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VARIANT PRESENTATION OF RHINOSCLEROMA: A CASE REPORT

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ABSTRACT: The name rhinoscleroma was first used by Von hebra & Kaposi. Most frequently effected chronic granulomatous lesion of the respiratory mucosa is rhinoscleroma. It mostly effects middle aged females. Commonly nose is involved spreading the disease to the other parts of the upper respiratory tract like larynx, nasopharynx, trachea and bronchi and sometimes maxillary antrum and orbit. It should be differentiated from other chronic granulomatous lesions like syphilis, midline granulomas and malignant lesions of the nose. Early diagnosis, surgical and medical treatment is likely to decrease the morbidity of the disease.

KEYWORDS: Klebsiella Rhinoscleromatis, nasal inflammatory disease, rhinoscleroma, upper respiratory tract infections.

INTRODUCTION: The name rhinoscleroma was first used by Von Hebra & Kaposi.⁽¹⁾ Rhinoscleroma is caused by gram negative encapsulated non- motile bacteria, klebsiella Rhinoscleromatis. Most cases are found in central America, Africa Middle East⁽²⁾ Indian subcontinent and Indonesia. Disease usually starts in the nose spreading to other parts like nasopharynx, oral cavity, larynx, trachea, bronchi, maxillary antrum and orbit. Laryngeal involvement may occur in almost half of the cases and hence the disease is also known as respiratory scleroma. Mikulicz established the non-neoplastic inflammatory nature of the disease.⁽³⁾ Von Frisch identified the causative agent of the lesion as gram negative-coccobacillus.⁽⁴⁾ Prolonged exposure is necessary for the manifestation of the disease. In the developing countries it is more common because of the poor hygiene and overcrowding, poor access of the antibiotics.

It occurs in 3 stages.⁽⁵⁾

Atrophic stage. 2. Granulomatous or proliferative stage.3. Cicatricial stage. Sites of predominance are nasal cavity, nasopharynx, larynx, trachea, bronchi and maxillary antrum.⁽⁶⁾ Here, we present a case with both nasal and maxillary antral extension. Histologically granulomatous tissue infiltrates the submucosa and is characterized by the presence of plasma cells, lymphocytes and eosinophils among which are scattered large foam cells (mikulicz cells)⁽⁷⁾ which have a central nucleus and vacuolated cytoplasm containing Frisch bacilli and russel bodies, the later resembling plasma cells and having eccentric nucleus and deep eosin- stained cytoplasm.

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CASE: A 34 year old male came with a complaint of right sided nasal obstruction since 14 months. Difficulty in breathing on the right side initially happened to be partial and progressed over 1 month to complete nasal block. It is associated with bloody discharge with associated pain in the right side of cheek. There is no history of difficulty in swallowing. On nasal endoscopy, irregular, pinkish mass of size 3x3x1 cm is seen in the floor of the right nasal cavity. Septum is in midline, turbinates couldn't be visualized.

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Fig 1

Fig. 2

Fig. 3

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Computerized tomography (CT) (Fig. 1 & 2) confirmed a unilateral mass in the right nasal cavity, no palatal bony destruction or lymphadenopathy was seen. Nasal endoscopy was done, biopsy was sent for histopathological examination which showed fragments of soft tissue showing several inflammatory cells and large histocytes with foamy cytoplasm without any evidence of infection, consistent with rhinoscleroma.

PROCEDURE: Under general anesthesia, patient kept in supine position, head end elevated. Endoscopic examination showed a single, pinkish irregular mass in the right nasal cavity, occupying the whole of the right nasal cavity extending into the right maxillary antrum. Endoscopically the mass was removed from the nasal cavity and maxillary antrum. Haemostasis was secured. Polythene tubes were kept in both nasal cavities for 2 weeks in order to maintain the patency of both nasal cavities. Post operatively clindamycin 150mg twice daily was given for 3 weeks. Airway symptoms showed a considerable improvement following surgical removal. Later, tetracycline 500mg BD was given for the next three weeks. In the next visit, patient's nasal cavity was very much patent as in a normal person.

DISCUSSION: Rhinoscleroma is caused by klebsiella Rhinoscleromatis. It manifests in 3 stages. Atrophic stage, proliferative stage and cicatricial stage. Clinically in the first stage patient complains of rhinitis and nasal discharge. Second stage is manifested as granulomas in the nasal mucosa with a possibility of spread to other areas of the airway tract. Scar formation is typically seen in the third stage which is otherwise called the cicatricial stage. Mode of transmission is by person to person contact. As the organism is of low infective nature, prolonged exposure is required for the disease manifestations to occur. In patients suffering from persistent rhinitis and nasal obstruction, rhinoscleroma should be suspected after excluding common diseases like tuberculosis, syphilis, wegeners granulomatosis and inverted papilloma. Histologically, Mikulicz cells are the histocytes that migrate to the places where neutrophil fail to destroy the klebsiella infection, there by engulfing the bacteria that causes the dilatation of the vacuoles. Russel bodies are the transformed plasma cells having a deeply eosin – stained cytoplasm with an eccentric nucleus. Positive culture of rhinoscleroma on MacConkey agar is diagnostic.^(8,9) MRI can be used in Hypertrophic stage which shows a soft tissue mass with mild to marked signal intensity in both T1 and T2 weighted images.⁽¹⁰⁾

As the disease is more common in young females with bilateral nasal block, this case report is being put forwarded due to uncommon presentation of unilateral nasal obstruction in a male patient. Medical treatment of rhinoscleroma is with tetracyclines, aminoglycosides and macrolide antibiotics like clindamycin. Initial surgical debridement prior to antibiotic therapy is reported to be useful in granulomatous stage. Dosage of the antibiotic is variable. But long term therapy is necessary for the adequate treatment of the disease. Treatment is prolonged for at least 6 weeks and must continue until at least 2 consecutive cultures from biopsied tissue are negative. Recurrence is common in 25% of cases in 10 years. Indications for the surgical de-bulking include, for airway patency and cosmetic purpose.

CONCLUSION: Rhinoscleroma is a disease caused by the klebsiella Rhinoscleromatis or Frisch bacilli which are gram negative coccobacilli. Infection usually presents either with chronic rhinitis or nasal obstruction. The disease is usually common in young middle aged females but can also occur in males

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as seen in this case. Obstruction caused by the presenting stage of the case could result in airway compromise. Histopathological examination is necessary for the conformation of the diagnosis, which typically shows macrophages and histiocytes. Medical treatment includes macrolides like clindamycin, tetracycline and aminoglycosides. Surgical de-bulking is necessary when the patency of the nasal cavity is compromised.

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