

# Idiopathic intraosseous ganglion of the distal tibia in a young adult male: A case report with review of literature



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## ABSTRACT

Intraosseous ganglion is a benign cystic bony lesion containing viscous and mucinous gelatinous material, which is usually seen in middle-aged individuals. It is similar to soft-tissue ganglion cysts present in other parts of the body. We, hereby, report a rare case of idiopathic intraosseous ganglion of the tibia in a young adult who presented with chronic pain in his lower leg. The patient was subjected to X-ray and MRI investigations and managed by excisional curettage of the lesion and histopathological examination. On histopathology, the diagnosis of intraosseous ganglion was confirmed. On 2-year follow-up, the patient was doing well without any complication and recurrence. This report describes the diagnosis and successful management of intraosseous ganglion at a very rare site in a young individual. More in-depth studies are required to understand the predisposing factors, underlying pathology, early diagnosis, and management of this lesion.

**Key words:** Intraosseous ganglion; Benign cystic bone lesion; Intraosseous mucoid cyst; Subchondral bone cyst; Curettage

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## INTRODUCTION

Intraosseous ganglion is a benign cystic bony lesion without degenerative arthritis and is similar to other ganglia found in soft tissues and tendons. It is also known as bible cyst, intraosseous mucoid cyst, juxta-articular bone cyst, and ganglionic cystic defect of bone. It is noninflammatory and nonneoplastic, and its histology is very similar to that of soft-tissue ganglion cysts.<sup>1-3</sup> Such ganglia is usually found in middle-aged individuals and is very rare in children, with maximum incidence observed in the age group of 40–50 years.<sup>4</sup>

Intraosseous ganglion is a unilocular or multilocular bone cyst that contains viscous and mucinous gelatinous material,

without any epithelial or synovial lining but is surrounded by a thin fibrous tissue. The most common presentation of intraosseous ganglion is mild localized pain without any visible swelling, deformity, or skin changes. Due to repeated microtrauma, intraosseous connective tissue undergoes mucoid degeneration, and there is herniation of juxtacortical soft-tissue ganglion into the underlying bone. The common sites of intraosseous ganglion are the epiphysis of long bones, such as medial malleolus, femoral head, proximal tibia, carpals (scaphoid and lunate), and acetabulum. Other uncommon sites are proximal humerus, ilium, talus, tarsal bones, distal ulna, metacarpals, olecranon, distal end radius, lateral malleolus, and proximal fibula. The distal tibia is a very rare site for intraosseous ganglion.<sup>5,6</sup>

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On X-ray, intraosseous ganglion typically presents as a well-demarcated, unilocular, or multilocular cystic lesion, with sclerotic margins. The bone scan shows increased radiotracer uptake.

## CASE PRESENTATION

A 25-year-old male was admitted to the Department of Orthopedics with the chief complaint of mild, localized pain over the distal tibia, persisting for 1 year, which was insidious on onset and could be initially managed with analgesics. The pain was progressive in nature, nonradiating, and was not associated with fever or other symptoms. No diurnal variation in pain was reported. The pain was relieved with medication and rest and aggravated upon prolonged walking, running, and other physical activity. The intensity of pain was reported to have increased from the past 2 months at admission. There was no history of trauma or preceding inflammatory disease.

On clinical examination, there was no visible swelling, deformity, or skin changes and no local increase in temperature. However, mild bony tenderness over the posterolateral side of the distal tibia was noted. There was no restriction of ankle movements or tenderness over the ankle. No vascular or distal neurological deficit was noted. The X-ray showed a small radiolytic lesion, with sclerotic margins, over the distal tibia (Figure 1).

There was no degenerative change in the ipsilateral adjacent joints, namely the ankle and knee. On MRI, we found a well-defined small intraosseous cystic lesion, measuring 9×11 mm, in the distal tibia along the posterolateral border, which was hyperintense on T2WI and hypointense on T1WI. On contrast MRI, there was a lack of gadolinium enhancement, confirming the cystic nature

of the bony lesion (Figures 2-5). No surrounding edema, periosteal thickening, or reaction was noted. Moreover, no pathological or stress fracture was noted at the affected site.

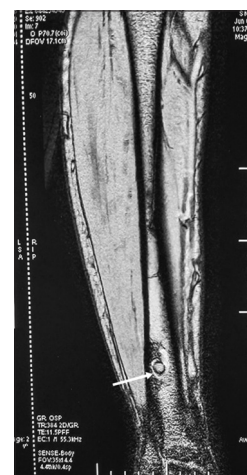
Blood investigations were performed for the complete blood count, erythrocyte sedimentation rate, C-reactive protein, rheumatoid arthritis factor, and serum uric acid. All inflammatory markers were found to be within the normal range. The initial treatment with analgesics for a period of 4 months failed to resolve the symptom. The patient was managed by excisional biopsy of the lesion under spinal anesthesia. An anterior incision was made over the distal tibia after localization of the cystic lesion under the guidance of C-arm. After soft-tissue dissection, bone was exposed and a cortical window was made; intraoperatively, gelatinous material was found surrounded by a capsule-like structure. The curettage was done, and after thorough washing, the wound was closed. The gross examination of the biopsied tissue showed irregular, grayish-white,



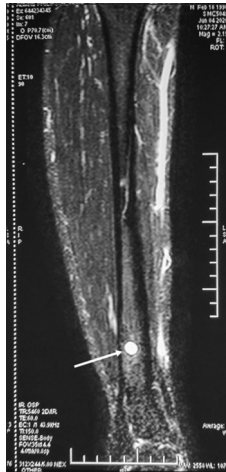
**Figure 2:** MRI showing well-defined small unilocular intraosseous cystic lesion along posterolateral border of the distal tibia, which is hyperintense on T2WI



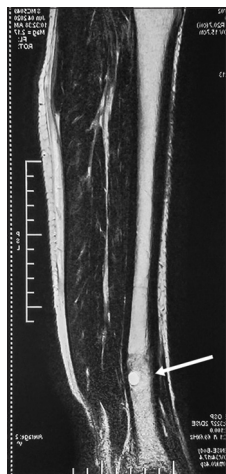
**Figure 1:** Preoperative X-ray showing small solitary unilocular lytic lesion with thin sclerotic margins, in the distal tibia



**Figure 3:** MRI showing a well-defined intraosseous lytic lesion in distal tibia with thin sclerotic rim, which is hypointense on T1WI



**Figure 4:** MRI showing a well-defined intraosseous lytic lesion in distal tibia, with a thin sclerotic rim on STIR



**Figure 5:** MRI showing a well-defined intraosseous lytic lesion in the distal tibia, hyperintense on T2WI

fragmented tissue bits, with slightly lobulated appearance on the outer surface. On sectioning, thick mucinous and gelatinous fluid were present in multilobulated fibrous tissue compartments. Microscopic analysis revealed degenerated bony trabeculae with hemorrhage and thick proteinaceous material; epithelial linings were not observed, giving an impression of a benign lesion of intraosseous ganglionic cyst of the distal tibia. Postoperatively, the patient was kept non-weight bearing. The pain was significantly reduced and the patient was discharged without any complication. On 2-year follow-up, the patient remained asymptomatic and showed progressive radiological healing of the lesion (Figure 6). The patient returned to work, and there was no sign of recurrence or complication.

## DISCUSSION

Intraosseous ganglion is a rare benign bony cystic lesion, which is histologically similar to the soft-tissue ganglion



**Figure 6:** Postoperative X-ray showing complete healing of the lesion

of tendon found in other parts of the body. The ganglia are very common near joints, tendons, and tendon sheaths; however, they are very rarely reported to occur inside the bone.<sup>1-3</sup> Intraosseous ganglion grows slowly and on radiography, it shows a well-defined rim of sclerosis, without any degenerative change in adjacent joints. Other terminologies used for intraosseous ganglion-type lesions are subchondral bone cysts, synovial bone cysts, intraosseous mucoid cysts, and juxta-articular bone cysts.

Intraosseous ganglion is a unilocular or multilocular lesion containing viscous and mucoid gelatinous material located in the subchondral bone adjacent to the joint.<sup>7,8</sup> Small bony lesions may be unilocular but large ones are often multilocular. Most of the intraosseous ganglionic cysts are 10–50 mm in size, however, a few are as large as 50 mm.<sup>5</sup> Sakamoto et al. reported the size of intraosseous ganglion to range from 6 to 40 mm, with a mean of 22.4 mm, in a series of 17 treated cases.<sup>9</sup> Sood et al. reported an unusual case of unilocular intraosseous ganglion of size 55×40×30 mm on anterior aspect of the right distal tibia.<sup>10</sup> In the present case, the size of the intraosseous ganglion was 9×11 mm.

Intraosseous ganglion cysts are very distinct from subchondral cysts. Subchondral cysts are located near the joints; they communicate with the joint cavity and are filled with synovial fluid whereas intraosseous ganglion cysts rarely communicate with the joint cavity and are filled with viscous and gelatinous mucoid material. Subchondral cysts are radiological features of degenerative changes in the joint in osteoarthritis and rheumatoid arthritis but intraosseous ganglionic cysts are rarely associated with degenerative changes in the joints.

Although the etiology of the intraosseous ganglion remains unknown, various theories have been postulated.<sup>11,12</sup> There are two types of intraosseous ganglionic cysts - those that originate by herniation of an extraosseous ganglion

**Table 1: Reported cases of intraosseous ganglion in the distal tibia**

S. No.	Author	Year	Description of the case	Size of the cyst	Management	Recurrence	Follow-up duration	Continuity with nearby joint	Remark
1	Schajowicz et al. <sup>5</sup>	1976	Reported 88 cases of intraosseous ganglia (age 14–73 years). Thirteen cases were located in the distal tibia	10–50 mm	Excision/ curettage with bone grafting of large cavity	One	NA	Absent	Described two types of intraosseous ganglion – Idiopathic and Penetrating
2	Dungan et al. <sup>14</sup>	1989	42-year-old female left distal tibia	20×16 mm	Surgical Excision with bone grafting	None	7 months	Absent	Patient was operated for parathyroid adenoma 2-year earlier
3	Buchler et al. <sup>12</sup>	2009	Reported 5 cases of intraosseous ganglia of the distal tibia (age 32–61 years)	8×9 mm to 20×22 mm	Curettage with bone grafting	None	24–69 month	Present in all cases	Arthroscopic assisted removal of all intraosseous ganglia
4	Sakamoto et al. <sup>9</sup>	2013	Reported 17 treated cases of intraosseous ganglia. Two cases were present in the distal tibia of right side (age 37 years and 40 years)	12 mm and 20 mm	Curettage with bone grafting	None	3 years	Absent	All cases were treated as an extension of biopsy.
5	Sedeek et al. <sup>13</sup>	2014	A 41-year-old female with recurrent right distal tibia intraosseous ganglion	10×20 mm	Curettage with bone grafting	None	2 years	Present	This case initially presented as a recurrent intraosseous ganglion Idiopathic type
6	Mohapatra et al. <sup>11</sup>	2018	A 20-year-old male right distal tibia	15×15 mm	Excision of the cyst with bone cement	None	7 years	Absent	
7	Sood et al. <sup>10</sup>	2019	A 60-year-old female anterior aspect of right distal tibia	55×40 mm	Curettage with bone grafting	None	2 months	Absent	Unusual large intraosseous ganglion with pathological fracture
8	Mei et al. <sup>15</sup>	2022	Reported 18 cases of intraosseous ganglion in the ankle region (age 22–74 years). Nine cases were located in distal tibia.	15 ×15 mm	Curettage with bone grafting	None	11 month–10 years	NA	Retrospective study on the diagnosis and treatment of intraosseous ganglion in the ankle region.
9	The current case	2023	A 25-year-old male right distal tibia	9×11mm	Surgical excision by curettage	None	2 years	Absent	Idiopathic type

into the underlying bone “penetrating” and those that are “idiopathic.”<sup>8</sup> The idiopathic type occurs due to chronic repetitive microtrauma or chronic ischemia, which leads to mucoid degeneration, and it is not associated with soft-tissue

extension. The repetitive microtrauma causes intramedullary vascular damage leading to aseptic necrosis of bone, as a consequence of which there is proliferation of histiocytes and fibroblasts and production of hyaluronic acid.

Intraosseous ganglionic cyst reported here was of the idiopathic type because there was no herniation of extraosseous ganglion into the distal tibia. It was not associated with soft-tissue ganglion or degenerative arthritis. The idiopathic type of intraosseous ganglion cyst originates from modified mesenchymal or synovial cells at the capsule-synovial interface in response to repeated microtrauma.

The presentation of benign cystic lesions within the tibia presents with nonspecific progressive leg pain and there are several differential diagnoses, including giant cell tumor, degenerative cyst, metastatic adenocarcinoma, and osteomyelitis. The radiological features of synovial and intraosseous ganglionic cysts are indistinguishable and pathologic evaluation is required to differentiate between the two. The giant cell tumor is usually large, with a less distinct osseous margin, compared with that in intraosseous ganglia.

On X-ray, intraosseous ganglia appear as well-defined lytic bony lesions with a rim of sclerosis in the juxta-articular or subchondral region, with or without cortical expansion and soft-tissue extension.<sup>7</sup>

The treatment of intraosseous ganglion is based on whether the lesion is painful or not. Many of these lesions are found incidentally on radiography. The surgical excision or curettage is the treatment of choice in most of the case to confirm the diagnosis as well as to relieve the pain. MRI is needed for diagnosis and operative planning and histopathological examination is required to confirm the diagnosis. Mohapatra et al. reported the management of the intraosseous ganglion of the distal tibia with curettage and bone cement and did not find recurrence or any complication at 7-year follow-up.<sup>11</sup> Buchler et al. reported arthroscopically assisted removal of five intraosseous ganglionic cysts of distal tibia. All patients underwent curettage and bone grafting through a cortical window made from a separate small incision. All patients had eventual relief of pain without recurrence and complication.<sup>12</sup> Sedek et al. reported a case of recurrent intraosseous ganglion of the distal tibia and treated effectively by curettage and autologous iliac cancellous bone graft.<sup>13</sup>

Considering the rarity of this disease in the tibia, more studies are required to understand the underlying pathology of the lesion and predisposing factors and to devise better strategies for early diagnosis and effective management (Table 1).

## CONCLUSION

Intraosseous ganglion is a benign cystic bony lesion filled with viscous and mucinous gelatinous materials, usually

found in middle-aged individuals. Intraosseous ganglion of the distal tibia is very rare. Herein, we have reported the presentation, diagnosis, and effective management of intraosseous ganglionic cyst in the distal tibia. The surgical excision or curettage was found to be an effective treatment for this rare condition; and large cavity was filled by bone graft or bone cement.

## CONSENT

The patient has provided informed consent of the case report to be published.

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**JA-** Concept and design, manuscript preparation, editing and revision, photographs, and treating surgeon; **BP-** Concept and design, manuscript preparation, editing, and revision; **AA-** Manuscript editing and revision; **VKS-** Manuscript editing; **MKS-** Manuscript editing.

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