Orofacial pain and headaches associated with exfoliation glaucoma

Noboru Noma, DDS, PhD; Mayumi Iwasa, MD, PhD; Andrew Young, DDS, MSD; Mariko Ikeda, DDS; Yung-Chu Hsu, MD, PhD; Maasa Yamamoto, DDS; Kenji Inoue, MD, PhD; Yoshiki Imamura, DDS, PhD

Glaucoma is the leading cause of blindness worldwide and is common among older adults. Glaucoma is defined as elevated intraocular pressure (IOP) (> 22 millimeters of mercury) in association with ocular damage and is classified as open angle or closed angle, depending on the configuration of the anterior chamber angle. Closed-angle glaucoma is a sight-threatening ophthalmic emergency. Patients typically have acutely painful red eyes, periocular headaches, vision loss, nausea, and vomiting, but extraocular symptoms and systemic manifestations such as headaches are the main symptoms in some patients. Pain is usually localized in the ocular region but may be referred to the temporal or hemifacial region, making the differential diagnosis somewhat difficult, as the pain may be similar to odontogenic, migrainous, and other forms of headaches, and to certain systemic disorders.

Open-angle glaucoma, the most common type of glaucoma, is painless, usually does not cause headaches, nausea, or vomiting, and progresses slowly. Peripheral vision may begin to decrease, leading to blindness if the condition is not treated. Exfoliation syndrome (XFS) is a systemic, age-related ocular disease in which abnormal extracellular material is produced and accumulates in tissues. XFS is the most common identifiable cause of open-angle glaucoma. Exfoliation glaucoma may result in rapid elevation of IOP, which can cause headaches. In this article, we report a case of exfoliation glaucoma in a patient with orofacial pain.

CASE REPORT
A 77-year-old woman went to an orofacial pain clinic reporting left-sided maxillary molar pain, facial pain, and headaches of 7 months’ duration. She reported eye

ABSTRACT

Background and Overview. Exfoliation syndrome is the most common identifiable cause of open-angle glaucoma. The authors report a case of exfoliation glaucoma in a patient who had orofacial pain.

Case Description. A 77-year-old woman was treated at the orofacial pain clinic for left-sided facial pain and headaches of 7 months’ duration. Her cataracts and open-angle glaucoma had been diagnosed approximately 3 years earlier. Her main symptoms were orofacial pain, eye redness, inflammation of the eyelids, and eyelid edema. Magnetic resonance imaging showed no evidence of intracranial or extracranial pathology. Hemicrania continua was considered as a possible diagnosis. Indomethacin was prescribed but did not affect her headaches. She then went to an ophthalmologist to rule out secondary headaches. Intraocular pressure was 13 millimeters of mercury in the right eye and 67 mm Hg in the left eye. The ophthalmologist made a diagnosis of exfoliation glaucoma, and the patient underwent surgical treatment for the glaucoma and cataracts. After surgery, she was free of symptoms, and intraocular pressure was 15 mm Hg in the left eye.

Conclusions and Practical Implications. During differential diagnosis, dentists need to consider intraoral and systemic conditions that can mimic odontogenic or orofacial pain disorders in the patient’s medical history and that have a higher incidence associated with the patient’s age.

Key Words. Exfoliation glaucoma; orofacial pain; intraocular pressure.

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redness, inflammation of the eyelids, and eyelid edema on the left side during pain exacerbations. Three years earlier, her cataracts and open-angle glaucoma had been diagnosed. An ophthalmologist had prescribed glaucoma medications (bimatoprost eye drops and dorzolamide hydrochloride–timolol maleate ophthalmic solution) to be instilled in both eyes at bedtime. Bimatoprost reduces IOP by increasing the rate of aqueous humor flow. Dorzolamide hydrochloride–timolol maleate ophthalmic solution is a combination of a topical carbonic anhydrase inhibitor and a topical β-adrenergic receptor blocking agent, and it reduces IOP. The patient had been taking the medications for 3 years.

She later developed intermittent left facial pain and headaches and swelling in the infraorbital region. She consulted her dentist for treatment. The pain intensity was usually mild (2-3 on a numerical rating scale; maximum 10), but she reported occasional exacerbations of moderate intensity, accompanied by eyelid edema and ocular pain (5-6 on the same scale). Exacerbations of pain occurred 2 to 3 times per month and lasted from 3 to 5 days. During exacerbations, the pain changed to a sensation of pressure. An intraoral examination by a general dentist revealed no visible abnormality, and she was thus referred to our orofacial pain clinic.

On physical examination, her blood pressure was 140/80 mmHg, and her body temperature was 35.5°C. Examination of the temporomandibular joint revealed a painless active range of motion of 45 mm and no joint noises such as clicking or crepitation. Masticatory muscle palpation revealed no tenderness. The patient’s medical history included diabetes mellitus, high cholesterol, and hypertension. She reported use of vildagliptin, olmesartan medoxomil and azelnidipine, and rosuvastatin calcium. Extraoral examination revealed mild conjunctival injection, lacrimation, and eyelid edema in the left eye, which the patient reported were ongoing for 5 months (Figure 1). Intraoral examination revealed no gingival tenderness on the edentulous sides of the maxillary left second premolar. A panoramic radiograph showed no abnormal findings in the maxillary sinuses (Figure 2), and magnetic resonance imaging results showed no evidence of intracranial or extracranial pathology (Figures 3 and 4).

Hemicrania continua (HC) was considered, and her dentist prescribed indomethacin. However, her symptoms persisted, and 2 weeks later, she began to report eye mucus and blurred vision. Her left eye showed signs of progressive inflammation, and she visited the ophthalmology department again to rule out headaches secondary to an ocular condition.

During the ophthalmologic examination, visual acuity was 30/50 for the right eye and 2/50 for the left eye, and IOP was 18 mm Hg in the right eye and 67 mm Hg in the left eye (normal IOP, ≤ 20 mm Hg). The ophthalmologist diagnosed exfoliation glaucoma and corneal edema from elevated IOP. The IOP immediately decreased to 41 mm Hg after administration of intravenous mannitol, and the pain subsided.

She used eye drops (bimatoprost, latanoprost, brimonidine tartrate, and dorzolamide hydrochloride salt plus timolol maleate) for 15 months, but the IOP in the left eye gradually increased. She underwent surgical treatment for glaucoma and cataracts, which she also had (cataracts are painless). After surgery, the patient report that she was free of symptoms. Postoperative IOP was 15 mm Hg in the left eye; visual acuity was 30/50 for the right eye and 30/50 for the left eye.

**DISCUSSION**

Compared with open-angle glaucoma, exfoliation glaucoma is associated with a worse clinical course and prognosis for several reasons. Mean IOP is higher in patients with XFS than in the general population and those with open-angle glaucoma. In addition, at a specific IOP, eyes with XFS are more likely to develop glaucomatous damage than are eyes without XFS. Optic nerve damage is more frequent and more severe at diagnosis, visual field damage and response to medications are worse, and surgical treatment is required more frequently.

Our patient’s open-angle glaucoma was originally diagnosed. When she developed eye mucus and visual disturbances, with progressive inflammation of the left eye, she was again referred to the ophthalmology department, where exfoliation glaucoma was diagnosed. Reports have described patients with orofacial pain secondary to closed-angle glaucoma. Hurwitz reported a case of closed-angle glaucoma with conjunctivitis in a patient who had consulted a dentist for facial pain.

however, in that report the patient also had eye pain, which was not the case for our patient. Joseph also reported an unusual case of closed-angle glaucoma in a patient with ocular pain and eye redness who had consulted her dentist for pain in the maxillary molar region. Although impacted maxillary second and third molars were extracted, pain in the maxillary jaw and ocular region was unchanged.

In the International Classification of Headache Disorders, 3rd edition (beta version), “headache attributed to acute glaucoma” is defined as headaches, usually unilateral, caused by closed-angle glaucoma, and associated with other symptoms and clinical signs of this disorder. However, conjunctivitis in our patient was mild and would thus not refer pain to the head, face, and mouth. Our patient’s third ophthalmic condition was exfoliation glaucoma, which is usually not painful. However, exfoliation glaucoma can cause a rapid increase in IOP, which may result in pain, as was the case in our patient.

Our patient had an uncommon case of pain localized to a maxillary molar—and referred to the facial, temporal, and parietal regions—originating in the eye. The ophthalmic division of the trigeminal nerve supplies sensory fibers to the eye. The maxillary division of the same nerve innervates the teeth of the maxillary jaw, the cheek, and maxillary sinuses. Any ocular source of pain can cause a headache in any region, by complex pain referral. When the ocular origin of pain is unilateral, the headache is ipsilateral.

Because a headache is sometimes the patient’s chief symptom in closed-angle glaucoma, it can be

**Figure 2.** Panoramic radiograph showing no abnormal findings on the intact floor of the maxillary sinus.

**Figure 3.** Magnetic resonance image showing no evidence of intracranial or extracranial pathology.

**Figure 4.** Magnetic resonance image revealing no abnormal findings in the maxillary sinuses.

would therefore not explain our patient’s pain. Conjunctivitis is sometimes painful and can cause headaches. “Headache attributed to ocular inflammatory disorder” is defined as a headache that is caused by ocular inflammatory conditions, such as iritis, uveitis, scleritis, or conjunctivitis, and is associated with other symptoms and clinical signs of the disorder. However, conjunctivitis in our patient was mild and would thus not refer pain to the head, face, and mouth. Our patient’s third ophthalmic condition was exfoliation glaucoma, which is usually not painful. However, exfoliation glaucoma can cause a rapid increase in IOP, which may result in pain, as was the case in our patient.

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Because a headache is sometimes the patient’s chief symptom in closed-angle glaucoma, it can be
misdiagnosed as migraine, cluster headache (CH), temporal arteritis, or atypical facial neuralgia. Sujatha and colleagues reported 2 cases for which migraine was the initial working diagnosis but the cause of symptoms was eventually identified as periodic acute IOP elevation. Prasad and colleagues reported the misdiagnosis of CH as closed-angle glaucoma. These findings indicate that comprehensive evaluation is warranted when narrow-angle glaucoma is suspected.

In the International Classification of Headache Disorders, the trigeminal autonomic cephalgias include CH, paroxysmal hemicrania, short-lasting unilateral neuralgiform headache attacks with conjunctival injection and tearing, as well as the short-lasting neuralgiform headache attacks with cranial autonomic features, and HC. HC is usually moderate, persistent, side-locked, and associated with ipsilateral conjunctival injection, lacrimation, nasal congestion, rhinorrhea, forehead and facial sweating, miosis, ptosis, and eyelid edema. The continuous moderate pain and absolute indomethacin response seen in HC differentiate it from CH, which is usually episodic and excruciating, and characterized by intense autonomic activation and nonresponse to indomethacin.

HC is absolutely sensitive to indomethacin, a nonsteroidal anti-inflammatory drug that acts on nitric oxide. Indomethacin is therefore used as a diagnostic test for HC. Indomethacin should be given at a dose of 25 milligrams on the first day, which can be increased, if necessary, to 25 mg 3 times per day for 3 days. Most patients respond within 24 hours. This regimen was used for our patient, but she was nonresponsive to treatment.

Horner syndrome, which is caused by damage to the sympathetic trunk supplying the face, might also be considered in the differential diagnosis for cases similar to our patient, because it too includes a decrease in eyelid opening. However, Horner syndrome results in drooping of the upper eyelid (ptosis) and elevation of the lower eyelid; our patient had eyelid edema.

In our patient, we initially considered diagnoses of migraine, CH, and HC. Migraines are usually preceded by prodromes that are episodic, intense, throbbing, and incapacitating—none of these characteristic findings were noted in our patient. Our patient’s facial pain and headaches were similarly mild to moderate, with eye redness, drooping eyelids, and eyelid edema. These characteristics are consistent with HC but not with CH; however, indomethacin testing resulted in no relief and thus we excluded HC. We were also able to exclude CH because she did not fulfill the criteria for location, attack frequency, duration, and accompanying signs and symptoms.

One study reported that severe Pancoast tumors (also known as pulmonary sulcus tumors or superior sulcus tumors) may induce complete Horner syndrome, which is characterized by miosis, anhidrosis, ptosis, and enophthalmos. Although it has features in common with those of our patient, Horner syndrome was not diagnosed, because the patient had eyelid inflammation (not ptosis) and no miosis. The absence of miosis also helps distinguish glaucoma from the trigeminal autonomic cephalgias.

Reiter syndrome is a type of autoimmune, reactive arthritis that develops as a reaction to bacterial infection. It can affect the intestines, genitals, and urinary tract; however, some patients experience multiple ocular symptoms, including blurred vision, conjunctivitis, and secondary glaucoma. We also excluded Reiter syndrome in our patient because of the absence of intestinal, genital, and urinary tract symptoms.

CONCLUSION

We described an unusual case of orofacial pain in a patient with exfoliation glaucoma and conjunctivitis. When these conditions induce orofacial pain, patients may seek dental treatment. Dentists, therefore, need to consider intraoral and systemic conditions, and the patient’s medical history and age when making a diagnosis. This case highlights the importance of a thorough knowledge of odontogenic and nonodontogenic causes of orofacial pain, and the value of consultation with or referral to appropriate medical specialists.

Dr. Noma is an associate professor, Department of Oral Diagnostic Sciences, and Division of Clinical Research, Dental Research Center, Nihon University School of Dentistry, Tokyo, Japan. Dr. Yamamoto is an assistant professor, Department of Dental Practice, Arthur Dugoni School of Dentistry, University of the Pacific, San Francisco, CA. Dr. Ikeda is an instructor, Department of Oral Diagnostic Sciences, Nihon University School of Dentistry, Tokyo, Japan. Dr. Hsu is an assistant professor, Division of Neurology, Department of Internal Medicine, Dimontmon Medical Foundation, Chia-Yi Christian Hospital, Chiai, Taiwan, China. Dr. Yamamoto is an instructor, Department of Oral Diagnostic Sciences, Nihon University School of Dentistry, Tokyo, Japan.

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