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Gastric teratoma — a rare benign tumour of neonates

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Summary We describe our experience of two neonates with gastric teratoma, one a low-birthweight, premature baby who had a massive, immature teratoma. Complete excision is the appropriate treatment.

Introduction

Teratomas are embryonal neoplasms containing tissues from all three germ layers and are derived from totipotential cells. They are the most common tumours of infancy, accounting for 38–46% of neonatal tumours.¹ Although teratomas have been identified in most body regions, the sacrococcygeal are the most frequent followed by cervicofacial, mediastinal, gonadal and retroperitoneal. The stomach is rarely involved, accounting for <1% of teratomas.^{1,2} Since the first report by Eusterman & Sentry in 1922, approximately 100 cases have been reported in the English literature.^{2,3}

This report presents our experience of two infants with this rare tumour. The importance of immature tumour elements in the management of such cases is discussed.

Case 1

A 25-year-old multigravida delivered a 1900 g baby boy at 32 weeks of gestation. Hypertension and polyhydramnios complicated the

pregnancy. The baby was referred on day 25 for evaluation of progressive abdominal distension and feeding difficulty noticed by the parents since the 1st week of life. Examination revealed a solid mass filling almost the whole abdomen. US and CT scans revealed a complex mass with coarse calcifications extending from the subphrenic to the pelvic region and displacing the other abdominal viscera. The alpha-fetoprotein (AFP) level was 45,974 ng/L (normal for age).⁴ On exploration, a mass arising from the posterior wall of the stomach was excised with a cuff of normal gastric wall. Pathological examination revealed a 343-g tumour with solid and cystic components, measuring 11 × 8 × 7 cm. Histology showed tissues from all three germ layers. There was a predominance of neuro-epithelial tissue together with immature connective tissue (grade III immature teratoma) (Fig. 1). No additional treatment was offered. At 5 years of age, physical and mental development is normal. Regular follow-up has not shown evidence of recurrence and AFP levels have remained within the normal range for age.

Case 2

A 2-day-old full-term infant weighing 3.2 kg presented for evaluation of a mobile epigastric mass noticed at birth. Results of

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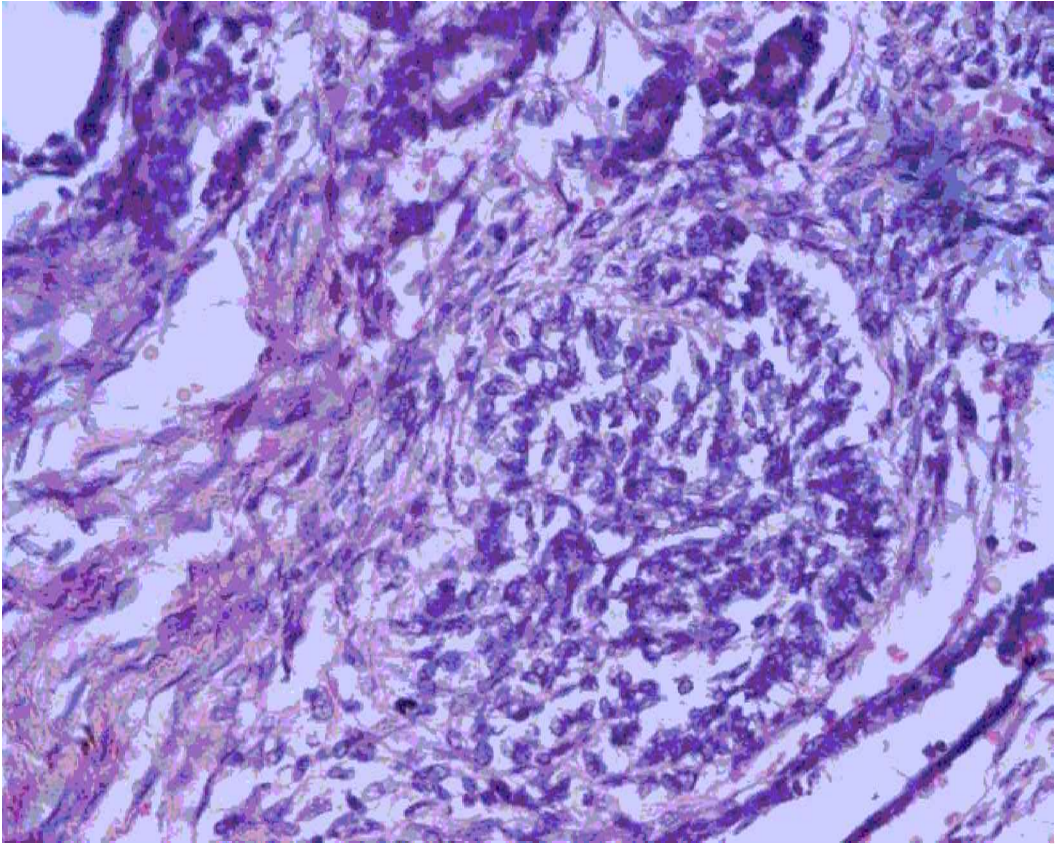


FIG. 1. Photomicrograph showing neuro-epithelial tissue with immature connective tissue ($\times 20$).

examination were otherwise unremarkable. Scan of the abdomen raised the possibility of gastric duplication. A CT scan showed a complex solid and cystic tumour with an intragastric component (Fig. 2). On exploration, an $8 \times 7 \times 6$ cm tumour arising from the greater curvature of the stomach was completely excised with a cuff of normal gastric wall. Histology revealed a mature teratoma comprising tissue from the three germinal layers. The postoperative course was uneventful and at 2.5 years of age he enjoys normal physical and mental health with no evidence of recurrence.

Discussion

Gastric teratomas (GT) are rare, accounting for $<1\%$ of all childhood teratomas.^{1,2}

They occur mostly in males (<3 months), in contrast with teratomas of other sites that predominate in females.² Fewer than 10% have been reported in the older age group (>10 years).^{2,3} One of our patients was pre-term and of low birthweight which, to our knowledge, has been reported only rarely.

Common presentations include abdominal mass and distension. Intra-gastric extension and erosion can lead to gastro-intestinal bleeding, perforation and intestinal obstruction.⁵⁻⁷ Abdomino-thoracic extension presenting with respiratory distress is rare. A large tumour can produce premature labour and dystocia. Investigations including plain radiograph of the abdomen show coarse calcification in over 50% of cases. Differentiation from neuroblastoma and meconium peritonitis is needed.⁶ An ultrasound and CT scan of the

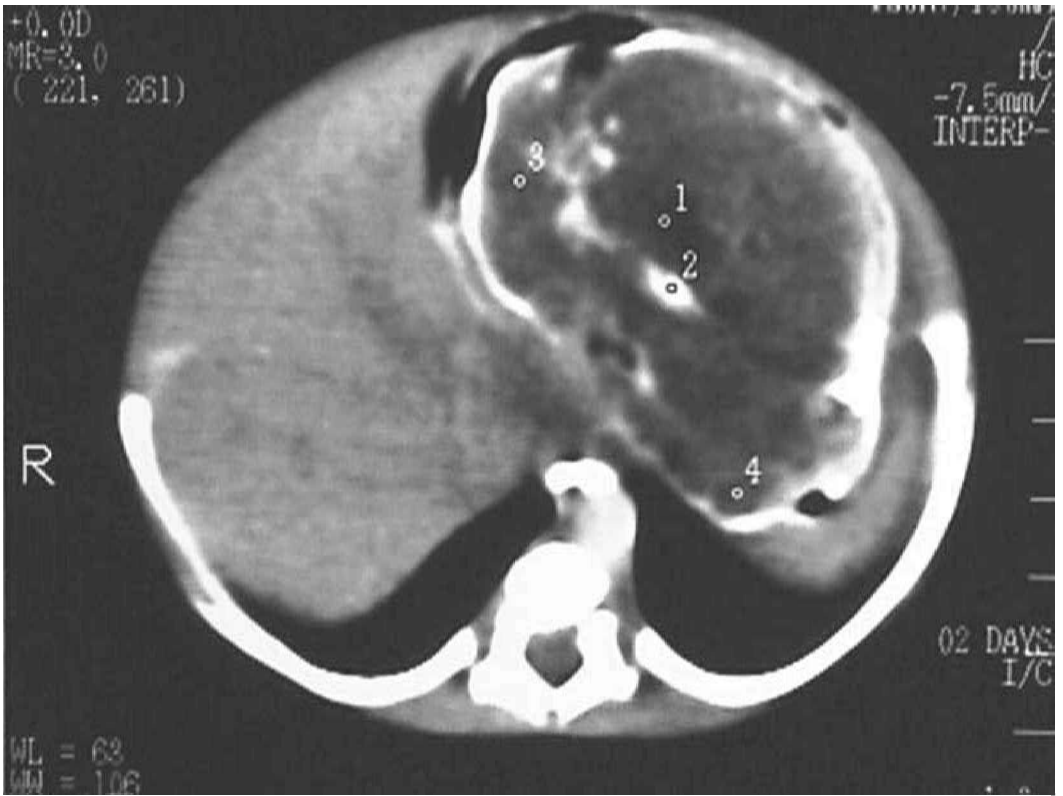


FIG. 2. CT scan of the abdomen showing a complex mass with coarse calcifications extending into the stomach lumen.

abdomen can identify the characteristic solid and cystic areas within the tumour, arising from the stomach, and extension (if any) into surrounding viscera. A CT scan is specific in detecting fat in the periphery of the tumour or within septations dividing the cystic and solid areas.^{1,3} Serum AFP is reported to be elevated in both mature and immature teratomas and is not a good predictor of malignancy. However, it has a definitive role in follow-up and may serve as a marker for early detection of recurrence.^{3,4,8}

The majority of lesions are mature teratomas, though immature and malignant elements have been reported.^{3,7-9} Immature teratomas contain various amounts of immature tissue, e.g. neuro-epithelium, in addition to tissues from all three germ layers. This causes concern among physicians involved in planning treatment as immature

teratomas may be mistakenly labelled malignant because the borders between disordered embryogenesis and oncogenesis are ill-defined at this age. Immature teratomas behave in a malignant fashion only if foci of malignant germ cell elements, such as yolk cell tumour, neuroblastoma or medullo-epithelioma, are present. As these clusters of malignant tissues are very small and frequently do not stain with AFP, they are easily overlooked on routine histology. The relationship between histology and recurrence of immature teratomas is not clear for all sites of teratomas.^{1,3,8} Because of their special physiology and developmental status, malignant tumours in neonates are pathologically, clinically and therapeutically distinct.¹ Behaviour cannot be directly inferred from the histological appearance, and risk stratification is difficult and complex. Chemoradiation, despite appropriate

dose reduction, has significant morbidity and mortality and its use is therefore severely restricted, unlike in adult practice.

Complete excision of gastric teratoma with a cuff of normal gastric wall is an adequate surgical treatment and so far no case of recurrence has been reported.¹⁻³ Extensive procedures such as gastrectomy (partial or total) carry high morbidity and should be avoided. The value of conservative surgery is well demonstrated by our patient with immature teratoma. He has remained tumour-free during 5 years of follow-up, without adjuvant therapy.

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