

“Surgical management of unusual large tophi in chronic tophaceous gout: A case report”

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Abstract

Gout is a disorder of purine metabolism a form of inflammatory arthritis characterized by recurrent attacks of a red, tender, hot, and swollen joint. Pain typically comes on rapidly, reaching maximal intensity in less than twelve hours. The first metatarsophalangeal joint is affected in about half of cases. It may also result in tophi, renal stones, or urate nephropathy.¹ Commonly, but very large tophi are unusual in chronic gout. Here we present 52 years male patient presenting with unusually large tophi over bilateral foot, with raised serum uric acid levels. In a untreated cases of chronic gout, patients can develop severe destructive poly articular involvement and tophi. Treatment aims at control of serum uric acid levels which can cause regression of tophi. And surgical excision of very large tophi.

Keywords: Gout, Arthritis, Tophi, Surgical excision.

Introduction

Gout is a metabolic disorder of purine degradation pathway usually affecting middle aged and elderly men and postmenopausal females. Gout occurs after prolonged periods of sustained hyperuricemia. Consequently, the epidemiology of gout closely parallels the epidemiology of hyperuricemia, some patients develop chronic polyarthritis similar to rheumatoid arthritis.^{2,3} Both disease entities have been reported to occur in adult population to the extent of approximately 1%.⁴ Other factors that result in sustained elevations of serum uric acid above the solubility product of mono sodium urate can lead to increased total-body urate and predispose to gout.^{3,5} Chronic tophaceous gout frequently occurs after 10 years or more of recurrent polyarticular gout.^{1,9}

Treatment aspect involve strict control of protein diet and drugs to reduce the uric acid levels and surgical excision⁶ of the tophi to relieve pain or to relieve secondary symptoms. Surgical intervention becomes inevitable when the overlying skin of tophus ulcerate and wound infection occur. However, the conventional curettage and debridement of tophi might not always provide the desired surgical result. The firmness of the tophi and the surrounding fibrosis often make it difficult to effectively debride the chalky mass manually. Furthermore, the large tophi can cause local ischemia to the surrounding Skin.^{7,8,10,16}

Case Report

A 52 years male patient named Shivananda from Jali Nagara, Davangere, presented with a 8 years long history of recurrent pain in both feet and a 5 years history of multiple swellings over both feet the pain was acute in onset initially which was throbbing type of pain predominantly in both feet at base of great toe, without any morning stiffness. Pain was severe and would partially subside with analgesics. Following first episode of acute arthritis patient had several episodes of such acute attacks and he would take analgesics from local hospital and symptoms would subside over a week. After three years of first episode of gouty arthritis

patient noticed small swellings over both feet at base of great toe, which were red and tender, over the years these swellings increased in size gradually. Later he developed similar swellings over the dorsum of both feet. At this point of time patient was informed about severity of the disease and was referred to higher centre. Patient consulted Department of Orthopaedics J.J.M. Medical college, Davangere. He presented with huge swellings over both feet which was painful [Fig. 1]. Patient had multiple tophi measuring 3*3*2 cm over both first metatarsophalangeal joints and 2*2*1 cm in size over the dorsum of both feet, 1*1*1 cm over right foot little toe [Fig. 1]. 1*1 cm over the 3rd middle phalanx of left hand and 4th digit middle phalanx of right hand. FAOS score was 58% at the time of presentation. Blood investigations serum uric acid was 10.7 mg/dl (normal value being 3.5-8.5 mg/dl in men), Hence correlating with his condition. Febuxostat 40 mg twice daily (xanthine oxidase inhibitor) was started at the time of presentation, pain and swelling did not subside. Patient was posted for surgical excision of tophi over both feet once fit for surgery. Post operatively colchicine 0.6mg tablet twice daily (inhibit multiple proinflammatory mechanisms, while enabling increased levels of anti-inflammatory mediators) was started for resolving small tophi over the digits, life style modification including diet was explained to the patient. Serum uric acid reduced to 6.2mg/dl after one month of oral uricosuric drugs.

X ray both feet AP and oblique views [Fig. 2] showed no evidence of bony involvement or arthritic changes in the MTP joint, only soft tissue shadow was visible in the region of swelling. There were no secondary skin changes. USG abdomen and pelvis showed normal study, renal involvement, urate stones were ruled out.



Fig. 1: Tophi over both feet 1st MTP joint, over lateral aspect of dorsum of feet and right 5th toe

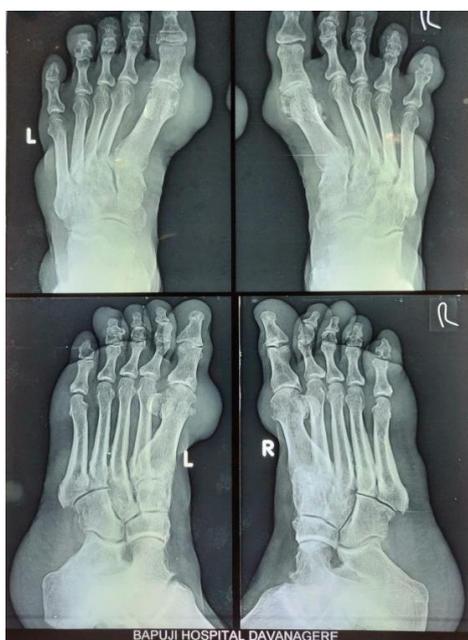


Fig. 2: X-Ray of both feet with multiple large tophi visible as soft tissue shadow over 1st MTP joints and dorsum of both feet and over the right 5th toe.



Fig. 3: Intra-Operative images showing tophi with areas of hard and chalky white fluid filled cavity; **A:** Skin incision Over tophi; **B:** Soft tissue dissection; **C:** Exposure of tophi; **D:** Post excision of tophi

Surgical Procedure

Under spinal anaesthesia, excision of gouty tophi over bilateral feet over the 1st MTP joints, 5th metatarsal bone and right little toe was performed, via linear incision over the tophi and blunt soft tissue dissection around the encapsulated mass. Intra operatively there were areas of hard and chalky whitish fluid filled cavities. All tophi were removed en-mass [Fig. 3]. Thorough wash given and skin closure was done with interrupted sutures Post-operatively, alternate day dressing done and sutures removed on 10th post-operative day, injectable antibiotics for 3 days followed by oral antibiotics for next 7 days till suture removal. Patient had superficial infection which resolved uneventfully. Patient had significant improvement in symptoms post surgery as assessed by Foot and Ankle Outcome Score FAOS. Patient was counselled for regularly follow-up every month for serum uric acid estimation.

On follow-up after one-month, Small tophi over the hands were reduced in size with no complaints of any pain. Patient was advised to continue oral uricosuric drugs. Tablet Febuxostat 40 mg twice daily and tablet colchicine 0.6 mg twice daily.

Follow-up of the patient was done at intervals 6,12,24 weeks FAOS score was 63% at 6 weeks, 72% at 12 weeks and 80% at 24 weeks follow-up. 6 months follow-up shows satisfactory wound healing and no recurrences of tophi clinically [Fig. 6] and radiologically [Fig. 7]. The chances of recurrence are less as the patient had serum uric acid levels of 6mg/dl at 6 months follow-up and ranging between 6-7mg/dl in regular follow-up and no new complaints of exacerbation of symptoms.

Histopathological examination report: [Fig. 4a,4b] haematoxylin and eosin staining of the tophi Showed bits of tissues composed of masses of amorphous material containing needle shaped crystals surrounded by giant cells , fibro blasts and lymphocytes.



Fig. 4: Gross specimen and histopathological microscopic picture; (4A): Gross specimen of tophi excised from both feet. (4B): Multinucleated giant cells; (4C): Amorphous material with needle shaped crystals.

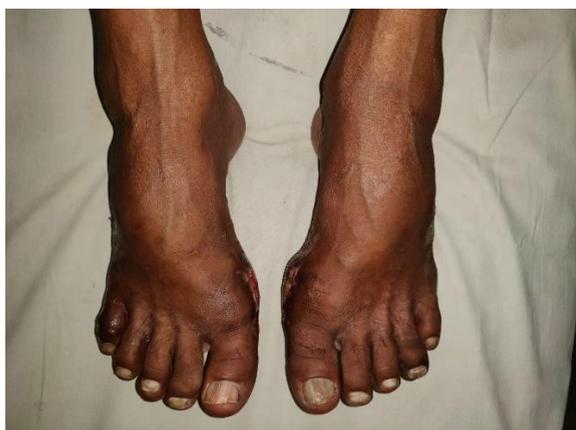


Fig. 5: Post-operative clinical picture showing tophi excision status



Fig. 6: 6 months follow-up showing no recurrence and satisfactory wound healing



Fig. 7: 6 months follow-up x-ray of both feet shows no recurrence of tophi

Discussion

Gout is a disorder of purine metabolism and results from longstanding hyperuricaemia and urate crystal deposition in various tissues. The essential abnormality in primary gout is increased formation of uric acid without intermediary incorporation into nucleic acids. In secondary gout, there is an increased breakdown of nucleic acids leading to an excess of the end-product, uric acid.

Purine Metabolism

The chief purines found in the nucleotides and nucleic acids are *adenine* and *guanine*. Uric acid is the final oxidation product (in man) of these purines. Purines combine with their 9-nitrogen position with sugar residues → nucleoside. If the sugar residue is also phosphorylated, then a nucleotide result. Purines are occasionally found as free bases, more usually as nucleosides and nucleotides, and as nucleic acids. The synthesis of purine nucleotides occurs through two pathways, referred as *de novo* and *salvage pathways*.

Gout affects 1-2% of adult men in western world, prevalence of gout in India is 0.12% as per International League Of Nations Against Rheumatism. In the first stage, it usually affects the first metatarsophalangeal joint in approximately 75% of patients during first attack and 90%

of patients sometime during the disease course and less commonly other joints. The next most frequent localizations are the midtarsal joints, ankles, knees.¹¹ In 3-14% the first attack is polyarticular. Old age, males are 10 times more affected than women, postmenopausal state and black race are related to a higher risk for development of the disease. Also, the use of certain medications may trigger gout (diuretics, cyclosporine, low doses of aspirin). In untreated patients, chronic tophaceous gout may develop, which is characterized by chronic destructive polyarticular involvement and tophi. Chronic tophaceous gout frequently occurs around 10 years or more of recurrent polyarticular gout. Tophi can occur in soft tissue, osseous tissues, ligaments and different organs either in presence or absence of gouty arthritis. Tophi are typically found on the helix of the ears, on fingers, toes, wrists and knees, olecranon bursa, Achilles tendons and also rarely on the corneal stroma, corneal epithelium, sclerae, subconjunctival involvement with 0.79% incidence of ocular involvement¹² and on the cardiac valves mainly aortic stenosis.¹³ A study showed history of gout in 21.4% of aortic stenosis subjects as compared to 12.5% of controls.¹⁸

Although the prevalence of tophaceous gout, principally the generalized form of it, has decreased in the past years, the disease still exists likely due to the absence of an accurate diagnosis and therapy.¹⁴ Our case had large tophi, which are unusual in chronic gout. If left untreated, hyper uricemic patients (serum urate level ≥ 68 mg/l or 400 $\mu\text{mol/l}$) can evolve from intermittent arthritis to polyarticular tophaceous gout with symptoms in between attacks. Lowering serum urate levels with xanthine oxidase inhibitors or uricosuric agents prevents acute flares and tophi development.¹⁵ The recommended target serum uric acid concentration is <60 mg/l (357 $\mu\text{mol/l}$).¹⁴ Although controversial, recommendations have been made to achieve a target serum urate level <50 mg/l (297 $\mu\text{mol/l}$) in severe chronic gout patients, as this concentration may be associated with greater depletion of synovial fluid crystals and a reduction in tophus size. Surgical treatment is seldom required for gout and is usually reserved for cases of recurrent attacks with deformities, severe pain, infection and joint destruction.¹⁶ It is also indicated when tophi are unsightly, painful; or when it interferes with tendon function or causes skin necrosis and ulceration; or encroach upon nerves causing symptoms of compression.¹⁷ The chances of recurrences are less if the patient is compliant with the life style modifications, and strict adherence to the medication and regular follow-up.

Conclusion

The treatment of gout should be undertaken early in order to avoid the evolution of the disease to the chronic tophaceous form responsible for joint deformities and their functional consequences. Our case is a rare form of large tophi complicating untreated gouty arthritis which subsided with both pharmacological and surgical management. Pain and

swelling reduced. Ankle score improved, excision specimen confirmed by gross and Histopathological examination. Lifestyle modification with regular follow-up and strict adherence to oral uricosuric drugs is the key for further control of the disease and prevent recurrence.

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