International Journal of Otorhinolaryngology and Head and Neck Surgery
Munjal M et al. Int J Otorhinolaryngol Head Neck Surg. 2022 Sep;8(9):779-782
http://www.ijorl.com
pISSN 2454-5929 | eISSN 2454-5937

DOI: https://dx.doi.org/10.18203/issn.2454-5929.ijohns20222176

Case Report

Subcutaneous canine fossa aspergilloma to intervene or not

Manish Munjal[1]*, Vikas Garg[2], Porshia Rishi[3], Harjinder Sidhu[3], Shubam Munjal[3], Shivam Talwar[3], Salony Sharma[3], Deeksha Chawla[3]

[1]Department of ENT, [2]Department of Medicine, [3]Dayananda Medical College and Hospital (DMCH), Ludhiana, Punjab, India

Received: 15 December 2020
Revised: 27 July 2022
Accepted: 28 July 2022

***Correspondence:**
Dr. Manish Munjal,
E-mail: manishmunjaldr@yahoo.com

ABSTRACT

The term "Aspergillosis" refers to an illness due to allergy, airway or lung invasion, cutaneous infection, or extrapulmonary dissemination caused by species of *Aspergillus*. A 51-year-old male with a history of swelling in the right maxillary region is presented. The diagnosis was subcutaneous aspergillosis in an immunocompromised patient, which was successfully treated with voriconazole and surgical excision of the mass. Possible clinical manifestations, diagnostic imaging techniques and treatment used are highlighted upon. Patient was taken up for premaxillary mass excision by sublabial approach. Sublabial incision was given in the upper gingivo-buccal sulcus from right canine upto the first molar, the canine fossa and a subcutaneous mass was dissected from anterior wall of maxilla Histopathological report was consistent with fungal granuloma with predominant hyphae of aspergillus. Patient was prescribed voriconazole 200 mg twice a day with weekly monitoring of liver functions.

Keywords: Aspergillosis, Canine fossa, Subcutaneous, Voriconazole, Debridement

INTRODUCTION

Invasive aspergillus infections can be either limited [chronic or indolent] or fulminant [acute], with a rapid malignant course advancing relentlessly to the destruction of the nasal cavity, the sinuses and the adjacent structures such as the orbit and the brain within a few days.[1] Circumscribed fungal mass in the subcutaneous tissue overlying the canine fossa of the cheek is usually seen as an associated or a late manifestation of a treated maxillary sinus pathology. The intervening bone many a time is intact and the likely portal of external spread is the infra orbital canal. A medical disease, the pathology many a time doesn't resolve with the imidazoles in use probably due to a fibrous pseudocapsule that prevents the medication from gaining access to the fungi. On the contrary if the fungi have been treated, the resultant "fibrous ball", disfigures the face. This necessitates usually a gingivo-labial approach with removal of the mass. An intriguing subject with such a persistent subcutaneous is being presented.

CASE REPORT

51 years old male presented to the ear, nose and throat (ENT) out patient department with chief complaint of diffuse swelling on the right side of the cheek for the last 1.5 months (Figure 1).

The physical examination revealed a diffuse swelling about 4×3 cm in size in the subcutaneous planes affecting the right side of cheek. It was extending superiorly to the lower eye lid and laterally to the zygomatic arch. The overlying skin was normal. There was no localised pain or tenderness. Clinically there was no involvement of the orbit. Patient was on antiepileptics with ventriculoperitoneal shunt in situ for hydrocephalous.

Patient had taken complete course of anti-tuberculosis therapy for pulmonary tuberculosis.

There was history of endoscopic sinus clearance 2 years back for sino-nasal polyposis. At that time computed tomography revealed bilateral pansinusitis with remodelling, sclerosis and erosion of sinus wall, nasal turbinates and nasal septum. Previous biopsy had reported fungal granuloma i.e. aspergilloma for which he was on irregular treatment. The present computed tomography revealed the lesion in right premaxillary area with well pneumatised bilateral paranasal sinuses (Figure 2).

Figure 1: Right sided cheek swelling (orange arrow).

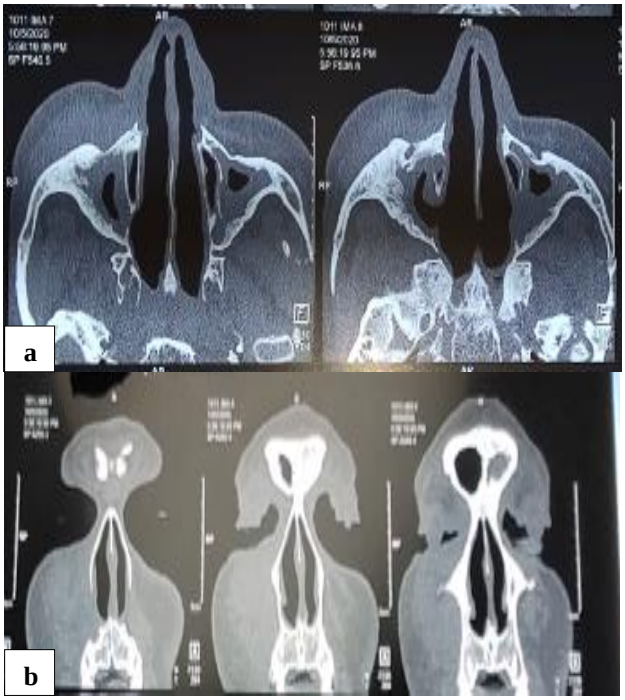

Figure 2: (a) Axial computed tomography section showing lesion in the premaxillary space right side (orange arrow); and (b) coronal computed tomographic section showing lesion in the premaxillary region (green narrow).

Patient was taken up for premaxillary mass excision by sublabial approach. A sublabial incision was given in the upper gingivo-buccal sulcus from right canine upto the first molar (Figure 3).

Figure 3: Sublabial incision made in upper gingivo-buccal sulcus right side from canine upto first molar.

Premaxillary mass was dissected in the subcutaneous plane upto the infraorbital rim (Figure 4).

Figure 4: Subcutaneous mass (black arrow) dissected from anterior wall of maxilla (orange arrow).

The lesion was 3×4 cm in size, pale white in color and firm in consistency. The anterior wall of maxilla was seen to be intact. Mass was dissected out in toto and sent for histopathological examination (Figure 5).

Figure 5: Subcutaneous mass removed in toto 4.5×3 cm in size.

Figure 6: Intact anterior wall of maxilla (yellow mark) post subcutaneous mass excision.

Figure 7: Sutured site at gingivobuccal sulcus.

Histopathological report was consistent with fungal granuloma with predominant hyphae of aspergillus. Patient was prescribed voriconazole 200 mg twice a day with weekly monitoring of liver functions.

DISCUSSION

Fungal rhinosinusitis is classified into an invasive and a non-invasive form, depending on invasion of the mucosal layer and destruction of the bone. Invasive aspergillus infections can be either limited (chronic or indolent) or fulminant (acute), with a rapid malignant course advancing relentlessly to the destruction of the nasal cavity, the sinuses and the adjacent structures such as the orbit and the brain within a few days.[1]

Aspergillus fumigatus is the most common species, followed by *Aspergillus flavus*.[2] The maxillary sinus is the most common sinus to be affected. The main route of infection is inhalation of fungal spores.[3]

Aspergillus shows septate hyphae that branch at 45º angles. The histology should be specific as to whether there is mucosal involvement (invasive) or the mucosa is intact (non-invasive disease). Fungal cultures on Sabouraud´s dextrose agar is needed to confirm the diagnosis.[4]

In disseminated or deep aspergillosis, serum galactomannan assay in conjunction with cultures and/or histologic examination can be used for diagnosis but it has limitations of having false positive and false negative results.[5]

The risk factors for aspergillosis include neutropenia, inappropriate use of antibiotics, immunosuppressive drugs, corticosteroids, uncontrolled diabetes mellitus, human immunodeficiency virus infection, trauma, burns, and radiation therapy.[6] In our case diabetes mellitus and past pulmonary tuberculosis made the patient immunocompromised thereby predisposing him to fungal aspergillosis.

The outcome of the patient with sinonasal fungal pathology depends on early diagnosis, Reversal of the predisposing risk factors, appropriate early surgical debridement and rapid antifungal therapy. Voriconazole (broad-spectrum triazole) has now become the drug of choice for invasive aspergillosis. The recommended dosing regimen of voriconazole is 6 mg/kg IV every 12 hours on day one followed by 4 mg/kg IV twice daily, and after that, 200 mg orally twice daily.[7,8] For most patients, antifungal therapy will continue for months or even years in some cases. Regular post-operative follow-up is recommended in all the cases with computed tomography scans and nasal endoscopy every 3-4 months.

Surgical debridement of abnormal tissue in the sinus is recommended, for pharmacological therapy to reach the infected area. The magnitude of the debridement depends on the extent of the pathology. Though tissue diagnosis is a must in all suspicious cases of subcutaneous aspergillosis but often medical treatment is initiated based on clinical suspicion, moribund patient, imaging findings/when the pathology is lying in inaccessible regions like the intraorbital, intracranial and the infratemporal fossae. Moreover, the report of fibrous tissue; only many times does not exclude an aspergilloma. It just means that site of the lesion has not been precisely accessed. Sometimes a space occupying lesion in the orbit which has healed by antifungals but restricts the extraocular movements needs to be removed.

In the present case the lesion could be delineated from the skin and anterior wall of maxilla as the plane could be found between them. Entire mass could be enucleated in toto without any buttonholing of the skin. The Cald well Luc procedure or anterior wall maxillary antrostomy was not performed as the anterior wall of maxilla was intact.

CONCLUSION

Any subcutaneous fungal manifestation usually seen in the canine fossa disfigures the face of the individual and affects the psyche. It necessitates an excision to reduce the

volume of the fungus and thereby the therapeutic dosage of the antifungal and its toxicity.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

REFERENCES

1. Sharma D, Mahajan N, Rao S, Khurana N, Jain S. Invasive maxillary aspergillosis masquerading as malignancy in two cases: Utility of cytology as a rapid diagnostic tool. J Cytol. 2012;29(3):194-6.
2. Akhaddar A, Gazzaz M, Albouzidi A, Lmimouni B, Elmostarchid B, Boucetta M. Invasive Aspergillus terreus sinusitis with orbitocranial extension: case report. Surg Neurol. 2008;69:490-5.
3. García-Reija MF, Crespo-Pinilla JI, Labayru-Echeveria C, Espeso-Ferrero A, Verrier-Hernández A. Invasive maxillary aspergillosis: report of a case and review of the literature. Med Oral. 2002;7:200-5.
4. Barton RC. Laboratory Diagnosis of Invasive Aspergillosis: From Diagnosis to Prediction of Outcome. Scientifica. 2013;459405.
5. Aquino VR, Goldani LZ, Pasqualotto AC. Update on the contribution of galactomannan for the diagnosis of invasive aspergillosis. Mycopathologia. 2007;163:191-202.
6. Singh N, Siddaraju N, Kumar S, Muniraj F, Bakshi S, Gopalakrishnan S. Fine-needle aspiration biopsy as an initial diagnostic modality in a clinically unsuspected case of invasive maxillary fungal sinusitis: A case report. Diagn Cytopathol. 2010;38:290-3.
7. Nakaya K, Oshima T, Kudo T, Aoyagi I, Katori Y, Ota J. New treatment for invasive fungal sinusitis: three cases of chronic invasive fungal sinusitis treated with surgery and voriconazole. Auris Nasus Larynx. 2010;37:244-9.
8. Arakawa H, Suto C, Notani H, Ishida T, Abe K, Ookubo Y. Selection of the antifungal agent decides prognosis of invasive aspergillosis: case report of a successful outcome with voriconazole. Int Ophthalmol. 2014;34:85-9in East Asia. Dis Colon Rectum. 2012;55(8):900-6.

Cite this article as: Munjal M, Garg V, Rishi P, Sidhu H, Munjal S, Talwar S, et al. Subcutaneous canine fossa aspergilloma to intervene or not. Int J Otorhinolaryngol Head Neck Surg 2022;8:779-82.