Case Report

Sinonasal Sarcoidosis: A Case Report

K Afroza[1], A Gulshan[2], K Nabila[3], C Nurun Nahar[4], R Abdur Mohammad[5], A Mohammad[6].

Abstract:

Sarcoidosis is a chronic granulomatous disease of unknown etiology which principally affects the lower respiratory tract & lungs. Sarcoidosis in the head & neck region is infrequent. Isolated sino nasal sarcoidosis without pulmonary involvement is rare. ***Case:*** An 18 years old male patient presented with the complaints of nasal blockage, purulent nasal discharge which was occasionally blood stained for 6 months, deformity of nose, swelling of face & lips for 4 months & watering of eyes for same duration. Endoscopy of nose revealed intra nasal mucosal thickening which was friable & bleeds on touch. The diagnosis of sino nasal sarcoidosis was made by histopathological examination of nasal biopsy specimen.***Conclusion:*** Sino nasal sarcoidosis is a disease of diagnostic challenge to the clinician as its mimicking clinical features may be misleading & cause delay in definitive diagnosis. In the current case report, we presented a case of sino nasal sarcoidosis presenting as chronic rhino sinusitis.

Bangladesh Journal of Medical Science Vol. 15 No. 04 October'16

Introduction:

Sarcoidosis is a multi system chronic granulomatous disease of unknown etiology which commonly affects young & middle aged females with involvement of multiple sites of body principally lower respiratory tract, skin, liver, eyes, spleen, peripheral lymph nodes & neural structures.[1]Sino nasal involvement in absence of pulmonary disease is extremely rare.[2] The characteristic features of sarcoidosis is non caseatinggranuloma detected by tissue biopsy.

Case Report:

Mr. Ripon, 18 years old farmer by occupation from Rajshahi, presented with the complaints of nasal blockage with purulent nasal discharge (occasionally blood stained) for 6 months, deformity of nose, swelling of face & lip & watering from right eye for 3 months, occasional dry cough & shortness of breath for same duration. With these complaints he initially received treatment from local doctors with no improvement of his symptoms. Then he got admitted at RMCH & consulted with an ENT specialist where he was operated (rt. lateral rhinotomy) & FNAC report of nasal tissue revealed giant cell tumor. FNAC was followed by histopathology the result of which showed chronic granulomatous inflammation & one of their differential diagnoses was tuberculosis. Due to controversy the patient was referred to NICRH for further management.

We first attended the patient at OPD at NICRH on 21.04.2011. On physical examination, patient had diffuse swelling over face more on rt. side. The swelling involved forehead, nose, lips & rt. chick. There was a depression just below the root of the nose. On palpation, the swelling was doughy, non tender & overlying skin was erythematous. Examination of nasal cavity showed mucosal swelling of nasal septum & lateral nasal wall. There was no lymphadenopathy. Examination of other

1. Dr. Afroza Khanam. Assistant Professor. Department of Otolaryngology. Green Life Medical College & Hospital.
2. Dr. Gulshan Akhtar. Associate Professor. Department of Paediatrics. Green Life Medical College & Hospital.
3. Dr. Nabila Khanduker. Assistant Professor. Department of Surgery. Green Life Medical College & Hospital.
4. Dr. Nurun Nahar Chowdhury. Associate Professor& Head. Department of Psychiatry. Green Life Medical College & Hospital.
5. Dr. Mohammad Abdur Rahman. Consultant, Medicine. Jinjira 20 bed Hospital, Keranigonj.
6. Dr. Mohammad Asifuzzaman. Associate Professor & Head. Department of Dermatology & Venereology. Green Life Medical College & Hospital.

Corresponds to: Dr. Afroza Khanam. Assistant Professor, Deptt. of Otolaryngology, Green Life Medical College & Hospital, Dhaka, Bangladesh. Email: afrozakhanam88@gmail.com

system revealed no abnormality.

At NIRCH, FNAC was done from nasal mucosa which showed significant number of giant cells with moderate number of fibroblast. The following investigations were also done:

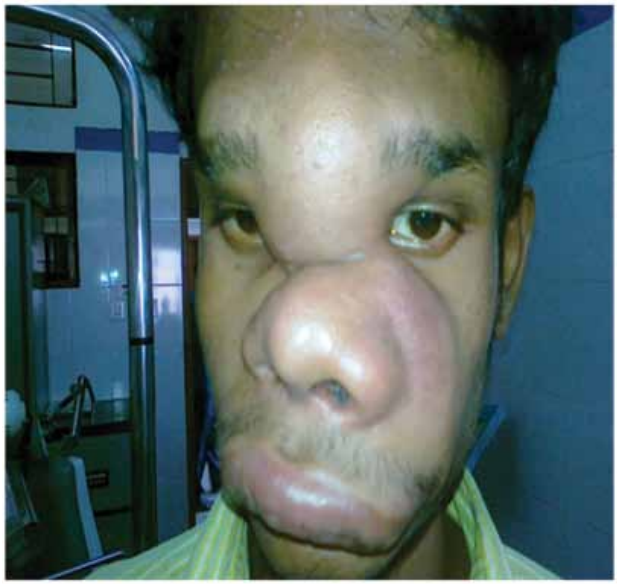

Figure 1: Sinonasalsarcoidosis (before treatment)

1. CBC with PBF: Normal
2. Liver function test: Normal
3. Renal function test: Normal
4. X ray chest P/A view: Normal study
5. X ray PNS (OM view): mucosal thickening of rt. maxillary antrum.
6. CT scan of nose & PNS: diffuse soft tissue swelling of upper lip, nose & fore head. No bony destruction & no cervical lymphadenopathy.
7. P- ANCA & C- ANCA: within normal limit
8. Serum calcium: within normal limit
9. MT: Negative

We also reviewed previous histopathology slide which showed reparative giant cell granuloma. Naso endoscopy showed intra nasal mucosal thickening which was friable & bleeds on touch. Nasopharynx was normal. Tissue was obtained from nasal cavity for histopathology which showed chronic non specific inflammation. So finally following history, physical examination & relevant investigations our diagnosis was sarcoidosis of nose & Para nasal sinus. The patient was treated with tab. Prednisolone 60 mg/day in divided doses for 1 month. Nasal douching with normal saline was advised to maintain nasal hygiene. The patient reported after 1 month of completion of corticosteroids. On examination during follow up visit there was complete resolution of previous symptoms. Then we advised him to continue prednisolone in same dose with tapering. The patient was advised to come to follow up visit after 2 months. However he didn't report after 2 months but informed us over phone that he was asymptomatic & better than before.

Discussion:

Sinonasal sarcoidosis is a rare, yet clinically challenging manifestation of the systemic disease of unknown etiology.Sarcoidosis can occur at any

Figure 2: Resolving torticollis after 1 week of IV Unasyn.

age but is most frequent in young adult &is rare among children.The disease typically involves the lower respiratory tract including lungs with rare involvement of head & neck region & can be of diagnostic challenge for the otolaryngologist.[1,2] Nasal involvement of sarcoidosis is described by Bocck in 1905.

In patients with sarcoidosis, sino nasal involvement may occur, but isolated involvement is rare.[3] Wilson et all at their study noted nasal involvement in 21 (2.8%) out of 750 patients with sarcoidosis confirmed by histopathology.[4] Whereas McCaffery&Mc Donald reviewed the record of 2319 patients diagnosed with sarcoidosis & found nasal mucosa involvement in 17 cases (<1%).[1] A recent report by Zeithin et al stated 4% incidence of nasal involvement in 159 patients with sarcoidosis.[5] The afore mentioned authors also noted that the actual incidence might be higher. The most frequent sites of nasal involvement are nasal septum, inferior turbinate followed by paranasal sinuses, nasal bone, cartilage & subcutaneous tissue.[3] Isolated sino nasal sarcoidosis with intra cranial extension was reported by OyDessouk in 2008.

The prognosis of patients with sarcoidosis is generally good, where as mortality rate are reported to vary from 5-7%. In our study, sarcoidosis is confined in nose & paranasal sinus with no other systemic organ

involvement. Regarding treatment, corticosteroids remain the mainstay of treatment. Nasal topical steroid application can help halt progression of isolated nasal involvement in certain cases.[6,8] However treatment with systemic corticosteroids is usually needed. Surgery should be the last treatment option. Patients should be followed up carefully over long term since there is tendency of recurrence & delayed systemic involvement.

Conclusion:

Sino nasal sarcoidosis is a rare condition. Otolaryngologist should consider this condition in differential diagnosis of chronic non remitting sino nasal complaints since signs & symptoms of sarcoidosis can mimic other common disorders. Further diagnostic tests are necessary to exclude other granulomatous disorder. Corticosteroids remain the mainstay of treatment. Relapse & chronicity are frequent after tapering or discontinuation of drug & thus these patients require long term follow up.

Further studies are necessary to determine the effectiveness of treatment to evaluate the prognostic factors & to access the natural history of sino nasal sarcoidosis.

Ethical Approval: This case report with photograph was published after getting approval from Ethics Committee of Green Life Medical College & Hospital.

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