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# **Brief Report**

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# Surgical resection is an effective adjunct to the management of an advanced metastatic osteosarcoma

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#### Abstract

Cardiac involvement of metastatic osteosarcoma is exceedingly rare and carries a dismal prognosis. Documentation of the surgical management of cardiac metastatic osteosarcoma is limited. We describe the successful resection of a  $\sim$  3.9 x 2.2 cm mass involving the right atrioventricular groove and right ventricular inferolateral wall, granting our patient 12 months of excellent cardiac function in the face of advanced metastatic disease.

#### Introduction

## **Case report**

## **Background**

Osteosarcoma, the 3<sup>rd</sup> most prevalent paediatric malignancy in the United States, has an overall 5-year survival rate of 68%. However, the mean 5-year survival is < 30% for those with metastatic disease. In this population, cardiac involvement of osteosarcoma is rare with < 2% of osteosarcoma lesions involving the heart since the 1980s. This statistic has been attributed to the advent of adjunctive chemotherapy and is supported by increased echocardiographic surveillance for anthracycline-induced cardiotoxicity.

To date, there is little evidence supporting the management of cardiac metastatic osteosarcoma. The current consensus for any cardiac lesion stipulates a comprehensive combined approach with 1) neoadjuvant chemotherapy, 2) resection of the primary tumour, 3) removal of all identified metastatic lesions, and 4) adjuvant chemotherapy.<sup>3</sup> In cases of rare and variable cardiac lesions, exhaustive excision of cardiac metastasis has improved survival.<sup>2,4</sup> In an extreme case, radicle resection of a right heart lesion and conversion to Fontan-like circulation was also documented.<sup>5</sup> We report a case of metastatic osteosarcoma with extensive involvement of the right myocardium, which was successfully resected with full reconstruction and good functional result.

## Clinical vignette

A 9-year-old male was diagnosed with expansile sclerotic osteosarcoma (stage III) of the right femoral diaphysis, with multiple metastatic lung lesions and a right ventricular mass. The patient underwent neoadjuvant chemotherapy, with reduction in primary tumour size and resolution of associated pain. He was anticoagulated with enoxaparin for a suspected thrombus in his right atrium and then underwent a resection of the involved femur with allograft reconstruction. Following discussion at a paediatric tumour board, it was recommended to prioritise surgery for the intra-cardiac mass, followed subsequently by a postero-lateral thoracotomy to resect lung nodules.

Echocardiography at presentation to our centre revealed normal cardiac segmental anatomy with a large, heterogeneous, multi-lobular mass along the right ventricular inflow portion. Its estimated dimensions were ~ 3.9 X 2.2 cm, and it appeared to invade into the inferior right atrioventricular groove and right ventricular inferolateral wall. It also contained a distinct peduncle attached to the posterior tricuspid leaflet and protruding into the right atrium (Figure 1A). No other masses were detected, and all other cardiac valves appeared normal.

Following sternotomy, cardiopulmonary bypass, and cardioplegic arrest, the tumour was approached and visualised through a right atriotomy (Figure 2A). The posterior leaflet of the tricuspid valve attached to the tumour was excised. The tumour was then followed through the ventricular wall, excising part of the free wall of the right ventricle and preserving all major coronary branches (Figure 2B). After complete resection (Figure 2C), an oval patch of bovine

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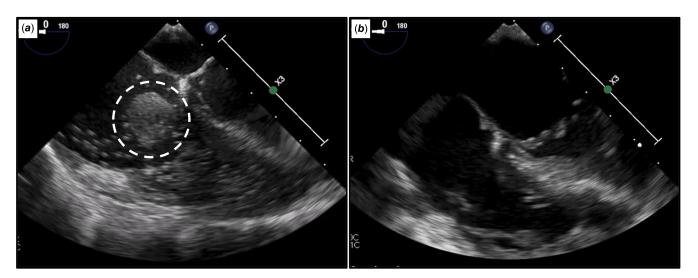
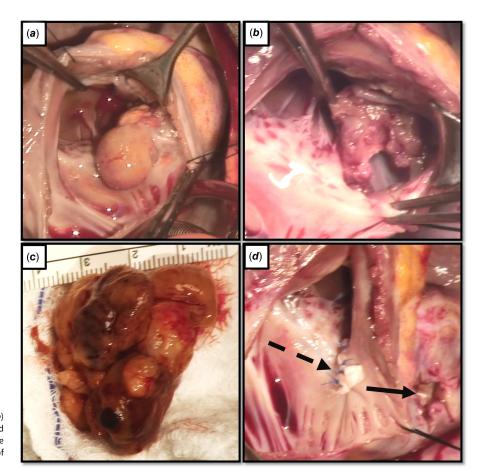


Figure 1. Transoesophageal ECHO mid-diastole (a) pre-operative ECHO depicting mass attached to the tricuspid valve. (b) post-operative ECHO revealing normal functioning tricuspid valve with no residual tumour.



**Figure 2.** (a) Right atriotomy visualising the tumour. (b) Excised portion of free right ventricle wall. (c) Resected tumour measuring 2 x 3 cm. (d) External view of bovine pericardial patch (black arrow) and cone repair of tricuspid valve (dashed arrow).

pericardium was sutured in place to close the right ventricular defect (Figure 2D, *black arrow*). The tricuspid valve was repaired by initially approximating the annulus, after which the anterior and septal leaflets were sutured together in a fashion similar to an Ebstein cone repair (Figure 2D, *dashed arrow*). Bypass was weaned with excellent haemodynamics. Postoperative echocardiography revealed a competent tricuspid valve with no residual tumour

(Figure 1B). Postoperative course was unremarkable, and the patient was discharged home on postoperative day 4.

Our patient remained in excellent cardiac health without cardiac recurrence or altered function for the next year. Unfortunately, 12 months following our care, a large metastatic spinal cord lesion was found. He underwent emergent palliative radiation therapy, was placed into hospice, and expired 2 months later.

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#### **Discussion**

Although exceedingly rare, the prognosis for metastatic paediatric osteosarcoma to the heart is dismal. Hartemayer and colleagues² identified 21 cases of direct cardiac involvement for paediatric osteosarcoma from 1893 to 2016. Of this cohort, only two patients remained cancer free 12 months following cardiac diagnosis. These two patients had undergone complete resection with multiagent chemotherapy, a strategy advocated since 1986. Due to the advanced metastatic disease in our patient, palliative resection achieved excellent cardiac function but was not curative. Our success was defined as an additional 14 months without cardiac tumour recurrence or symptoms.

As demonstrated in this case, successful surgical excision of right ventricular tumours extending into the free wall and posterior tricuspid leaflet can be accomplished with favourable haemodynamic outcomes. As metastatic osteosarcoma to the heart carries a poor prognosis,<sup>2</sup> our management was not curative. Even so, our strategy granted this patient excellent cardiac health and an additional year of life with family and friends. We advocate for the removal of potentially resectable tumours as a strategy to prolong life and provide patients invaluable time in the face of advanced metastatic disease.

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#### Competing interests. None.

Ethical standards. This report does not involve human and/or animal experimentation.

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