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Case report

Isolated left upper eyelid ptosis with pansinusitis and contralateral otitis media in a 9-year-old boy

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ABSTRACT

Purpose: Upper eyelid ptosis has different etiologies in children and adults. In children, the common causes include orbital cellulitis, congenital ptosis, Cranial Nerve (CN) III palsy, and Horner's syndrome. The purpose of this report is to discuss an unusual presentation of ptosis.

Observations: We describe a case of a 9-year-old boy with left-sided ptosis with no apparent clinical signs of orbital or preseptal infection. Magnetic resonance imaging (MRI) revealed pansinusitis and contralateral otitis media with direct extension into the superior aspect of the left orbit affecting the levator palpebrae superioris muscle.

Conclusions and importance: This finding on imaging disclosed the etiology of an otherwise unexplained case of upper lid ptosis.

1. Introduction

Acquired upper eyelid ptosis can have many different etiologies, including traumatic, mechanical, neurogenic, and myogenic.[1] The most common cause of ptosis in the older population is aponeurotic due to levator dehiscence.[1] In the pediatric population, common causes include congenital ptosis, Cranial Nerve (CN) III palsy, Horner's syndrome, and mechanical ptosis due to a space-occupying lesion or orbital cellulitis. It is exceedingly rare for orbital cellulitis to cause isolated upper lid ptosis without any other associated neuro-ophthalmological findings or orbital signs such as chemosis, limited motility, proptosis, or afferent pupillary defect.

Orbital cellulitis is a rather commonly encountered pathology in the pediatric population and delineating between preseptal and orbital cellulitis is critical to determine diagnostic studies and management. Findings on the history and physical examination should heighten the suspicion for the location of the infection. Preseptal cellulitis more often occurs in young children and following minor trauma, while orbital cellulitis more often occurs in older children, concomitantly with acute sinusitis. The hallmark features of orbital cellulitis include fever, diffuse bulbar chemosis, diplopia due to ophthalmoplegia, and proptosis.[2] Herein, we report a unique presentation of orbital cellulitis with isolated left upper eyelid ptosis in a 9-year-old boy with pansinusitis and otitis media. The collection and evaluation of protected patient health information was HIPAA-compliant.

2. Case report

A 9-year-old boy with no past ocular history and with a past medical history of long QT syndrome on daily nadolol (Corgard) presented to an outside hospital Emergency Department (ED) with a chief complaint of left upper eyelid (LUL) drooping, left brow pain, and nasal congestion for 3 days. He was seen at his pediatrician's office 3 days prior and was started on a course of amoxicillin for a right ear infection. The vital signs revealed a temperature of 98 °F (36.7 °C), a pulse of 66, a respiratory rate of 20; the blood pressure was 118/68 with an oxygen saturation of 98%. The physical exam was significant for a LUL ptosis and bilateral opaque middle ear effusions, worse on the right than the left. Complete blood count (CBC) with differential and chest X-ray were unremarkable. A computed tomography (CT) of the head without contrast was performed and was negative for any acute intracranial process. It did show, however, significant paranasal sinus disease including involvement of the sphenoid sinus and ethmoid air cells, bilaterally. After receiving a dose of intravenous ceftriaxone, the patient was transferred to the University of Virginia (UVA) ED due to unexplained ptosis.

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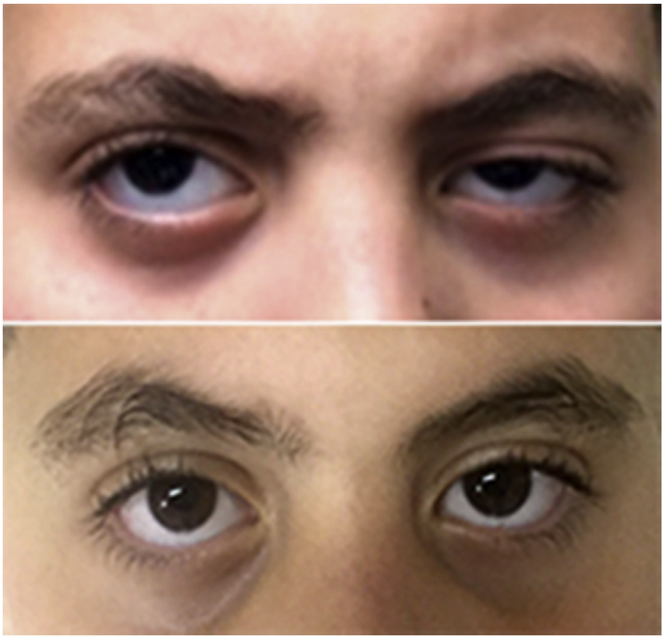

Fig. 1. Patient Photographs: Photograph at initial presentation depicting 3 mm of left upper eyelid (LUL) ptosis (upper frame). Photograph sent from the father a few weeks after completing antibiotics showing resolution of ptosis (lower frame).

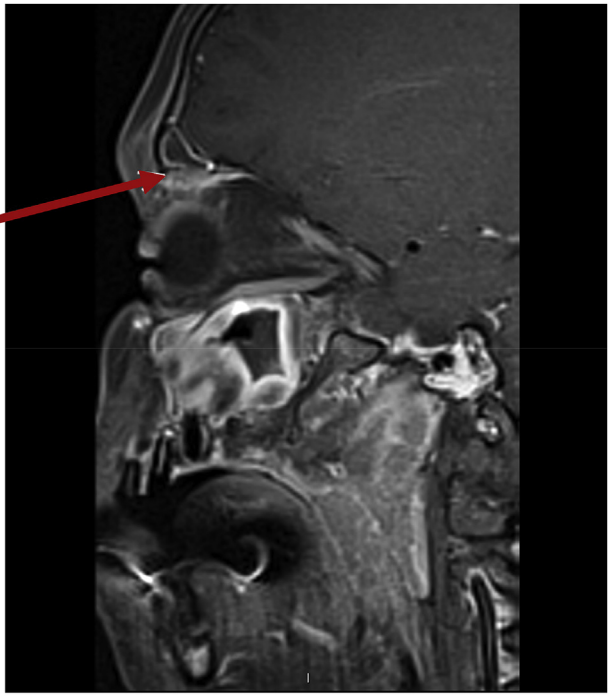

Fig. 3. Sagittal T1 Image: Sagittal T1 post contrast image with fat saturation through the left orbit demonstrating frontal sinus mucosal thickening with enhancing inflammation extending through the bony orbital roof into the superior extraconal space (arrow).

The Ear, Nose, and Throat (ENT) examination revealed a bulging and hyperemic right tympanic membrane with purulent fluid in the middle ear space. The left tympanic membrane was clear and intact with normal anatomic landmarks. No evidence of orbital cellulitis or abscess was detected. The CT performed previously was interpreted at UVA similarly emphasizing “right acute otitis media and pansinusitis with no signs of preseptal or orbital cellulitis”. A dose of intravenous Unasyn was given in the ED and the patient was prescribed oral Augmentin for 14 days; nasal saline irrigations qid and Flonase bid were also recommended.

The patient was then referred to UVA Ophthalmology Clinic 4 hours after his ENT examination due to unexplained LUL ptosis (Fig. 1). His uncorrected visual acuity was 20/25 OU. The pupils were equal in light and dark with no afferent pupillary defect which was confirmed by 2 ophthalmology residents and faculty on duty. The intraocular pressure (IOP) by rebound tonometry was 21 mm/Hg OU. The visual fields were full to confrontation bilaterally. He had full motility in both eyes with no strabismus. He was found to have at least 100 seconds of stereopsis. On external exam, no periorbital edema, erythema, or warmth was detected but he had mild tenderness over his left brow area. The palpebral fissures were 9 and 6 mm OD and OS respectively. Margin-reflex distance test 1 (MRD1) was 4 mm OD, 1 mm OS. The levator functions were 17 and 16mm in OD and OS respectively. There was no lagophthalmos. By Hertel exophthalmometry, axial global protrusions were symmetrical at 17 mm OU at a base of 120 mm. The eyelids, lashes, and lacrimal system were normal on the right side. On the left, there was LUL ptosis but no eyelid or periorbital edema, erythema, or warmth. Biomicroscopically, the conjunctiva and sclera were normal with no injection or abnormal pigmentation OU. The remaining anterior segment examination, dilated indirect ophthalmoscopy, and neuro-ophthalmic examination did not reveal any pathology.

The patient had symmetric pupil sizes and reactions with full motility, making Horner's syndrome or a CN III palsy unlikely. By examination and CT he had pansinusitis and an ear infection, but the primary involvement was on the right side. ENT examination showed a “clear and intact left tympanic membrane with normal anatomic landmarks.” Moreover, he had no clinical evidence of orbital inflammation. A magnetic resonance imaging (MRI) study was requested

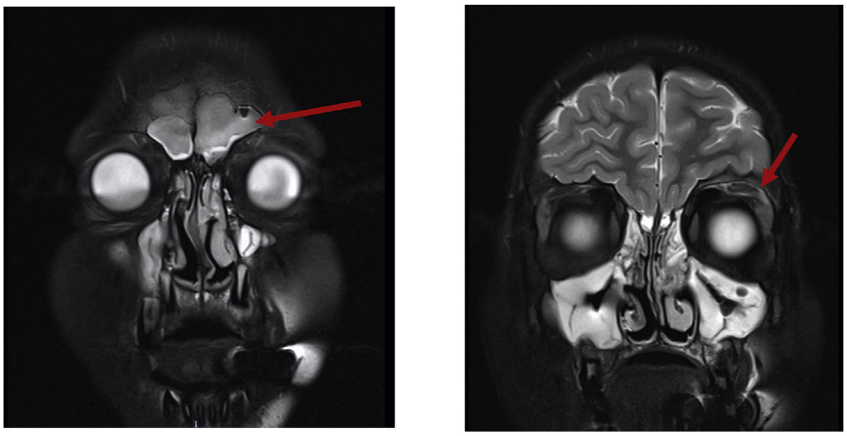

Fig. 2. Coronal T2 Images: Coronal T2 image (left) with fat saturation through the frontal sinuses demonstrating mucosal thickening and fluid opacifying the sinuses (arrow). Coronal T2 image (right) with fat saturation demonstrates extensive maxillary and ethmoid mucosal thickening and edema involving the superolateral extraconal space of the left orbit (arrow).

to rule out an unexpected space-occupying lesion or subclinical evidence of orbital inflammation such as localized subperiosteal inflammation.

MRI orbits with and without contrast depicted pansinusitis with involvement of the extraconal space in the superior aspect of the left orbit. There was mild marrow edema and enhancement in the left orbital bony plate which was reported to possibly represent reactive changes or early osteomyelitis. There was minimal dural enhancement adjacent to the posterior wall of the left frontal sinus also possibly representing reactive change. Contrast enhancement revealed fluid in the mastoid air cells and middle ears, and was suspicious of otomastoiditis bilaterally.

The patient failed to keep his follow-up appointments. However, we were able to interview the patient and the father over the phone in detail and learned that the patient was doing well with resolved LUL ptosis after completing his course of oral Augmentin. A photograph (Fig. 1) was sent to us confirming the resolution of the ptosis.

3. Discussion

There are not many cases reported in the literature describing isolated ptosis from orbital cellulitis with no ophthalmoplegia, or pupillary involvement, and no obvious mechanical cause from inflammation. Coker et al. described a case in 1996 of an adolescent with 5 mm of LUL ptosis without lid edema, erythema, ophthalmoplegia, or pupillary disturbance.[3] The authors reviewed the pertinent anatomy of the course of the superior ramus of CN III after it penetrates the orbit through the superior orbital fissure, and based on the absence of the fat signal around the superior muscle complex on coronal views of CT images, they concluded that the nerve/muscle complex was compromised due to “localized inflammation or congestion”. Although possible, this does not seem to be very likely considering the close proximity of the inferior ramus which innervates the medial and inferior recti, the inferior oblique, and sends parasympathetic fibers to the pupillary constrictors and all of these functions were normal in their patient. Another interesting feature of the Coker and Ros paper was the rapid resolution of the LUL function which was described as “5 mm of left eye ptosis”. Of course, the authors were at the mercy of rather limited CT resolution in 1996. With the advances of imaging over the last 20 years, our report is able to reveal high-resolution MR images highlighting the pathology in the orbit as described below.

Our case describes a unique presentation of orbital cellulitis as isolated ptosis in the absence of orbital signs and symptoms including ophthalmoplegia, pain with eye movements, bulbar chemosis, proptosis, and an afferent pupillary defect (APD) in some cases.[2] Most non-traumatic cases of orbital cellulitis in this age group are secondary to rhinosinusitis. Ethmoid sinusitis is the most common cause, being found in 86–98% of cases due to contiguous spread via the lamina papyracea.[4] Otitis media is an infrequent, yet notable, origin for orbital cellulitis.[5] In both scenarios, rhinosinusitis and otitis media, orbital cellulitis most often develops ipsilateral to the site of infection. Although rare, cases of bilateral orbital cellulitis in the absence of cavernous sinus thrombosis have been reported, all of which were associated with subperiosteal abscesses; however, contralateral disease without ipsilateral involvement was not found upon reviewing the literature.[6] Our patient developed ptosis in the setting of bilateral pansinusitis but contralateral to the site of otitis media, although, later in the course of the patient, the involvement of the left ear with infection was also documented.

Orbital cellulitis, particularly in adults can have significant consequences, including loss of sight and even loss of life.[7] Therefore, early diagnosis is essential and is facilitated by features of the history and physical examination and confirmed with imaging. Both CT scan and MRI are suitable modalities for investigating orbital cellulitis, but in children, MRI should be preferred because of the lack of radiation to the skull and brain.[8]

Current recommendations for imaging include: proptosis, ophthalmoplegia, pain with eye movements, limitation of eye movements, diplopia, vision loss, APD, edema extending beyond the eyelid margin, marked bulbar chemosis, absolute neutrophil count (ANC) > 10,000 cell/microL, signs or symptoms of central nervous system (CNS) involvement, inability to examine the patient fully, and patients who do not begin to show improvement within 24–48 hours of initiating appropriate therapy.[9] In our case, initial imaging would not have been indicated with these guidelines. However, it was elected to be done because of the unusual presentation of the patient with pupil-sparing LUL ptosis on the contralateral side. When considering which factors might contribute to such a focal finding without more prominent inflammatory orbital signs, timing of patient presentation likely played a role. The inflammation had just begun to invade the orbit directly from the frontal sinus. Had our patient been left untreated with a later presentation, he may have demonstrated more classic and severe signs of orbital infection.

Superior orbital fissure syndrome is reported in the literature but is rare, especially with pupil-sparing ptosis in orbital cellulitis. Infectious involvement in the posterior orbit can affect structures that cross the anatomic region near the superior orbital fissure.[10] As it was briefed in the Coker and Ros paper, this foramen transmits many important structures including the lacrimal, frontal, and nasociliary branches of CN V-1, the superior and inferior divisions of the oculomotor nerve, the trochlear nerve, the abducens nerve, sympathetic fibers from the cavernous plexus, and the superior ophthalmic vein. Isolated superior division palsy of the oculomotor nerve is reported which causes ptosis and limited upgaze due to disruption of innervation to the levator and superior rectus, respectively.[11] Bodily et al. describe a case of invasive Streptococcus viridans sphenoethmoiditis leading to an orbital apex syndrome in an immunosuppressed older female with acute myelogenous leukemia.[12] On careful review of the imaging in our case, there is no superior fissure inflammation at the orbital apex. Rather, it shows a direct extension of inflammation into the soft tissues of the LUL, affecting the levator superioris but with relative sparing of the superior rectus. This explains why our patient had such dramatic ptosis but with preserved ocular motility and no other signs or symptoms of orbital cellulitis. This demonstrates that the ptosis was actually mechanical in nature but only visible through imaging with no external evidence of inflammation clinically.

Another possibility that should be on the differential diagnosis when evaluating these patients is myositis. Court and Janicek reported a case of isolated levator palpebrae myositis.[13] This can present very similarly to the way our patient did with unilateral ptosis in the absence of ophthalmoplegia, pain with eye movements, and APD. However, it is more often found in the older population and is rare. Additionally, imaging reveals isolated inflammation of the levator palpebrae superioris muscle in myositis without the contiguous extension of inflammation from the frontal sinus as in our case. Arat et al. described 5 patients with transient, acute, unilateral blepharoptosis of unknown etiologies.[14] Only 2 of the patients had imaging of the orbits and both revealed no pathology. All of their patients had flu-like illnesses preceding their episodes of ptotis, and the authors postulated that the viral infection could be an etiologic factor that could affect “the terminal branch of the oculomotor nerve innervating the levator muscle, neuromuscular junction, or the muscle itself.” Our case was quite different in that the infectious etiology was presumed to be bacterial, and there was imaging revealing the pathology in the orbit.

Thin section, multi-planar MR imaging of the orbits is capable of delineating the extent of sinus disease and orbital involvement in exquisite detail, particularly with the use of fat saturated sequences, while avoiding the use of ionizing radiation. The opacified and inflamed frontal sinuses (Fig. 2) with edema and inflammation extending through the bony orbital roof into the superior left extraconal space (Figs. 2 and 3) are readily apparent in this case. More posterior images demonstrate sparing of the orbital apices and optic nerves.

4. Conclusions

As a summary, this case demonstrates an unusual presentation of orbital cellulitis that may have implications for clinical decision-making for isolated upper lid ptosis in the pediatric population. With new onset, unilateral ptosis in the setting of acute sinusitis and/or otitis media, diagnostic imaging studies, such as CT scan and MRI, may be beneficial to pursue early in the management in certain cases. Despite concerns for radiation exposure in the pediatric population, the potentially detrimental outcomes from overlooking orbital cellulitis should warrant further investigation with appropriate imaging.[15] This patient also serves as a reminder that neurogenic upper lid ptosis may develop ipsilaterally due to orbital cellulitis and sinusitis if the inflammation is located near the orbital apex, but it can also manifests itself following other mechanisms as it is shown in our case. It highlights the importance of maintaining a high level of suspicion in cases of suspected orbital cellulitis even in the absence of typical signs and symptoms.

Patient consent

The patient's legal guardian consented to publication of the case orally. This report does not include any personal information that could lead to the identification of the patient.

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Conflicts of interest

All authors have no financial disclosures.

Authorship

All authors attest that they meet the current ICMJE criteria for Authorship.

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