JMH
Jurnal Medika Hutama
Vol 04 No 04, Juli 2023
http://jurnalmedikahutama.com

e-ISSN. 2715-9728
p-ISSN. 2715-8039

Open Acces

A CASE REPORT: ADJACENT ORGAN INFECTION CAUSING ORBITAL CELLULITIS IN AN IMMUNOCOMPROMISED PATIENT

Annisa Ayu Rachmawati[1], Ade Irawan[1]

[1]Internship General Practitioner, Cilegon General Hospital, Banten, Indonesia
[1]Department of Ophthalmology, Cilegon General Hospital, Banten, Indonesia

Corresponding Author: Annisa Ayu Rachmawati. Cilegon General Hospital. Banten

Received 27 April 2023; **Accepted** 31 Mei 2023; **Online Published** 07 Juli 2023

Abstract

Introduction : Orbital cellulitis (OC) is a vision-threatening condition caused by an infection of the tissue behind the orbital septum. It can easily occur, especially when the individual's condition is accompanied by a weakness of the normal host defenses or because of hypervirulence of the causative microbe. Patients with comorbid or underlying conditions, may have higher risk. If left untreated, OC can seriously result in vision loss and life-threatening conditions. Case report: We reported a 46-year-old untreated diabetic male came to the ER with unilateral right eye pain, proptosis and oedema of the infected eye, headache, also fever. A diagnosis of OC with several comorbidities such as diabetic mellitus, and reactive anti-HIV was made. The diagnosis was also suspected by his condition before the symptoms, odonto-sinusitis that could be a primary infection of his right OC. A culture examination of the pus found coccus, Gram positive. The patient was admitted for seven days. However, the patient had died when outpatient schedule control. Conclusion: This complex case demonstrates the importance of a multidisciplinary approach to the management of OC starting from the risk factors present to prevent serious complications and permanent sequelae. Immunocompromised condition in our patient may have contributed his rapid deterioration.

Keywords: *Rapid Impairment, Adjacent Organ, Orbital Cellulitis, Immunocompromised*

Introduction

Orbital cellulitis is one of several eye emergencies that cause inflammation of behind the septum, affecting the orbit and its contents such as the periorbital adipose tissue, extraocular muscles, and neurovascular bundles, and delay in treatment can lead to blindness and even death.[1-3] It also known as postseptal cellulitis and it is a rare complication of sinusitis, but sinusitis is the most common cause of orbital cellulitis.[4-6] In developed countries, secondary infections extending from the paranasal sinuses are the most frequent predisposing factors.[7] Infection may also arise from hematogenous spread

from distant sources of infection, the eyelids, face, foreign bodies retained in the orbit.[3]

The symptoms of orbital cellulitis are red eye, pain, blurred vision, double vision, eyelid and periorbital swelling, nasal congestion or discharge, sinus headache, tooth pain, infraorbital and supraorbital pain, or hypesthesia.[8] Orbital cellulitis can easily occur, especially when the individual's condition is accompanied by a weakness of the normal host defenses or because of hypervirulence of the causative microbe. Natural host defenses, such as the immune system or an anatomical barrier like the orbital septum, both can help keep tissues free from microbes in the orbit. The number of risk factors include recent infection, such as sinus, upper respiratory infection, dacryocystitis, bacteremia, tooth abscess, trauma, or facial surgery which is also leading to the infection.[9]

The orbit, the skin of the eyelids, and the adjacent sinuses surrounded by millions of microbes. The most frequently isolated organisms are Staphylococcus aureus, Streptococcus pneumoniae, Staphylococcus epidermidis, Haemophilus influenzae, Moraxella catarrhalis, Streptococcus pyogenes, Group A streptococci and upper respiratory anaerobes.[3,8,10]

The sinuses (frontal, sphenoid, maxillary, and ethmoid) are air-filled cavities lined with pseudostratified ciliated epithelium. All of these sinuses open into the nasal cavity through the ostium. The maxillary sinus is the largest of the sinuses, and maxillary sinusitis is a relatively common condition.[11]

Odontogenic orbital cellulitis is a rare but serious infection with a high risk of vision loss. Intensive surveillance, serial imaging, multidisciplinary management, and surgical intervention may be required for this infection.[12]

Potential risk factors for developmental complications have been described in numerous uncontrolled observational studies. These include gender, age and comorbidities. Male sex and younger age are the strongest risk factors. Patients with underlying and comorbid conditions such as allergic rhinitis or diabetes that compromise the immune system may be at increased risk. However, due to lack of control in previous studies, the risk factors for developing orbital complications in sinusitis patients have not been precisely defined.[13]

If left untreated, orbital cellulitis can become more serious and lead to vision loss and life-threatening conditions.[6] There is a consensus that surgical drainage must be performed in cases of complete

ophthalmoplegia or significant visual impairment or large abscesses formation.[10] This major complication of sinusitis is considered an uncommon but serious condition, requiring early attention and prompt treatment. The incidence of complications is approximately 2-5 cases per million general population per year, for precisely 95% of it.[10,13] Postseptal cellulitis can cause blindness and death if the infection spreads to the skull.[4]

Case Illustration

A 46-year-old male patient presented to the emergency room with pain in his right eye 4 days before his hospitalization. The complaint was accompanied by swelling, difficulty opening, and blurring vision in his right eye (Figure 1).

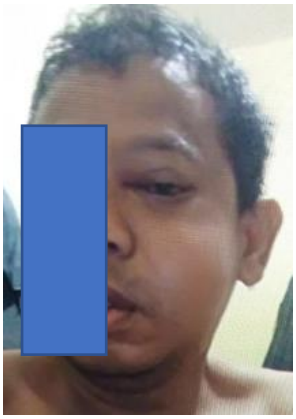

Figure 1. Patient's symptoms

The previous complaint was preceded by swollen right cheek from 1 week before admission, which gradually increased. The complaint was accompanied by pain in the right cheek. Currently, the appearances of the symptoms were the discharge of pus and blood from the nose and mouth, throbbing headache, fever, swollen right side of the patient's lips so that the patient becomes difficult to drink, to eat and to speak (Figure 2). The patient also complains of nausea, decreased appetite and weakness. The patient previously had a history of pain in the upper right tooth and cavities. On ophthalmological examination, there was proptosis, edema and erythema on the lids, conjunctival chemosis, impaired movement of the eyeball and decreased vision acuity with no light perception (Figure 3&4). The intraocular pressure is measured just by palpation of the palpebral and it feels high. Intraoral examination found cavities on the upper teeth.

Figure 2. Nasal discharge and edema

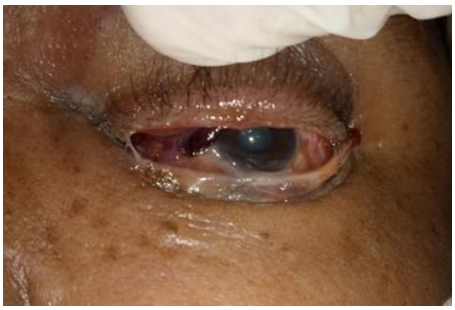

Figure 3. Chemosis conjuntiva

Figure 4. Ophthalmology examination

The patient has treated his complaint with unknown medicine, but the complaints have not reduced. The complaints are not accompanied by glare in the eyes or history of previous trauma to the eye. This is the first time this patient has experienced this. The patient has had diabetes mellitus since 5 years ago, but he does not routinely get treatment, when in the ER the blood glucose examination was 273 mg / dL. The patient did not know a history of hypertension, but when in the inpatient room his blood pressure was 150/90 mmHg. The patient's anti-HIV result was found reactive, so he had just received therapy with triple antiretroviral once a day.

His psychosocial history worked as a security at a company. The patient had a history of having multiple sexual partners and drinking alcohol when he was young.

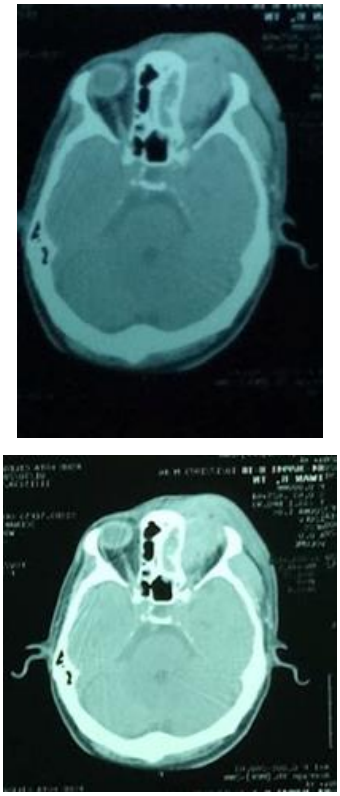

Figure 5 & 6. CT-scan examination

In the laboratory results obtained leukocytes 12.89 x 103 / L, on the 3rd day of treatment there was a decrease in leukocytes after antibiotic therapy. The results of the culture examination of the pus specimen, found coccus Gram positive with the type of bacteria *Staphylococcus aureus* but do not include Methicillin for resistance examination. CT-scan examination revealed maxillary and ethmoid sinusitis, right sphenoid with a differential diagnosis of mass infiltration (Figure 4&5). The patient underwent maxillary abscess drainage and obtained 50 ml of pus and blood.

Case Discussion

The diagnosis of this patient was made from various aspects such as history taking, physical examination and supporting examinations including CT-scan and bacterial culture. Through these steps, we try to rule out possible causes such as non-infective inflammation, orbital tumors or vascular malformations. Our patient's symptoms were pain in his right eye, discharge of pus and blood from the nose and mouth, throbbing headache, fever, swollen right side of the patient`s lip. ophthalmology examination found proptosis, edema and erythema on the lids, conjunctival chemosis, impaired movement

of the eyeball. In the study of Zhaoxin et al (2022) diagnostic criteria for OC was used the presence of ophthalmoplegia, proptosis, chemosis, decreased visual acuity, or radiologic evidence of orbital inflammation or abscess.[7] Sevlia et al (2017) reported in their orbital cellulitis group study seven cases had ophthalmoplegia, three of them also had proptosis.[6,7] Based on the study by Benjamin (2019) symptoms of eye pain were documented in four of six patients. Proptosis, conjunctival congestion, and chemosis and limitation of ocular motility were present in all six patients and the duration between the onset of symptoms and the first ophthalmic evaluations by ophthalmologists ranged from 1 to 7 days, with an average of 2.5 days.[14] Another study by Harvey et al (2007) reported proptosis was the most frequent clinical finding related to loss of vision.[14,15] Yi-Sheng et al (2017) found that fever was present in 21 (43.8%) of 48 adults.[16]

Laboratory and radiology findings also help confirm the diagnosis. A good history of illness and a general medical history of the patient are required. Blood tests, including HIV status, complete and differential blood counts, and blood cultures are very important to rule out other differential diagnosis. The patient's diagnosis is most likely due to infection, which can originate from sinusitis, and possibly from cavities then spread into orbital tissue leading to an infection, especially cellulitis. Based on Sevliya et al (2017) reported in their study that sinusitis is causing orbital cellulitis in 10 of 12, other predisposing factors in this study are conjunctivitis, dental abscess, varicella-zoster infection, and herpes simplex virus infection of adjacent skin.[6] Chieh-Hung et al (2018) also reported that four from six patients had a sinus source of infection.[14] However, another study Zhaoxin et al (2022) reported that ocular trauma is an increasingly common cause of orbital cellulitis in developing regions and may present features quite different from the reports which is predominance of sinusitis.[7] Kornkiat et al (2020) found clinical findings classified by stage of sinusitis-related orbital complications on their study founding the most common orbital complication was subperiosteal abscess (42.1%), followed by orbital cellulitis (15.8%).[13] Based on Allegrini et al (2017) reported infection can also originate from a very specific site like dental infection.[17]

Based on the ophthalmology examination patient’s found decreased visual acuity with no light perception and the IOP was high, it showed orbital involvement in the patient's disease so that the diagnosis was directed to orbital cellulitis. Regarding visual acuity measured at admission, 50% of the cases were NLP on the study in Guangzhou.[7] However, another study stated four patients

had decreased visual acuities (VA) of various degrees, three recovered to normal, and one ended up with hand motion due to compressive optic neuropathy.[14]Among the 35 patients with postseptal cellulitis, 10 (29%) experienced permanent severe visual loss.[15]

Palate of the ethmoid sinuses is the anterior cranium base, and the ethmoid cells are divided from the medial orbital wall by a thin bony plate called the lamina papyracea. The lateral lamella, which follows the fovea ethmoidalis to the cribriform plate, is also a thin bone. The anterior ethmoid artery penetrates the olfactory fossa in the thinnest part of this bone. Therefore, an abscess can expand directly from the ethmoid cells through these thin bony walls, causing orbital complications. Secondly, an infection in the ethmoid cells can spread through the preformed pathways that connect the ethmoid cells to the eye balls.[13] The first axis of this structure is the anatomic location of pathology resulting in visual loss and includes the anterior segment, retina, choroid, optic nerve, optic chiasm or tract, and cerebral cortex. The second includes the most usual disease processes related to orbital infection, which may affect the anatomical portions of the visual pathway and lead to compressive effects, infiltration, inflammation, and thrombosis.[13,18] The mechanisms of visual loss may involve optic neuritis as a reaction to adjacent infection, ischemia resulting from thrombophlebitis, compressive ischemia possibly resulting in artery occlusion, or elevated in intraorbital pressure.[3,19] Intraocular pressures were all elevated at initial presentation.[14]

In addition to orbital cellulitis, the diagnosis of maxillary abscess, type 2 diabetes mellitus was also diagnosed according to high blood glucose examination and his history of diabetes mellitus 5 years ago. Patients do not take anti diabetic oral routinely, so the possibility of infection other Staphylococcus aureus, as evidenced by culture examination, fungal infection can also occur in HIV-reactive patients. Research results in Turkey of 36 cases of blood culture only four of them were positive. Three of four positive cultures were obtained from the patients in the orbital cellulitis group.[6] Another study in Thailand stated that the most common pathogens identified in the complicated group were Staphylococcus aureus 25.0% of 37,2% who was positive for blood culture.[13] However, according to Meyer (2020) Staphylococcus aureus was the most frequently cultured pathogen 50% of 92 isolates, a polymicrobial infection was present in 38.1% of episodes.[20] Another study that had the same patient characteristics as us, having diabetes mellitus but no surgical intervention was performed, did not know the causative

agent of the orbital cellulitis, they suspected a fungal infection like mucormycosis which could worse their patient condition.[5] Individuals with diabetes mellitus often experience fungal infections. Impaired effective immunological response to infection in patients with diabetes mellitus due to low granulocyte phagocytic activity with impaired polymorphonuclear leukocyte cell response. orofacial or sinuses are usually areas of co-infection. In fact, rhino-cerebral localization predominates in patients with diabetes mellitus.[21] In a case series according to Singh (2017) two out of 4 cases found out Staphylococcus aureus when bacterial culture was carried.[22]

The results of our patient laboratory examination showed leukocytosis, so the diagnosis of bacterial infection was getting stronger. A research study stated mean white blood cell counts of the patients with orbital cellulitis was 14483±5816/mm3 .[6] Another study also found leukocytosis in all five patients, with an average count 12.6 ×1000/μL.[14] Leukocytosis was also seen in 68.7% cases in the study but not associated with disease severity.[14,16]

Our patient CT scan result is also supported by showing orbital involvement. There were other foci of infection presented by maxillary, ethmoidal and sphenoidal sinusitis. Several studies reported the same result, according to Chieh-Hung (2018) the six adult patients had proven unilateral involvements and CT-scan with orbital cellulitis.[14] Yi-Sheng (2017) reported that CT of orbits and sinuses was conducted in 85.5%, and sinusitis and preseptal/orbital inflammation was evident in all these cases.[16]

Another study also stated the etiology of orbital cellulitis 64.7% cases had radiological evidence of paranasal sinusitis, 36 patients had ethmoidal sinusitis, 6 patients had frontal sinusitis, and 24 patients had multiple sinuses affection, two of them had fungal orbital cellulitis (mucormycosis).[3]

The psychosocial history of the patient who used to have multiple partners or promiscuity could be a risk factor for HIV, which is an additional comorbidity in this case in addition to diabetes mellitus. Based on Chieh-Hung (2018), three of six patients had underlying diabetes mellitus, and two of them had never been diagnosed of diabetes until the episode of orbital cellulitis.[14] Singh et al (2017), found that two out of four cases had type 2 Diabetes Mellitus and 1 case out of 4 was found to be HIV positive.[22] Another study in Taiwan shared 24.1% had diabetes mellitus, which is associated with disease stage, with 10.3% in stage I, 28.1% in stages II–IV, and 58.3% in stage V.[16] Orbital involvement in the presence of diabetes, especially in ketoacidosis should always raise the suspicion of life-threatening fungal

infection of the order Mucorales that can involve the sino-orbito-cerebral region. Although the pathophysiology is not entirely understood, it has been hypothesized that during acidosis, impairment of iron metabolism is leading to compromised cell-mediated immunity.[23] The rapid deterioration of the disease may also be due to the patient's weakened immune condition and the adjacent infected organs that facilitate the direct spread of bacteria. Immunocompromised conditions like HIV and diabetes can alter the course of even mild disease processes in the body. Mild sinusitis in the presence of such immuno-compromising diseases can easily end up in orbital cellulitis.[24] A study stated 50% of individuals with HIV positive and orbital cellulitis died within 1 year of the infection, mostly due to the intracranial spread. Cellulitis and soft tissue infection anywhere on the body are often underestimated complications of HIV disease. Orbital cellulitis remains a cause of morbidity and mortality in HIV positive cases in Sub-Saharan Africa, in spite of the availability of antiretroviral.[20]

Considering the condition of orbital cellulitis can develop various complications that can cause blindness, the treatment for the patient is hospitalized. Therefore, early management and prevention are necessary. Orbital cellulitis is treated with medical treatment. The antibiotics used in the patient were injection of meropenem 1 gram t.i.d and metronidazole 500 mg t.i.d. In addition, symptomatic and topical drugs were given, chloramphenicol eye ointment, levofloxacin eye drops and artificial tear. In a study by Berdouk et al (2018) their patient was given with IV antibiotics and insulin therapy along with supportive management.[5] Orbital cellulitis is an acute and aggressive disease requiring immediate and effective treatment. In the current study, intravenous antibiotics and glucocorticoids were administered to all cases after decision of the diagnosis, except in cases of suspected fungal infection.[5,7] The management of orbital cellulitis requires a multidisciplinary approach. It also depends on the level or class of the disease and the cause of the disease. Antibiotherapy with ampicillin-sulbactam with or without ceftriaxone was successful in 95.2% of the patients.[6] Antibiotic selection may later be modified once culture and sensitivity results become available. Local trends in antimicrobial susceptibility are a critically important consideration, as different communities may have distinctive flora with varied resistance profiles. Treatment should be modified based on culture/sensitivity results and local resistance profiles, and consultation with the infectious disease service may be valuable.[6,25] Our patient was given the diabetes treatment while in the

ward is metformin 500 mg once daily. In the study with the same diabetic patient also controlled the glycemic blood serum successfully managed with metformin 100 mg bd.[23] On the 7th day of treatment our patient asked to go home due to cost issues, so the examination and treatment that we gave can not be maximally. According to the study by Sevliya (2017) median length of hospital stay was 20.5 days for orbital cellulitis.[6] Orbital cellulitis has been associated with serious complications including visual loss, cavernous sinus thrombosis, meningitis, frontal abscess, osteomyelitis, and even death.[3] One of the significant complications of an orbital abscess is blindness of the eye affected by the several processes.[26]

Seeing the progress of the patient, the prognosis of this case is dubia ad malam because immunocompromised as a high risk factor in our patient. Immunocompromised individuals commonly have higher rates of complications and an overall poor outcome.[22] The management of orbital cellulitis requires a multidisciplinary approach, our patient hospitalized with several departments, such as, ophthalmology, ENT, Odontology, and Internal medicine.

Conclusion

Our case highlights that doctors need to have high suspicion regarding complications that are quite severe in cases of orbital cellulitis so that management can be given appropriately and quickly. Our threshold for investigating such patients should be low, especially if they are immunocompromised, or come from a less affluent background with access to healthcare and a history of poor patient habits. Our patient had a long-term history of untreated diabetes mellitus as well as HIV reactivity and this is a suspicion that it may have contributed to its rapid deterioration. Indeed, there are not many studies that explain whether this immunocompromised state can make cases rapidly worsen.

The weakness of our study was that we did not perform fungal cultures, which could be another cause considering the immunocompromised condition of our patients. We recommend the use of contrast-enhanced imaging studies (CT / MRI) early in the management of suspected OC, especially orbital CT, and CT scan of teeth and paranasal sinuses as well as MRI to assess the onset of infection and whether there are complications reaching the brain. In addition, the weakness of this case report is that the patient was forced to go home because of financial problems, there was no guarantor, so that after being traced again,

the patient died not long after returning from the hospital.

References

1. Clarke CL, Mitchell KT. Orbital Cellulitis in West Texas. Southwest respir crit care chron. 2021 Jul 4;9(40):9–13.
2. Chaudhry IA, Al-Rashed W, Arat YO. The hot orbit: orbital cellulitis. Middle East Afr J Ophthalmol. 2012 Jan;19(1):34–42.
3. Elshafei AMK, Sayed MF, Abdallah RMA. Clinical profile and outcomes of management of orbital cellulitis in Upper Egypt. J Ophthalmic Inflamm Infect. 2017 Dec;7(1):8.
4. Stead TG, Retana A, Houck J, Sleigh BC, Ganti L. Preseptal and Postseptal Orbital Cellulitis of Odontogenic Origin. Cureus. 2019 Jul 6;11(7):e5087.
5. Berdouk S, Pinto N. Fatal orbital cellulitis with intracranial complications: a case report. Int J Emerg Med. 2018 Nov 22;11(1):51.
6. Demir SÖ. Clinical features and outcome of preseptal and orbital cellulitis in hospitalized children: Four years experience [Internet]. Medeniyet Medical Journal. 2017. Available from: http://dx.doi.org/10.5222/mmj.2017.007
7. Jiang Z, Zhongliu X, Ding X, Yang Y, Duan F, Lin X. Clinical Characteristics and Risk Factors for Evisceration in Trauma-Dominant Orbital Cellulitis: A 10-Year Review. Front Med. 2022 Jun 17;9:935022.
8. GERVASIO. The Wills Eye Manual: Office and Emergency Room Diagnosis and Treatment of Eye Disease. LWW; 2021. 496 p.
9. American Academy Ophthalmology. San Francisco: Orbital cellulitis; 2022. [updated 2022 Nov 1; cited 2022 Nov 28]. AAO EyeWiki; Available from: https://eyewiki.aao.org/Orbital_Cellulitis
10. Akçay E. Preseptal and orbital cellulitis. J Microbiol Infect Dis. 2014 Sep 1;4(3):123–7.
11. Youssef OH, Stefanyszyn MA, Bilyk JR. Odontogenic orbital cellulitis. Ophthal Plast Reconstr Surg. 2008 Jan-Feb;24(1):29–35.
12. DeCroos FC, Liao JC, Ramey NA, and Li I. Management of odontogenic orbital cellulitis. Journal of Medicine and Life. 2011 July; 4(3):314-17.
13. Snidvongs K, Chitsuthipakorn W, Akarapas C, Aeumjaturapat S,

Chusakul S, Kanjanaumporn J, et al. Risk factors of orbital complications in outpatients presenting with severe rhinosinusitis: A case-control study. Clin Otolaryngol. 2021 May;46(3):587–93.

14. Yen CH, Wu SY, Liao YL. *Klebsiella pneumoniae* Orbital Cellulitis: Clinical Manifestations and Outcomes in a Tertiary Medical Center in Taiwan. J Ophthalmol [Internet]. 2018 Oct 2 [cited 2022 Nov 30];2018. Available from: https://doi.org/10.1155/2018/4237573
15. Uy HS, Tuano PMC. Preseptal and orbital cellulitis in a developing country. Orbit. 2007 Mar;26(1):33–7.
16. Chang YS, Chen PL, Hung JH, Chen HY, Lai CC, Ou CY, et al. Orbital complications of paranasal sinusitis in Taiwan, 1988 through 2015: Acute ophthalmological manifestations, diagnosis, and management. PLoS One. 2017 Oct 3;12(10):e0184477.
17. Allegrini D, Reposi S, Nocerino E, Pece A. Odontogenic orbital cellulitis associated with cavernous sinus thrombosis and pulmonary embolism: a case report. J Med Case Rep. 2017 Jun 20;11(1):164.
18. Chamberlain PD, Foroozan R. Mechanisms of visual loss from orbital cellulitis. In: Orbital Cellulitis and Periorbital Infections. Cham: Springer International Publishing; 2018. p. 23–34.
19. Gupta S, Sharma S. Orbital cellulitis: defining multidisciplinary approach as the need of the hour. Indian J Otolaryngol Head Neck Surg. 2019 Mar 16;71(4):464–9.
20. Meyer D, Smit DP. Eyelid and Orbital Involvement in HIV Infection - An African Perspective. Ocul Immunol Inflamm. 2020 Oct 2;28(7):1022–30.
21. Cerebro-rhino-orbital mucormycosis and aspergillosis coinfection in a patient with diabetes mellitus: A case report. IDCases. 2021 Jan 1;23:e01022.
22. I D Singh, Lt Col V Gupta, S Gupta, Anubhav. Acute orbital cellulitis: a case series. Glob J Otolaryngol. 2017 January;3(3):1-5
23. Colapinto P, Aslam SA, Frangouli O, Joshi N. Undiagnosed type 2 diabetes mellitus presenting with orbital cellulitis. Orbit. 2008;27(5):380–2.
24. Kator C, Dominic C, Sunday OA. Management of Bilateral Orbital Cellulitis in 41-Year-Old Man. Nigerian Journal of

Ophthalmology. 2014 January;1(1):50-5

25. Lee S, Yen MT. Management of preseptal and orbital cellulitis. Saudi J Ophthalmol. 2011 Jan;25(1):21–9.
26. Sikora M, Wróbel K, Stąpor A, Sielski M, Chlubek D. Odontogenic orbital cellulitis in a young man with complete vision loss – a case report. Pomeranian Journal of Life Sciences. 2020 Dec 1;66(4):41–5.