

Idiopathic Recurrent Bilateral Orbital Emphysema: A Case Report

Burak Simsek, Hakan Yildirim, Hamdi Tasli

Burak Simsek, Hakan Yildirim, Birecik Government Hospital Department of Ophthalmology, Sanliurfa, Turkey.

Hamdi Tasli, Birecik Government Hospital Department of Otorhinolaryngology, Sanliurfa, Turkey.

Conflict-of-interest statement: The author(s) declare(s) that there is no conflict of interest regarding the publication of this paper.

Open-Access: This article is an open-access article which was selected by an in-house editor and fully peer-reviewed by external reviewers. It is distributed in accordance with the Creative Commons Attribution Non Commercial (CC BY-NC 4.0) license, which permits others to distribute, remix, adapt, build upon this work non-commercially, and license their derivative works on different terms, provided the original work is properly cited and the use is non-commercial. See: <http://creativecommons.org/licenses/by-nc/4.0/>

Correspondence to: Burak Simsek, MD, FEBO, Birecik Government Hospital Department of Ophthalmology, Sanliurfa, Turkey.

Email: drburaksimsek@gmail.com

Telephone: +904146521083

Received: November 11, 2017

Revised: January 20, 2018

Accepted: January 23, 2018

Published online: February 27, 2018

ABSTRACT

A 17-year old female applied with acute bilateral swelling in her eyelids with no trauma history or previous medical disorder. She mentioned in last 9 months, this situation occurred very often: Acute onset of bilateral swelling and spontaneous recovery in a few days. Severe bilateral periorbital swelling and subcutaneous crepitations without ecchymosis was seen. Body temperature, eye movements, reactions of pupils in light were normal. No proptosis and diplopia were noted. Visual acuity (VA) in right eye could not be determined but the VA of left eye was full. Anterior and posterior segments were seen normal. In orbital computerized tomography (CT) bilateral intraorbital and preseptal emphysema were remarkable. No fracture line was detected. The patient was accepted as preseptal cellulitis and treatment was arranged. After 3 days it was seen that the swelling

in left eye was lessened and interpalpebral aperture was noticeable. However right eye's condition was the same. Control orbital CT was also same. Clinical partial recovery in left eye was accepted significant and present treatment was maintained. In second control visit, no further recovery findings were remarkable. Routine blood tests were normal except for immunoglobulin E (IgE). Most common reasons of orbital emphysema are severe sneezing, nose blowing or occult minor periorbital traumas. But in this case none of these had happened. Nontraumatic orbital emphysema is a benign disorder which rarely proceeds with complications and has no response to systemical therapy. Even if it is bilateral and recurrent, because it heals spontaneously, it is appropriate to follow these cases without giving systemical treatment.

Key words: Bilateral; Recurrent; Orbital Emphysema; Orbita; Emphysema

© 2018 The Author(s). Published by ACT Publishing Group Ltd. All rights reserved.

Simsek B, Yildirim H, Tasli H. Idiopathic Recurrent Bilateral Orbital Emphysema: A Case Report. *International Journal of Ophthalmic Research* 2018; **4(1)**: 265-267 Available from: URL: <http://www.ghrnet.org/index.php/ijor/article/view/2192>

INTRODUCTION

Orbital emphysema is a clinical condition defined as the existence of air in orbita. The most common cause is orbital wall fracture secondary to periorbital trauma. Other common reasons are history of sinusitis and surgery. However nontraumatic etiology is quite rare: For instance vigorous nose blowing, sneezing and pressure changes during air travels^[1-3].

Orbital emphysema is generally a mild and self-limiting disorder^[3-7]. Nevertheless it has been reported that it could rarely cause some severe advanced complications and need surgical intervention^[7, 8]. These uncommon and severe complications are occlusion of central retinal artery, compressive optic neuropathy and, most commonly, ischemic optic neuropathy^[9].

In this article we aimed to present a recurrent, bilateral, nontraumatic orbital emphysema case which is not seen in literature before.

CASE REPORT

A 17-year old female applied to our clinic with acute bilateral swelling in her eyelids without itching or secretion. There was no trauma history or previous medical disorder. She mentioned in last 9 months, this situation occurred very often: Acute onset of bilateral swelling and spontaneous recovery in a few days. Additionally she mentioned that she applied several hospitals because of the same reason and despite performing some investigations and treatments there were no change in her clinic.

In physical examination severe bilateral periorbital swelling was seen. No ecchymosis were accompanied. Severe bilateral subcutaneous crepitations were palpable. Body temperature was normal. On superior eyelids' skin there were some ambiguous dots like insect bites. However the patient was denied to be exposed to insect bite. The examination of globes and their functions were quite hard because of the swellings: Eye movements were checked hardly and seen normal. Reactions of pupils in light were also normal. No proptosis and diplopia were noted. Due to the severity of swelling in right eye VA could not be determined but the VA of left eye was full. With several insistent examination attempts in biomicroscopy, anterior and posterior segments were seen normal. No signs of optic nerve or retinal vessel compression was remarkable.

In addition to physical examination only orbital CT was requested. Bilateral intraorbital and preseptal emphysema were remarkable in CT and the emphysema was more prominent in right eye (Figure 1). No fracture line was also detected. The soft tissue density in preseptal area was augmented. Hence the patient was accepted as preseptal cellulitis and treatment was arranged in terms of this diagnosis: Oral ampicillin+sulbactam combination, oral nonsteroidal anti-inflammatory drug (NSAID), topical antibiotics and due to both vague insect bite impression and severe emphysema oral metronidazole were given. Patient was called to control examination 3 days later.

In control visit, it was seen that the swelling in left eye was lessened and interpalpebral aperture was noticeable. However right eye's condition was totally the same as previous visit (Figure 2, 3). Severe subcutaneous crepitations were still palpable. In control orbital CT there were no significant difference in comparison with the previous one (Figure 4, 5). Biomicroscopic examination showed no abnormal findings again. Nevertheless the clinical partial recovery in left eye was accepted significant and it was decided to maintain the present treatment. The patient was called to second control 5 days later.

In second control visit, no further recovery findings were remarkable. According to the recommendation of radiology department, contrasty orbital CT was taken but, again, no further information about the etiology could be aquired. Bone abnormality or fistula were not also detected. Furthermore, the routine blood tests including complete blood count (CBC), sedimentation, C-reactive protein (CRP) of the patient were normal. The only elevated blood parameter was IgE, eosinophil number was normal though.

Because no further clinical recovery, medical history was questioned comprehensively one more time. In the literature, it is pointed out in orbital emphysema case reports that the most common reasons of this situation are severe sneezing, nose blowing or occult minor periorbital traumas. Hence the patient was questioned carefully about these conditions, but she denied all. Moreover no allergic findings or history were present to explain the elevated IgE in blood test.

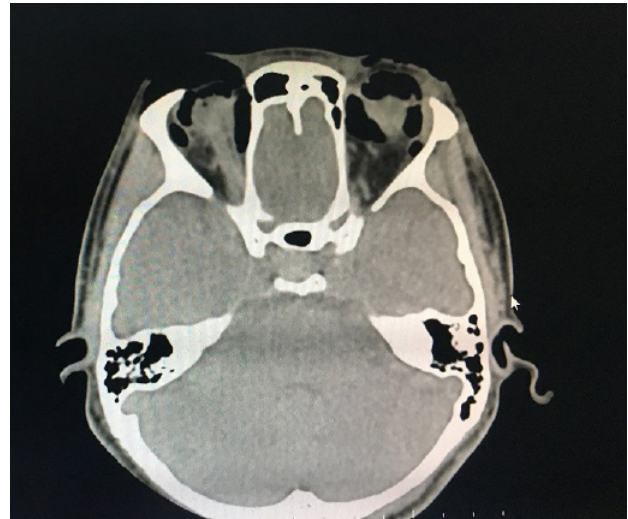


Figure 1 CT image of first visit. Severe air signals in both orbita and preseptal areas are remarkable without bone fracture.



Figure 2



Figure 3

Figure 2-3 Patient's photography in first control day. The swelling in left eye is less than before. However the right eye has no change and palpebral rima is still totally closed.

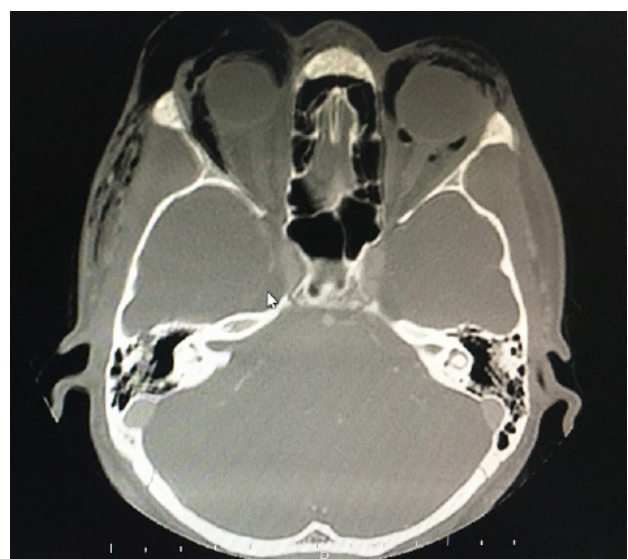


Figure 4 CT image of third control visit. In comparison with previous control no significant difference is remarkable.



Figure 5 CT image of the lacrimal bone section of third control visit.



Figure 6 Patient's photography before discharge. No further recovery findings are remarkable.

Due to no further clinical healing were obtained, systemic steroid therapy was planned under the protective effect of antimicrobial therapy. The patient was hospitalized for parenteral therapy for 3 days. Twice 1 gram of ceftazidime per day (2×1 gr), 2×500 mg metronidazole, 1 mg/kg/day prednol, 2×1 ampula of pheniramine and topical antibiotics and steroid were performed. However after these 3 days, one more time, no significant recovery was remarkable (Figure 6). Therefore the parenteral therapy was terminated and the swelling and emphysema were left spontaneous healing just as the previous recurrences mentioned in medical history. To avoid vigorous sneezing or nose blowing, patient were warned.

DISCUSSION

The most important differences of this case are recurrence and bilaterality in comparison with the other orbital emphysema cases in the literature. There is no similar case reports in databases. However, in terms of response to the treatment, our case is similar to the other orbital emphysema case reports in which systemic treatment are not recommend because it is useless^[3-7].

When orbital CTs of this patient were investigated again comprehensively, it was noticed that ethmoid sinus was hypocellular especially in anterior segments and the lacrimal bones on medial orbital walls had severe dehiscence. This dehiscence is particularly noticeable in Figure 5. However no cut off signs were remarkable. Hence the underlying etiology can be bone structure variation. If a higher quality orbital CT with thinner sections are taken to similar cases, such bone variations can be detected.

CONCLUSION

Nontraumatic orbital emphysema is a benign disorder which rarely proceeds with complications and has no response to systemical therapy. Even if it is bilateral and recurrent, because it heals spontaneously, it is appropriate to follow these cases without giving systemical treatment.

REFERENCES

1. Mohan B, Singh KP. Bilateral subcutaneous emphysema of the orbits following nose blowing. *J Laryngol Otol.* 2001 Apr; **115**(4): 319-20.
2. Shah N. Spontaneous subcutaneous orbital emphysema following forceful nose blowing: treatment options. *Indian J Ophthalmol.* 2007 Sep-Oct; **55**(5): 395.
3. Moon H, Kim Y, Wi JM, Chi M. Morphological characteristics and clinical manifestations of orbital emphysema caused by isolated medial orbital wall fractures. *Eye (Lond).* 2016 Apr; **30**(4): 582-7.
4. Bařterzi Y, Sari A, Yavuzer R, Atabay K. Periorbital subcutaneous emphysema: an unusual clinical presentation of medial orbital wall fracture. *Plast Reconstr Surg* 2001; **108**: 2156-2157.
5. Van Issum C, Courvoisier DS, Scolozzi P. Posttraumatic orbital emphysema: incidence, topographic classification and possible pathophysiologic mechanisms. A retrospective study of 137 patients. *Oral Surg Oral Med Oral Pathol Oral Radiol* 2013; **115**: 737-742.
6. Ozdemir O. Orbital Emphysema Occurring During Weight Lifting. *Semin Ophthalmol.* 2015; **30**(5-6): 426-8.
7. Zimmer-Galler IE, Bartley GB. Orbital emphysema: case reports and review of the literature. *Mayo Clin Proc.* 1994 Feb; **69**(2): 115-21.
8. Key SJ, Ryba F, Holmes S, Manisali M. Orbital emphysema-the need for surgical intervention. *J Craniomaxillofac Surg* 2008; **36**: 473-476.
9. Ahnood D, Toft PB. Recurrent orbital compartment syndrome caused by a blow-out fracture and accumulation of air; management by orbital punctures. *Acta Ophthalmologica* 2012; **90**: 199-200

Peer Reviewer: Yoshihiko Usui