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(CASE REPORT)

Giant subcutaneous infantile hemangioma: A case report

Dennis Donath Mosha [1,*] and Pius David Muzzazzi [2]

[1] *Department of Radiology, Tumbi Regional Referral Hospital, Kibaha, Pwani, Tanzania.*
[2] *Department of Paediatrics, Tumbi Regional Referral Hospital, Kibaha, Pwani, Tanzania.*

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Abstract

Infantile hemangiomas are the most common benign vascular tumors of infancy, affecting approximately 5–10% of infants, and occur more frequently in females. Although most infantile hemangiomas undergo spontaneous involution without requiring treatment, some may result in ulceration, bleeding, functional impairment, cosmetic deformity, and life-threatening complications depending on their size, depth, and anatomical location. Giant infantile hemangiomas are particularly uncommon and present significant diagnostic and therapeutic challenges, especially in resource-limited settings.

We report a rare case of a giant subcutaneous infantile hemangioma in a male infant who presented with a progressively enlarging truncal mass. Imaging with ultrasound and magnetic resonance imaging (MRI) played a crucial role in defining the extent of the lesion and narrowing the differential diagnosis. Despite treatment with propranolol followed by vincristine because of poor response, the patient developed severe pneumonia with respiratory failure and subsequently died. This case highlights the importance of early recognition, appropriate imaging, multidisciplinary management, and close follow-up of infants presenting with extensive infantile hemangiomas.

Keywords: Infantile hemangioma; Giant hemangioma; Propranolol; Magnetic resonance imaging

1. Introduction

Infantile hemangiomas are the most common benign vascular tumors of infancy, affecting approximately 5–10% of infants. They were distinguished as a distinct clinical entity from other vascular anomalies by Mulliken and Glowacki in 1982 based on their unique biological behavior and natural history (1). Unlike congenital vascular lesions, which are fully developed at birth, infantile hemangiomas usually become clinically apparent during the first few weeks of life, undergo a rapid proliferative phase during infancy, and subsequently regress through a gradual involution phase (2,3).

Although most infantile hemangiomas are uncomplicated and resolve spontaneously, some may cause considerable morbidity depending on their size, depth, and anatomical location. Potential complications include ulceration, bleeding, permanent skin changes, functional impairment, and, in severe cases, airway obstruction or high-output cardiac failure (2,7). Early recognition is therefore essential to identify high-risk lesions requiring prompt intervention.

Imaging plays an important role in the evaluation of large, atypical, or deep infantile hemangiomas. Ultrasound provides valuable information regarding lesion vascularity and internal characteristics, whereas Magnetic Resonance Imaging (MRI) accurately delineates lesion extent and its relationship to adjacent structures, thereby facilitating diagnosis and treatment planning (2,10).

*Corresponding author: Dennis Donath Mosha

Oral propranolol is currently regarded as the first-line treatment for problematic infantile hemangiomas because of its proven efficacy and favorable safety profile. However, patients with extensive, refractory, or life-threatening lesions may require additional therapies, including systemic corticosteroids, vincristine, or other multidisciplinary interventions (2,7).

Herein, we report a rare case of a giant subcutaneous infantile hemangioma in a male infant, highlighting the diagnostic value of multimodality imaging and the therapeutic challenges encountered during management.

2. Case Report

We report a 40-day-old male infant who was referred from a peripheral hospital to Muhimbili National Hospital for further evaluation and management of a progressively enlarging truncal mass first noticed at 7 days of life. The swelling involved the right supramammary region and extended to the right iliac fossa.

He was born at term by Caesarean section because of a previous maternal uterine scar. He cried immediately after birth, with Apgar scores of 9 and 10 at one and five minutes, respectively, and had a birth weight of 3.8 kg. The pregnancy was uneventful and there was no family history of similar lesions or congenital abnormalities.

On admission, the infant was alert, afebrile, and not pale, jaundiced, cyanosed, or in respiratory distress. Local examination revealed a large soft tissue mass extending from the right supramammary region along the lateral chest wall to the right iliac fossa.The overlying skin demonstrated a port-wine stain-like discoloration. The lesion measured approximately 20 × 24 cm**,** had irregular margins, and was fluctuant with focal firm areas on palpation (Figure 1). The remainder of the systemic examination was unremarkable.

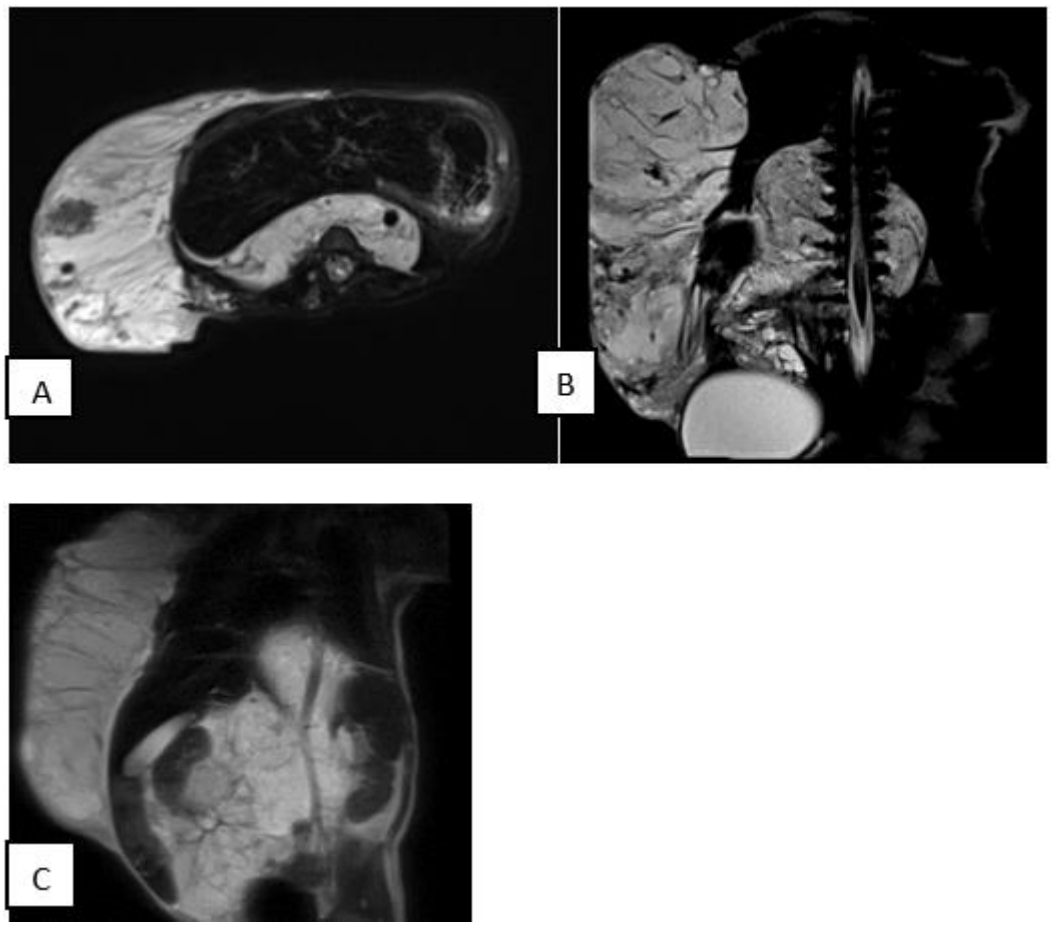

Figure 1 A–C. Axial and coronal T2-weighted MRI images of the abdomen at the level of the liver demonstrate a well-defined, encapsulated giant mass that is predominantly hyperintense with internal signal voids. The mass is confined to the subcutaneous tissues, extending from the anterior, lateral, and posterior chest wall to the pelvic region

Initial laboratory investigations were largely unremarkable except for thrombocytopenia (77×10^9/L). C-reactive protein, renal function tests, thyroid function tests, and coagulation studies were within normal limits. Peripheral blood smear demonstrated features suggestive of hemolysis and an infectious process.

Abdominal ultrasound demonstrated a large multiloculated cystic lesion displacing the liver, with lymphangioma considered as the initial differential diagnosis. Soft tissue ultrasound subsequently demonstrated an extensive multiloculated cystic lesion extending from the right clavicular region along the lateral chest wall to the right iliac fossa, with lymphangioma remaining the leading differential diagnosis. Magnetic resonance imaging (MRI) was therefore performed for further characterization.

MRI demonstrated a well-circumscribed, encapsulated subcutaneous soft tissue mass extending from the right lateral chest wall to the abdomen and pelvis **(Figures 1 and 2)**. The lesion was confined to the subcutaneous tissues without invasion of the underlying muscles or bones. It contained multiple septations and signal voids, demonstrating low signal intensity on T1-weighted images, high signal intensity on T2-weighted images, and homogeneous enhancement following contrast administration. These findings were suggestive of a vascular lesion, with infantile hemangioma and a peripheral nerve sheath tumor considered the principal differential diagnoses.

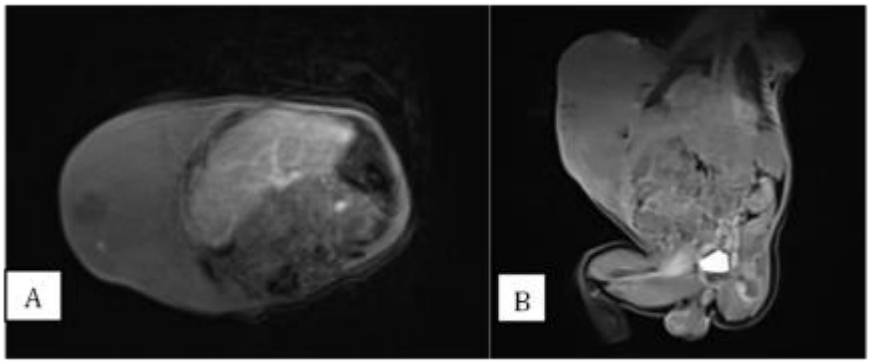

Figure 2A–B. Contrast-enhanced T1-weighted MRI images in the axial and coronal planes demonstrate homogeneous enhancement of a subcutaneous mass involving the right chest wall and extending to the pelvic region. The imaging findings are suggestive of an infantile hemangioma.

Following multidisciplinary review by the pediatric hemato-oncology team, radiologists, and tumour board, the lesion was considered most consistent with a giant subcutaneous infantile hemangioma. Because of the extensive vascular lesion, associated thrombocytopenia, and laboratory evidence of hemolysis, Kasabach-Merritt phenomenon was also considered, although lymphatic malformation and peripheral nerve sheath tumor remained alternative differential diagnoses.

The patient was commenced on oral propranolol at 2 mg daily for one week, which was increased to 4 mg daily for two weeks and subsequently 7 mg daily as the maintenance dose. Despite this treatment, there was minimal reduction in lesion size. Owing to the extensive nature of the lesion and inadequate response to propranolol, the patient subsequently received 12 cycles of vincristine (1.0 mg/m^2 every two weeks) with close inpatient monitoring for treatment-related adverse effects.

Four days after completing the twelfth cycle of vincristine, he was readmitted with a four-day history of fever and a one-day history of difficulty in breathing associated with poor breastfeeding. On examination, he was febrile and in respiratory distress. Oxygen saturation was 88% on room air, improving to 99% on oxygen supplementation, temperature was 38.7°C, pulse rate 130 beats/min, blood pressure 65/37 mmHg, and random blood glucose 5.7 mmol/L. Respiratory examination demonstrated lower chest wall indrawing, nasal flaring, inspiratory wheezing, and bilateral vesicular breath sounds. Abdominal examination revealed a distended but non-tender abdomen with normal bowel sounds.

A provisional diagnosis of severe pneumonia with sepsis was made. Malaria rapid diagnostic testing was negative, and treatment with intravenous amoxicillin-clavulanic acid, paracetamol, and supplemental oxygen was initiated.

Chest radiography demonstrated a wedge-shaped homogeneous opacity in the right lower lung zone, consistent with right lower lobe collapse (Figure 3). Repeat abdominal ultrasound demonstrated a highly vascularized predominantly hyperechoic subcutaneous mass with cystic components measuring approximately 20 × 24 × 13 cm, extending from the right chest wall to the pelvic region. Free intraperitoneal fluid was present, with infantile hemangioma and hemoperitoneum considered as differential diagnoses.

Antibiotic therapy was escalated to intravenous piperacillin-tazobactam, while oral propranolol was continued initially together with prednisolone. The patient subsequently developed severe anemia with features of circulatory compromise, requiring packed red blood cell transfusion (170 mL) and insertion of a nasogastric tube for nutritional support.

His clinical condition continued to deteriorate with worsening respiratory distress and respiratory acidosis, necessitating transfer to the pediatric intensive care unit, where he was intubated and mechanically ventilated. Propranolol was discontinued because of the patient's deteriorating clinical status. A repeat chest radiograph demonstrated persistent right lower lobe collapse with new left lower zone opacities consistent with pneumonia (Figure 3).

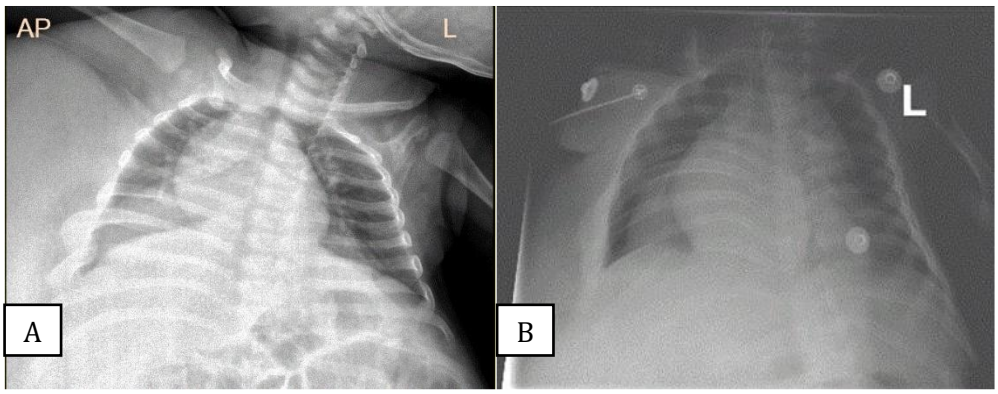

Figure 3A - B AP chest radiographs obtained one week apart. The first image demonstrates a wedge-shaped opacity predominating in the right lower zone, suggestive of right lower lobe collapse. A follow-up portable chest X-ray shows similar findings with additional hazy opacities in the left lower zone. These appearances are consistent with pneumonia. A soft-tissue mass is visible along the right lateral chest wall

Despite aggressive supportive management, the infant developed progressive circulatory failure with bradycardia and cold peripheries. Cardiopulmonary resuscitation was unsuccessful, and he died four days after admission to the intensive care unit. The cause of death was certified as respiratory failure secondary to severe pneumonia**.**

3. Discussion

Infantile hemangiomas are the most common benign vascular tumors of infancy, affecting approximately 5–10% of infants, whereas giant infantile hemangiomas are uncommon (1,13). Most lesions undergo spontaneous involution and do not require treatment (1,2,7,15). They typically become apparent within the first 2–4 weeks after birth, undergo rapid proliferation during early infancy, and gradually involute over several years (3). Large lesions are more likely to develop ulceration, bleeding, functional impairment, cosmetic deformity, and other life-threatening complications depending on their size, depth, and anatomical location (1,2,15).

In this case, the patient was a male infant with a giant subcutaneous infantile hemangioma involving the right chest wall, abdominal wall, and pelvic region. Infantile hemangiomas are more frequently observed among white infants and occur approximately three times more often in females than in males (1,7). They are classified according to their depth as superficial, deep, or mixed lesions and may also be categorized based on their anatomical distribution and extent of involvement (2,6,13). The extensive subcutaneous involvement observed in our patient represented an uncommon presentation and contributed to significant diagnostic and therapeutic challenges.

Most infantile hemangiomas do not require intervention because they undergo spontaneous involution. However, treatment is indicated for clinically significant lesions associated with functional impairment, ulceration, risk of

permanent disfigurement, or life-threatening complications (2,15). Oral propranolol remains the recommended first-line treatment for problematic infantile hemangiomas because of its proven efficacy and favorable safety profile, as demonstrated in randomized clinical trials (2,4,5,14,15). Topical timolol may be effective for selected superficial lesions, whereas systemic corticosteroids may be considered in patients who do not respond to or cannot tolerate propranolol (7,15). In severe or refractory cases, additional therapies such as vincristine may be considered as part of multidisciplinary management (7,8,15). In our patient, propranolol failed to achieve significant regression of the lesion, necessitating treatment escalation with vincristine because of the lesion's extensive size and poor response to first-line therapy. Surgical excision was not considered appropriate because of the extensive vascular nature of the lesion and the substantial risk of intraoperative bleeding.

The lesion became clinically apparent at 7 days of life, which is consistent with the typical presentation of infantile hemangioma (3). There were no reported abnormalities during antenatal ultrasonography. This emphasizes the importance of careful postnatal examination and early recognition of infantile hemangiomas, particularly large or atypical lesions, to facilitate timely referral, appropriate investigation, and early initiation of treatment (2,15).

Imaging played a pivotal role in establishing the diagnosis in this patient. Ultrasound is useful for assessing lesion vascularity and internal characteristics, whereas magnetic resonance imaging (MRI) provides superior delineation of lesion extent, tissue planes, and involvement of adjacent structures (10,15). In our patient, the initial ultrasound suggested an intra-abdominal multiloculated cystic lesion with a differential diagnosis of lymphangioma. However, MRI demonstrated that the lesion was confined to the subcutaneous tissues, extending from the right chest wall to the pelvic region without invasion of the underlying muscles or bones. These findings were instrumental in narrowing the differential diagnosis and guiding subsequent management. This case highlights the complementary roles of ultrasound and MRI in evaluating large and complex infantile hemangiomas.

The extensive nature of the lesion, together with thrombocytopenia and laboratory evidence of hemolysis, raised concern for Kasabach-Merritt phenomenon (KMP). KMP is a rare but life-threatening consumptive coagulopathy characterized by profound thrombocytopenia, microangiopathic hemolytic anemia, and consumptive coagulopathy. It is classically associated with kaposiform hemangioendothelioma and tufted angioma rather than typical infantile hemangiomas (11,12). Although our patient demonstrated thrombocytopenia and hemolytic features, the clinical presentation, radiological findings, and multidisciplinary review favored the diagnosis of a giant subcutaneous infantile hemangioma. Nevertheless, close hematological monitoring was warranted because of the potential risk of bleeding, progressive coagulopathy, and clinical deterioration.

This case highlights the diagnostic and therapeutic challenges associated with giant infantile hemangiomas, particularly in resource-limited settings. Early recognition, appropriate imaging, multidisciplinary collaboration, and close clinical monitoring are essential for optimizing patient outcomes. Furthermore, this case underscores that although propranolol remains the cornerstone of treatment, some patients with extensive or refractory lesions may require escalation to alternative therapies and intensive supportive care

4. Conclusion

Giant infantile hemangioma is a rare vascular tumor that presents significant diagnostic and therapeutic challenges because of its size, extent, and potential complications. Accurate diagnosis and assessment of lesion characteristics are essential for appropriate management. Imaging modalities such as magnetic resonance imaging (MRI) provide detailed visualization of lesion extent and anatomical relationships, while ultrasound is valuable for evaluating vascularity and monitoring changes over time. Biopsy is generally avoided in typical cases because the diagnosis can usually be established clinically and radiologically, while biopsy may carry additional risks, including bleeding.

Compliance with ethical standards

Acknowledgement

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Disclosure of conflict of interest

The authors declare that they have no competing interests. No funding was received for this work.

Statement of ethical approval

Ethical clearance and approval were obtained from the Institutional Review Board of Muhimbili National Hospital

Statement of informed consent

Written informed consent for publication was obtained from the patient's parents. Patient confidentiality has been maintained throughout the preparation of this report. Administrative permission to publish the case was granted by Muhimbili National Hospital.

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