CASE REPORT

AGGRESSIVE ALLERGIC FUNGAL SINUSITIS MANAGED COMPLETELY BY ENDOSCOPY

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ABSTRACT: Aggressive Allergic fungal sinusitis with bony destruction, deviation of eye, proptosis & anosmia required extensive surgical approach. However with newer modalities like CT scan & MRI imaging, Endoscopic staging & biopsy, FESS with powered instrumentation (microdebrider) external approaches can be avoided. CT & MRI imaging helps to determine nature, extent of disease & planning of surgical approach. Conservative endoscopic approach is now the treatment of choice for such aggressive fungal sinusitis. **CASE REPORT:** We are presenting a case of 29 year old male with aggressive AFRS with nasal obstruction, anosmia, bony destruction, deviation of eye & proptosis. CT, Biopsy & histopathology were suggestive of AFRS. Excision of AFRS was done by Endoscopic sinus surgery with post op steroids. **CONCLUSION:** Aggressive fungal rhinosinusitis presenting with nasal mass, bony destruction, deviation of eye & proptosis can be managed best with Endoscopic sinus surgery. CT, Endoscopic staging, biopsy & Histopathology are important diagnostic tools.
KEYWORDS: AFRS, fungal, allergy, sinus endoscopy.

INTRODUCTION: Allergic fungal rhinosinusitis is defined as an immunocompetent patient with an allergy to fungus.[8,9] It is non invasive fungal sinusitis. 7% of all chronic rhinosinusitis cases requiring surgery have been diagnosed as AFRS.

The fungi responsible for this hypersensitivity reaction reside in mucin & provide continued stimulation for the same. Has a similar etiology like allergic bronchopulmonary aspergillosis. Common pathogen responsible are Dematiceous species, bipolaris (Curvularia, Alternaria) rarely Aspergillus.[8]

Commonly occur where the climate is warm and humid but warm dry climate can also cause. History, clinical evaluation, blood eosinophilia, IgE level, allergic skin test, CT scan, MRI, endoscopy & biopsy are required for diagnosis. Surgery is treatment of choice for such extensive disease with or without systemic / local steroids. We are presenting a case of allergic fungal rhinosinusitis in a 29 year old male with diagnosis and complete endoscopic treatment.

CASE REPORT: 29 year Muslim male (fig. 1) leather industry worker presented with bilateral nasal obstruction, mucopurulent nasal discharge, recurrent rhinitis, anosmia from 8 years. Swelling and downward, lateral & outward deviation of eye, watering from eyes, frontal headache from 6 years. No blurring, diplopia or diminished vision. He was immunocompetent, no addiction in history. Physical examination showed swelling over nose, pinkish polypoidal mass involving left nostril and right nostril multiple polyps, S shaped septal deviation, mucopurulent discharge present. Air blast was absent on left side and diminished on right side. Cottle's test negative.

Smell test was suggestive of anosmia. Probe test revealed sensitive mass, not bleeding on touch. Probe cannot be passed laterally and above. Posterior rhinoscopy showed mass just reaching nasopharynx. Sensation over face present bilaterally. Left eye was deviated downward, laterally and outward. Proptosis present with normal extraocular movements, normal vision on Snellen's chart.

His routine blood investigation showed peripheral eosinophilia rest all normal. CT PNS with Orbit revealed soft tissue density opacification of bilateral maxillary sinuses along with ethmoid, frontal and sphenoid sinusitis. Left frontal sinus is hyperpneumatised, large and fully opacified. Both nasal cavities involved, mass extending to posterior choana and nasopharynx. Erosion of floor of sella turcica, sphenoethmoidal recess, both lamina papyracea with subtle protrusion of abnormal soft tissue into left orbital cavity. Bilateral panpolyposis with superadded fungal infection inspissated secretions (double density) (Fig. 2). Nasal endoscopy showed polypoidal mass (Fig. 3) more on left side than right. Biopsy taken was suggestive of allergic fungal sinusitis. Final surgery was done under GA by endoscopic sinus surgery & polypoidal mass removed along with allergic fungal muck (secretion) (Fig. 4) completely from all the sinuses. Left frontal sinus was very large and full of allergic muck cleaned completely. Histopathology confirmed the diagnosis of allergic fungal sinusitis (Fig. 5). No fungal growths on KOH mount.

Patient was given course of antibiotics, anti inflammatory, antihistaminics, oral steroids in tapering doses for 2 week. Nasal pack removed after 2 days, alkaline nasal douching given for one month. Patient followed on 1 wk, 3 wk, 1 mth, 3 mth, 6 mth, 1 year later; cavity is well epithealised no evidence of residual or recurrence noted. The patient fully recovered of its nasal and ocular symptoms.

DISCUSSION: The role of fungi in the nose & sinus cavities is unclear, even if in some healthy individuals fungi can be cultured from nasal secretion.[4,5,6] However there is a poor understanding of fungi simply as part of normal flora.[7] Fungi are normally found in nature as spores so it is reasonable to have fungal colonization in the nasal cavity. Ponikau et al [5] discovered > 90% of controls have fungi present in the nasal mucosa. De shazo[1] classification suggests AFS frequently occur in immunocompetent atopic patients. Mostly caused by Bipolaris species, curvularia lunata, aspergillus fumigatous. In histopathology fungal elements are present in eosinophil rich dense mucoid material.

Pathophysiology two hypothesis 1) inhalation of ubiquitous fungi in atopic patient provokes an antigenic stimulus & on inflammatory response of mucous membrane[5] results oedema and associated with production of allergic mucin (defined as thick green to grey lamellate of dense inflammatory cells mostly eosinophils in various stages of degranulation, charcot leyden crystals & fungal hyphae).[10] Eosinophilic mucin with or without fungus is more appropriate term.
2) Hypothesis concerning the presence & production of microbial T cell superantigen.[12]

Staging system for rhinosinusitis most commonly described by Lund & Kennedy, Mackay.[22] Diagnostic criterias are still under debate[13]

Stage	Endoscopic findings	Diagnostic criteria
0	No mucosal oedema/ allergic mucin	Type I hypersensitivity on history, skin test, serology
1	Mucosal oedema +- allergic mucin	Nasal polyposis with characteristic CT signs
2	Polypoid oedema +- mucin	Positive fungal stain/ culture
3	Polypswith fungal debris or mucin	Eosinophilic mucus with fungus. No tissue invasion, charcot layden crystals, peripheral eosinophilia, radiographic bone erosion

AFRS is common in young immunocompetent adults approx. 30 year of age. Male and female equally affected. Bilateral disease more common than unilateral. Patient presents with multiple polyposis, nasal obstruction. Obstructed airflow to olfactory area leads to hyposmia or anosmia.

RAST and skin test appears to be minimal allergy test for diagnosis. Total eosinophil count, total serum IgE, antigen specific IgE & IgG are suggested to reinforce the diagnosis.[2, 17, 18] culture is necessary to identify the actual fungal agent but no growth is frequently observed. Presence of hyphae in mucin associated with eosinophils is one of the main criteria for diagnosis. CT & MRI are preferred modality of imaging.[20] CT gives more bone details than MRI.[21] MRI is only indicated when complications like intracranial or orbital extension & skull base erosion. MRI also differentiates the tumor from retained secretions or inflammation. CT shows polyps involving sinonasal cavity, opacification of sinuses bone expansion & erosion, expanding inflammatory lesion, no direct invasion of dura/ periorbita.

Endoscopy supported by appropriate imaging to confirm the diagnosis, relevant anatomy & disease extent. Endoscopy gives excellent visualization of disease, origin, extent and anatomical abnormalities. Culture of discharge and biopsy for diagnosis is possible at same time.[20]

Treatment of choice for non invasive AFRS is preferably surgical. Endoscopic approach is very helpful i.e., attempting to reverse pathophysiological process by conservative surgery in defined areas dictated by disease. Treatment for invasive fungal sinusitis requires radical surgery and systemic antifungals. Treatment is controversial removal of all mucin is recommended by all in combination with medical therapy. Recurrence is quite common. Recent advances in FESS involve computer assisted navigation and powered instrumentation (microdebrider),[26] image guided surgery. Microdebrider minimizes inadvertent mucosal trauma & stripping.

Removal of mass without altering the specimen for hisopathology, rapid post operative healing, minimal crust & synaeche formation are possible by microdebrider. Medical line of management includes systemic and local steroids. Prednisolone is a commonly used systemic steroid and topical steroids can be given for at least 1 year. Corticosteroids have broad range of anti-inflammatory effects, inhibits total lymphocytes & eosinophils, cytokine synthesis.[23] Systemic steroids reaches all parts of nose, sinuses, olfactory cleft & middle meatus improves the sense of smell.[24] Before surgery oral steroids are given for about 5–7 days to shrink and decrease the

vascularity of polypoidal mass. Long term post-operative steroid sprays increases recurrence time.[24] Topical and systemic antifungal therapy are not actually recommended.

Immunotherapy has shown to give beneficial effects in patients with AFRS & recurrent polyposis.[25] Immunotherapy was studied by Mabry et al[19] in the form of intranasal injection of fungal antigens, positive non fungal antigens every weekly in first year, then monthly for 3 years. This reduces necessity of systemic and local steroids.[20]

CONCLUSION: Allergic fungal rhinosinusitis rarely present with complications but aggressive ARFS mostly present with complications. Our case had bilateral sinonasal polypoidal masses with erosion of floor of sphenoethmoidal recess, floor of sella turcica, lamina pyparacea with subtle protrusion of abnormal soft tissue into left orbital cavity. Patient have large hyper pneumatised left frontal sinus full of disease. CT showed soft tissue mass with superadded fungal inspissated secretions in left nostril along bilateral panpolyposis and large diseased left frontal sinus. CT was conclusive of AFRS with bony & orbital erosion. Definitive diagnosis was possible on CT, endoscopic biopsy & histopathology. In this case we performed endoscopic surgery for complete excision of disease.

Peripheral eosinophilia, Allergic tests, CT, MRI, endoscopy, biopsy & histopathology are important diagnostic tools. Endoscopic staging of disease with biopsy, CT prevents extensive external surgical approach.

This concludes that aggressive AFRS with bone erosion and orbital complications can be safely and completely managed by endoscopic sinus surgery approach.

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FIG. 1: Patient of AFRS with downward, outward & lateral deviation and proptosis of left eye

FIG. 2: CT PNS: shows soft tissue mass With double density (concretions), large hyperpneumatised Left frontal sinus, orbital erosion & invasion

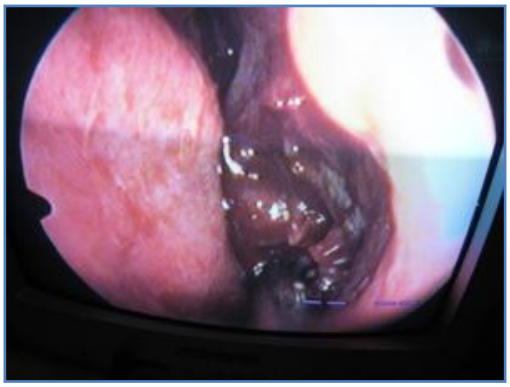

FIG. 3: Endoscopic view of left nasal polypoidal mass

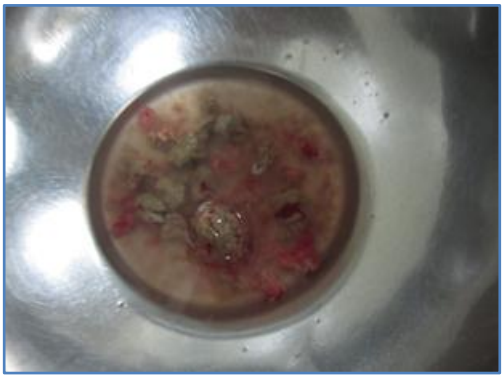

FIG. 4: Surgical specimen of sinonasal mass with allergic fungal secretions

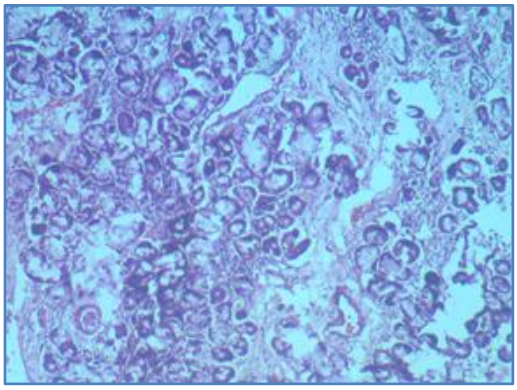

FIG. 5: Final histopathology slide showing Inflammatory eosinophil infiltrate with allergic fungal mucin

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