CASE REPORT

Primary osteosarcoma of the clavicle and the perils of bone biopsy

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SUMMARY

We present a rare case of delayed diagnosis of osteosarcoma of the medial clavicle in a young man. He presented following a pathological fracture with a falsely reassuring core and fine-needle aspiration biopsy. The initial biopsy was suggestive of an aneurysmal bone cyst and was therefore treated conservatively without further follow-up. The rapid increase in size over the next 8 months triggered a repeat presentation and subsequent repeat biopsy. The open biopsy confirmed high-grade osteosarcoma and the patient underwent claviculectomy with neoadjuvant chemotherapy.

BACKGROUND

This case highlights the importance of appropriately selected biopsy techniques in primary bone malignancies. Despite the impressively large superficial mass, this patient had a significant delay to diagnosis. The case serves as an important reminder of the principles of bone biopsy to the surgical community.

CASE PRESENTATION

A 27-year-old man with a 3-month history of an expanding growth to the medial third of the right clavicle was referred by his family doctor to a musculoskeletal tumour service. He was clinically well with no recent weight loss, lethargy or fevers. On examination, the 10 cm mass was fixed, rounded, firm and mildly tender to palpation. The patient's medical history was significant for a pathological fracture of his right clavicle 10 months prior to presentation, sustained by a low mechanism of force injury while abroad. Plain film radiographs at the time of initial injury identified an undisplaced fracture of medial third of right clavicle, with suspicious features for an underlying aneurysmal bone cyst. The fracture was managed conservatively.

INVESTIGATIONS

Initial assessment by the patient's local general practitioner in consultation with a general thoracic surgeon led to arrangement of a CT-guided bone biopsy (figure 1A); fine-needle aspiration (FNA) and core biopsies were both performed. Histopathological examination reported a giant cell-rich cellular spindle cell neoplasm, suggestive of an aneurysmal bone cyst. No cytologically malignant cells were seen.

Following the biopsy, the patient reported ongoing slow growth of the mass for a period of 8 months before a sudden phase of rapid growth, which prompted re-presentation (figure 2). Urgent imaging was obtained, with plain radiographs

showing a completely eroded medial end of the right clavicle, replaced by a large mass with peripheral calcification. Further CT imaging demonstrated a large intraosseous partially vascular soft tissue lesion measuring 9.7×7.8×7.2 cm, with no evidence of lymphadenopathy. A whole body bone scan confirmed no features of distant metastatic disease. MRI demonstrated a large soft tissue mass with osteoid matrix with locally aggressive features and internal fluid levels. Repeat MRI 6 weeks later revealed increasing size and mass effect compression of the right subclavian and inferior internal jugular veins as well as mild midline shift of the trachea (figure 1B).

TREATMENT

Open incisional bone biopsy revealed a bone-forming malignant spindle cell tumour consistent with osteosarcoma. The patient received two cycles

Figure 1 (A) CT-guided bone biopsy 20 days following pathological fracture and (B) axial plane T1-weighted MRI 404 days postinitial injury, demonstrating extrinsic mass effect on the upper airway and local vasculature.

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Figure 2 Clinical appearance at presentation to the musculoskeletal tumour service 404 days following initial pathological fracture of the right medial clavicle.

of neoadjuvant chemotherapy consisting of cisplatin and doxorubicin, as well as high-dose methotrexate and folinic acid before proceeding to partial claviclectomy. The resected specimen confirmed the pathology to be high-grade conventional

osteosarcoma of osteoblastic type with intramedullary expansion of the periosteum and focal infiltration into surrounding soft tissue (figure 3A–C). The percentage of tumour showing necrosis secondary to neoadjuvant treatment was less than 5%.

OUTCOME AND FOLLOW-UP

Postclaviclectomy, the patient was continued on a further 6 months of adjuvant chemotherapy. CT and MRI at 9 months postoperatively demonstrated no recurrence of malignancy.

DISCUSSION

Primary tumours of the clavicle are exceedingly rare, comprising less than 1% of primary bone tumours.¹ Despite being anatomically superficial and therefore presumably readily apparent to prompt patients to seek early clinical review, delays in diagnosis are not unusual.² Reasons for diagnostic delay are likely to be due to an insidious and generally asymptomatic onset, low index of suspicion for malignancy and features that are often indeterminate on plain radiographs.³ The diagnosis and management of primary malignant tumours of the clavicle are made challenging by the frequency of benign shoulder girdle pain symptoms, their clinically aggressive natural history and their variable response rate (50–80%) to chemotherapy.⁴ FNA biopsy is an appealing investigative option in this circumstance, due to ease of access, lower degree of invasiveness and small risk of procedure-related complication. FNA is feasible for osteolytic lesions of the clavicle with an eroded cortex⁵ and is reported to provide a variable sensitivity of 79–96%.^{6–7} However, FNA biopsy specimens from primary bone tumours are more likely to be insufficient or non-diagnostic, which may lead to diagnostic delays.² Core biopsy is minimally invasive and maintains tissue architecture, with a reported diagnostic accuracy of over 95%.⁸ FNA and core biopsy modalities have potential to seed the tumour cells. In this case, the tract used for open incisional biopsy was reused and excised during the definitive tumour resection. Open biopsy permits a larger biopsy sample, allows a wider panel of ancillary tests and is associated with a diagnostic accuracy nearing 100%.^{7–9} The differing biopsy techniques have merits and disadvantages. A general biopsy principle of ‘bigger is better’ to achieve improved sensitivity, is maintained and recommended.

The FNA biopsy in this case was acellular and therefore non-contributory for diagnostic purposes. The concurrent core biopsy revealed non-specific histopathology with a giant and spindle cell pattern, not necessarily confirmatory of aneurysmal bone cyst. Osteosarcomas frequently have a heterogeneous histomorphology and may contain cystic, haemorrhagic, necrotic, chondroid or fibrous areas, which can be misleading in

Figure 3 Macroscopic appearances of the (A) intact en bloc resected tumour, (B) transversely (sagittally) sliced specimen demonstrating two small areas of necrosis and one cystic focus and (C) longitudinal (coronal) slice demonstrating tumour erupting from the medial clavicle.

Learning points

- Our experience highlights the importance of prompt and satisfactory biopsy specimens to confirm osteosarcoma diagnosis and expedite definitive management.
- Reliable pathological assessment is dependent on the provision of adequate well-preserved material that is fully representative of the lesion.
- The limitations of small biopsy specimens and the potential for sampling error therefore must be understood.
- Lesions of the clavicle should be considered clinically malignant until proven otherwise and promptly referred to a bone oncology unit.

diagnostic assessment. If the biopsied areas are osseous and/or the overtly malignant areas are not included in the specimen,⁴ as occurred in our case, the diagnosis may be missed.

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