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A Recurrent Benign Osteochondroma of ventral scapula associated with Snapping Scapula and Pseudoparalysis - A case report

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ABSTRACT
Introduction: Osteochondromas (often referred to as exostosis) are usually an incidental finding and are characterized by a benign bony protuberance covered with a cartilaginous surface in the metaphyseal region of long bones. Though they do not affect the patients directly; but cause indirect complications of varying degree of severity such as bursitis, neurovascular impingement, fracture of peduncle or malignant transformation. Imaging studies such as an MRI or CT, are useful aids in the diagnosis; however, biopsy is a mandatory requisite for a definitive diagnosis. Treatment modalities include open surgical or an arthroscopic resection and has good prognosis. Recurrence follows incomplete excision.
Presentation of case: In this report, we present a rare case of a recurrent, symptomatic scapular osteochondroma associated with scapular pseudowinging and snapping scapular syndrome in a 16-year-old female. Radiological findings exhibited by this tumour was highly characteristic. The tumour was then managed with surgical resection thereby resulting in an entire resolution of the patient's symptoms.
Discussion: This case was unique because she presented with recurrence 5 years after her previous surgery as she neared her age of skeletal maturity raising a suspicion of malignant transformation. And also she was having associated pain, pseudowinging and snapping scapula syndrome.
Conclusion: By reporting this rare case of a scapular osteochondroma that presented with scapular winging, we aim to increase the awareness of the unusual manifestations of osteochondroma, particularly atypical sites, signs, and symptoms. Furthermore, we've given a comprehensive description of this case and its management to help other surgeons who face similar cases.

Keywords: Osteochondroma, Snapping Scapula, Pseudoparalysis

INTRODUCTION
Two different schools of thought categorizing osteochondromas exists. Controversy exists if is to be considered as a pseudotumour or a neoplasm. Osteochondromas are the most common benign tumour constituting about 20%–50% of all benign bone tumors and 9% of all bone tumors and the location of a solitary exostosis in the scapula is relatively rare: comprising about 6.4% of cases as majority of them are seen in appendicular skeleton.[1-4] Though the debate continues, it certainly is an exostosis and usually seen during rapid

skeletal growth in children and in adolescence. Etiology remains unknown[5]. It is usually noted in patients, below 30 years of age, with a slighly male predilection; male: female ratio of > 1.5: 1.[6] Arising from the metaphysis, it typically projects away from the epiphysis. They can be either sessile or pedunculated, often with broadened metaphysis. Lesions usually present as a solitary lesion with an approximate size of about 4cm [7]. But multiple lesions also do occur.

It is to be noted that a separation of a fragment of growth cartilage occurs which suffers herniation. The loose piece of cartilage continues to grow and its subsequent endochondral ossification forms a cartilage covered bony salience.

This lesion is characterized by a cartilaginous cover that bears a close resemblance to the physis and depicted as a normal growth of a growth plate in an abnormal direction. Also, the fact that the lesion ceases to grow on skeletal maturity corroborates of a possible correlation[8].

Nonetheless, if the growth continues into adulthood, it should alarm the surgeon for possible malignancy. The cartilage cap shows variegated appearance ; could be be thick with rings and arcs, or thin and arduous to recognize, calcification and irregular subchondral bone. In most instances, expectant management would suffice.

Malignant transformation occurs in around ~1% in solitary osteochondromas and about ~5-25% with hereditary multiple exostoses; usually resulting in low grade chondrosarcoma (67-85% of cases), and surgery is usually curative (70-90%). Limb-sparing wide local excision usually suffices. [9-12]

Hallmarks of sarcomatous transformation are soft tissue mass that grows after skeletal maturity, pain after puberty, lucency (new), additional scintigraphic activity, destruction (cortical), thickened cartilage cap greater than 1.5 cm. Local recurrence could be attributed to spillage of cartilage cells into the resection bed with estimated recurrence rates at 2% and up to 15% respectively [11,13-15]. Both benign as well as malignant lesions show recurrence which is a characteristic trait. Consequently, it is imperative that the lesion be excised at the earliest, lest transformation of the mass into a malignant chondrosarcoma ensues.

CASE PRESENTATION

At the Orthopaedic and Trauma Clinic of Rajah Muthaiah Medical College and Hospital, Annamalai University, we observed a case of osteochondroma that was interesting from both the radiographic and clinical points of view, and the diagnosis was challenging because of the atypical location with nonspecific shoulder pain. A healthy 16-year-old female was admitted with recurrent swelling at the inner border of Right Scapula; which was noted by her mother 5 months ago and has grown in volume since then.

She had a history of similar complaints at the same site 5 years ago and was diagnosed to have Benign Osteochondroma and was surgically resected. Otherwise, the patient's history was unremarkable. At presentation, she had terminal restriction of glenohumeral abduction and bony outgrowth at the medial border of Right Scapula.

Clinically, a uniform, rounded, protuberant, bony mass with a diameter of about 4 cm was seen and palpated in the medial region of the dorsal aspect of the right scapular blade.

The large size of the lesion changed the profile of the medial scapula resulting in it appearing higher than the contralateral one. The lesion was painful on palpation.

On careful physical examination, Pseudo winging of scapula was evident from the marked uneveness in the symmetry of the upper body upon full abduction of the Right arm. (figure 1) On moving the joint to different positions, we could hear and feel grinding and clicking movements through auscultation and palpation. She was found to have an associated Snapping Scapula.

No neurovascular compromise of her upper limbs was found on assessment. She chose to seek medical attention for cosmesis, reduced range of motion and to exclude a possible recurrence and malignant transformation.

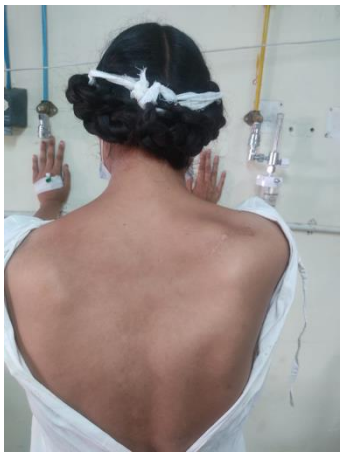
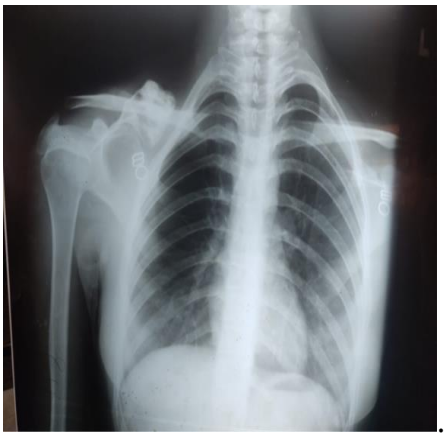


Figure 1: Clinical photograph showing pseudo-winging and deformity of the right scapula.



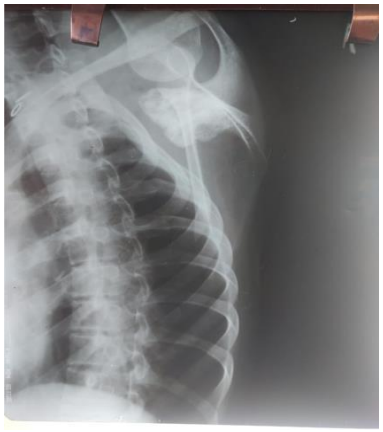
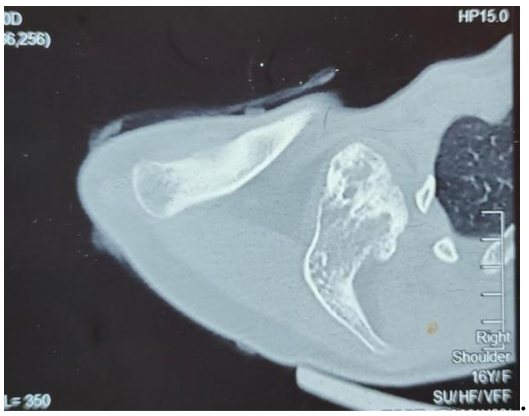


Figure 2 **Figure 3**

An Anteroposterior X-ray of the chest & Scapular Y views showed a well-defined bony outgrowth from the superomedial portion of the Right scapula.



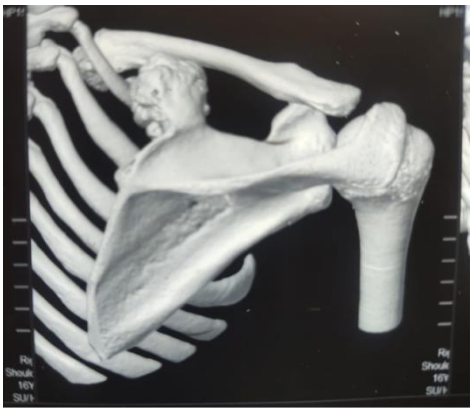


Figure 4 **Figure 5**

Fig-4, Fig-5: Three-dimensional CT scan images (reconstructed) showing a large bony tumour arising from the medial border of the body of the right scapula.

A CT scan revealed an anteriorly directed expansile, exophytic bone growth with scerotic, lytic regions and calcified cartilaginous portion at superior angle of medial border of the right scapula, measuring 4×2.9×1.5 cm. Characteristically, the medullary cavity showed continuity with the parent bone. (Figures 4 & 5)

The imaging studies were consistent with findings of a benign bone tumor the radiographic appearance of a solitary osteochondroma was pathognomonic. (figures 2–5) Also, protuberance of cortical and medullary bone from the underlying bone was seen in the imaging which substantiated the diagnosis. Yet, benign bone tumors like chondroblastoma, osteoid osteoma, osteoblastoma, enchondroma, fibroma, and periosteal chondroma were briefly considered as part of differential diagnoses.

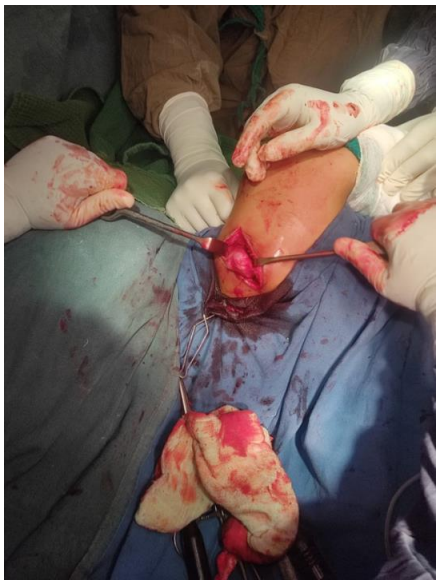


Figure 6: Exposure of the Osteochondroma

A bony hard tumour (4×2 cm) was excised in toto from the right scapula under general anaesthesia, by an utilitatrian incision over the previous surgical scar. Trapezius was incised and Levator Scapulae was erased. Delicate subperiosteal dissection done. The pedunculated osteochondroma was retracted to expose the full extent of the cartilage cap and excised from its base; thereby it was removed in toto. (Figure 6)

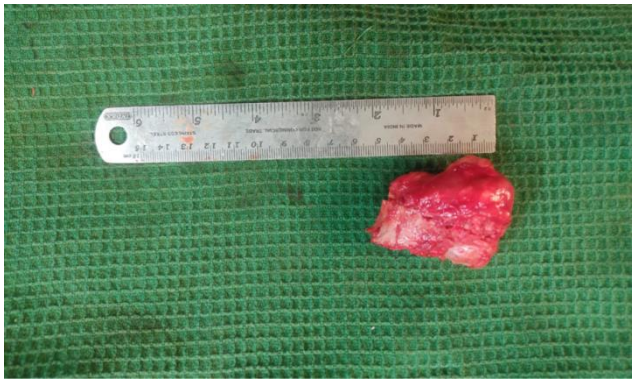


Figure 7: Excised specimen showing the bony tumour with cartilaginous cap.

The single lobular bony mass measuring 4 x 2 x 1.5 cm was sent for histopathology, which revealed no malignant transformation at the cartilage cap (Figure 7)

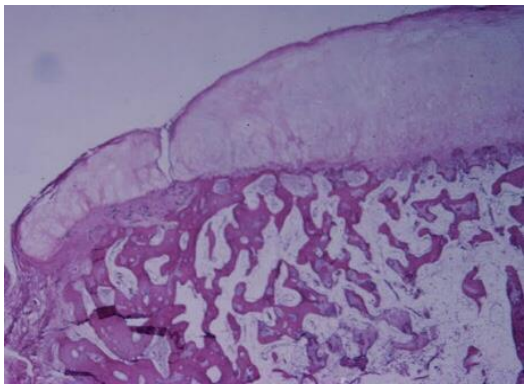


Figure 8: Histopathological section of the tumour showing a well-formed cartilage cap on the surface with a prominent enchondral ossification at the base, which continues into trabeculae of mature lamellar bone.

A microscopic examination showed a benign chondrogenic tumour composed of hyaline cartilaginous cap with overlying perichondrium and Enchondral ossification and mature bone trabeculae seen underneath the cap. Also, mature adipose tissue was seen interspersed between the bone trabeculae. Neither an atypical mitosis nor necrosis noted. Cartilaginous cap thickness measured 1.5cm (Figure 8)

Figure 9: Post Operative Clinical Photograph

Her postoperative course was uneventful. The complete lesion resection was ascertained radiographically, and the patient's deformity disappeared completely and she regained full range of glenohumeral and scapulothoracic movements with no pain, swelling, or scapular winging. She commenced her rehabilitation therapy in her immediate post operative period which she pursued on an outpatient basis (Figure 9)

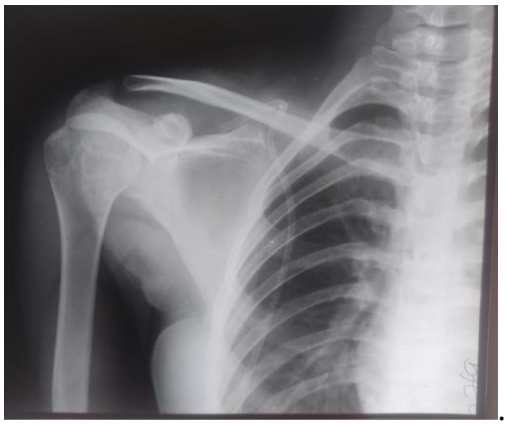
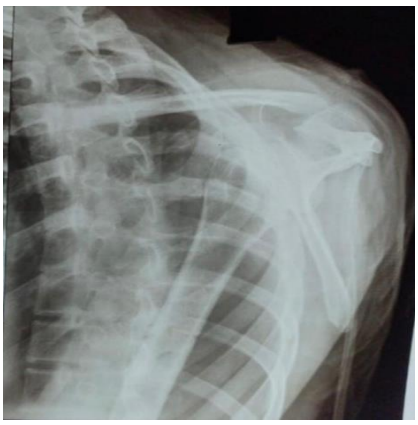



Figure 10 **Figure 11**

Fig-10, Fig-11: Post Operative X rays - Anteroposterior & Lateral X rays

DISCUSSION

Osteochondroma is defined as a cartilage-capped bony projection on the external surface of a bone that arises because of herniated epiphyseal cartilage through a periosteal defect during the endochondral ossification and contains a marrow cavity from the growth plate[5] explaining the lower predilection of osteochondroms in bones formed through intramembranous ossification.

As per the World Health Organisation affects 3% of the general population and >30% of all benign bone tumours. Metaphysis of tubular long bones such as the distal femur, proximal tibia, and proximal humerus (90%) are the most common sites for osteochondromas[1]. Scapular involvement is seen in only 4% of cases[2]. Of the overall scapular tumors,14.4% are osteochondromas arising mostly from ventral surface.[2]

The overwhelming majority of osteochondromas are recognized in the first or second decade of life, as most are asymptomatic and the tumour growth typically ends with physis closure[3].

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Osteochondroma in long bones can be easily discerned from plain radiographs such as X-rays; however, diagnosis of osteochondroma on flat bones demands more specialized modalities such as MRI. Only very few reports exist in literature about the involvement of ventral surface of the scapula. Boinet in 1867 was the first to describe Snapping Scapula syndrome where in an audible and/or palpable crepitus of scapula on scapulothoracic movement is accompanied with pain[4]. A Solitary exostosis on the ventral side of scapula may cause snapping shoulder syndrome and static winging.

Inappropriate treatment leads to subsequent complications such as pseudo-winging, restricted movement of the shoulder due to fibrosis, snapping, and bursa formation. Nevertheless, one could adopt a wait and watch approach in a small sized osteochondroma in age groups ranging from pediatric to young adults. Careful monitoring is mandatory.

Supportive management includes Immobilization, anti-inflammatory medications & physiotherapy. Osteochondromas are treated with Open or arthroscopic excision.

Osteochondromas are usually asymptomatic, slow-growing masses on the involved bonefound inadvertently. Still, some complications do occur, such as a bursal inflammation, or bony deformity, fracture at the base of the peduncle causing pain, swelling, or joint problems[5]. Patients with Osteochondroma of the scapula are usually symptomatic; The

common presentations are weakness of shoulder girdle muscles, pain on movements, crepitus, restriction of movements and other compressive effects on the surrounding anatomical structures. Additionally, bursa formation, pseudo winging, snapping scapula, chronic pain, and cosmetic deformities can also occur [6 - 8].

At the time of presentation, our patient had pseudowinging of scapula, restricted abduction along with pain on palpation. On reviewing the literature, most lesions were located along the scapular equator and those arising at the inferior part of scapula tend to be larger in size due to a lack of space restriction[9]. Furthermore, an internally projecting osteochondroma are misdiagnosed as scapular winging in some cases. In our case, the scapular lesion grew on the superior angle on the medial border on the dorsal aspect and was relatively larger than other reported osteochondromas. It is rather unusual to encounter a sarcomatous change in a osteochondroma, and is infrequent in a solitary osteochondroma compared to multiple osteochondromas. However, sudden growth after the third decade of life is a red flag sign and must raise suspicion of its malignant transformation into chondrosarcoma, which necessitates appropriate management. This change is evinced by the measurement of cartilage thickness of greater than 2cm on a CT scan or MRI[2,6]. Other significant signs are myxoid changes, fibrous banding, lobulations, and gadolinium enhancement. Our patient had recurrence and presented at an age nearing her skeletal maturity warranting us to suspect malignant transformation. A diagnosis of osteochondroma is gleaned from on clinical examination and radiological imaging studies, and substantiated via histopathology[4,10].

In our patient, histology divulged endochondral ossification with a cartilage cap, and CT revealed a large, mushroom-shaped exostosis in the dorsal aspect of the right scapula. Osteochondromas are usually managed surgically unless the skeleton is immature. Literature describes different resection techniques for scapular lesions and all concur that resection should be in its entirety along with its stalk, if present. A minimally invasive approach as described by Nascimento involves a small incision, minimal dissection, and preservation of muscle attachments reducing the need for immobilization postoperatively and during rehabilitation[9]. By contrast, Kwon demonstrated that excision of a lesion at the ventral aspect of the scapula following open technique surgery resulted in improvement of mechanical symptoms in his patients [4]. A similar technique to that of Kwon[4] was employed to treat our patient successfully. She was relieved of her symptoms, attained complete resolution and regained a full range of motion in his affected shoulder. Nevertheless, with larger tumors, it may be impossible to salvage the scapula by performing partial or complete scapulectomy.

CONCLUSION

Osteochondroma being infrequent over scapula presents a diagnostic challenge in itself for a clinican.[2] Fortunately our patient had a recognizable pseudowinging of the scapula, an apparent bony nidus on the ventral aspect of the scapula impeding her full abduction coupled with a past history of osteochondroma at the same site presenting with similar complaints necessitating surgical removal pointed towards a diagonosis of a possible recurrence of osteochondroma. It is therefore put forth that any bony hard swelling over the scapula causing mechanical obstruction to movements or neurovascular structures or pain could be very well due to osteochondroma and this entity should be kept in mind while figuring out the differential diagnoses.

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