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Ophthalmoplegia considered to be Tolosa-Hunt syndrome: A case report

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ABSTRACT

Tolosa-Hunt syndrome (THS) is a type of painful ophthalmoplegia characterized by unilateral orbital pain and oculomotor paresis. This report describes a case of painful ophthalmoplegia considered to be THS. The patient was a 72-year-old woman. After dental implant placement in the left maxillary first molar site, she was referred to our hospital for persistent left periorbital and facial pain, diplopia, and taste disorder. With an initial diagnosis of trigeminal neuralgia, diplopia, and taste disorder, we began administration of carbamazepine and polaprezinc, and asked the department of ophthalmology in our hospital to examine her diplopia. With the increase in serum zinc levels, the taste disorder resolved, but her periorbital pain and diplopia were aggravated, and she had to be hospitalized. As a result of extensive examinations, we suspected her painful ophthalmoplegia was due to cavernous sinus abnormalities; finally, THS was diagnosed. There was significant improvement in the periorbital pain within 48 h after the start of corticosteroid therapy. Since then, she has never suffered from painful ophthalmoplegia. This case indicates that careful evaluation and accurate diagnosis are critical in cases of painful ophthalmoplegia, and the possibility of THS should always be kept in mind.

1. Introduction

Painful ophthalmoplegia is a rare pathologic condition characterized by different combinations of ocular conditions, including unilateral periorbital or hemicranial pain, ipsilateral oculomotor paralysis, and oculosympathetic dysfunction. Notably, it can result from various etiologies, such as neoplasms, vascular abnormalities, inflammatory disorders, or infections, as well as other miscellaneous conditions [1].

Tolosa-Hunt syndrome (THS) is a painful ophthalmoplegia disorder, described in the 2013 International Classification of Headache Disorders (Third Edition, ICHD-3 beta) as unilateral orbital pain in association with paralysis of one or more of the third, fourth, and/or sixth cranial nerves [2]. The ocular symptoms are caused by extrinsic compression and secondary dysfunction of neurovascular structures within the cavernous sinus by a nonspecific inflammatory granuloma. Infrequently, when the inflammation extends into the orbital apex and/or superior orbital fissure, dysfunction of the optic, trigeminal, and facial nerves and sympathetic innervation of the pupil can ensue [3,4].

Oral surgeons frequently encounter neurological diseases, such as trigeminal neuralgia, in their day-to-day clinical practice, but they rarely encounter painful ophthalmoplegia. Notably, the clinical presentation of painful ophthalmoplegia has a wide differential diagnosis, which can greatly assist oral surgeons in its early accurate diagnosis and management. This report describes the onset, diagnosis, and treatment in a case of painful ophthalmoplegia considered to be THS that developed after dental implant placement.

2. Case report

A 72-year-old woman had undergone dental implant placement in the left maxillary first molar region in June 2011 (Fig. 1). She experienced spontaneous pain in the left cheek immediately after surgery. The next day, she developed severe left periorbital pain and visited her dentist. He prescribed loxoprofen sodium 180 mg/day for postoperative pain associated with surgical invasion, which did not provide any pain relief within the next 1 week. At 3 weeks after surgery, she developed diplopia and taste disorder. After consultation with her physician, periorbital pain with diplopia and taste disorder, attributed to the dental implant placement, was diagnosed, and she was referred to our hospital in August 2011.

Her medical history was unremarkable and did not include any head trauma or infection. She complained of constant, spontaneous periorbital pain with an intensity of 7/10 on the Numerical Rating Scale (NRS). There was no swelling on her face (Fig. 2A), and hypoesthesia

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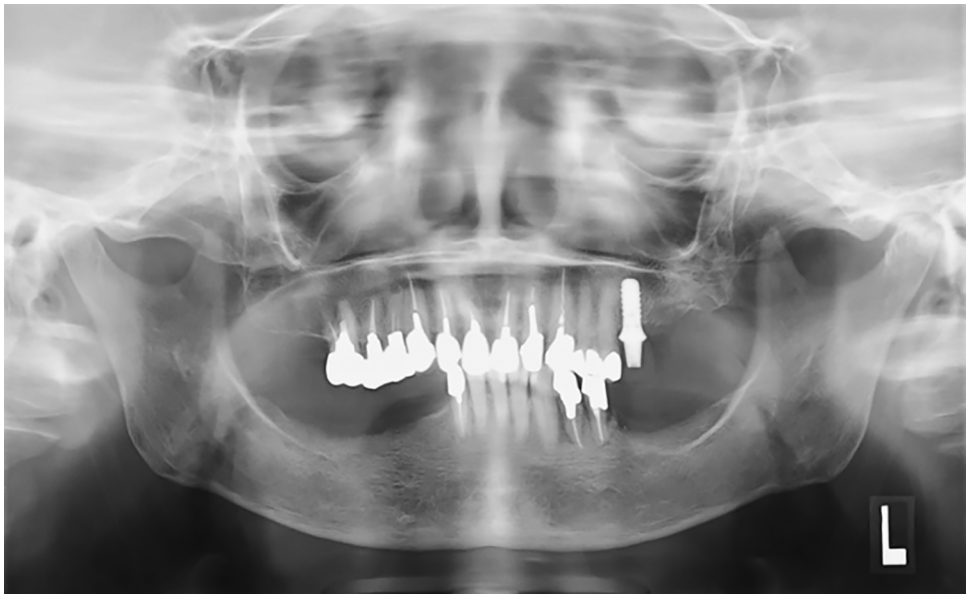

Fig. 1. Panoramic radiograph immediately after dental implant placement in the left maxillary first molar region.

was not observed in the Semmes-Weinstein sensory test. An intraoral examination did not reveal any local inflammation such as abscess at the site of the dental implant (Fig. 2B). She underwent a thorough radiographic examination, including computed tomography (CT) and technetium 99 m-methyl diphosphonate (99 mTc-MDP) bone scintigraphy. These examinations did not reveal any relevant abnormalities (Fig. 2C, D). Her body temperature was 36.5 °C, and a blood examination revealed little inflammatory reaction. The peripheral white blood cell count was 5900/μL and C-reactive protein level was 1.2 mg/dL. A taste examination using the filter paper disc method was performed to examine the taste recognition threshold, because the serum zinc level was low (50 μg/dL). In brief, recognition thresholds for four basic tastes (sweet, salty, sour, and bitter) were evaluated using the same chemical solutions (sucrose, NaCl, tartaric acid, and quinine, respectively). The solutions were sequentially diluted with distilled water into five grades. Concentration number 1 is the lowest and 5 is the highest (0.3, 2.5, 10, 20, and 80% for sucrose; 0.3, 1.25, 5, 10, and 20% for NaCl; 0.02, 0.2, 2, 4, and 8%, for tartaric acid; 0.001, 0.02, 0.1, 0.5, and 4% for quinine). The threshold level of sweet in the left chorda tympani nerve field was higher compared with that on the right side. As other neurological symptoms, facial palsy, tinnitus, vertigo and nystagmus were not observed.

Because of the presence of pain focused in the left trigeminal region, absence of any radiographic or hematological abnormalities, and the failure of the nonsteroidal anti-inflammatory drugs prescribed after dental implant placement, we suspected trigeminal neuralgia and prescribed carbamazepine 200 mg/day. In addition, we prescribed polaprezinc 150 mg/day to treat the taste disorder by increasing serum zinc levels. Furthermore, we consulted the department of ophthalmology in our hospital regarding her diplopia, but no organic or neurological abnormalities other than bilateral esotropia were observed in ophthalmic examinations. The taste disorder resolved as the zinc levels increased. In spite of the increased dosage of carbamazepine, her periorbital pain became more severe (NRS 9/10), and was accompanied by an increase in diplopia. In addition, nausea caused by the periorbital pain led to an eating disorder, and she was admitted to our hospital in September 2011.

Because of the exacerbation of the periorbital pain and diplopia, we consulted the department of ophthalmology in our hospital again. It was discovered that the diplopia was due to impairment of ocular excursion (Fig. 3A) and ptosis, suggesting a diagnosis of painful ophthalmoplegia due to cranial nerve abnormality. We subsequently consulted the department of neurosurgery and neurology in our hospital to identify the cause of painful ophthalmoplegia. Magnetic resonance angiography (MRA) and magnetic resonance imaging (MRI) revealed only mild, residual cerebral infarction, but no tumors or vascular disorders that could have caused the painful ophthalmoplegia (Fig. 3B). Gadolinium-enhanced MRI identified an abnormal lesion in the left cavernous sinus (Fig. 3C, D). Hematological examinations also did not reveal any abnormal findings. These included complete blood count; glucose, electrolyte, hemoglobin A1c, anti-nuclear antibody, angiotensin-converting enzyme, C-reactive protein, and cytoplasmic anti-neutrophil cytoplasmic antibody levels; liver and renal function; serum protein electrophoresis results; and erythrocyte sedimentation rate. Lumbar puncture was performed, and the cerebrospinal fluid was acicular with normal protein and glucose levels; negative bacterial, fungal, and mycobacterial cultures; and negative cytology results (Table 1). Although the clinical course did not fulfill the criteria indicated by ICHD-3 beta (Table 2), the radiological and hematological findings suggested THS. Systemic symptoms associated with THS, such as back pain, chronic fatigue, arthralgia, and gut problems [5], had not occurred.

Two days after admission to our department, the severity of periorbital pain had increased (10/10 on the NRS). After consultations with the departments of neurology, neurosurgery, and ophthalmology, corticosteroid therapy was started: 1000 mg/day pulse intravenous methylprednisolone for 3 days, followed by 60 mg/day oral methylprednisolone. There was significant improvement in the periorbital pain within 48 h (NRS 3/10) after initiation of corticosteroid therapy. Periorbital pain resolved (NRS 0/10) in 7 days, ptosis in 26 days, and diplopia in 40 days. Corticosteroid therapy was tapered gradually and stopped at the end of 5 months. A follow-up MRI performed at 6 months after initiation of corticosteroid therapy was normal (Fig. 4A, B). There was no recurrence of painful ophthalmoplegia for the next 5 years.

3. Discussion

The differential diagnosis of painful ophthalmoplegia is extensive and includes numerous serious etiologies, such as neoplasms (primary intracranial tumors, and local or distant metastasis), vascular abnormalities (aneurysm, carotid dissection, and carotid-cavernous fistula), inflammatory disorders (orbital pseudotumor, giant cell arteritis, sarcoidosis, and THS), infections (fungal and mycobacterial), as well as other miscellaneous conditions (ocular migraine and microvascular