Case Report

Secondary chondrosarcoma from a solitary osteochondroma of the fibula head: a case report

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Keywords: osteochondroma, secondary chondrosarcoma, fibula head tumour

Introduction

Osteochondromas are the most common primary, benign tumors of the bone. They account for nearly 35% of all primary benign bone tumors [1,2]. Osteochondroma is a disease of the young and usually presents during the second and third decades of life. Malignant transformation is considered as a serious complication of an osteochondroma. Less than 1% of all osteochondromas undergo malignant transformation into secondary chondrosarcoma and literature reporting such cases is rare. In fact, no such cases have been reported in Sri Lanka. In this report, we discuss the surgical management and outcome of chondrosarcoma secondary to osteochondroma of the proximal fibula.

Case report

A 24-year-old female presented to the National Hospital of Sri Lanka with mild knee pain and an incidental finding of a lump over the posterolateral aspect of the upper end of the left leg for one year duration. The lump had progressively enlarged over time and was associated with mild movement restriction of the L/S knee joint. However, no atrophy of muscles, fever or weight loss was seen. There was no significant medical history or family history of bone tumors.

Physical examination of the patient revealed an intact neurovascular status in both lower extremities. Mild tenderness over the posterolateral aspect of the left knee joint was elicited on direct palpation over the fibular head. The range of motion of the knee joint was limited secondary to antalgic guarding. No palpable lymph nodes or lumps elsewhere in the body were noted. The rest of the physical examination was normal.
A plain radiograph showed a large, irregular, exophytic, postero-lateral growth localized to the head of the fibula with sclerotic margins of adjacent tibia (Figure 1). MRI showed a bony outgrowth of the proximal part of the left fibula, measuring 24mm x 34mm x 33mm in size, with continuation of both the cortex and medulla of the outgrowth with the native bone. Focal cortical breaching was seen in the posterior inferior region. Associated soft tissue component was seen with extension to the soleus muscle. Neurovascular bundle was seen separately and the distal femur and proximal tibia appeared normal. CT angiogram, which was performed to identify possible involvement of vascular structures, showed a speared popliteal artery with marked medial displacement.

![Figure 1: X Rays, anteroposterior view (a) and lateral view (b) of knee joint and proximal fibula showing an exophytic tumor mass arising from the head of the fibula](image)

A surgical decision for a wide local excision of the tumor was made. During the surgery, an irregular, firm, bony mass was seen to have extended from the fibular head posteriorly towards the popliteal fossa displacing the popliteal artery medially (Figure 2.1). The tumor was indenting into the posterior cortex of tibia with a clear plane of dissection (Figures 2.2(a) and 2.2(b)).
Intraoperative illustrations of tumor

| Figure 2.1 | Figure 2.2(a) | Figure 2.2(b) |
|------------|---------------|---------------|
| Intraoperative illustrations of tumor | Showing tumor bed with clear plane of dissection |

The popliteal artery and joint space were not involved. The lateral collateral ligament (LCL) was detached from its fibular attachment and a saw cut of the fibula made 4cm below its neck. Fibula head with bony tumor and its tibia tumor bed was resected out with part of the soleus muscle and sent for histology. LCL reconstruction was not done during the surgery. AP and lateral view radiographs of the right knee after surgery showed evidence of a completely resected tumor mass with intact normal joint space. Patient recovery was unremarkable apart from foot drop noted during the immediate post-operative period. We kept the patient non-weight bearing for 3 months and knee and ankle joint physiotherapy was started along with a hinge knee brace and foot drop splint.

Pathological findings of the specimen revealed a nodular mass of bony tissue covered by a cartilaginous cap measuring 50mm x 50mm x 30mm, an attached bony stalk measuring 45mm in length and multiple bony tissue fragments measuring 25mm x 15mm x 8mm. Sections from the lesion showed a chondroid cap composed of nodules of cartilages separated by wide fibrous bands showing no hypercellularity, crowding or significant nuclear pleomorphism in most areas. The maximum cartilaginous cap thickness was 20mm [Figure 3.1(a)] and reached the resection margin fully. However, there were foci showing mild cellular crowding and occasional bi-nucleated cells with minimal cellular atypia. [Figure 3.1(b)] A focus showing infiltration of cartilaginous tissue into the surrounding soft tissue was also seen. [Figure 3.1(c). Although morphological features were of an osteochondroma, findings such as focal areas of cellular atypia, crowding and fibrous bands, along with a 20mm thick cartilaginous cap, which are beyond the benign spectrum, are in favor of possible low grade chondro-sarcomatous changes.
Follow up examination of the patient ten months after surgery revealed no knee swelling or pain. However, increased varus movement with a positive varus stress test was demonstrated. Range of movement of the joint was normal and there was no foot drop. Although high demand activities were restricted, patient could bear her weight during daily activities at home without significant difficulty. MRI was found to be normal one year after surgery.

**Discussion**

Osteochondroma is the most common "tumor" of the bones. Although chondrosarcoma is an unusual sequel of osteochondroma, the conditions are frequent enough to deserve discussion. The risk of malignant transformation in solitary osteochondromas is only 1%–2% [3]. However, it can be higher in multiple exostosis. Fortunately, most of the chondrosarcomas secondary to osteochondromas have proved to be low-grade. Ahmed et al reported 107 secondary chondrosarcomas arising in osteochondromas and found that 97 tumors were grade 1 and the rest were grade 2 [4]. Our patient also reinforces the theory that chondrosarcomas arising in osteochondromas are mostly low-grade neoplasms. However, other malignant histology has also been reported, less frequently, including osteosarcoma and pleomorphic high-grade sarcoma. In addition, a single case of Ewing sarcoma of the proximal fibula secondary to osteochondroma in a patient with multiple osteochondromas has been reported in the literature. The head of the fibula is a relatively rare site for secondary chondrosarcoma as it tends to occur most commonly in the pelvis, trunk and proximal femur.

Diagnosing secondary chondrosarcoma following osteochondroma is challenging. For such a diagnosis, unusual pain, growth of a long standing bony lesion beyond the bone’s skeletal maturity and images showing a bulky cartilaginous cap (more than 20mm), surrounding bony erosions, adjacent soft tissue mass with or without calcifications and T2 weighted MRI
showing high metabolic activity in the cartilage as shown by uptake of gadolinium is helpful [5,6,7]. Our patient showed focal area of cortical breaching in the posterior inferior part with an associated soft tissue component extending into the soleus muscle and early erosion into the adjacent tibia, suggesting malignant behavior.

Irrespective of the location, wide local surgical resection is considered as the treatment of choice when secondary chondrosarcoma is suspected. Primary biopsy for diagnosis is often found to be difficult to interpret histologically and excision may be an unnecessary additional procedure [6]. Moreover, if the biopsied lesions prove to be malignant, simple excision may result in an intra-lesional tissue sample, which carries a high risk of recurrence and metastasis. Chondrosarcomas are generally not considered sensitive to radiotherapy or chemotherapy [4]. Many studies confirm that the most important factor determining the rate of local recurrence is adequate primary surgical excision rather than histological type. Patients with lesions of the trunk are found to have higher local recurrence compared to patients with tumors in the limb bones [4]. Difficulty in achieving tumor free margins might be a contributing factor for the higher recurrence rate seen in the trunk. Most of the recurrences after excision of secondary chondrosarcoma were reported within 5 years of surgery. The rate of distant metastasis of secondary chondrosarcoma was very low. One reason may be that most of tumors are low grade. Ahamed et al reported that none of the grade 1 tumors metastasized during follow up. A few patients diagnosed with high grade tumors had metastases. As with chondrosarcoma in general, the lungs were the most common site for metastasis [4].

Literature reporting fibular head chondrosarcoma is rare. Our patient had secondary chondrosarcoma of the fibular head with possible involvement of the proximal tibia-fibular joint and tibia. Close proximity to the deep peroneal nerve and popliteal artery may interfere with wide local excision of tumors involving the proximal fibula. Literature recommends wide excision of the proximal fibula with 2–3 cm of the diaphysis and muscle cuff in all directions with preservation of the common peroneal nerve (Malawer’s type I resection) or wider extra-compartmental resection of 6 cm of normal diaphysis, parts of the anterior and lateral muscle compartments, and the tibio-fibular joint extra-articularly with deep peroneal nerve (Type 2) depending on tumor size [8]. In this case, we could perform wide local excision while preserving most of the soft tissue as early adjacent invasions showed clear planes of dissection and tumor behavior was mostly benign.

Although we couldn’t perform LCL reconstruction during primary surgery, we planned to proceed with reconstruction later. Unlike traumatic disruption of the lateral ligamentous structure of the knee, Einoder et al reported that reconstruction might not be required following elective resection of the fibular head secondary to tumor. Furthermore, they reported that function was satisfactory in 6 such cases where fibular head resection was done without ligamentous reconstruction after 14 years of follow-up [9]. On the contrary, even with satisfactory tumor management, some research encouraged precise ligament and
muscle reconstruction as they found variable functional recovery without it [10]. One year after surgery, our patient revealed minimum or no functional disability or pain. However, increased varus movement with varus stress testing was detected and further follow up in view of a future need for LCL reconstruction was planned.

Conclusion
We report a rare case of chondrosarcoma that has arisen within an osteochondroma of the fibula head. Although osteochondroma is a common finding that may be subjected to routine assessment, this case report highlights the value of careful gross and histological examination of tumors to avoid missing unexpected comorbidity.

Consent for publication
Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

Acknowledgements
Dr. Modini Jayawickrama, Consultant Histopathologist, National Hospital of Sri Lanka, Colombo
Dr. P G N L Chandrasiri, Registrar (Histopathology), Post Graduate Institute of Medicine, University of Colombo, Sri Lanka
Dr. A Pallewattha, Consultant Radiologist, National Hospital of Sri Lanka, Colombo

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