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Orbito-Masticatory Syndrome

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Abstract

We describe two unique cases of visual symptoms occurring during mastication in patients with lateral orbital wall defects. A 57-year old male reported intermittent double vision and oscillopsia after a right fronto-temporal-orbito-zygomatic craniotomy with osteotomy of the lesser wing of the sphenoid for a complex invasive pituitary adenoma. Proptosis of the right globe was present only during mastication. Computed tomography (CT) revealed a bony defect in the right lateral orbital wall. A 48-year-old man presented with transient diplopia and scotoma in the right eye elicited by chewing. CT and magnetic resonance imaging demonstrated a bilobed lesion connecting the temporal fossa to the orbit through a defect in the right lateral orbital wall. The regional neuroanatomy and pathophysiology as pertaining to the cases are discussed.

Keywords

proptosis; orbit; craniotomy; diplopia; oscillopsia; mastication

Intermittent visual symptoms (oscillopsia, double vision, vision loss) are common presenting complaints to the clinician and often due to ocular, systemic, orbital or central nervous system diseases. In this report, we describe two unique patients in whom intermittent visual complaints were related to the transmission of forces generated by contraction of the temporalis muscle against the soft tissue contents of the orbit. The relevant neuroanatomy is reviewed in order to explain the pathomechanism of the visual symptoms.

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Figure 5 illustrations by Rob Flewell, CMI.

Case 1

A 57-year-old man was seen for episodes of double vision and oscillopsia four months following a right fronto-temporal-orbito-zygomatic craniotomy with osteotomy of the lesser wing of the sphenoid for an invasive pituitary adenoma. Approximately 2 months after surgery, his daughter noticed that his right eye was moving while eating dinner. He indicated that on one occasion he had experienced double vision and oscillopsia while chewing.

Visual acuity was 20/20 in both eyes. Color vision was reduced in the right eye with a right relative afferent pupillary defect. Slit lamp examination and intraocular pressures were normal. Confrontation visual fields were full in each eye. Amsler grid testing was normal in each eye. There was atrophy of the right temporalis muscle. Ophthalmoscopy revealed mild pallor of the right optic nerve while the remainder of the posterior segment was normal in each eye. At rest, there was no proptosis. However, when the patient was given a piece of gum to chew in the clinic, he developed intermittent proptosis with inferior and nasal displacement of the right eye. The displacement corresponded with contracture of the right temporalis muscle (Fig 1 and Video 1, Supplemental Digital Content). Postoperative computed tomography (CT) demonstrated a bony defect of the right lateral orbital wall (Fig 2A).

Case 2

A 48-year-old man was referred for evaluation of swelling of the right temporalis fossa and intermittent double vision that began a year prior. Initially, the symptoms occurred rarely upon awakening, but increased in frequency to occur every morning for 30 minutes. He could also elicit diplopia and a superonasal grey spot in his right visual field by chewing or exerting pressure on the right temple. He reported a history of blunt trauma to the right cheek 30 years previously that was treated with temporary placement of a balloon in the nasal passages that was removed after a few weeks.

Visual acuity was 20/20 in both eyes. Kinetic perimetry was normal, as were slit-lamp and dilated ophthalmoscopic examinations. The area immediately superior to the right zygoma was swollen without discoloration, tenderness, or a bruit. There was 2 mm of right proptosis. By Maddox rod testing, there was a 2-4 prism diopter right hyperphoria in all directions of gaze. Jaw clenching induced a small, gray, superonasal scotoma in the right eye and caused the hyperphoria to increase to 14 prism diopters in downgaze. Manual pressure applied to the right temple also elicited vertical diplopia.

Orbital magnetic resonance imaging (MRI) revealed a 26 × 35 × 15 mm bilobed mass in the extraconal space of the right inferolateral orbit and right temporalis muscle (Fig 3A-3D). The signal intensity of the lesion was heterogeneous with peripheral enhancement. CT of the orbits confirmed that the mass passed through a bony defect in the lateral orbital wall (Fig 2B and 2C). Cranial magnetic resonance angiography demonstrated two questionable areas of extracranial flow-related signal inferior to the lesion (Fig 3E). Ophthalmic arteries were visualized bilaterally and the superior ophthalmic veins were not enlarged.

An anterior orbitotomy revealed a fleshy mass with surrounding muscle fibers passing through a defect in the lateral orbital wall. Biopsies were taken and contents of the intraorbital portion of the lesion were aspirated. The defect in the lateral orbital wall was not reconstructed. Histopathologic examination of the biopsy specimens revealed a mixture of small and medium-sized arteries and veins within a fibrous stroma (Fig 4). Some of the vessels had very thin walls and were dilated and ectatic. There was focal hemosiderin deposition. No cytologic atypic or lymphoid aggregates were seen. CD31 immunohistochemistry highlighted the endothelium of the vascular channels. A pathological diagnosis was made of a vascular lesion, either arteriovenous malformation (AVM) or arteriovenous fistula (AVF).

Following surgery, catheter angiography was performed and no persistent arteriovenous communication was found. The patient's symptoms resolved following the orbitotomy

Discussion

Chewing-induced oscillopsia and diplopia are rare visual complaints. These symptoms have been reported following eyelid surgery (1) as well as surgery involving the orbital walls. Fayers et al (2) found that 34 of 98 patients (35%) reported oscillopsia following lateral orbital wall decompression for thyroid eye disease (2). The majority of these patients experienced oscillopsia during mastication due to temporalis contraction against the orbital contents. Visual symptoms while chewing also have been described in non-surgical settings. This has been reported after zygomaticomaxillary complex fracture with adhesion of the temporalis muscle to the periorbital (3) and in the setting of dermoid cyst eroding through the lateral orbital wall (4). The pathomechanism of masticatory induced visual symptoms in these cases is due to the contraction forces generated by the temporalis muscle transmitted to the orbital soft tissue through a bony defect in the lateral orbital wall (Figure 5).

Another potential cause of chewing-induced diplopia is a change in volume of a cystic orbital lesion spanning the lateral orbital wall. This has been described in the setting of dumbbell shaped dermoid cysts and is likely a contributing factor in our case 2 (6, 7). The change in volume may be secondary to vascular congestion in the supine position, occlusion of extracranial venous drainage by temporalis contraction, or a shift of blood from the extracranial to orbital part of the lesion by contracting temporalis muscle or manual pressure. In our case 2, the hyper - deviation can be attributed to the mechanical elevation of the eye and the superior nasal scotoma likely reflects pressure on the inferior temporal retina by the orbital portion of the lesion.

A third mechanism of chewing-induced diplopia, not illustrated by our cases, is cranial nerve VII synkinesis. In a patient with prior facial nerve palsy, voluntary contraction of the orbicularis oris was accompanied by synkinetic contraction of the ipsilateral orbicularis oculi, pulling the globe inward and upward to cause visual symptoms (8).

Case 1 highlights the neuro-ophthalmic complications orbitozygomatic craniotomy. This approach often is combined with a fronto-temporal craniotomy to increase visualization and minimize brain retraction, allowing better access to the orbit, anterior fossa, middle fossa,

cavernous sinus, and the superior aspect of the posterior fossa (9). After soft tissue dissection, a fronto-temporal craniotomy is performed by drilling burr holes, which are connected by placing cuts in the bone and then raising a bone flap. Osteotomies are placed in the superior orbital roof, lateral orbital wall, and zygoma to perform an orbitozygomatic craniotomy and residual bone can be removed as needed. During closure, the orbitozygomatic craniotomy is first plated back into place. Because the osteotomies result in bone loss, orbital defects often are reconstructed with a number of materials including porous polyethylene implants or bone cement. The fronto-temporal bone flap is then plated into place.

Reported neuro-ophthalmic complications in the setting of orbitozygomatic and fronto-temporal craniotomy include cosmetic deformity, enophthalmos, pulsatile exophthalmos, ptosis, diplopia, vertical gaze limitations and blindness (10, 11). Poor cosmetic and structural outcomes often stem from suboptimal reconstruction of a defect in the temporal region (12) and we add masticatory proptosis and oscillopsia to the list of complications.

The precise etiology of the vascular lesion in case 2 is unclear. The lack of orbital vascular symptoms or imaging findings suggests that the feeding and draining vessels originated from the extracranial circulation; however pre-operative catheter angiography was not performed to confirm this hypothesis. The history of right facial trauma may have been a contributing factor. Histopathological appearance of the vascular lesion is consistent with either an AVM or AVF. AVMs are benign, congenital hamartomas, which are well-known to occur in the orbit (13, 14). Occurrence in the temporalis muscle is rare (15, 16), and we were unable to find any reports of AVMs passing through a defect in the lateral orbital wall. Possibly the patient's prior facial trauma may have stimulated the lesion to grow in size or caused an orbital wall defect through which it herniated (17). AVFs are acquired lesions that can occur spontaneously or following trauma (18), but again, we could not find any reports of an AVF crossing the lateral orbital wall. If the lesion was an AVF, we speculate that it may have occurred following facial trauma and either involved a vessel that traversed the orbital wall or herniated through an acquired defect in the wall.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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Figure 1.

Appearance of patient at rest. D-F. With mastication, there is anterior movement of the right eye leading to increased fullness of the right lower lid (E compared to B). In addition there is inferior displacement of the right eye during mastication causing increased distance from the superior pupillary border to the right upper lid margin (F compared to C).

Figure 2.

Case 1. A. Axial computed tomography (CT) demonstrates a bony defect (arrow) in the right lateral orbital wall. Case 2. Axial (B) and coronal (C) CT reveals a bony defect (arrows) in the right lateral orbital wall.

Figure 3.

Case 2. Axial (A-C) and coronal (D) magnetic resonance images of dumbbell-shaped lesion with extracranial component in the temporalis muscle and extraconal orbital component. E. Magnetic resonance angiography demonstrates possible flow-related signal in the region inferior to the extracranial portion of the dumbbell-shaped lesion.

Figure 4.

Case 2. Histopathologic appearance of orbital and temporal muscle lesion. A. Arteries and veins of various sizes are present in a fibrous stroma (hematoxylin & eosin, × 50). B. A more magnified view shows no evidence of cytologic atypia or lymphoid aggregates (hematoxylin & eosin, × 400).

Figure 5. Following orbitozygomatic craniotomy (A), a defect in the greater wing of the sphenoid (inset) allows the force of the temporalis muscle to be transmitted into the orbit (inset and B) causing proptosis (arrow).