

A RARE CASE OF LOCALISED EWINGS SARCOMA OF THE SCAPULA: A MULTIDISCIPLINARY APPROACH

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ABSTRACT

Ewing's sarcoma (ES) of is a rare case that mostly occurs in young adults with male predominance. Ewing's sarcoma mostly affects the skeletal system, and in rarer cases, it affects the scapula region. This paper reports a rare case of the scapula region and its multidisciplinary approach with VAC/IE regimen causing toxicity and measure to taken. **Presentation:** This case reports highlights the multidisciplinary approach of Ewing's sarcoma and highlights the VAC/IE regimen toxicities and its measures to avoid the toxicities in patient. **Conclusion:** The case report emphasized the need of early detection of sarcoma on scapula region and understand the treatment process and its early detection of toxicities to ensure the patients quality of life.

INTRODUCTION

Ewing's sarcoma is the second most common primary malignant bone tumor after osteosarcoma, accounting for three per cent of all childhood malignancies.^[1] In Ewings sarcoma family tumour (ESFT) Primary bone tumour account for 5% of all child and adolescent cancers, and ESFT is the second most common primary bone tumour.^[2] Primary bone tumour and tumour like lesions of scapula are uncommon. Primary bone tumour of scapula is more likely to be malignant than benign. To the best of our knowledge less than 15 cases of Ewing's sarcoma of scapula have been reported.^[3] The scapula is an extremely rare site for primary bone tumors, with approximately only 3% of bone tumors arising from this site. Extra-skeletal ES mainly occurs in the paravertebral area

and lower extremities and rarely in upper extremities.^[4]

There is no well-established association between Ewing sarcoma and environmental risk factors, drug exposure, radiation history, or cancer history in the family. Studies have been limited to small retrospective, case-control studies.^[5] The most common regions of metastasis are the lungs and pleural cavity, the skeletal system, and bone marrow, or a combination of these.^[6]

VAC/IE regimen includes vincristine, doxorubicin, and cyclophosphamide (VAC) with ifosfamide and etoposide (IE). Doxorubicin-induced cardiotoxicity (DCT) is defined by a decline in left ventricular ejection fraction of more than 10% to a value smaller than 53%.^[7]

According to the most recent statistics, up to 48% of patients receiving doxorubicin can develop heart failure.^[8]

CASE REPORT

Basic information of patient

Age- 17 years

Gender- Male

Chief complaints

Pain at the site of right scapula region.

History of present illness

A-17 years old male presented to hospital with complains of pain at the right shoulder or upper back worsens at night or with activity, swelling, restricted shoulder movement.

History of past illness

No past medical history.

Family history

No Known Family History of Malignancies.

General examination

- The patient was conscious, oriented, and cooperative
- Well-nourished, moderately built
- No pallor, icterus, cyanosis, clubbing, pedal edema, or lymphadenopathy

- No signs of dehydration or cachexia.

Systemic Examination

Cardiovascular system

- Heart sounds are heard clearly and are normal with no murmurs.
- The pulse in both wrists (radial arteries) is felt at the same time indicate normal blood flow to both arms.
- The wrist pulse and groin pulse (femoral artery) occurs at the same time indicates normal blood flow

Respiratory system

- Normal bilateral Air Entry with Normal vesicular breath sounds.

Central nervous system

- Glasgow comma scale: 15/15- the patient is fully conscious and alert and
- The neurological findings are normal with no focal neurological deficits.

Abdominal examination

- Soft, non-tender and bowel sounds are present from heart to the lower body.

LABORATORY INVESTIGATIONS

IMMUNOHISTOPATHOLOGY OF RIGHT POSTERIOR SHOULDER LESION BIOPSY SHOWS

Malignant Small round cell tumor.

Table 1: Immunohistochemistry.

IMMUNOHISTOCHEMISTRY	
CD99 -Diffuse	strong positive
Synaptophysin	Negative
Vimentin	Diffuse strong positive
NKX2.2	Diffuse strong positive
FLI-1	Weak positive
ERG	Positive

Immunohistochemistry suggest CD99-diffuse positive indicate the small round cell blue tumour, vimentin diffuse strong positive suggest the tumour is mesenchymal origin, NKX2.diffuse strong positive is a specific marker for Ewings sarcoma, ERG positive indicates vascular tumor.

2D ECHO

IMPRESSION: EF 65%

NO LV RWMA

TRIVIAL TR WITH NO PAH

NORMAL LV/RV FUNCTION

NO CLOTS /PE

FDG PET CT SCAN SHOWS

Hyper metabolic lytic lesion involving right scapula with adjacent periosteal reaction and heterogeneously enhancing soft tissue component and suggestive of biopsy proven primary malignancy and Multiple minimally hypermetabolic bilateral cervical lymph nodes are likely inflammatory.

The above makers highly suggestive the Ewing's sarcoma

DIAGNOSIS

Ewing's Sarcoma Localized: Right Scapula Region.

Treatment protocol

The patient is diagnosed with Ewing's sarcoma Localized at Right Scapula Region and planned for Neo Adjuvant Chemotherapy with VAC/IE 3 Weekly. The patient gone through the 4 cycles of planned chemotherapy cycles of regimen VAC/IE. After the 4 Cycle of regimen VAC/IE patient gone through the WLE (wide local excision). S/P: Partial scapulectomy.

After the successful removal of tumour PET CT shows Complete metabolic response. HPE shows Margins free, Residual disease present, 20% necrosis with Involvement of Infraspinus muscle. Patient was referred for Adjuvant Radiation therapy. Patient Tolerated Treatment Well.

Radiation therapy

Patient was treated with External RT of 5sessions to a dose of 45 Gray in 25 Fractions to tumor bed with adequate margins.

AFTER 9 CYCLES

Patient went through the induction phase of VAC/IE regimen where the main goal is to remove the disease and tumour. After the induction phase the patient whole body pet CT

finding that is likely a benign (non-cancerous) condition, such as a healed injury or a chronic degenerative change, in the bony prominence at the top of the right shoulder blade.

Comparison of FDG avid reports to previous reports shows that the patient showing the complete and effective response to the treatment.

Table 2: Induction phase of treatment.

INDUCTION PHASE OF TREATMENT	
CYCLE 1	DAY 1 VINCRISTINE 2MG +DOXORUBICIN75 MG +CYCLOPHOSPHAMIDE 1500MG
CYCLE 2	DAY 1-5 CHEMOTHERAPY WITH ETOPOSIDE120MG +IFOSFAMIDE2200MG +MESNA700MG
CYCLE 3	DAY 1 CHEMOTHERAPY WITH VINCRISTINE1.9MG +DOXORUBICIN 75MG +MESNA 350MG +CYCLOPHOSPHAMIDE 1400MG
CYCLE 4	DAY 1-5 CHEMOTHERAPY WITH ETOPOSIDE110MG +IFOSFAMIDE 2000MG +MESNA 600MG
RADIATION	Patient was treated with External RT to a dose of 45 Gy in 25 Fractions for 5 days, to tumor bed with adequate margins.
CYCLE 5	DAY 1 CHEMOTHERAPY WITH VINCRISTINE 2MG +DOXORUBICIN 75MG + MESNA 350MG +CYCLOPHOSPHAMIDE 1500MG
CYCLE 6	DAY 1-5 CHEMOTHERAPY WITH ETOPOSIDE120MG +IFOSFAMIDE 2000 MG+MESNA 600MG
CYCLE 7	DAY 1 CHEMOTHERAPY WITH INJ VINCRISTINE2MG + INJ DOXORUBICIN 75MG + INJ MESA 350MG + INJ CYCLOPHOSPHAMIDE 1500MG
CYCLE 8	DAY 1-3 CHEMOTHERAPY WITH INJ ETOPOSIDE120MG + INJ IFOSFAMIDE 2000MG + INJ MESNA 600MG
	DAY 4-5 CHEMOTHERAPY WITH INJ ETOPOSIDE100MG + INJ IFOSFAMIDE 1800MG + INJ MESNA 600MG
CYCLE 9	DAY 1 CHEMOTHERAPY WITH INJ VINCRISTINE 2MG + INJ DOXORUBICIN 75MG + INJ MESA 350MG + INJ CYCLOPHOSPHAMIDE 1500MG

Patient started the consolidation phase of VAC/IE regimen where the main goal is to maintain and strength the treatment response.

Table 3: Consolidation phase of treatment.

CONSOLIDATION PHASE	
CYCLE 10	DAY 1-3 CHEMOTHERAPY WITH INJ ETOPOSIDE120MG + INJ IFOSFAMIDE 2000MG + INJ MESNA 600MG DAY 4-5 CHEMOTHERAPY WITH INJ ETOPOSIDE100MG + INJ IFOSFAMIDE 1800MG + INJ MESNA 600MG
CYCLE 11	DAY 1 CHEMOTHERAPY WITH INJ VINCRISTINE 2MG + INJ DACTINOMYCIN 1.3MG + INJ MESA350MG + INJ CYCLOPHOSPHAMIDE 1500MG
CYCLE 12	DAY 1-3 CHEMOTHERAPY WITH INJ ETOPOSIDE130MG + INJ IFOSFAMIDE2000MG + INJ MESNA 600MG DAY 4-5 CHEMOTHERAPY WITH INJ ETOPOSIDE110MG + INJ IFOSFAMIDE1800MG + INJ MESNA 600MG
CYCLE 13	DAY 1 CHEMOTHERAPY WITH INJ VINCRISTINE 2MG + INJ DACTINOMYCIN 1.3MG + INJ MESNA400MG + INJ CYCLOPHOSPHAMIDE 1400MG
CYCLE 14	DAY 1-3 CHEMOTHERAPY WITH INJ ETOPOSIDE130MG + INJ IFOSFAMIDE2000MG + INJ MESNA 600MG DAY 4-5 CHEMOTHERAPY WITH INJ ETOPOSIDE110MG + INJ IFOSFAMIDE1800MG + INJ MESNA 600MG
CYCLE CONTINUATION	
CYCLE 15	DAY 1 CHEMOTHERAPY WITH INJ VINCRISTINE 2MG + INJ DACTINOMYCIN 1.3MG + INJ MESA 400MG + INJ CYCLOPHOSPHAMIDE1400MG
CYCLE 16	DAY 1-3 CHEMOTHERAPY WITH INJ ETOPOSIDE 110MG + INJ IFOSFAMIDE 2000MG + INJ MESNA 600MG DAY 4-5 CHEMOTHERAPY WITH INJ ETOPOSIDE110MG + INJ IFOSFAMIDE1800MG + INJ MESNA 600MG

The above table summarizes the consolidation phase of the chemotherapy cycle, including the administered drug dosages. The variation in the dosing schedule of etoposide and ifosfamide between days 1–3 and days 4–5 of the VAC/IE regimen is designed to minimize treatment-related toxicity. Fractionated administration helps reduce the risk of ifosfamide-induced hemorrhagic cystitis, bladder toxicity and limits the likelihood of etoposide-associated hypotension during infusion.

Furthermore, substitution of doxorubicin with dactinomycin was undertaken to mitigate the risk of anthracycline-induced cardio-toxicity. This modification highlights the importance of individualized treatment planning and careful toxicity monitoring during chemotherapy in patients with Ewing sarcoma.

“Although 14 cycles of interval-compressed VAC/IE chemotherapy constitute the standard protocol for localized Ewing sarcoma, additional cycles were administered in our case to

ensure sustained disease control and minimize the risk of recurrence, considering the tumour characteristics and multidisciplinary team decision.”

Table 4: 2D echo impression during the course of treatment.

2D ECHO	IMPRESSION
Before chemotherapy started	EF normal
After cycle 4	EF normal
After cycle 11	EF normal
After cycle 15	EF normal

CALCULATION OF DOXORUBICIN CUMULATIVE DOSE

$$\begin{aligned} \text{Cumulative dose} &= \text{dose per cycle} \times \text{Number of cycles} \\ &= 75 \times 5 = 375 \text{ mg/m}^2 \end{aligned}$$

According to table 4 2D reports remained within normal limits throughout the treatment course. However, considering the cumulative cardiotoxic potential of doxorubicin, early substitution with dactinomycin was performed as a preventive measure to minimize the risk of long-term cardiac complications.”

In above case we get total cumulative dose is 375mg which is a warning sign (<300mg/m²) of Cardio toxicity. So, the substitution of doxorubicin with dactinomycin was undertaken in many paediatric Ewings protocol proactively switch earlier to protect the heart.

DISCUSSIONS

Ewings sarcoma of a scapula region is a rare presentation that predominantly occurs in children and adolescence. It is a highly malignant tumour composed of small, round cells that primarily affects the skeletal system. Ewings predominantly affects the lower extremities, while occurrence in the upper appendicular skeleton is relatively uncommon. The most frequent clinical presentation is localized pain at the affected site, often associated with swelling and restricted movement.

All though study on Ewings sarcoma family tumour is a well recognises entity, studies specifically addressing the scapula region is limited. Ewings sarcoma standard treatment follows VAC/IE regimen protocol which consists induction and consolidation phase. Ewings sarcoma management required multi model approach including chemotherapy, radiation therapy and surgical resection for optimal therapeutic response and improve survival outcomes. Early diagnosis of scapular involvement in the Ewings sarcoma is a crucial to

prevent disease progression and distant metastasis. Prompt recognition and timely initiation of therapy significantly improve prognosis, particularly in paediatrics and adolescent patients.

During treatment with the VAC/IE chemotherapy regimen, careful cardiovascular monitoring is essential, as anthracyclines such as doxorubicin are associated with dose-dependent cardiotoxicity. This is especially important in children and adolescents, who are more vulnerable to long term treatment related complication.

The present case highlights the importance of a multimodal management approach and emphasizes the need for early cardiology evaluation to detect and manage chemotherapy-induced toxicity. In cases where cardiac toxicity is suspected or confirmed, modification of the chemotherapy protocol may be warranted. Substitution of doxorubicin with alternative agents such as dactinomycin can be considered to reduce further cardiovascular risk while maintaining therapeutic efficacy. Proactive treatment adjustment plays a critical role in preventing long-term cardiac sequel and ensuring treatment continuity.\

CONCLUSION

Ewing's sarcoma is a extremely rare in the scapula and should be considered a differential diagnosis for any patients with inflammation of scapular region. Early detection of the disease is vital to reduce the risk of metastasis. Comprehensive multidisciplinary care is required throughout the course to ensure early identification and management of chemotherapy related toxicity.

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