

CASE REPORT

Aspergilloma of Infra Orbital Nerve in an Immunocompetent Female: A Case Report

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Abstract:

Aspergilloma of the paranasal sinus is a non-invasive form of aspergillosis. We report a 50 year old female with chief complaints of swelling in left infra orbital area without involving maxillary sinus. Computed tomography revealed enlarged infraorbital nerve foramen with clear left maxillary sinus. Microscopy of the excised mass showed, numerous fungal hyphae, which branch at acute angle consistent with features of aspergilloma. Early diagnosis and treatment is important in aspergilloma. Since the symptom is usually non-specific, radiographic study is nonspecific; excision biopsy should be performed for all cases.

Keywords: Aspergilloma, Infraorbital nerve, Maxillary Sinus.

Introduction:

Aspergillosis is a fungal disease caused by *Aspergillus* species which is the most common fungal infection of the paranasal sinuses and is characterized as invasive and non-invasive forms. Non-invasive aspergillosis is the more common form, appearing either as an allergic reaction or cluster of fungal hyphae. Low-grade sinus infection results into a mass of fungal hyphae, called aspergilloma [1]. Most patients with aspergilloma presents with non-specific symptoms. The maxillary sinus followed by the sphenoid sinus is the most common site for the formation of an aspergilloma [1].

Case report:

A 50-year-old south Indian female presented with slow growing swelling below left infra orbital region of 4 months duration. Swelling was associated with intermittent pain on applying pressure and tingling sensation over the left cheek. She did not complain of any ocular and nasal symptoms. On examination a solitary swelling was found measuring 3 x 2.5 cms in size extending downwards about 3cms from left infraorbital margin (Fig.1).



Fig. 1: Preoperative Photograph showing a Diffuse Swelling below the Left Infraorbital Margin with Normal Overlying Skin.

The swelling was non-tender, solid, firm to hard consistency, mobile which could be moved in side to side direction. The swelling was not pulsatile and transillumination test was negative. The skin over swelling was normal with hypoesthesia. Diagnostic nasal endoscopy, ear and throat examination were normal. Visual acuity in both eyes was 20/20, N6 with glasses. The pupillary reactions, colour vision, ocular fundi and visual fields were normal. Corneal sensation was intact. There was no globe displacement or proptosis and ocular motility was full. The rest of the cranial nerves and neurological examination were normal. Provisional diagnosis of lipoma or schwannoma was made. Fine Needle Aspiration Cytology [FNAC] of the swelling was done which revealed lipoma. Routine blood investigations were within normal limits. CT scan of the orbit and paranasal sinus revealed soft tissue mass over left maxilla and infraorbital foramen was enlarged and eroded suggestive of infraorbital nerve tumor (Fig. 2).

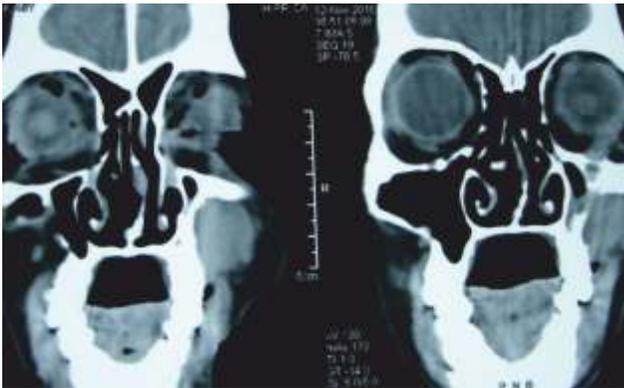


Fig. 2: Coronal CT- Scan Imaging Enlarged and Erosion Left Infraorbital Foramen

Excision biopsy of the mass was planned under general anesthesia by sublabial approach. Well encapsulated pedunculated mass was identified along the course of infraorbital nerve extending towards the floor of the orbit which was traced using microdrill with diamond burr after opening the infraorbital foramen (Fig. 3).



Fig. 3: Intraoperative Photograph showing Infraorbital Foramen

After infiltrating the skin with 2% Xylocaine with adrenaline an inferior lid crease incision placed below the eyelashes paralleling the lid margin. Pretarsal skin flap was separated and intact muscular portion of the orbicularis oculi was undermined and a dissection plane between the muscle and the septum orbitale was created. Incision was placed over muscle over the bony infraorbital rim. Periosteal incision was made after skin muscle flap is retracted inferiorly over the anterior edge of the infraorbital rim. Periosteal elevators used to strip the periosteum from the underlying bone and the infraorbital nerve was located below the medial portion of the infraorbital rim. After identifying the infraorbital foramen and the nerve exit zone, the periosteum over the inferior orbital rim was reflected and the nerve was traced along the floor of the orbit as posteriorly as possible. The pedunculated mass was excised at the stalk and sent for histopathological examination (Fig. 4).



Fig. 4: Excised Mass along with Pedicle of Infra-Orbital Nerve

Postoperative period was uneventful except for minimal numbness over left cheek area. Histopathological examination revealed epitheloid granuloma with foreign body and Langhans type giant cell surrounded by dense hyalinized fibrous tissue and many giant cells containing septate hyphae (Fig. 5).

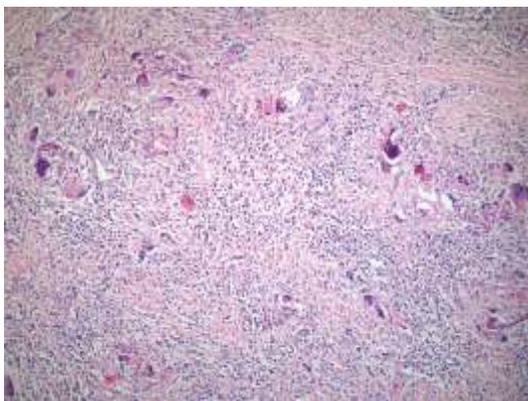


Fig. 5: Histopathological Picture showing Epitheloid Granuloma with Giant Cells surrounded by Dense Hyalinized Fibrous Tissue and Giant Cells containing Septate Hyphae.

Large nerve fibers entrapped in inflammatory mass was seen. It was diagnosed as aspergilloma involving infraorbital nerve based on the histopathologic findings. The patient was started

on oral itraconazole 200 mg twice daily for four weeks. Patient was followed up for period of 2 years without any recurrence (Fig. 6).



Fig. 6: Post Operative Photograph showing No Recurrence.

Discussion:

Aspergillus is a spore-forming filamentous fungus which occurs as a saprophyte in soil and decaying vegetable matter and is spread by airborne transmission which is recognized histologically by its septate hyphae, which branch at a 45 ° angle [1]. Aspergillosis of the paranasal sinuses and orbit can be noninvasive or invasive. Noninvasive aspergillus infection manifests as allergic sinusitis or mycetoma, is confined to the sinuses and is usually seen in immuno competent individuals. It usually causes bony expansion, without invasion of tissue or bone destruction [2]. Aspergillosis of brain in immunocompetent hosts has been recently reported mainly from India, Pakistan,

Saudi Arabia, and other African countries [3, 7]. The reason in immunocompetent hosts is thought to be related to tropical hot and dry conditions, bad hygiene, and poor socioeconomic status. These environmental changes encourage proliferation of the *Aspergillus* organism with high rate of allergic and infectious rhinosinusitis in host may contribute to tissue invasiveness in immunocompetent patients [3]. The mechanism causing invasiveness of aspergillosis in immunologically competent hosts still remains unclear, but is thought to be unrecognized or poorly characterized qualitative cellular or sub cellular immunodeficiency. Our patient was an immunocompetent lady of low socioeconomic status having environmental exposure of dust.

Paraesthesia or anaesthesia in the area of the trigeminal nerve distribution is one of symptoms of perineural extension [5]. Yohai RA *et al* have in 1994 reported facial numbness as the first symptom in 7% of patients with mucormycosis in large case series and advocated paraesthesia as a less recognised but an early symptom of fungal infection which was similar to our case [4].

Safdar A *et al* in 2002 have reported a case of perineural extension of invasive mycosis by *Aspergillus fumigatus* in an immunocompetent

patient [6]. Perineural spread is a known entity in head and neck malignancies, in which the tumour spread along the endoneurium or perineurium. It is rarely described as a mode of spread in invasive aspergillosis [5].

The prognosis of aspergilloma is generally good [1]. Medical therapy has limited role in treatment of aspergilloma [8]. Antifungal chemotherapy with itraconazole, voriconazole, or posaconazole provides some therapeutic benefit with minimal risk and also improvement in symptoms and stabilization of radiologic findings [9]. In cases of single aspergilloma surgical resection or intracavitary antifungal therapy may be appropriate in selected patients. Long-term, perhaps life long, antifungal treatment is required for chronic cavitary pulmonary aspergillosis [10].

Conclusion:

Early diagnosis and treatment is important in aspergilloma. Since the symptom is usually non-specific, radiographic study is nonspecific; excision biopsy should be performed for all cases. Aspergilloma should be kept as one of the differential diagnosis of any infraorbital mass lesions even in immunocompetent patients. Long term follow up is required as chances of recurrence are there in fungal infections.

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