diagnosed by detecting viral specific antigen in the pharyngeal mucous membrane, J. Laryngol. Otol. 121 (2007) 163–165.
[7] J. Clark, Herpes zoster of right glossopharyngeal nerve, Lancet. 1 (8106) (1979) 38–39.
[8] M. Adachi, A case of varicella zoster virus polyneuropathy: involvement of the glossopharyngeal and vagus nerves mimicking a tumor, AJNR Am. J. Neuroradiol. 29 (2008) 1743–1745.
[9] H. Sugimoto, K. Furukawa, Milian's ear sign: erysipelas, IDCases. 14 (2018), e00449.
[10] J.W. Lance, The red ear syndrome, Neurology. 47 (1996) 617–620.
[11] R.W. Evans, J.W. Lance, The red ear syndrome: an auriculo-autonomic cephalgia, Headache. 44 (2004) 835–836.
[12] K.M. Grundfast, E. Harley, Vocal cord paralysis, Otolaryngol. Clin. N. Am. 22 (1989) 569–597.
[13] M. Yamada, M. Hirano, H. Ohkubo, Recurrent laryngeal nerve paralysis. A 10-year review of 564 patients, Auris Nasus Larynx 10 (S1–15) (1983).
[14] Website: Neuromuscularwustl: Vocal Cord Disorders. https://neuromuscularwustledu/musdist/bulbarhtml#vocalcord, 2021. Access 2/2021.
[15] G.H. Bateman, S.R. Mawson, A case of herpes zoster confined to the tenth cranial nerve, J. Laryngol. Otol. 64 (1) (1950) 17.
[16] T. Ohashi, M. Fujimoto, H. Shimizu, T. Atsumi, A case of isolated vagus nerve palsy with herpes zoster, Rinsho Shinkeigaku 34 (1994) 928–929.
[17] P.W. Tsau, M.F. Liao, J.L. Hsu, H.C. Hsu, C.H. Peng, Y.C. Lin, et al., Clinical presentations and outcome studies of cranial nerve involvement in Herpes Zoster infection: a retrospective single-center analysis, J. Clin. Med. 9 (2020) 4.
[18] N.C. Utheim, R. Josefsen, P.H. Nakstad, T. Solgaard, O. Roise, Occipital condyle fracture and lower cranial nerve palsy after blunt head trauma - a literature review and case report, J. Trauma Manag. Outcomes 9 (2015) 2.
[19] S.C. Tang, J.S. Jeng, H.M. Liu, P.K. Yip, Isolated vagus nerve palsy probably associated with herpes simplex virus infection, Acta Neurol. Scand. 104 (2001) 174–177.
[20] J.M.M.R. Velázquez, J.A. Garrido, A.A. Tejerina, Lower cranial nerve involvement as the initial manifestation of Lyme borreliosis, Neurologia (Barcelona, Spain) 14 (1999) 36–37.
[21] R.S.M.K. Fuyuki Tateno, Emina Ogawa, Yohei Tsuyuzaki, Yoshida M.Y. Tomoe, Zoster lower cranial neuropathy: a case report and a review, Am. J. Case Rep. 12 (2011) 137–139.
[22] T. Mengi, Y. Secil, T.K. Incesu, S. Arici, Z.O. Akkiraz, N. Gurgor, et al., Guillain-Barre syndrome and swallowing dysfunction, J. Clin. Neurophysiol. 34 (2017) 393–399.
[23] M. Sturzenegger, P. Huber, Cranial nerve palsies in spontaneous carotid artery dissection, JNNP 56 (1993) 1191–1199.
[24] J.D. Green Jr., K.D. Olsen, L.W. DeSanto, B.W. Scheithauer, Neoplasms of the vagus nerve, Laryngoscope 98 (1988) 648–654.
[25] J. Kapoor, A. Trinidade, G. Mochloulis, W. Mohamid, Plasmacytoma of the atlas presenting as hoarseness: a rare cause of unilateral vocal fold palsy, J. Laryngol. Otol. 126 (2012) 870–872.
[26] S. Janssen, Ch. Glanzmann, B. Yousefi, K. Loewenich, G. Huber, S. Schmid, G. Studer, Radiation-induced lower cranial nerve palsy in patients with head and neck carcinoma, Mol. Clin. Oncol. 3 (2015 Jul) 811–816.