Idiopathic Recurrent Bilateral Orbital Emphysema: A Case Report

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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 aquity (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 systemic 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

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-limitating disorder[4-5]. Nevertheless it has been reported that it could rarely cause some severe advanced complications and need surgical intervention[6-9]. These uncommon and severe complications are occlusion of central retinal artery, compressive optic neuropathy and, most commonly, ischemic optic neuropathy[10].

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 noticable. 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 acquired. 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.
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 in 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 systematic therapy. Even if it is bilateral and recurrent, because it heals spontaneously, it is appropriate to follow these cases without giving systematic treatment.

REFERENCES

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 recommended because it is useless.

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.

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