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Understanding paediatric allergic fungal sinusitis: Is it more aggressive?

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ABSTRACT

Objective: To study and characterize the features of AFRS in children as compared to adults.
Methods: 50 consecutive patients of AFRS attending our outpatient department were included in the study from July 2011 to December 2013. They were divided into two groups (A and B) according to age being ≤14 years and >14 years. Clinical history and examination included anterior rhinoscopy, SNOT 20 scores, CT of Nose and PNS (para nasal sinuses) (Lund Mackay scores), diagnostic nasal endoscopy (Kupferberg's grades), punch biopsy from nasal polyp, serum IgE, absolute eosinophil counts (AEC) and Aspergillus skin hypersensitivity test was done in all patients for conformation of AFRS. Bent and Kuhn's criteria were used for diagnosis. Sweat chloride levels were done in all patients of group A.
Results: Group A had 12 patients and group B had 38. Mean duration of symptoms was significantly less in children as compared to adults ($p < 0.05$). All patients of both groups had nasal polyposis at presentation. Unilateral disease and multisinus involvement was more common in children (6/12) as compared to adults. Proptosis (2/12) and telecanthus (4/12) was more common in children (group A) as compared to adults (group B). LM (Lund Mackay) scores and serum IgE were significantly high in children as compared to adults. Follow up CT scans showed early evidence of recurrence in children as compared to adults ($p < 0.05$).
Conclusion: AFRS was seen to be more aggressive in children with increased fungal load when compared with adults. Typically, AFRS in children was less responsive to treatment with increased recurrence rates.

1. Introduction

Allergic fungal rhinosinusitis (AFRS), initially described by Katzenstein [1], is caused by a hypersensitive response to fungi present in the paranasal sinuses. It occurs in 5%–10% of adults with chronic rhinosinusitis who require surgery [2]. Initially it was thought that *Aspergillus* was the primary causative agent, but later dematiaceous fungi like *Alternaria*, *Bipolaris*, *Curvularia*, and *Drechslera* were also implicated in causation of AFRS [3,4]. This is mostly seen in young atopic adults in warm and humid climates [5].

Patients of allergic fungal rhino sinusitis present with nasal polyposis at time of diagnosis. They also complain of symptoms like nasal obstruction, congestion, purulent or clear rhinorrhoea, anosmia, and headache and allergic mucin in nasal endoscopic examination [6,7]. 63% of AFRS patients have history of allergic rhinitis and 53% have associated history of asthma [8]. Characteristic radiologic findings include serpentine patterns of hyper dense areas in non-contrast scan of PNS [9]. Histopathological findings include eosinophil, allergic mucin, Charcot Layden crystals and presence of fungal hyphae without any evidence of mucosal and vascular invasion [6].

Surgery followed by adjuvant therapy has always been considered the gold standard in the therapy of AFRS. Adjuvant therapy in form of topical and oral steroids, topical and oral antifungal and immunotherapy have been useful adjuncts in the treatment of AFRS. Recurrence rates vary from 10 to 79% in adults [9].

The behaviour of the disease is well studied and documented among adults. AFRS also present in the paediatric age group. There is paucity of data in literature regarding nature, clinical course and its recurrence in children. In this article we intend to study and characterize the features of AFRS in children as compared to adults.

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2. Material and methods

This study was a single centre prospective study done from July 2011 to December 2013. It included 50 consecutive histologically diagnosed cases of AFRS. Informed consent was taken for the study and treatment from all patients. The patients were divided into two groups: paediatric group or Gr A (age ≤14 years) and adult group Gr B (age >14 years). All the cases of AFRS were evaluated with a detailed clinical history and head neck examination including anterior rhinoscopy. The symptomatology scores were evaluated as per SNOT scores. Computerized tomography in axial and coronal planes of Nose and PNS was done in all patients, evaluated as per Lund Mackay scores. Diagnostic nasal endoscopy with punch biopsy from nasal polyp was done in all patients. Nasal endoscopy findings were graded based on Kupferberg's grades [15]. Additionally total serum IgE, absolute eosinophil counts (AEC) and Aspergillus skin hypersensitivity test was done in all patients. Skin test showing a wheal of diameter >8 mm and AEC >400 were taken as significant. Final diagnosis was done using Bent and Kuhn's criteria [10]. Sweat chloride levels were done in all the patients of paediatric age group, i.e. group A to look for any evidence of cystic fibrosis which is an important cause of nasal polyposis in children.

Both the groups were taken for functional endoscopic sinus surgery after diagnosis and preliminary investigations. In all cases the surgical procedure included adequate decongestion of the nasal cavities, opening of all the sinuses namely maxillary, anterior and posterior ethmoids, frontal and sphenoid sinuses with removal of polyps and the fungal muck and mucin from all these sinuses with preservation of the normal mucosa. Operative findings were recorded and surgical specimens were sent for fungal smear and histopathological examination. Histological evidence for presence of allergic mucin, eosinophils and fungal hyphae and tissue invasion was recorded. Any patient having evidence of tissue invasion were excluded from analysis.

Postoperatively both the groups were given [6] weeks of oral steroids in tapering doses. Patient in both the groups were given oral prednisolone 1 mg/kg for 7 days, 0.8 mg/kg for 7 days, 0.6 mg/kg for 7 days, 0.4 mg/kg for 7 days, followed by 0.2 mg/kg for 14 days. This constituted the 6 weeks of steroid therapy which was common in both the groups. All patients in both the groups were advised for regular alkaline nasal douching twice a day.

Follow up was done at 1st, 6th, 12th, 24th weeks post-surgery. On each visit all patients were taken for office based nasal endoscopies to look for the cavity conditions and signs of recurrence. SNOT 20 symptoms scores and nasal endoscopic grades were determined at 1st, 6th, 12th week follow up in the post-operative period. Computerized tomographic scans of nose and paranasal sinuses were done at 12 weeks, 24 weeks in the postoperative period. Total IgE levels were done at 6 weeks post-operatively.

Both the groups were compared for the above parameters to see for the effect of therapy and to compare their responses in children as compared to that of adults. Patients in both the groups are under our continuous follow up till the present date.

Statistical analysis was done using IBM SPSS statistics V20.0. Parametric data were compared with t test, nonparametric data were compared with chi χ^2 test and Fischer exact test.

3. Results

A total of 50 patients of AFRS were included in the study from July 2011 to December 2013. Group A (Paediatric group) included 12 patients and group B (Adult group) included 38. Mean age of patients in paediatric group was 10.17 (6–14 years) and in adult group was 35.92 years (15–65 years). Paediatric group had 8 male and 4 female while adult group had 21 males and 17 females.

Mean age of duration of symptoms was 8.67 months in paediatric group and 28.50 months in adult group. This suggests an early progression of disease in paediatric age group. This difference was statistically significant (p value 0.000). Nasal discharge, post nasal drip, nasal obstruction and allergic symptoms were the most common symptoms. One patient in paediatric group had h/o sensitivity to aspirin while no patient in the adult group had hyper sensitivity to aspirin. Nasal polyposis was the most common sign. Unilateral nasal polyposis was seen in 6/12 (50%) patients of the paediatric age group, while in the adult group only 9/38 (23.6%) patients had unilateral nasal polyposis at time of presentation ($p = 0.145$). Bilateral nasal polyposis was thus found to be more common in adults with AFRS. Proptosis was seen in 4/50 (8%) patients of AFRS (2 in each group). Tele canthus was seen in 6/50 (12%). Tele canthus was more common in paediatric group 4/12 (33%) while in adults only 2/38 (5.2%) had telecanthus ($p = 0.023$), suggesting an aggressive course of disease in children (Table 1).

Mean SNOT score was 30.58 ± 13.15 (6–51) in the paediatric group and was 33.05 ± 10.36 (10–56) in the adult group. The groups were statistically similar in terms of SNOT scores ($p = 0.504$). Nasal endoscopic grades of the patients at the time of presentation was also similar in both adult and paediatric groups ($p = 0.247$). Mean Lund Mackay radiologic score in the paediatric group was 14.17 ± 4.42 (8–21) while it was 18.26 ± 4.88 (3–24) in the adult group. However it was interesting to find increased evidence of skull base and orbital erosions among the computerized tomographic scans in the paediatric age group compared to the adults (Figs. 1 and 2). Serum IgE values was also found to be significantly higher in the paediatric group with mean value of 6143 ± 3049.86 (1400–10,000) as compared to adult group having mean value of 4154 ± 2834.04 (70–9600) ($p = 0.043$). The above findings suggest an aggressive nature of disease in the children as compared to adults. We also found no significant difference for absolute eosinophil counts between paediatric group and adult group ($p = 0.429$) (Table 2). Sweat chloride levels was done in all the patients of paediatric group as a screening for cystic fibrosis. It was found to be in the normal range in all the patients which suggest that none of our children's with AFRS had coexisting cystic fibrosis.

Endoscopic sinus surgery for clearance of disease was done in all 50 patients. Post-operative fungal smear was positive in 10/12 (83%) in paediatric group while in adults 36/38 (94%) was positive for septate hyphae though this difference between the two groups was found to be statistically non-significant (p value 0.179). All 50 patients had histopathologic diagnosis of AFRS which reinforced initial diagnosis obtained by office based diagnostic punch biopsies. Fungal culture was positive for Aspergillus in 10/12 (83.3%) in paediatric group as compared to 24/38 (63.2%) in the adult group. This difference in fungal culture was statistically significant ($p = 0.045$) suggesting higher fungal load in children as compared to adults (Table 1). All the positive fungal cultures were

Table 1
Presentation and mycological results in both the groups.

	Group A		Group B	
Age	<=14 years		>14 years	
No. of patients	12		38	
Proptosis	2		2	
Telecanthus	4		2	
Unilateral disease	6		9	
Post op fungal smear	+	10	+	36
($p = 0.179$)	–	2	–	2
Post op fungal culture	+	10	+	24
($p = 0.045$)	–	2	–	14