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PALATAL PUZZLE – A CASE REPORT OF ACTINOMYCOSIS

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ABSTRACT

Actinomycosis is per se one rare disease encountered today, even in the Developing World – Thanks to the widespread use of antibiotics. Cervicofacial actinomycosis is the most common form of disease and insofar its only presentation is in the form of a mass or lump in the jaw or neck which presents silently or as discharging sinuses. This 75yr old gentleman presented with a palatal lesion which turned out to be an Actinomycotic Ulcer – a totally unheard presentation. This stresses the importance of HPE in the diagnosis of even apparently simple cases.

Keywords: Palatal Puzzle, Cervicofacial Actinomycosis

CASE REPORT

A 75 yr old gentleman presented with complaints of pain and swelling of the Left cheek for over a month duration. The swelling had been almost static and not increasing much in size but the pain has been getting worse. He had noticed a Palatal ulcer at about the same time which was causing him pain on swallowing his food and during chewing. He also started having Disharge from his left nasal cavity which was not foul smelling or blood stained, which was more mucoid in nature. However there was no history of nasal obstruction, blurring of vision, loosening of teeth, restricted mouth opening.

There was also no recent history of trauma or fever. Patient was a diabetic for the past 7yrs on irregular treatment with oral hypoglycaemics. He denied history of

hypertension, contact with tuberculosis, asthma, CAD or other systemic diseases.

He also denied history of previous hospitalization and any history of blood transfucions or drug allergies.

- On General examination, his vitals were within normal limits.
- ENT Examination:
  - **External nose:** minimal swelling of the left cheek was noted. It was of a diffuse nature can be described as more of a cellulitis and no mass was made out separately. The swelling was not warm however deep tenderness was present on palpation. Sensation over the swelling and of the Infraorbital Nerve were intact.
- **Anterior Rhinoscopy**: minimal septal deviation to right was seen with the middle and inferior turbinatesappearing normal. The inferior meati appeared normal and no mass or eschar was seen.
- **Posterior Rhinoscopy**: Both choanae and the Eusttachian Tube orifices on both sides appeared normal and no mass or other lesions were seen.
- **Oral cavity:** A 4*4cm whitish slough covered, punched out, Pale eschar-like lesion was seen on the hard palate. This was not covered by mucosa, thus exposing the underlying bone. It was tender on palpation and there was no discharge from the lesion. Noother swelling or mass was seen. Oral hygiene was adequate and patient was edentulous.
- The oropharynx and the throat were normal with normal IDL findings.
- Diagnostic Nasal Endoscopy showed a deviated nasal septum to the right and a normal Right nasal cavity. In the Left nasal cavity there was defect in the nasal floor corresponding to the palatal ulcer and a similar pale slough covered raw area which exposed the underlying bone, had no mucosal covering, which was tender on palpation was encountered. Otherwise the Left nasal cavity also appeared normal. No blackish eschar or mass was seen inside the nose.
- CT scan showed regions of multiple fractures in the anterolateral, inferior and medial walls of both maxilla with thickening and sclerosis of the maxillary bone, The radiologist had suggested a Maxillary Osteomyelitis likely of Fungal etiology.

- With all these in mind, we also came to the working diagnosis of Nasal Mucormycosis or Invasive Fungal sinusitis.

We started the patient on IV high grade broad spectrum antibiotics like Piperacillin – Tazobactam and Metronidazole. His blood sugar (217mg/dl, urine acetone negative) was brought under control with Inj. Insulin – R12/ R12/ R8N10.

We were debating Starting Inj. Amphotericin empirically but were dissuaded by his creatinine values at 2.4mg/dl and Bl. urea at 58mg/dl.

We had the following differential diagnoses in mind:

- Maxillary osteomyelitis
- Mucormycosis
- Maxillary tumor
- Granulomatous diseases

We decided to continue the conservative management and to withhold antifungals till conclusive proof was obtained. Hydration was improved using IV fluids and his creatinine value started to fall.

Patient tested negative for HIV and HBSAg. However he was VDRL positive in dilution of1:21. His blood was processed for TPHA testing.

At the same time a swab was taken from his palatal lesion and it came out positive for *Candida albicans* with features of pseudohyphae and yeast forms.

And finally left with no other clues we scraped a portion of the periphery of the ulcer including the sloughed portions and sent it for biopsy where it turned out to be actinomycotic colonies which were very characteristic on HPE.

We continued these antibiotics for 2weeks. The patient responded with reduction in the severity of pain and apparent reduction in the size of swelling.

We started the patient on Inj. Crystalline penicillin 9million units per day as tds dosage and also on T. Doxycycline 100mg bd. We planned for a FESS surgery for exploratory and ventilatory purposes but patient declined consent and requested discharge. We sent him with T. Phenyl Methyl Penicillin 250mgtds for 2weeks, to continue T. Doxycycline and also suggested Betadiene gargles.

The Patient reported after 3weeks with his ulcer marginally better but his sugar and renal parameters and symptoms were normal. He was asked to continue the same treatment.

CLINICAL PICTURES

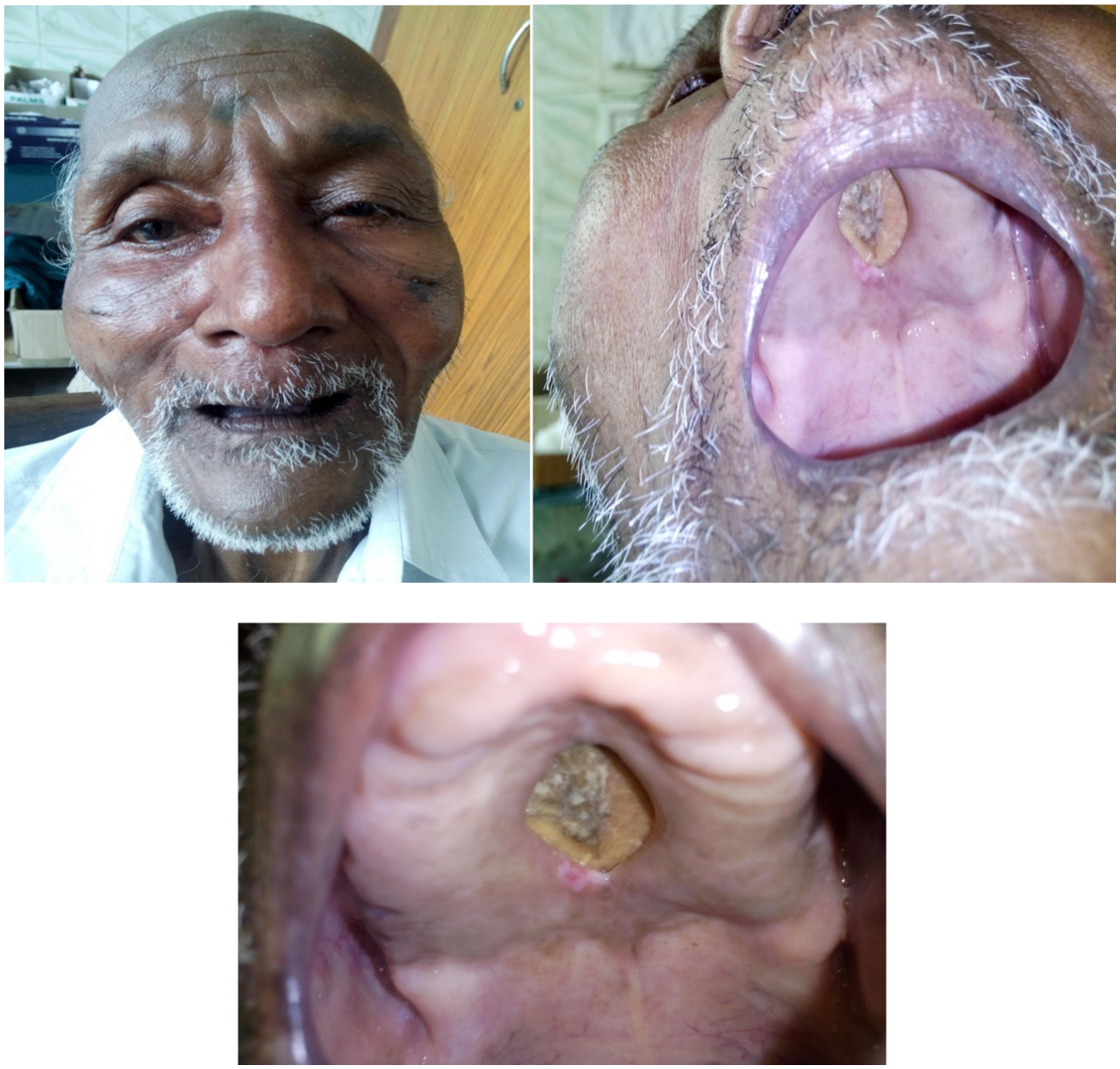


Figure 1: Pictures of the patient A, B, C: From top Left, A: picture of minimal and diffuse swelling Left cheek, B: Edentulous Patient with the palatal ulcer, C; Close-up view of the palatal ulcer – 4*4cm punched out pale ulcerative eschar like slough covered lesion.

CT IMAGING

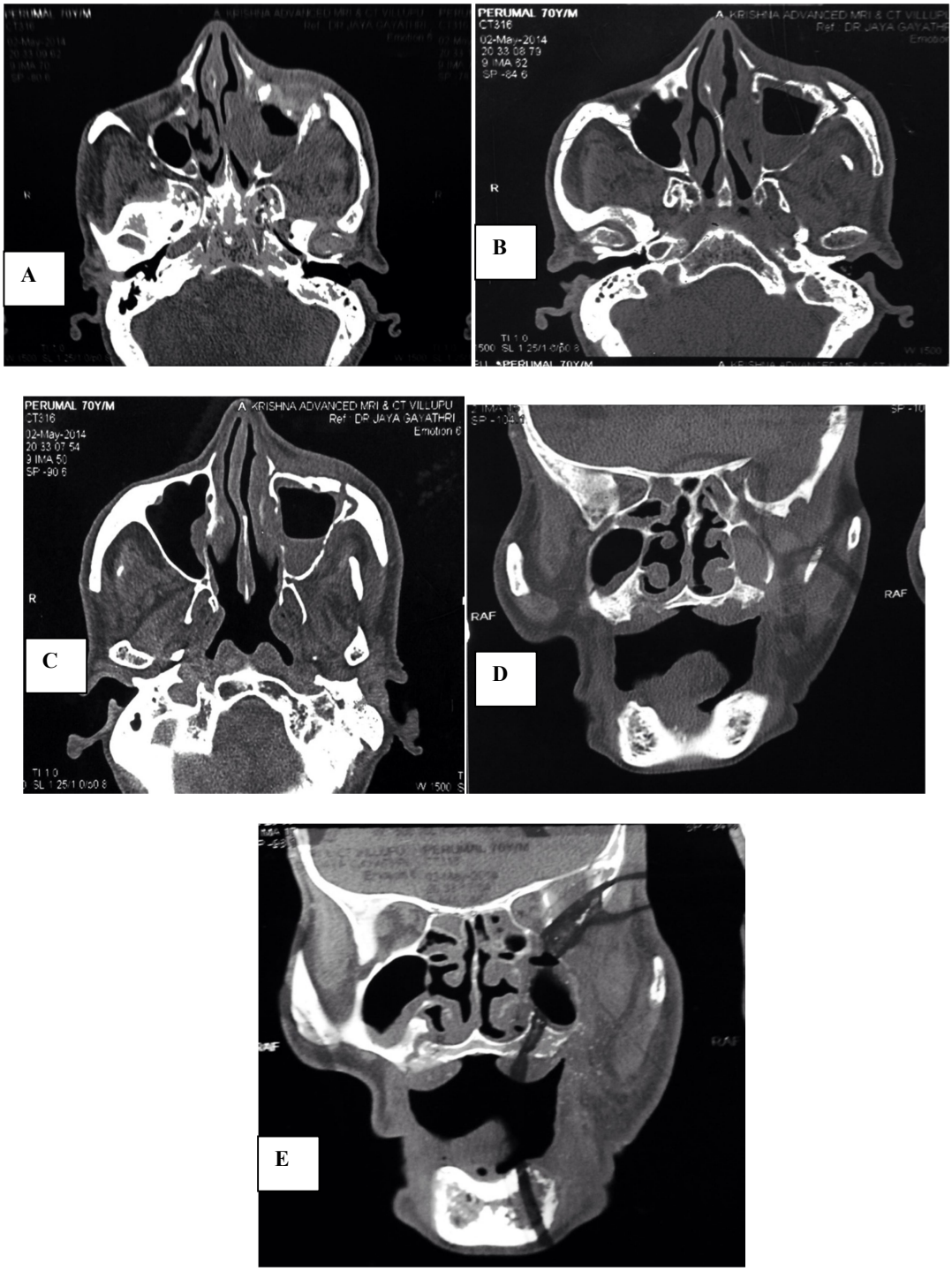


Figure 2: From Top left Clockwise, CT images of the patient showing:

- **thickening, bony sclerosis seen in B/L maxillary bones – alveolar process, palate, posterior wall**
- **Areas of fracture - ?pathological fractures – seen on Rt hard palate and lateral walls of both maxillae**
- **Areas of necrotic bone also seen**
- **Maxillary and ethmoid sinusitis**

DNE FINDINGS

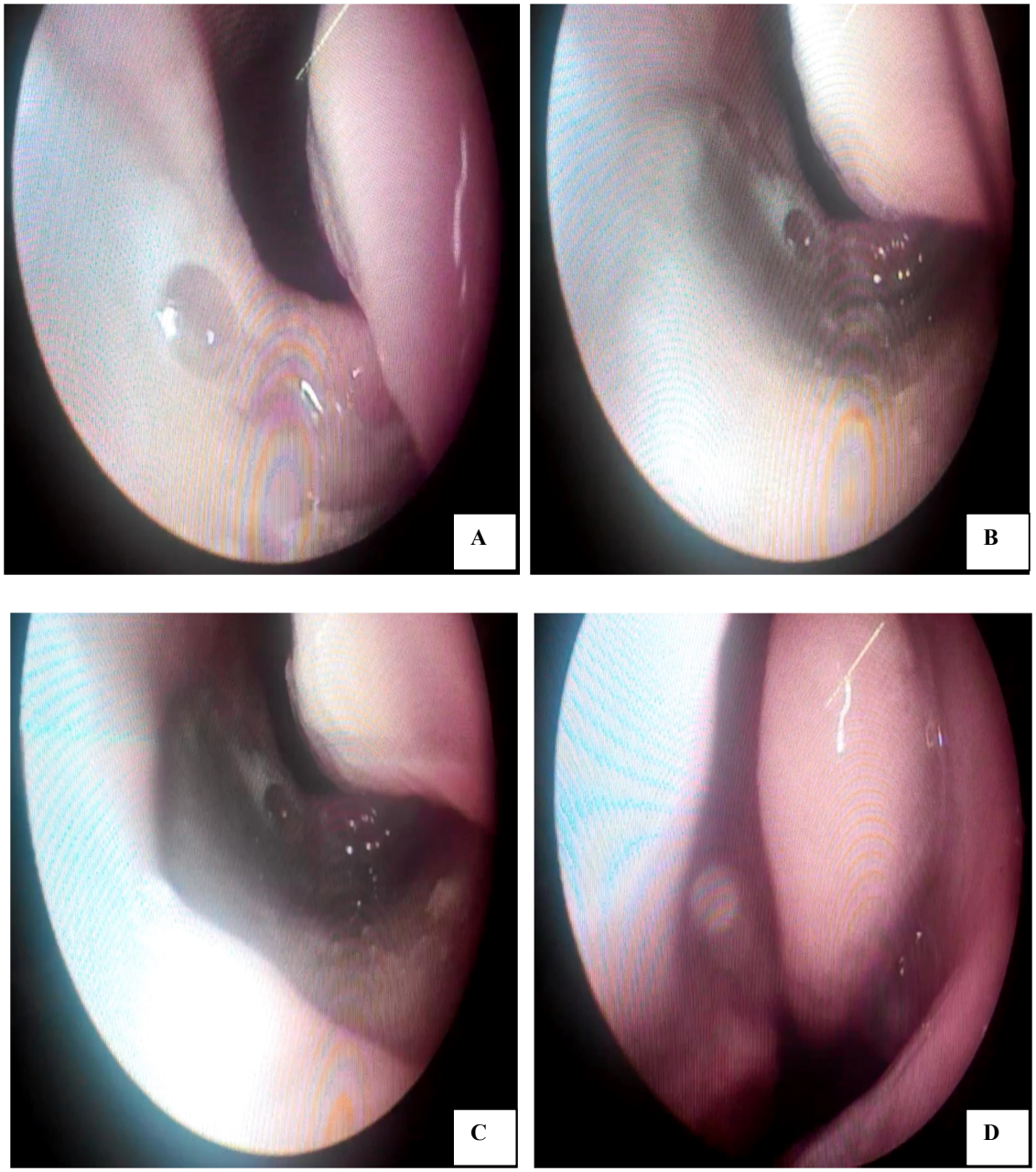
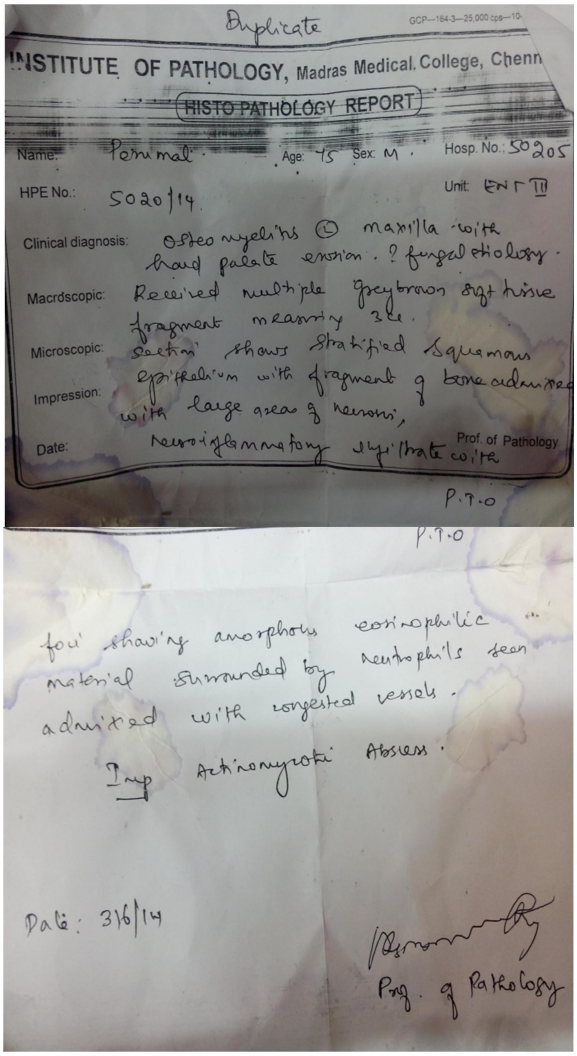


Figure 3: Deviated nasal septum to right, Right nasal cavity normal
Left nasal cavity: Defect in the nasal floor corresponding to the palatal ulcer. Similar raw area, exposing bone, tender on palpation, no mucosal covering, pale slough covered, no blackish eschar or mass seen. Left middle meatus normal

HPE PICTURE


Duplicate

INSTITUTE OF PATHOLOGY, Madras Medical College, Chenn

HISTO PATHOLOGY REPORT

Name: Perumal Age: 45 Sex: M Hosp. No.: 50205

HPE No.: 5020/14 Unit: ENT III

Clinical diagnosis: Osteomyelitis ⓛ maxilla with hard palate erosion. ? fungal etiology.

Macrdscopic: Received multiple greybrown soft tissue fragment measuring 3cc.

Microscopic: Section shows Stratified Squamous epithelium with fragment of bone admixed with large areas of necrosis,

Impression: Necroinflammatory hyperplastic c/w

Date: Prof. of Pathology

P.T.O

P.T.O

foci showing amorphous eosinophilic material surrounded by neutrophils seen admixed with congested vessels.

Imp Actinomycotic Abscess.

Date: 3/6/14

Prof. of Pathology

Figure 4: Duplicate of HPE REPORT,

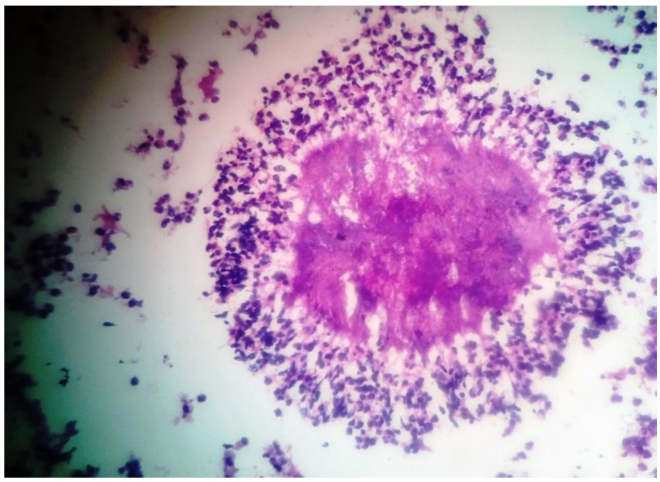


Figure 5: The picture of the slide from the microscope

DISCUSSION

Actinomycosis is a rare and tricky diagnosis to make in the head and neck region. It has been called "The Great Masquerader of the Head and Neck" with, 10% cases being diagnosed correctly.

Actinomycosis is a chronic granulomatous infectious disease caused by Gram Positive, anaerobic/ microaerophilic filamentous bacteria. These are fastidious organisms which are a part of the normal flora of the Aero digestive tract Most common species: A. *Israelii*; less commonly, infection is caused by A. *Propionica*, A. *Naeslundii*, A. *Viscosus* and A. *Odontolyticus*. Dental caries, dental manipulations and oromaxillofacial trauma are predisposing factors through which the bacteria enter the submucosa and establish infection over time.

Classification (COPE 1938)

Cervicofacial (50%)

Pulmonothoracic (30%)

Abdominopelvic (20%)

Clinical features:

Swelling in the face (more common is mandible) – progressive increase in size, fluctuant (initially hard and indurated and painful, later softens due to necrosis), as mass increases further in size, many sinuses discharging purulent material are seen. Is the characteristic lesion.

Low grade fever in <50% patients

Diagnosis:

Clinical diagnosis with the symptoms and signs.

Microbiological diagnosis: The purulent discharge from the sinuses is actually bacterial colonies and they can be cultured. However actinomyces is an extremely fastidious organism with culture rates <50% even in appropriate anaerobic culture media.Hence it is extremely difficult to establish the diagnosis.

Histopathology: However tissue diagnosis is possible with a picture of Chromic Granulomatous infection along with the typical Actinomycosis colonies in the Sunray Appearance. The typical finding of an outer zone of granulation and a central zone of necrosis which contains multiple basophilic granules that represent lobulated micro-colonies of Actinomyces.

CT and MRI are more important to rule out other malignant or granulomatous causes and no specific characteristics are attributed. However they help to estimate the extent of disease, other foci in hidden areas and also determine the dimensions of surgery.

Treatment:

Surgical debridement of involved tissue is important in cases of abscess and discharging sinuses. It is best if superimposed with medical therapy, thereby it helps to reduce the infective load and hasten recovery.

Medical Treatment:

Penicillin G – Drug of choice - Cervicofacial cases: 1-6 million units/day IV x 6weeks, may follow with penicillin V(forup to 6mnths – even 1yr if needed)

Thoracic and abdominal disease: 10-20 million units/day IV divided q4-6hr x 6 weeks, may follow with penicillin V

Penicillin V - Mild: 2-4 g/day PO divided q6hr for 8 weeks

Surgical: 2-4 g/day PO divided q6hr for 6-12 months

In cases of Penicillin Allergy, Doxycycline is to be preferred - 100-200 mg/day twice daily oral/ IV

Clindamycin, Cefotaxime or Imipenem are also useful in patients with resistance (Resistance per se in actinomycosis is rare and due to co infection with other resistant organisms)

OURS IS A UNIQUE CASE:

- This case that presented to us was of a 71yr old gentleman who had a painful palatal ulcer type of lesion which resembled a mucor eschar-like lesion. There has been a description of Ulcerative lesion due to osteomyelitis which has been described similar to aphthous ulcers showing erythema, edema, tenderness and a pseudomembrane but it progressed rapidly to nearby areas on the palate and patient presented with signs of progression barely five days later. However this patient was static in his disease course and later improved with antibiotics and conservative management.
- Most cases described in literature have the characteristic indurated mass and the discharging sinus as flag signs for clinching the diagnosis – both of these were absent in our case.
- As in most cases described in literature, HPE diagnosis was made by the presence of characteristic colonies and culture was negative.
- Surgical treatment was not undertaken in this patient in consideration of the patients wish, symptoms and signs that responded to medical treatment.
- This presentation is quite rare and lives up to the name of " The Great Masquerader of Head and Neck"

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