CASE REPORT

Pulmonary Metastasis in Infantile Choriocarcinoma: Successful Outcome

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Background: Infantile choriocarcinoma is usually fatal without appropriate treatment. **Case characteristics:** A 3-month-old boy who presented with respiratory distress, hepatomegaly, amemia and bilateral nodular lesions on chest *X*-ray. **Observation:** Fine-needle liver aspiration revealed necrotic tumour cells. The serum β-hCG level was very high (2057 mIU/L), supporting a diagnosis of infantile choriocarcinoma of the liver. Surgical resection after cisplatin-based multiagent chemotherapy afforded successful remission. **Message:** Early treatment of infantile choriocarcinoma can yield a successful outcome.

Keywords: *Computed tomography, Diagnosis, Metastasis.*

Infantile choriocarcinoma is an extremely rare disease characterized by severe anemia, failure to thrive, hepatomegaly and seizures. Less commonly hemoptysis, respiratory distress, and/or signs of precocious puberty may be evident. A marked elevation in the β-hCG level is typical (often to $>10^6$ IU/L) [1]. If not treated appropriately, death usually occurs within three weeks of initial presentation [2]. Recently, multiagent chemotherapy combined with surgical resection has yielded successful outcomes [3]. We report a case of respiratory distress reflecting pulmonary involvement of infantile choriocarcinoma.

CASE REPORT

A 3-month-old boy was admitted to emergency department of our hospital with a history of fever, fatigue (2 days in duration), and cough (2 months in duration). He exhibited significant respiratory distress, but no abnormal sounds were evident bilaterally on auscultation. His liver was enlarged 6 cm below the costal margin. Laboratory tests revealed a hemoglobin level of 7.5 g/dL, a platelet count of 503.000/μL, a white blood cell count of 12.5×10^3/μL, and a C-reactive protein level of 120 mg/L (normal, 0-5 mg/L). Aspartate aminotransferase level was 103 IU/L, and chest *X*-ray revealed bilateral, disseminated nodular lesions (***Fig.* 1**). The patient was admitted to the pediatric intensive care unit and antibiotics (ceftriaxone and clarithromycin) were started to treat the pneumonia.

Thoracic computed tomography (CT) revealed multiple small, bilateral pulmonary nodules (***Fig.* 2*a*, 2*b***) and a mass of 49×63 mm in dimension, with indistinct cystic components, in the right lobe of the liver (***Fig.* 2c**). Magnetic resonance imaging (MRI) identified an irregular heterogeneous mass (50×60×60 mm) in the right lobe of the liver (***Fig.* 2d**). Fine-needle aspiration of the liver mass revealed necrotic tumor cells. The serum a-hCG and alpha fetoprotein (AFP) levels were 2057 mIU/mL (normal 0-25 mIU/mL) and 18 IU/mL, respectively, which suggested a diagnosis of infantile choriocarcinoma of the liver. The mother's placenta was normal.

Chemotherapy was initiated with bleomycin (0.5 mg/kg; 1 day), cisplatin (0.7 mg/kg; 5 days) and etoposide (3 mg/kg; 5 days); the BEP protocol. The patient tolerated six cycles of chemotherapy well. Repeat testing of the α-hCG level showed that it had decreased to <0.1 mIU/mL.

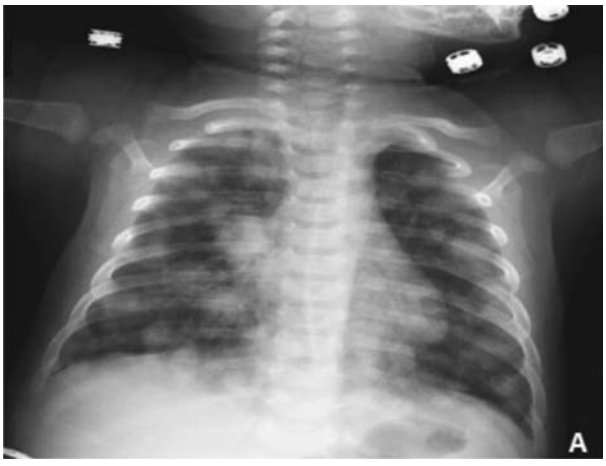

FIG. 1 *Chest X-ray: Bilateral pulmonary nodules evident at initial presentation.*

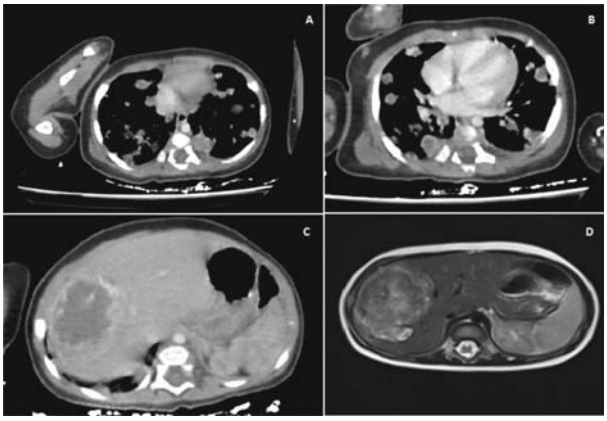

FIG. 2 *Bilateral pulmonary nodules on computed tomography (a and b); hepatic mass in right lobe of liver on computed tomography (c), and in magnetic resonance imaging (d).*

Radiological evaluation revealed no further evidence of metastatic lung disease. Repeat CT showed a decrease in liver size; complete tumor resection was achieved *via* right partial hepatectomy. Histopathological examination revealed a few syncytiotrophoblasts and necrotic cells. The child has been in complete remission for the past 3 years and remains under regular follow-up. His growth and development are normal.

DISCUSSION

Most cases of infantile choriocarcinoma are metastases of gestational choriocarcinoma of the placenta, but primary liver tumors have also been reported [4,5]. Choriocarcinoma can be aggressive and destructive. Although relatively few cases have been reported, they include cases cured with chemotherapy [3,6,7] .

The disease can rapidly become fatal if left unrecognized. Of 30 cases reviewed by Blohm and Gobel [8], two were diagnosed in stillborn children and 19 were diagnosed after death (on postmortem examination). Among children who succumbed, the median time from initial symptoms to death was 21 days. Such a rapid progression indicates that treatment must not be delayed (to allow for histological confirmation) in infants with a markedly elevated α-hCG level and a clinical presentation consistent with infantile choriocarcinoma. We commenced multiagent chemotherapy in the absence of clear histological evidence. It is recommended that all infants presenting with anaemia and a hepatic mass be screened for α-hCG levels [3].

Single-agent chemotherapy (methotrexate) usually cures maternal gestational trophoblastic disease but has never been curative in an infant with choriocarcinoma; multiagent chemotherapy is required [9,10]. In the Blohm and Gobel series of 30 children [8], five were successfully treated with a platinum compound (cisplatin or carboplatin) combined with etoposide. In addition, because of the rapid tumor growth, the platinum-plus-etoposide combination was often further combined with methotrexate given for 1 week either before or after the platinum-containing regimen. Surgical resection of the residual mass is usually performed at the end of platinum-based therapy. Early surgical intervention is not advisable; tumour fragility increases the risk of uncontrolled bleeding, thus delaying chemotherapy [1]. The serum β-hCG level is an ideal marker for monitoring therapeutic success [8]. Although the tumour is very aggressive, early cisplatin-based chemotherapy can yield a successful outcome but must commence early.

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