CASE REPORT

Rare case of disseminated rhinosporidiosis with chronic osteomyelitis of the calcaneum treated by a simple technique of negative pressure wound therapy

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SUMMARY

Occurrence of nasal, cutaneous and disseminated rhinosporidiosis in the same patient is uncommon and involvement of calcaneum is rare. Rhinosporidial osteomyelitis is treated surgically, but multiple recurrences and local spread are common. Many a times, surgical ablation or amputation of the involved area of the limb is carried out to prevent recurrences. We present a case of rhinosporidial osteomyelitis of the calcaneum with a chronic discharging sinus and large lytic lesion in the calcaneum treated by an alternative surgical option in the form of debridement and negative pressure wound therapy.

BACKGROUND

Rhinosporidiosis is a chronic granulomatous infection caused by *Rhinosporidium seeberi* that predominantly affects the nasal and nasopharyngeal mucosa. Disseminated cutaneous and visceral lesions can occur due to autoinoculation or due to haematogenous spread. Osteoarticular rhinosporidiosis is uncommon and combined presentation of disseminated cutaneous lesions associated with osteomyelitis of the calcaneum is extremely rare.

Treatment of rhinosporidiosis is difficult and when it presents as chronic osteomyelitis, it becomes a therapeutic challenge. Even after optimal surgical treatment and medication, recurrence is quite common. Some authors have advocated ablation of the involved limb as a definitive treatment in resistant cases. We report a case of disseminated cutaneous rhinosporidiosis presenting with chronic osteomyelitis of the calcaneum. We describe an alternate simple operative treatment in the form of debridement and negative pressure wound therapy (NPWT) for such refractory cases.

CASE PRESENTATION

A 75-year-old male patient presented with a painful swelling and chronic discharging sinus over the lateral aspect of his left heel of 6 months duration. On local examination, an active sinus discharging thick purulent material and of approximately 0.5 cm in circumference was present, which was fixed to the underlying calcaneum. Subtalar joint movements were painful, whereas ankle movements were normal. He also had multiple subcutaneous

swellings around left knee, left leg and medial border of his left foot. They were approximately of the size 4×2 cm, 3×2 cm and 2×1 cm, respectively. The swellings were subcutaneous, mobile and cystic in consistency.

On detailed enquiry, he gave a history of having undergone excision of a nasal mass 10 years ago and with treatment remained symptom free for 7 years. Two and half years back, he noticed multiple swellings over the anterior abdominal wall and right lower limb which were excised. The histopathological features were consistent with cutaneous rhinosporidiosis. Postsurgery, he was treated with dapsone until this date. However, 18 months before, he had an ulcer over the dorsum of left foot, for which he underwent debridement followed by cryotherapy. On physical examination at the time of presentation, there was no hepatosplenomegaly or other visceral lesions. The pain in the heel had significantly restricted his mobility and activities of daily living.

INVESTIGATIONS

Haemogram showed a haemoglobin level of 10.7 g/dL; total and differential counts were within normal limits. Erythrocyte sedimentation rate was 23 mm/hour and C-reactive protein was 11.6 mg/L. Radiographs revealed a lytic lesion in the left calcaneum. The CT scan showed a large destructive lesion involving the left calcaneum (figure 1).

TREATMENT

The sinus was excised and the cavity was thoroughly curetted. The wound was left open and a negative-pressure wound therapy (NPWT) dressing was applied (figure 2). The subcutaneous swellings were excised (figure 3). Culture of the curetted material did not show any fungal or bacterial growth. Histopathological examination of the curetted material as well as the excised lesions revealed numerous sporangia with endospores along with areas of infarction. Focal fibrocollagenous stroma with dense neutrophilic lymphoplasmacytic infiltrate and foreign body giant cell reaction were seen. It was consistent with rhinosporidiosis (figure 4). The NPWT dressing was changed every 4–5 days in the first 2 weeks. After 2 weeks of vacuum-assisted NPWT dressing, the wound was found to be clean and the floor culture did not show any

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Figure 1 (A) Discharging sinus over the lateral aspect of left heel. (B) Plain radiograph lateral view of the left heel showing a lytic lesion in the left calcaneum. (C and D) CT scan of the left calcaneum showing destruction lesion involving the entire calcaneum with destruction of the lateral cortex. Soft tissue lump on the medial aspect of the foot is also seen in D.

micro-organism. A below knee walking cast was applied for 6 weeks. On removal of the cast, the cavity was found to be filled with healthy granulation tissue. The wound healed by secondary intention (figure 5).

Figure 2 (A) Destruction of the lateral cortex of the calcaneum. (B) The necrotic curetted material from the cavity. (C) The deep cavity inside the calcaneum after debridement. (D) The negative-pressure wound therapy dressing applied over the wound.

Figure 3 Figure showing the well-encapsulated subcutaneous swellings which were excised. Inset shows the cut surface of the swelling with creamy gelatinous material inside.

Our technique of NPWT dressing

We use freely available and cheap poly vinyl alcohol (PVA) foam as filler. We use burn mesh as wound contact layer to prevent adhesion of the foam to the wound bed. A 16 French Ryle's tube was used for suction which is placed superficial to the PVA foam and the wound sealed with transparent waterproof adhesive film. The Ryle's tube is connected to a simple universal suction device which deliver a pressure of 50–80 mm Hg. This dressing is connected to the suction for 20 min and continued cyclically every 2 hours. The NPWT dressing is changed every 4–5 days.

OUTCOME AND FOLLOW-UP

At 6 months follow-up, the wound had healed completely and the X-rays showed healing with new trabeculae formation (figure 6). The patient was pain free and walking weight bearing with the aid of a walking stick.

DISCUSSION

Rhinosporidiosis is a recurrent granulomatous infection commonly affecting the skin, mucous membrane of nose and conjunctiva of the eye. The causative organism is *R. seeberi*.

Figure 4 H&E stained photomicrograph (10x) showing numerous sporangia with endospores and fibrocollagenous stroma with areas of infarction and foreign body giant cells. Inset shows the high power (40x) photomicrograph with multiple sporangia containing endospores.

Figure 5 (A) Picture of the wound after 2 weeks of negative pressure wound therapy dressing. (B) Wound at 6 weeks follow-up. (C) Wound at 3 months follow-up. (D) Fully healed wound at 6 months follow-up with a healthy scar.

which is an aquatic organism presently considered to be in the class Mesomycetozoa.¹ It is endemic in many Asian countries, but has also been reported from nearly 70 other countries across the globe. The transmission of infection is by direct contact with the organism or its spores through air or fomites. Extranasal involvement is rare, which can involve the skin, subcutaneous tissue, muscles and very rarely bone. Dissemination to

Figure 6 Radiographs at 6 months follow-up showing healing of the calcaneal lesion with trabeculae formation and marginal sclerosis.

Table 1 Table showing the review of literature of rhinosporidial osteomyelitis

Serial number	Study	Year	Number of cases	Bone involvement	Management
1	Chatterjee <i>et al</i> ⁹	1977	1	Hand, foot	Not mentioned
2	Aravindan <i>et al</i> ¹⁰	1989	1	Scapula	Excision
3	Mitra <i>et al</i> ³	1996	1	Calcaneum	Not mentioned
4	Gokhale <i>et al</i> ¹¹	1997	1	Hand, third toe	Partial amputation
5	Adiga <i>et al</i> ¹²	1997	1	Tibia	Excision
6	Makannavar <i>et al</i> ¹³	2001	1	Tibia	Not mentioned
7	Dash <i>et al</i> ¹⁴	2005	1	femur	Not mentioned
8	Amritanand <i>et al</i> ⁴	2008	1	Talus, calcaneum, tibia, fibula, tarsals, metatarsals	Below knee amputation
9	Suryavanshi <i>et al</i> ¹⁵	2011	1	Clavicle	Wide excision
10	Mondal <i>et al</i> ¹⁶	2013	1	Wrist	Below elbow amputation
11	Kundu <i>et al</i> ¹⁷	2013	1	Little finger	Ray amputation
12	Bandyopadhyay <i>et al</i> ⁵	2015	1	Distal radius	Not mentioned
13	Present case	2017	1	Calcaneum	Debridement, negative pressure wound therapy

atypical sites is mainly by haematogenous spread or by direct implantation.²

Atypical presentations of disseminated rhinosporidiosis can cause a dilemma in diagnosis. However, a proper history and necessary microscopic examination will help in arriving at a final diagnosis. Osteoarticular rhinosporidiosis is exceedingly rare. Only 12 cases of disseminated rhinosporidial osteomyelitis have been documented in the literature out of which only two cases had involvement of the calcaneum.^{3,4} In a series of 119 cases of rhinosporidiosis by Bandyopadhyay *et al*, there was only one case involving the bone.⁵ In the 12 previously reported cases, treatment given was described only in seven cases (table 1). Amputation of the involved limb was carried out in four out of those seven patients, whereas three patients underwent excision of the affected part. Operative treatment is the method of choice in cases with osteomyelitis. However, recurrence is common. Following debridement, wound healing is a major concern and recovery is time-consuming. Therefore, many surgeons have advocated amputation of the part of the limb in cases of prolonged resistant cases. Antimicrobial therapy with numerous drugs like dapsone, ketoconazole and ciprofloxacin have been tried for rhinosporidiosis but believed to have doubtful efficacy.^{6,7}

Our patient presented with disseminated cutaneous rhinosporidiosis associated with osteomyelitis of the calcaneum. The cutaneous lesions were not painful. However, the osteomyelitis in the calcaneum was disabling and he was not able to bear weight. To salvage the limb, we planned for curettage and debridement

of the infected cavity which was managed postoperatively by NPWT dressing, which utilises vacuum pressure suction at the wound site. NPWT functions in the following ways: wound being left open, collection of the infected material does not occur; the residual infective material is continuously being drained out by the negative suction; the wound being sealed by sterile material, prevents the entry of external infection; in an optimal non-infected environment, healthy granulation tissue fills the cavity, allowing the wound to heal gradually and NPWT draws the wound edges closer to each other.

The vacuum suction was used till the wound was satisfactorily healthy and the cavity was filled with healthy granulation tissue. A below knee walking cast further helped in wound healing. The patient was kept non-weight bearing for the entire period. NPWT has been used in the treatment of chronic osteomyelitis and non-healing ulcers.⁸ We believe that the same procedure can effectively be used in the treatment of refractory atypical osteomyelitis such as fungal osteomyelitis Madura foot. However, a prolonged follow-up is necessary to assess the long-term efficacy of the treatment modality, for such slow-growing diseases.

Nasal, cutaneous and disseminated rhinosporidiosis to occur together in the same patient is unusual. Rhinosporidial osteomyelitis is a rare and difficult condition to treat. Recurrences and local spread are common. The patient should be counselled regarding the advantages and limitations of each surgical procedures. Limb salvage can be achieved using the technique described in this patient. Further studies are required to understand the behaviour of the causative organism and to find an effective cure for rhinosporidiosis.

Learning points

- The present case is being reported to highlight a rare presentation of rhinosporidial osteomyelitis of the calcaneum.
- To describe an innovative method of treating this highly refractory infection by negative pressure wound therapy successfully.
- Physicians and surgeons should keep a high degree of suspicion to diagnose atypical presentations of disseminated rhinosporidiosis.

Contributors JKN has done the data collection, preparation of manuscript and editing. RS has done the data collection. SPM was involved in the preparation, proof reading and final editing of the manuscript.

Competing interests None declared.

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