CASE REPORT

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Orbital subperiosteal hematoma associated with sinus infection*

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SUMMARY

Objectives: *There are few reports of orbital subperiosteal hematomas as a result of sinusitis complications. The present study reports on 2 such cases and presents a literature review regarding this condition.*

Methods: *We report 2 cases of orbital subperiosteal hematoma, and reviewed previous literature reports.*

Results: *Including the 2 current patients, 11 cases of orbital subperiosteal hematoma involving sinusitis appear in the literature. The current cases involved 2 older females presenting with proptosis. One had frontal sinusitis and the other a postoperative frontoethmoid mucocele. Both patients underwent a superior orbitotomy and sinus surgery, which resolved the orbital symptoms.*

Conclusion: *Orbital subperiosteal hematomas can develop associated with sinus infection. Such a condition should be treated as a sinusitis complication and the associated sinus infection must be treated concomitantly.*

Key words: hematoma, complication, sinusitis

INTRODUCTION

Orbital subperiosteal hematomas occur infrequently. While usually associated with trauma, they can also be associated with sinus infection, albeit rarely [1,2]. Seven cases of orbital subperiosteal hematoma as complications of paranasal sinusitis have been reported [2-8], as have 2 cases associated with a paranasal sinus mucoecele [7,9]. Of the reported types of orbital complications of sinusitis, none have mentioned a subperiosteal hematoma. While orbital subperiosteal hematomas occur rarely as a complication of sinus infection, they can result in more severe complications such as blindness [10]. Orbital subperiosteal hematoma associated with sinusitis should be treated as a complication of sinusitis.

The present report describes 2 cases of orbital subperiosteal hematoma associated with sinus infection: one associated with sinusitis and the other with paranasal sinus mucocele. In addition, we identify another 9 such cases that have been reported previously [2-9]. We review the literature and discuss the pathogenesis, diagnosis and treatment of this disease.

CASE REPORTS

Case 1

A 57-year-old female arrived at the emergency room and was admitted with right periorbital swelling and proptosis. A severe headache in the right frontal area was experienced 2 days prior to admission, and proptosis rapidly developed the day before arrival at the clinic. There were no diplopia symptoms, vision was good, and there was no fever. She experienced the symptoms of an upper respiratory infection (URI) 10 days before admission. There were no signs of rhinorrhea or nasal obstruction, and no history of trauma or sinus surgery. However, 3 years previously the patient had undergone external drainage due to a right orbital subperiosteal hematoma at another ophthalmology clinic. Review of the previously taken computed tomography (CT) images showed combined right frontal sinusitis which had not been treated at the time. Following the previous surgery, a lacunar infarction occurred and aspirin medication was initiated. The patient also had hypertension.

We found the patient to have a best corrected visual acuity of 0.8/0.8, full extraocular muscle movements in 6 cardinal directions, and prompt light reflexes. There was no chemosis or conjunctival injection, and rhinological examination showed no specific abnormality. A complete blood cell count showed no abnormality, with a white cell concentration of 5900/mm3. The coagulation profile and blood glucose levels were normal. C-reactive protein (CRP) level and erythrocyte sedimentation rate (ESR) were within normal ranges.

Contrast-enhanced computed tomography (CT) revealed a well-demarcated biconvex non-enhancing soft tissue density

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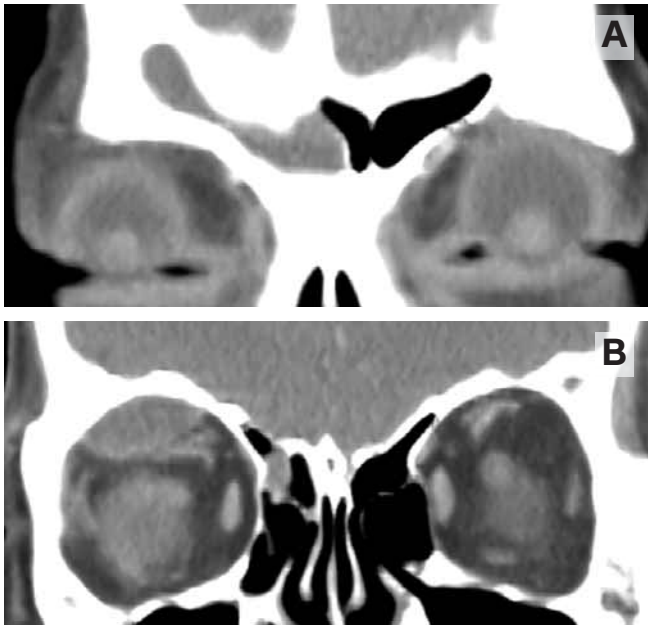

Figure 1. Case 1. (A) Contrast-enhanced computed tomography showing a clouded right frontal sinus and (B) a well-demarcated biconvex non-enhancing soft tissue density lesion in the right supraobital area.

lesion in the right supraorbital area (Figure 1). The right frontal sinus and a part of the ethmoid sinus were clouded.

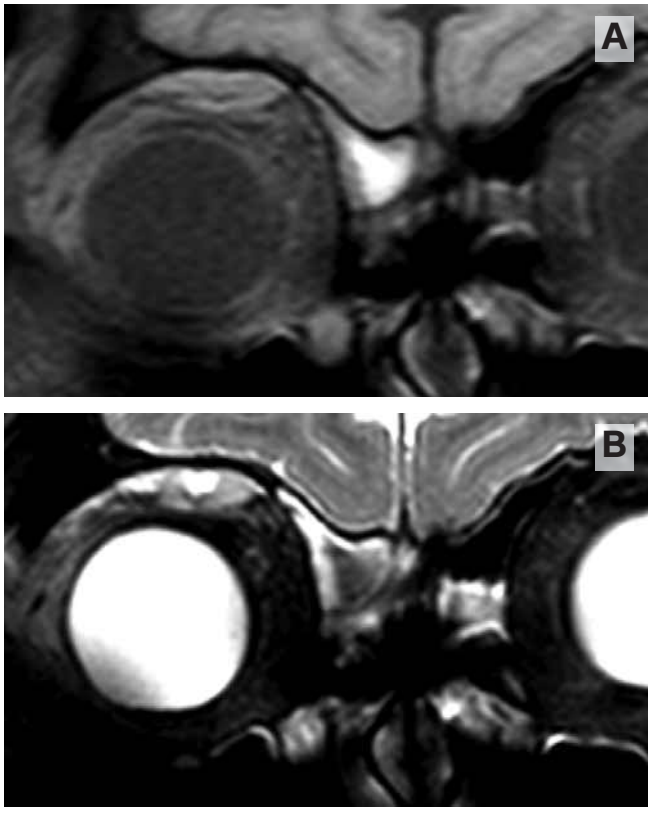

Figure 2. Case 1. (A) Magnetic resonance imaging. T1-weighted image showing a high-to-intermediate signal intensity lesion in the right supraorbital area. (B) T2-weighted image showing a high signal intensity. Right frontal sinusitis is also apparent.

There were no bony defects in the orbital roof or frontal sinus wall, and no connection between the orbital lesion and the frontal sinusitis. Magnetic resonance (MR) imaging T1-weighted images showed a high-to-intermediate signal intensity lesion in the right supraorbital area, and T2-weighted images showed a high signal intensity. Right frontal sinusitis was observed (Figure 2).

Our initial diagnosis was a subperiosteal abscess associated with frontal sinusitis. Ampicillin/sulbactam was administered intravenously. While this treatment slightly improved the eye symptoms, CT imaging showed an increase in the size of the supraorbital lesion. Endoscopic frontal sinus surgery and a superior orbitotomy via an eyebrow incision were performed. Purulent discharge was drained from the right frontal sinus and samples sent for laboratory culture. Following incision of the periosteum and elevation of the periorbita from the superior orbital wall, a brown serous discharge was drained from the right subperiosteal space. The periorbita was intact and there appeared to be no connection with the frontal sinus. The hematoma was completely evacuated and a silastic drain inserted. Proptosis improved following surgery. A CT on postoperative day 13 showed a remaining half-sized orbital lesion, but no sinusitis. The patient was discharged on postoperative day 20 without symptoms. No microorganisms grew in cultures of either the orbital or frontal sinus discharges. The patient underwent revision endoscopic frontal sinus surgery after 2 months due to a re-obstructed frontal sinus ostium. CT images at that time showed recurrent frontal sinusitis but no orbital hematoma. Following that surgery, the patient was free of symptoms and showed no remaining disease.

Case 2

A 62-year-old female presented complaining of left eyeball pain and proptosis for 15 days. She had a headache, and diplopia that had begun 8 days previously. She was afebrile, did not display rhinorrhea or nasal obstruction, and had no history of URI or trauma. Five years prior she had undergone endoscopic sinus surgery due to chronic sinusitis. There was no specific medication history and no history of systemic disease such as diabetes, hypertension or vascular disorders.

Hertel's exophthalmometry demonstrated a 5 mm exophthalmos in the left eye, and there was marked upgaze restriction of ocular movement in the same eye. Best corrected visual acuity was 0.6/0.4, and light reflexes were prompt. Funduscopic examination showed an elevated retinal lesion in the superonasal aspect possibly due to a mass lesion in the left eye. Nasal cavity examination revealed no specific abnormality. A complete blood cell count showed no specific abnormality, with a white cell concentration of 5400/mm^3. CRP and ESR were within the normal ranges, as were the coagulation profile and blood glucose levels.

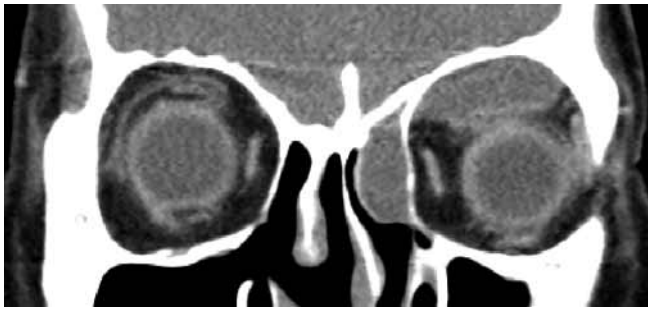

Figure 3. Case 2. Contrast-enhanced computed tomography showing a well-demarcated low density non-enhancing lesion in the left supraorbital area. A left frontoethmoid mucoeceleis apparent.

Contrast-enhanced CT revealed a low-density non-enhancing lesion in the left supraorbital area (Figure 3). The lesion was well demarcated at the inferior margin and was considered to be in the subperiosteal area. A cyst-like lesion considered a postoperative mucoecele was noted in the left anterior ethmoid sinus, revealing a bony change. There was no connection between the supraorbital lesion and the mucocele. Obstructive sinusitis was observed in both frontal sinuses. A subperiosteal abscess due to the left frontoethmoid postoperative mucoecele was suspected and endoscopic surgery was performed. The left frontoethmoid mucoecele was marsupialized and the pus discharge drained. A superior orbitotomy via an eyebrow incision was performed and a brown serous discharge was drained (Figure 4). The periorbita and orbital bony wall were intact. A tube was positioned to drain the remaining hematoma. The proptosis and eyeball pain were relieved following surgery. Staphylococcus epidermidis grew in cultures of the frontoethmoid mucoecelefluid, while no microorganisms grew from the orbital fluid. A CT on postoperative day 8 revealed the orbital lesion had decreased in size. The patient was discharged on postoperative day 9 with improved symptoms. By postoperative day 14, exophthalmos had completely subsided and there was full extraocular movement. The patient was followed postoperatively for 8 months during which time there was no evidence of disease recurrence.

DISCUSSION

Orbital subperiosteal hematomas can be divided into traumatic and non-traumatic types, with most being the former [1,3,7]. Such hematomas occasionally follow orbital injury, and occur due to mechanical rupture of small vessels under the periosteum [3]. Non-traumatic orbital subperiosteal hematomas have been associated with sudden elevations in cranial venous pressure and venous congestion (violent coughing, vomiting, straining, weight lifting and labor) [1,11,12], systemic diseases associated with bleeding tendencies (coagulopathy with liver disease, scurvy) [13,14] and paranasal sinusitis [1]. Occasionally, no associated cause has been found [1]. Orbital subperiosteal hematomas associated with sinusitis occur very rarely, and are believed to be a sinus infection complication [1,2].

Sinus infections can extend to adjacent structures via the venous system, moving between the nose, paranasal sinuses, orbits and cavernous sinuses [1-4,7]. The venous system lacks valves allowing infections to more easily spread to the orbit [1-3,12]. In the orbit, veins are considered to have the major role in the spread of infections, rather than arteries, lymphatics or bony dehiscences [1,3]. Most orbital hemorrhages are reported to occur due to the extension of phlebitis [1-3,7]. Harris et al. [2] stated that phlebitis in the sinus mucosa may extend to veins in the periorbita, resulting in subsequent rupture of the vessels. Vessel rupture may result from congestion or erosion of the vessel due to infection [3].

Including the two present cases, there are only 11 reported cases of orbital subperiosteal hematomas associated with sinusitis [2-8] or paranasal sinus mucoecele [7,9] (Table 1). These involved 8 females and 3 males with a mean age of 46.5 years (range, 4 to 68 years). It was reported that the cases of the non-traumatic type of orbital hematoma were nearly all female [1]. But these reported series involved three male patients of 11.

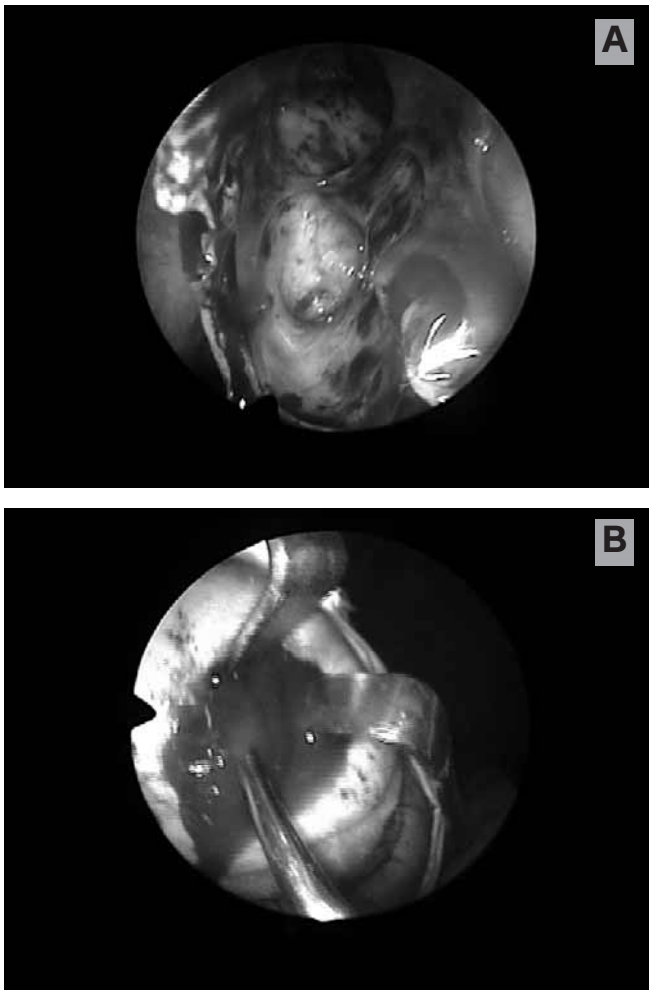

Figure 4. Case 2. (A) Intraoperative photograph showing the frontal sinus opening after marsupialization of the frontoethmoid mucocele. (B) Brown serous discharge drained from the superior orbitotomy site.

Four of the 11 reported patients were right-sided and 7 left-sided. 5 of the subjects had antecedent histories of upper respiratory tract infection (URI) (including one patient in the present report), suggesting inflammation may initiate hematoma development. Two patients had undergone prior Caldwell-Luc surgery, while one of the present patients had undergone previous endoscopic sinus surgery. Most patients had frontal or ethmoid sinusitis in the same side as the orbital hematoma. Three patients had frontoethmoid mucoeceles on the lesion side and one of these (the present case 2) had a postoperative mucoecele after endoscopic sinus surgery. Two mucocoele cases [7] had no connections between the mucoecele and hematoma despite bony dehiscence, suggesting the hematoma may be caused by phlebitis but not direct extension of inflammation. However, one frontoethmoid mucoecele case [9] had a connection between the frontal sinus and the subperiosteal hematoma through a bony orbital roof erosion, in addition to a frontal sinus hematoma. In that case, direct extension of the frontal sinus hemorrhage due to a frontoethmoid mucoecele might have caused the subperiosteal hematoma. A craniotomy was performed and the frontal sinus hematoma and orbit were evacuated using a subfrontal extradural approach.

In the present report, the first patient had an aspirin medication history that did not appear to cause hematoma development. At the time of undergoing her first operation at another clinic, the frontal sinusitis and hematoma existed concomitantly. Even though frontal sinusitis might initiate development of an orbital hematoma, hematoma evacuation but not frontal sinusitis treatment was carried out at that time. Although this treatment improved her orbital symptoms, frontal sinusitis persisted, and subsequently a lacunar infarction occurred initiating aspirin medication. On arrival at our clinic the patient showed evidence of both frontal sinusitis and a hematoma. We believe that the initial frontal sinusitis persisted and caused another hematoma, and that aspirin medication had little effect. While other reports do not indicate patients had a history of aspirin or antiplatelet medication, it can be assumed that such drugs might also cause hemorrhage [1].

Others report that the most common site of non-traumatic orbital subperiosteal hematoma is the superior orbital wall [1,3], and this was the case with all the reported series. This site may be common due to the frontal sinus providing the largest bony surface on the orbit [3] and there being some irregular diploic vessels connecting the frontal sinus and the subperiosteal space [1-3]. Such diploic vessels may be the source of hemorrhage. Other authors [15] suggest that loose adhesion between the periosteum and orbital bones allows for hemorrhage development. The orbit periosteum is loosely attached to the underlying bone except around the orbital fissures, the optic canal, the orbital margin, suture lines, and foramina for blood vessels and nerves that cross the subperiosteal space [1,15]. The orbital roof, being the widest area in which the periosteum is not interrupted by firm adhesions to the bony orbit, is the most common site for development of spontaneous hemorrhage [1,15]. This might be the cause of frequent hematoma occurrence in the superior orbital wall [1,15].

In most reported cases, symptom onset is sudden [2-9], proptosis, diplopia and ocular pain are common [3,4], and the globe is usually displaced downward and laterally [3,16]. These symptoms are caused by the mass effect of an orbital lesion [16]. Sudden development of proptosis may indicate the presence of hematoma [2]. Other signs such as periorbital swelling, restriction of ocular movement and discoloration of the eyelids or conjunctiva are also commonly observed [3,4]. Severe loss of vision is rare, and the visual prognosis after orbital hemorrhage is reported to be excellent [3,4]. However, prompt action is important as optic nerve involvement can occur if the condition is left untreated and can lead to loss of vision and other complications [10]. While these symptoms and signs are similar to those of abscesses, they lack the inflammation characteristic of abscesses [2,3,8,9]. The two present patients did not show fever, marked leukocytosis or elevated CRP or ESR, similar to other reported cases [2,3,8,9].

While contrast-enhanced CT is useful for examining intraorbital lesions, it does not differentiate between hematomas and abscesses. MR can aid in discriminating hematomas from abscesses [18]. The CT definition of an acute orbital subperiosteal hematoma is a biconvex, well-defined, non-enhancing mass lesion of homogeneous density slightly higher than that of the brain [18]. CT can also reveal the inferior displacement of the orbital contents and associated sinusitis or mucoecele. There are usually no connections between hematoma and sinus disease.

In the present case 2, the T1-weighted MR image showed a high-to-intermediate signal intensity and the T2-weighted image showed a high signal intensity (Figure 1). These findings were compatible with a subacute-chronic (from 7 days to weeks) stage of hematoma [18]. The MR definition of a subperiosteal hematoma is a biconvex, well-defined mass of varied signal intensity, depending on the age of the hematoma. It is best seen in coronal and oblique sagittal sections. The T1-weighted images will show low signal in the hyperacute stage (fresh blood) and high signal in the subacute stage (3-7 days, before cell lysis). The T2-weighted images will show a high signal in the hyperacute stage and a low signal in the subacute stage. Both T1- and T2-weighted images will show high signal in the subacute to chronic stage (7 days to weeks, after cell lysis) and low signal in the chronic stage (months to years, organized hematoma). These differences between MR images are assumed to be the result of the accumulation of different hemoglobin metabolic products and the effect of osmotic fluid shift by the blood clot [18].

In the present case 1, neither orbital hematoma nor sinus fluid

Table 1. Previously reported cases of orbital subperiosteal hematoma associated with sinus infection.

Author, Year	Sex	Age	Sinus surgery	URI history	Proptosis (mm)	Vision (R, L)	Location	Site (in orbit)	Treatment	Associated sinusitis	Sinus bony wall	Culture (sinus, orbit)
Wheeler, 1937[3]	M	29	N	N	8	WNL	R	superior	Superior orbitotomy	Frontal sinusitis	NA	NA
Harris, et al, 1978[2]	F	30	N	N	10	20/20 20/30	L	superior	Lateral orbitotomy, external frontal sinusotomy	Frontal sinusitis	intact	*Eubacterim* sp, *Eubacterim* sp
Leopold, et al, 1980[4]	F	56	N	Y	4	20/15, 20/15	L	superior	Frontal sinus trephination, external fronto-ethmoidectomy	Frontal, maxillary sinusitis	NA	No organism, no organism
Ichino, et al, 1985[5]	F	57	C-L op (5 yrs)	N	7	0.6, 0.5	L	superior	External fronto-ethmoidectomy	Ethmoid, frontal sinusitis	intact	NA
Choi, et al, 1988[6]	M	62	N	N	6	20/70, 20/70	R	superior	External ethmoidectomy	Supra-orbital Infection	intact air cell	*S. epidermidis* *S. epidermidis*
Zalzal, 1991[7]	F	4	N	Y	NA	20/70, 20/50	R	superior	External ethmoidectomy	Ethmoid, maxillary sinusitis	intact	NA (maxillary sinus aspirate; *M. catarrhalis)*
Woo, et al, 1997[8]	F	44	N	Y	NA	20/20, finger count before 20cm	L	superior orbitotomy, Endoscopic	Superior ethmoid mucocele marsupialization	Fronto-medial wall defect	Orbit no organism	No organism,
	M	42	C-L op (10 yrs)	N	11	20/20, 20/40	L	superior	Superior orbitotomy, Endoscopic ethmoidectomy and maxillary antrostomy	Ethmoid, maxillary sinusitis	intact	*S. epidermidis,* no organism
Aoki, et al, 1997[9]	F	68	N	Y	NA	NA	L	superior	Subfrontal extradural approach	Fronto-ethmoid mucocele	Frontal sinus floor eroded	NA
Our case #1	F	57	N	Y	NA	0.8/0.8	R	superior	Superior orbitotomy, Endoscopic frontal sinusotomy	Frontal sinusitis	intact	No organism, no organism
Our case #2	F	62	Y	N	5	0.6/0.4	L	superior	Superior orbitotomy, Endoscopic marsupialization	Fronto-ethmoid mucocele	intact	*S. epidermidis*, no organism

M = male; F = female; Y = yes; N = no; C-L op = Caldwell-Luc operation; yrs = years; R = right; L = left;
WNL = within normal limit; NA = not available
Eubacterim sp = *Eubacterim* species; *S. epidermidis* = *Staphylococcus epidermidis; M. catarrhalis* = *Moraxella catarrhalis*

cultures grew microorganisms, possibly due to the use of antibiotics. In case 2, *Staphylococcus epidermidis* grew in cultures of the frontoethmoid mucoecelefluid, and no microorganisms in orbital fluid cultures. Other reports [2,3,5,7] showed that cultures of both orbital and sinus contents usually grew the same microorganisms, most commonly *Staphylococcus epidermidis*. Some reports showed no growth [5,7] and one report identified growth of anaerobes [2] in the sinus and orbit culture despite preoperative intravenous antibiotic therapy.

The three most likely treatment options for orbital hematoma are waiting for resolution, needle aspiration, and surgical exploration [1-4,7]. Waiting was not a suitable option for the present cases as we believed there was some risk of optic nerve damage. Needle aspiration was also not viable as sinus infections cannot be treated in such a manner. In most cases, surgery is preferable in order to evacuate both the hematoma and sinusitis simultaneously [2-4,7]. Surgery can also confirm the diagnosis [3,4]. In the present cases, the hematomas were drained using external approaches involving an incision around the eyebrow and a superior orbitotomy. While this approach can evacuate the hematoma under direct vision by the surgeon and more completely than other approaches [7], a scar around the eyebrow may remain. The treatment methods used in other reported cases are summarized in Table 1. These generally involved external frontoethmoidectomy [3-5,8], and orbitotomy with sinus surgery [2,7] such as endoscopic marsupialization, although one case [9]. involved a transcranial approach.

In conclusion, orbital subperiosteal hematomas can develop as an orbital complication of a sinus infection. Such a hematoma should be treated as a sinus infection complication and the infection should be treated simultaneously. Clinicians must be aware of the possibility of orbital hematomas when presented with sinus infection patients, and should treat these patients accordingly.

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