

CASE REPORT/OPIS PRZYPADKU

Idiopathic trigeminal sensory neuropathy. A case report

Idiopatyczna czuciowa neuropatia trójdzielna. Opis przypadku

Piotr Szczudlik¹, Biruta Kierdaszuk¹, Leopold Bakoń², Edyta Maj², Anna Kamińska¹

¹Katedra i Klinika Neurologii, Warszawski Uniwersytet Medyczny

²II Zakład Radiologii, Warszawski Uniwersytet Medyczny

Neurologia i Neurochirurgia Polska 2013; 47, 5: 499-501

DOI: 10.5114/ninp.2013.34694

Abstract

Idiopathic trigeminal sensory neuropathy is a rare clinical condition characterized by sensory disturbances on the face. Its symptoms may be permanent or temporary and a wide variety of diagnostic procedures is usually required to establish the diagnosis. Frequently, it is the first manifestation of a systemic disorder. In the majority of cases causal treatment is not possible, even though patients with trigeminal sensory neuropathy should be carefully monitored by physicians.

Key words: idiopathic trigeminal sensory neuropathy, trigeminal sensory neuropathy, trigeminal nerve.

Case report

A 43-year-old woman was admitted for the first time to the Department of Neurology, Medical University of Warsaw in January 2012. She complained of a sudden loss of sensation on the left side of her face. A few weeks before she suffered from mild upper respiratory tract infection. The medical history was unremarkable except for meningitis when she was 16 years old.

Neurological examination revealed only impairment of superficial sensation on the left side of her face in the area of innervation of all three branches of the left trigeminal nerve. Computed tomography performed at admission presented no abnormalities. All laboratory investigations, including C-reactive protein, procalcitonin and erythrocyte sedimentation rate, were within

Streszczenie

Idiopatyczna czuciowa neuropatia trójdzielna to rzadka choroba dotycząca zaburzeń czucia w obrębie twarzy. Zjawisko to może mieć charakter przejściowy lub stały, diagnostyka zaś wymaga zazwyczaj wykonania wielu różnych badań. Często neuropatia trójdzielna jest pierwszym objawem choroby układowej. Najczęściej leczenie przyczynowe nie jest możliwe, jednak pacjenci z idiopatyczną neuropatią trójdzielną powinni być pod stałą opieką medyczną.

Słowa kluczowe: idiopatyczna czuciowa neuropatia trójdzielna, czuciowa neuropatia trójdzielna, nerw trójdzielny.

normal limits. The levels of immunoglobulins, thyroid hormones, proteinogram, rheumatoid factor, and antibodies against Lyme disease in serum were in the normal ranges. Additionally, thrombophilia, the presence of lupus anticoagulant, anti-neutrophil cytoplasmic antibodies, antinuclear and anticardiolipin antibodies, increased angiotensin converting enzyme activity, HIV infection and syphilis were excluded by appropriate tests. The level of antithrombin III was within normal limits. Visual-, somatosensory- and brain stem auditory-evoked potentials as well as blink reflex potentials presented no abnormalities. Aneurysms and vascular malformations were excluded by the use of computed tomography angiography. Moreover, the computed tomography of the skull base and nasal sinuses revealed chronic sinusitis of the left maxillary sinus and in both sphenoid sinuses.

Correspondence address: dr Piotr Szczudlik, Klinika Neurologii WUM, ul. Banacha 1 A, 02-097 Warszawa, Polska,

e-mail: pszczudlik@wum.edu.pl

Received: 29.11.2012; Accepted: 30.01.2013

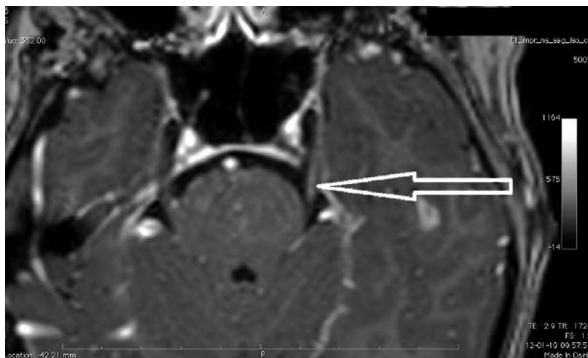


Fig. 1. T1-weighted images of head MRI with gadolinium enhancement of trigeminal nerve (arrow)

Table 1. Differential diagnosis of trigeminal sensory neuropathy (adapted from Dumas and Pérusse [4])

Central origin	Primary and metastatic cerebral tumors (schwannoma, meningioma, cerebral metastases, leptomeningeal infiltration)
	Other cerebral conditions (demyelinating diseases, cerebrovascular diseases, syringobulbia, conversion disorders)
Peripheral origin	Iatrogenic (postinjection or postsurgery)
	Infection (odontogenic infection, osteomyelitis, endocarditis)
	Primary tumors of jaw
	Metastatic tumors of jaw
	Perineural spread of squamous cell carcinoma
	Peripheral neuropathy
Central and/or peripheral origin	Lymphoreticular malignancies
	Lupus
	Systemic sclerosis
	Sjögren syndrome
	Mixed connective tissue disorder
	Sarcoidosis
	Amyloidosis
	Wegener granulomatosis
	Neuropathy
	Syphilis
	Lyme disease
	AIDS neuropathy
	Sickle-cell anemia
	Drug-induced neuropathy
Idiopathic trigeminal sensory neuropathy	

The patient was consulted by a laryngologist who recommended the treatment of chronic sinusitis. Magnetic resonance imaging (MRI) presented abnormal enhancement of the left trigeminal nerve in the segment between the brain stem and the Gasserian ganglion (Fig. 1). Additionally, lumbar puncture showed a slightly elevated protein level (65 mg/dL; normal range 15-45 mg/dL), without oligoclonal protein bands, and absence of anti-*Borrelia* antibodies. The only abnormality was an elevated serum level of chicken-pox antibodies in class G (16.02; results above 11 are considered positive) but not in class M, which may be interpreted as a sign of past infection. The patient was also consulted by a rheumatologist, who did not find any features of connective tissue disorder.

The patient was admitted for the second time to the Department of Neurology, Medical University of Warsaw, in July 2012. During the last months she experienced a slight decrease in the sensation disturbances on the left side of her face. Laboratory investigations were again normal. MRI revealed no contrast enhancement or any other abnormalities within the cranial nerves and brain.

Discussion

Our case represents the infrequent clinical condition of idiopathic trigeminal sensory neuropathy. In differential diagnosis we took into consideration primary or metastatic brain tumors, demyelinating diseases, connective tissue diseases, systemic infection and sarcoidosis as possible causes of trigeminal neuropathy. However, none of the mentioned disorders was confirmed in additional tests. The diagnosis was clarified by suggestive MRI of the left trigeminal nerve. After a few months MRI showed complete resolution of abnormalities. These findings are comparable with literature data where similar changes were described in MRI [2].

Trigeminal sensory neuropathy comprises sensation alterations within one or more branches of the fifth cranial nerve. Generally, it occurs after the second decade of life, and the corneal reflex should be preserved [3]. Diagnosis of idiopathic trigeminal sensory neuropathy is challenging and usually there is a heterogeneous group of disorders which have to be first excluded (Table 1) [4]. Some authors have distinguished acute primary forms with a rather painless beginning and the possibility of full recovery. Chronic forms of idiopathic trigeminal sensory neuropathy are characterized by a complicated course and sometimes pain and are more often connect-

ed with connective tissue diseases [1]. Detailed medical and dental history of the patient is necessary as in some cases the onset of disease may be preceded by infection [4]. Among infectious agents, herpes zoster virus, herpes simplex, syphilis and leprosy should be mentioned [1]. It was also reported that trigeminal sensory neuropathy was provoked by facial dermatitis caused by exposure to the well-known anthurium flower (*Anthurium* sp.) [5]. On the other hand, the patient may be unaware of some symptoms of a systemic disorder. When sudden beginning and rapid progression are observed, a neoplastic process should be excluded (or considered in the differential diagnosis) [1]. The involvement of several nerves or dermatomes indicates a central origin. When symptoms are associated exclusively with the third branch of the trigeminal nerve, mental nerve neuropathy is more likely. It is mainly connected with metastatic lesions in the mandible, which may originate from the breast, lung, prostate, thyroid or kidney cancer. As mentioned in Table 1, perineural spread of squamous cell carcinoma is another condition related to trigeminal neuropathy. The nasopharynx area is very difficult to assess by imaging investigations; however, clinicians should remember direct spread of cancer cells through the cranial nerves. Trigeminal sensory neuropathy may be the first manifestation of multiple sclerosis; nevertheless, in the majority of cases some other abnormalities are disclosed on neurological examinations. When connective tissue disorder is suspected, various immunological tests are helpful to establish the correct diagnosis. The etiology of selective trigeminal nerve involvement in connective tissue diseases remains unknown; however, mixed motor-sensory neuropathy, sensory neuropathy and mononeuritis multiplex were described in systemic sclerosis [6]. Posttraumatic origin of trigeminal sensory neuropathy is also possible, and it is often connected with lower third molar extraction, minor fractures or root canal treatment [7]. It may comprise up to 40% of all causes and results from anatomical arrangement. As the nerves are damaged, the recovery process sometimes takes up to 6 months or even 2 years [1]. Recently published data indicate that faster recovery is connected with younger age of patients and less severe pain [8]. The group of drugs involved in trigeminal sensory neuropathy includes for example phenytoin, labetalol, HIV protease inhibitors, acetazolamide, ergotamine, nitrofurantoin, propranolol, monoamine oxidase inhibitors, tricyclic antidepressants, isoniazid and propofol [3,7]. Apart from these, some case reports have suggested a relationship between facial numbness and mefloquine, a drug used for treat-

ment and prophylaxis of malaria [3]. It was reported that detection of enhancement of the trigeminal nerve on MRI led to surgical procedures in order to exclude neoplastic lesions; however, no pathological tissue was found, and sequential MRI showed progressive resolution of changes [9]. Literature data also indicate that in some cases a special advanced method of MRI called CISS (constructive interference steady state – a type of gradient echo sequence) should be performed to better visualize the trigeminal nerve [10].

In conclusion, it is worth remembering that identification of idiopathic origin of numbness or paresthesia on the face may be established only after detailed clinical examination. A lack of evidence of primary or metastatic tumors, systemic disorder or iatrogenic injury allows the clinicians to diagnose idiopathic trigeminal sensory neuropathy.

Disclosure

Authors report no conflict of interest.

References

1. Peñarrocha M., Cervelló M.A., Martí E., et al. Trigeminal neuropathy. *Oral Dis* 2007; 13: 141-150.
2. Chan L.-L., DeMonte F., Ginsberg L.E. Thermally induced transient trigeminal sensory neuropathy: imaging findings. *Am J Neuroradiol* 2000; 21: 201-202.
3. Watt-Smith S., Mehta K., Scully C. Mefloquine-induced trigeminal sensory neuropathy. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2001; 92: 163-165.
4. Dumas M., Pérusse R. Trigeminal sensory neuropathy. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1999; 87: 577-582.
5. Twardowsky C.A., Teive H.A.G., Siquineli F., et al. Trigeminal sensory neuropathy and facial contact dermatitis due to Anthurium sp. *Arq Neuropsiquiatr* 2007; 65: 822-825.
6. Vincent F.M., Van Houzen R.N. Trigeminal sensory neuropathy and bilateral carpal tunnel syndrome: the initial manifestation of mixed connective tissue disease. *J Neurol Neurosurg Psychiatry* 1980; 43: 458-460.
7. Shotts R.H., Porter S.R., Kumar N., et al. Longstanding trigeminal neuropathy of nontraumatic cause. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1999; 87: 572-576.
8. Peñarrocha M., Peñarrocha D., Bagán J.V., et al. Post-traumatic trigeminal neuropathy. A study of 63 cases. *Med Oral Patol Oral Cir Bucal* 2012; 17: e297-e300.
9. Dominguez J., Lobato R.D., Madero S., et al. Surgical findings in idiopathic trigeminal neuropathy mimicking a trigeminal neurinoma. *Acta Neurochir* 1999; 141: 269-272.
10. Kuroki N., Ohyagi Y., Kawano Y., et al. Acute onset facial numbness. *J Clin Neurosci* 2008; 15: 1077.