



## "TWO RARE CASES OF PRIMARY CUTANEOUS ACTINOMYCOSIS AT UNUSUAL SITES SUCCESSFULLY TREATED BY COMBINED SURGICAL EXCISION AND MEDICAL MANAGEMENT".

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### ABSTRACT

Actinomycosis is a rare subacute or chronic suppurative,granulomatous infection caused by facultative pathogens belonging to Actinobacteria class[1].The term actinomycosis was derived from Greek term "aktino" which refers to radiating appearance of sulfur granules and "mycos" labels the condition as a mycotic lesion.This is predominantly endogenous infection.These organisms are normal commensals in the oral cavity,respiratory mucous membrane,colon and female genital tract and any breach in the lining membrane may cause tissue infiltration and growth and pathological lesions are produced with typical yellow sulfur granules[2].On the contrary Primary cutaneous actinomycosis is extremely rare and the organisms are exogenous introduced into the body by trauma. Iam presenting two cases;one in a female aged 38 yrs who sustained road traffic accident with injury around the left orbital region of the face;and another male aged 28 yrs who works in fish ponds developed 3 lesions in the left thumb without any obvious cutaneous injury ,most likely due to the fish bites.Both the cases were confirmed by microbiological and pathological investigations and were proved actinomycotic lesions and were successfully treated by combined surgical excision and prolonged penicillin -G administration and were followed for more than 2 ½ years and had complete cure from the disease.

**KEYWORDS :** primary cutaneous actinomycosis—injury left orbit region face –left thumb-Aquaculture-fish bite.

### CASE REPORTS

CASE ONE: Missamma F/38 yrs.

Referred from cancer hospital,vangayagudem,near Eluru on 19-08-2018.She is suffering from multiple nodules confined to skin and subcutaneous tissue, some with seropurulent discharge over left orbital and zygomatic area of face over the last 3 months.

She sustained road traffic accident due to fall from a two wheeler on 10<sup>th</sup> may 2017.She was treated at a private nursing home at palakollu, where primary wound suture was done.

Wound healed well but she developed few vesicular lesions at the site after 8 months.She went to the same doctor where she was given some medication and the vesicles subsided.She had no complaints for one year.Later she developed vesicles at the same site again and she was referred to dermatologist who prescribed some medicines.There was no relief and the patient went to Tanuku where she had some medical treatment including anti tuberculous treatment without any relief.The nodules were extending in size and number.In 2018 she was referred to cancer hospital at vangayagudem near Eluru with a suspicion of malignancy ,where wedge biopsy was done.After the histopathology report she was told it is not a cancerous condition and was referred to Asram medical college hospital.She attended ASRAM Hospital,general surgery and later plastic surgery outpatient department on 19-08-2018.After preliminary investigations she was admitted in plastic surgery ward on 20-08-2018.clinical exam: Multiple nodular lesions over the left maxillary and zygomatic region of the face.some sinuses are discharging clear serous fluid and the inflammation is extending from the nose on left side upto preauricular region and from upper eyelid,outer canthal region to just above the base of the mandible on the left side of face. There is no enlargement of regional lymph nodes. clinical diagnosis of actinomycosis was made and planned for excision+primary skin grafting and medical treatment after evaluation with CT AND MRI FACE.

x-ray skull AP& lt lateral view was taken on 25-8-2018 soft tissue shadow left orbital and zygomatic area with dense areas(?calcifications) (photograph-2a)

CT Scan skull (photograph-2c)

Report:

- 1) Left periorbital cellulitis with periostitis
- 2) Multiple small hyperdense areas are seen likely calcification
- 3) Left maxillary,bilateral ethmoid,rt sphenoidal and rt frontal sinusitis.

MRI FACE :(photograph-2b) Suggestive of soft tissue lesion left orbital and zygomatic region with dense calcification.

She was operated on 22-08-2018 under general anaesthesia.Complete excision was done.Calcified plaques were seen during excision.Primary split skin grafting was done.Healed well and was discharged on 01-09-2018 with advise to continue the drugs and follow up once in two weeks.

### HP REPORT OF EXCISED LESION

HPE/7632/18 DT:31-8-2018 F/38 YRS R. Missamma

"Features are that of Actinomycotic granuloma extending into subcutaneous fat".

### MEDICAL TREATMENT AFTER SURGERY

Cap Amoxycilline 500 mg twice a day Inj.Penidure LA 12 (Benzathine penicillinG 1.2 million units per vial) once in 3 weeks deep im.

On 4-9-19 fresh lesions were seen on the left temporal region,upper eyelid lt and preauricular inflammation.

Penidure LA 12 is changed to once a week instead of once in 3 weeks and Cap.amoxycilline 500mg twice a day continued.

14/10/19—most of the lesions are controlled and the patient was feeling very comfortable.

### Followup

18/11/19 (1 yr 3 months after surgery)

All the lesions healed completely.Antibiotics completely stopped.

07/06/21(Two years 10 months after surgery) Complete healing,no deformity or scarring and no recurrence Pt. completely recovered from the lesions and going to regular work.

Photograph-1 to photograph-5



**PHOTOGRAPH 1: CASE 1: PREOPERATIVE**



**PHOTOGRAPH 2: CASE 1: A: X-Ray skull (AP & It lateral views)**

**B: MRI face**

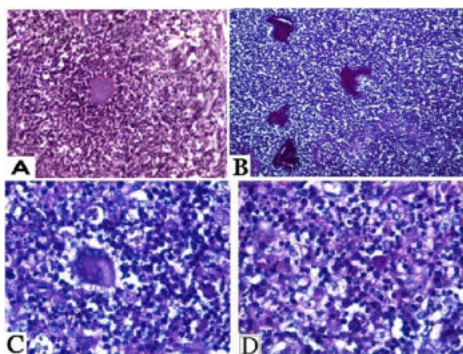
**C: CT Scan skull**



**PHOTOGRAPH 3: CASE 1: A: Preop surgical marking**

**B: After complete excision**

**C: Five days postop after skin grafting**



**PHOTOGRAPH 4: CASE1:**

**A.** Hematoxylin & eosin stained section, Histopathological examination X40 magnification showing amorphous basophilic material with splendore Hoeplic phenomenon (actinomycotic granuloma)

**B.** HISTOPHOTOMICROGRAPH PAS POSITIVE 10X

**C.** HISTOPHOTOMICROGRAPH Giant cells

**D.** HISTOPHOTOMICROGRAPH Plasma cells



**PHOTOGRAPH 5: CASE 1: A: PREOPERATIVE**

**B: POSTOPERATIVE**

**(TWO YEARS TEN MONTHS)**

## CASE TWO :

B.Avinash/Male 28 YRS. works in Fish Ponds.(Aquaculture).

Noticed three painful nodules over the dorsal aspect of the left thumb 3 months back. There was no history of any injury to the finger. They are gradually increasing in size and painful. One week back one of the nodules near the interphalangeal joint ulcerated with serous discharge. The nodules are in the subcutaneous plane, not attached to deeper structures and firm in consistency, mobile over underlying structures. No other lesions elsewhere in the body.

Admission: 7-12-2018.

Operation: 8-12-2018 (complete excision of the 3 nodules and primary repair was done under regional anesthesia).

Discharged on 17-12-2018.

He was given cap amoxycilline 500 mg twice a day and Inj Penidure-LA 12/once in 3 weeks/deep IM.

HP REPORT: Actinomycosis (photograph 8 a,b,c ) Cap Amoxycilline stopped after 3 months and penidure LA-12 continued once in three weeks deep IM.

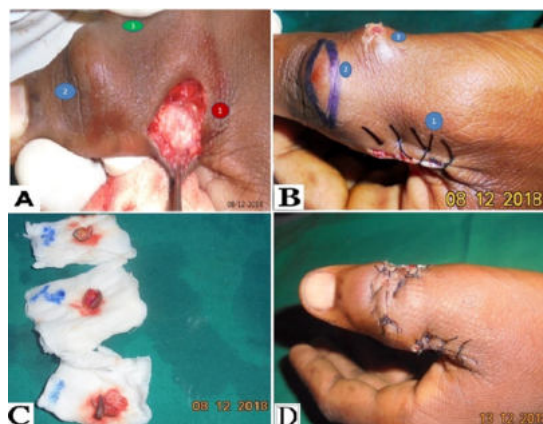
completely healed-no recurrence and is going to his work regularly. PENIDURE-LA also stopped after 3 months.

Followup :03-06-2021 (2 yrs 7 months), completely healed, good movements of the lt thumb and going to his regular work. stopped all drugs.

## Photographs- 6-9.



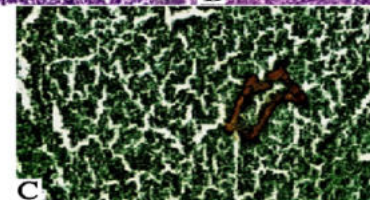
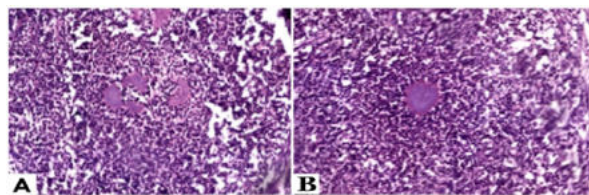
**PHOTOGRAPH 6: CASE 2: A & B : PREOPERATIVE**



**PHOTOGRAPH 7: CASE 2: A,B,C,D. Peroperative**

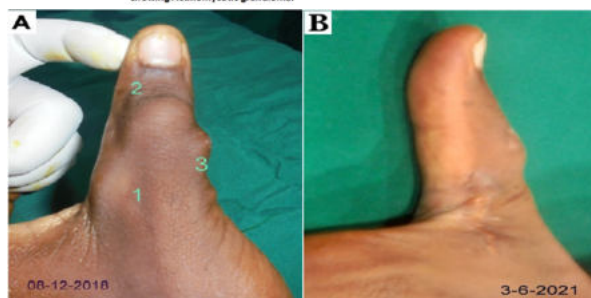
**excision of all the lesions and repair of the skin defect**





PHOTOGRAPH 8: CASE2:

- A. Haematoxylin & eosin stained section, Histopathological examination X 40 magnification showing amorphous basophilic material with splendore Hoeglic phenomenon (actinomycotic granuloma).  
 B. Hematoxylin & eosin stained section, Histopathological examination X40 magnification showing amorphous basophilic material with splendore Hoeglic phenomenon (actinomycotic granuloma).  
 C. GMS (methamine silver stain), Histopathological examination X40 showing Actinomycotic granuloma.



PHOTOGRAPH 9: CASE 2: A &amp; B Pre and 2 yrs &amp; 7 months postop

## DISCUSSION:

Actinomycosis is a rare, slowly progressive infectious bacterial disease [3], caused by anaerobic or microaerophilic bacteria, primarily of the genus *Actinomyces*, which colonize in the mouth, colon, and vagina[4]. These are non-spore-forming, anaerobic gram +ve bacilli. The pathogenic Actinomycotic species are only isolated in human beings. *Actinomyces israeli* is the most common human pathogen and is seen in most of the clinical presentations. Actinomycosis has been called the most misdiagnosed disease and listed as a rare disease by the Office of Rare Diseases of the National Institutes of Health[5].

Disease incidence is greater in males between the ages of 20 and 60 years than in females. Before the discovery of antibiotics, the incidence in the Netherlands and Germany was one per 100,000 people/year. The incidence in the U.S. in the 1970s was one per 300,000 people/year, while in Germany in 1984, it was estimated to be one per 40,000 people/year in females the incidence of genitourinary Actinomycosis has increased by increasing the use of intrauterine devices[6].

There are five main clinical types depending upon the anatomical distribution; 1) cervicofacial-60% 2) thoracic-20% 3) abdominal,-15% 4) pelvic and primary cutaneous.-5%[7]

The most common symptoms of cutaneous actinomycosis are having progressive skin and soft-tissue inflammation, which can become an abscess or cold mass, or nodular lesions with fistulas that need to be differentiated from chronic inflammatory skin disease, cutaneous mycobacterial infections, and sporotrichosis[8]. Basically the diagnosis of Cutaneous Actinomycosis is made by the following tests and exams [9]. i) A thorough physical examination and assessment of symptoms ii) Evaluation of the affected individual's medical history. iii) Microscopic observation of pus or tissue samples, to check for the presence of sulfur granules (which are round and yellow and are named for their characteristic appearance; but, they do not contain sulfur).

The tissue samples may have to be obtained surgically. iv) The culture of fluid or tissue from the infected area: These bacteria are slow to grow in culture, and it may take over 3 weeks to obtain a (positive) culture result. V) X-ray, CT, or magnetic resonance imaging scans of the affected area, to ascertain location and number of abscesses, as well as to differentiate inflammatory masses due to infection from the tumors.

The treatment of cutaneous actinomycosis is primarily based on antimicrobial therapy. Drug of choice is Penicillin G. High-dose Penicillin G - 12 - 24 million U/d intravenous by continuous infusion or in divided doses is given followed by oral amoxycillin, ampicillin, or penicillin V which is administered over a prolonged period (6 months to 1 year). An alternative to penicillins, if the patient is hypersensitive to penicillin or the causative organisms are resistant to penicillin, are Ceftriaxone, Imipenem/Cilastatin, Clindamycin, Amoxycillin/Clavulanic acid, Doxycycline, Tetracycline, Lincomycin and Macrolides (erythromycin, Carbomycin, Spiramycin and Oleandomycin). Surgery is indicated mainly for taking a biopsy specimen, to drain abscesses, to extirpate a fibrotic sinus tract or a refractory fistulous tract which are not responding to the conservative medical treatment[10]. The follow-up should be adequate and meticulous to have a complete cure of the disease as the prognosis is usually excellent, especially when it is diagnosed early and treated with the appropriate antibiotic therapy[11].

Wherever feasible surgical excision of the affected area and continued antibiotic therapy will hasten healing and take lesser time for complete recovery. In these two patients with extensive lesions one over the face and around the orbit and another with three nodules in the left thumb, one already ulcerating, gave very good results because of combined surgical excision and continued penicillin-G. Because of this and primary skin grafting over the face, the scarring and deformities could be prevented. In the second case also primary repair after excision enabled restoration of normal movements in the left thumb without any deformities.

## CONCLUSIONS:

In this paper I presented two unusual cases of Primary Actinomycosis of skin. In the first case history of Road traffic accident is present and that was responsible for the mycotic infection. But in the second case there is no injury to the skin, but he developed cutaneous actinomycosis as 3 subcutaneous nodules. Since his occupation is Aquaculture and regularly working in fish ponds, I postulate that mycotic infection was likely due to fish bites, just like transmission of actinomycosis by human bite was reported[12]. This needs further study to confirm, since such cases were not reported earlier in the literature. This patient told me after further interrogation, that one type of fish occasionally bites the skin through a sharp thorn.

Both the patients because of combined surgical and medical treatment recovered very well in short time possible without any further recurrence.

## Acknowledgements:

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