Cervicofacial emphysema following unilateral external dacryocystorhinostomies: A case report

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Cervicofacial emphysema (CFE), mostly seen after trauma or dental procedures, is an unexpected, extremely rare condition after uncomplicated dacryocystorhinostomy (DCR). It may be misdiagnosed as angioedema or necrotizing fasciitis. In this article, we present the case of a 40-year-old female with CFE twice after uncomplicated unilateral DCR for left and right sides on different operative days. CFE was confirmed by computed tomography, demonstrating extensive air within subcutaneous tissues of the face, neck, and orbital cavity. Subcutaneous crepitation supported the diagnosis. This is the first case report, to the best of our knowledge, describing a patient with recurrent massive CFE after each unilateral DCR.

Key words: Dacryocystorhinostomy, emphysema, cervicofacial emphysema, external dacryocystorhinostomy

Access this article online

Quick Response Code:	Website: www.ijo.in
	DOI: 10.4103/ijo.IJO_1107_17

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Manuscript received: 15.11.17; Revision accepted: 06.01.18

Cervicofacial emphysema (CFE) is a rare condition which involves abnormal air flow and entrapment into subcutaneous layers of the head and neck through disrupted mucosal or bony barriers. When air has a high positive pressure, it flows through the least resistant loose connective tissue planes and reaches distant areas easily. There are many predisposing factors such as trauma, dental procedures, maxillofacial or orbital surgeries, tracheal intubations, pneumomediastinum, and infections that result in cervicofacial subcutaneous emphysema.[1-4] Although CFE is generally accepted as a self-limited situation, it may threaten life by airway obstruction due to neck involvement. Moreover, it may cause loss of vision in case of extensive orbital emphysema.[5-7] In our case, massive CFE occurred twice, following unilateral external dacryocystorhinostomy (DCR) procedures.

Cite this article as: Akingol Z, Karslioglu S, Serin D. Cervicofacial emphysema following unilateral external dacryocystorhinostomies: A case report. Indian J Ophthalmol 2018;66:722-4.

Case Report

A 40-year-old female presented to our clinic with watery eyes on both sides for 6 years. Obstruction of both nasolacrimal ducts was diagnosed with lacrimal irrigation. Her ophthalmological findings were otherwise insignificant. She had no history of systemic diseases. Ear-nose-throat (ENT) examination was normal. The patient underwent left external DCR with bicanalicular silicone tube insertion under general anesthesia without any complications. During and after surgery, the patient did not need any facial mask ventilation. On the 1st postoperative day, she developed severe swelling and ecchymosis of the left upper and lower eyelids, severe periorbital swelling, and crepitation extending to the whole face and neck with mild respiratory discomfort [Fig. 1a]. Steroids and antibiotics were started immediately to control inflammation on airway and the risk of infection. For further investigation, blood samples were collected and radiological imaging was planned. The patient was referred to internal medicine and ENT departments for further investigation to determine if any anatomical and systemic pathology may cause CFE. When swelling and ecchymosis became prominent on both sides, steroid treatment was immediately stopped because of the risk of necrotizing fasciitis or soft-tissue infection. On her computed tomography (CT) scans, there was massive air accumulation within subcutaneous tissue layers on both sides and left orbital cavity, confirming massive CFE. Blood tests were normal. Bilateral periorbital crepitation were palpated. Fever and lymphadenopathy were not observed. With the diagnosis of CFE and exclusion of infectious disorders, systemic steroid treatment was restarted. Emphysema limited itself on the 3rd day and regression had started. Steroid treatment was tapered daily. There was no vision deterioration or pathology on fundoscopic examination during follow-ups. The swelling decreased every day during hospitalization period and resolved by the 5th day. The patient was discharged with normal ophthalmological examination findings on the 7th day.

One month later, the patient was hospitalized again for right-sided DCR. The patient was informed about DCR under local anesthesia option but she did not accept, so we had to do DCR under general anesthesia again. Operation was performed under general anesthesia without any complications. On the 1st postoperative day, the patient presented with more prominent swelling of the right upper and lower eyelids spreading toward the other side of the face, periorbital region, and neck [Fig. 1b]. Crepitation were detected over swollen areas. This time, airway obstruction was more obvious because of massive emphysema around the neck. ENT consultation, blood sampling, and radiological imaging were performed. CT scan revealed CFE with extensive air within subcutaneous tissues and orbital cavity [Fig. 1c and d]. Appropriate medical treatment was started and the airway was kept open under close observation in Intensive Care Unit. Emphysema and respiratory distress gradually diminished and, on the 13th postoperative day, emphysema was totally resolved. Ophthalmological examination including visual acuity and slit-lamp, and fundoscopy was normal. The patient was discharged from the hospital and followed up monthly during the first 3 months. No ophthalmological or cosmetic sequelae were encountered on her either side during follow-ups.

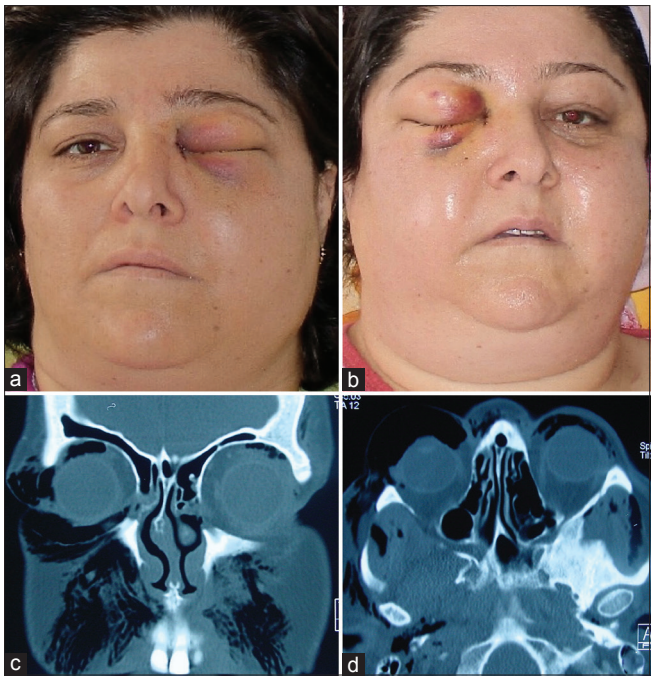

Figure 1: (a) Cervicofacial emphysema after left-sided dacryocystorhinostomy and (b) right-sided dacryocystorhinostomy. Air is widely seen on (c)coronal and (d) axial planes of orbita computed tomography imaging

Discussion

In an external DCR procedure, creation of an ostium between nasal cavity and nasolacrimal system disrupts the mucosal barrier. During the early postoperative period, manual ventilation with facial mask, sneezing, violent coughing, or nose blowing creates positive high pressure. Therefore, to prevent CFE, anesthetists and patient should be warned to prevent pressure increase in intranasal cavity.[8,9]

CFE is usually a self-limiting condition with swelling and crepitation. However, pain and tenderness are generally not observed. Vision loss may accompany CFE in rare cases of orbital involvement.[4-7] Orbital emphysema develops mostly after orbital trauma, but is an extremely rare complication of external DCR. Orbital air entrapment occurs as a result of a communication between the orbit and the nasal sinuses. If the patient has a history of DCR, blow-out trauma, medial wall fracture or functional endoscopic sinus surgery, sneezing, nose blowing, Valsalva maneuver, flying, and diving can push high pressure air into the orbital cavity through a communication between the orbit and the nasal cavity. Air in the orbital cavity increases the intraorbital pressure that can compress the optic nerve, resulting in loss of vision.[2,8-10] Therefore, air should be discharged through surgical incision area or directly from the orbital cavity as fast as possible in the operating room.

In cases of CFE, it is important to exclude infectious disorders that may be life-threatening. CFE may be difficult to differentiate from life-threatening conditions such as necrotizing fasciitis in most of the cases, but fever and erythema which accompany fasciitis do not exist in CFE. Necrotizing fasciitis is an emergency condition which requires intensive care and antibiotherapy; however, CFE is mostly a

self-limiting situation for which close observation is usually sufficient. CFE can easily be diagnosed with the help of a CT scan, normal blood test results, and crepitation palpated under the swollen area.

Conclusion

Among numerous reported cases of CFE, we present, to the best of our knowledge, the first case of recurrent massive CFE after unilateral external DCR surgeries on the same patient.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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