European Review for Medical and Pharmacological Sciences 2014; 18: 3831-3833

Experience with the treatment of testicular yolk sac tumor in children: a report of 14 cases

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Abstract. – OBJECTIVE: This paper discusses the optimal treatment for testicular yolk sac tumor at stage I in children.

PATIENTS AND METHODS: Fourteen children with testicular yolk sac tumor (including 10 cases of stage I and 4 cases of stage II) underwent high ligation of internal spermatic cord vein and orchiectomy. Among these, seven cases of stage I were below 1 year of age. Retroperitoneal lymph node dissection without postoperative systemic chemotherapy was implemented in 9 cases (5 cases of stage I and 4 cases of stage II), and only one was positive.

RESULTS: Among the 12 cases followed, 9 cases were alive (of these, 5 children < 1 year old, in stage I, underwent high ligation of internal spermatic cord vein and orchiectomy, with a survival time of 25 months to 10 years and 4 cases with radical retroperitoneal lymph node dissection). Three cases older than 1 year died of retroperitoneal lymph node and lung metastases.

CONCLUSIONS: For the high ligation of internal spermatic cord vein, orchiectomy is a kind of simple and effective treatment for children younger than 1 year with stage I, without recurrence and metastases. However, attention to the accuracy of staging and close observation are important aspects of the treatment.

Key Words:

Testicular yolk sac tumor, Mesonephroma, Orchiectomy, Chemotherapy.

Introduction

At present there is no consensus for the treatment of the children with testicular yolk sac tumor in stage I. Epidemiological studies suggested that the incidence is most among Caucasian children in comparison to Asian and African children[1]. There are different views regarding whether to perform radical retroperitoneal lymph node dissection and to administer postoperative chemotherapy[2,3]. In the present report we summarize our treatment case load with 14 cases of pediatric testicular yolk sac tumor, confirmed by pathology in our hospital during 15 years, combined with a brief literature survey of this disease.

Patients and Methods

All the studies were performed with Institutional Ethics Committee approval from our hospital, Central Hospital of Xuzhou. Among 14 cases of pediatric testicular yolk sac tumor, 10 patients were younger than 2 years age (7 were under age 1), and 4 patients above 4 years age, with an age range of 16 days to 12 years old. The time from the onset of disease to treatment varied from 16 days to 12 months, mostly within six months with an average of 3.8 months. Most of the children presented with painless scrotal mass on one side are unilateral disease, with nine cases of the right side and five cases of left side. Most of the hard and solid masses are smooth maintaining the original morphology of the testis with negative light test. Four cases were presented with inguinal palpable enlargement of the lymph nodes, in which 2 cases of the masses were not smooth with scrotum adhesions. As with the clinical staging, 10 cases were in stage I and 4 cases in stage II. Five cases underwent examination by centesis outside the hospital due to hydrocele of testis and hematoma of scrotum. One of these cases had injury in virilia before the onset and underwent 3 times of examination by centesis due to hematoma of testis and simultaneously presented with inguinal palpable and hard swollen lymph nodes. Eleven of 14 cases underwent preoperative alpha-fetoprotein (AFP) examination and seven cases were positive. There preoperative X-ray chest films were negative for all the cases. Lymph node metastasis was found in 2 cases conducted by retroperitoneal CT.

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All patients of this group underwent high ligation of internal spermatic cord vein and orchiectomy. Out of the 9 cases that received radical retroperitoneal lymph node dissection, five cases of stage I and four cases of stage II including one of phase II patient were confirmed as retroperitoneal lymph node metastasis by pathological examination. All the cases received one course of preoperative chemotherapy and were treated with 15 g/kg of dactinomycin daily (intravenous infusion at the first and the fifth day) and 1.5 mg/m^2 of leurocristine each day (intravenous infusion at the first and the fifth day). After surgery, most of the patients were not treated by the regular chemotherapy. Twelve out of 14 cases (86%) were followed up, of which were 9 cases of stage I. These included 8 cases of survival of which five cases were < 1 year age and underwent high ligation of internal spermatic cord vein and orchiectomy, with average survival of 5.6 years and three cases with radical retroperitoneal lymph node dissection. One death occurred. Among the three followed up cases in stage II, one survival case was still followed up at 18 months after operation, and the other two patients died from retroperitoneal lymph node and lung metastases, respectively, at 11 and 16 months after surgery.

Discussion

The treatment on pediatric testicular yolk sac tumor in clinical phase II and III has basically reached a consensus, but there are still different opinions on treatment for children in clinical Phase II. Xie et al[4] advocated that all the patients underwent radical peritoneal lymph node dissection regardless of clinical stage and age. This disease mainly results from the hematogenous metastasis and even retroperitoneal lymph node dissection can only get rid of 75% of the lymph node in this region, and the pathological examination of the cleared lymph node showed a very low positive rate. Griffin et al[5] summarized 95 cases of the disease, and only 11 cases were confirmed as retroperitoneal lymph node metastasis by pathology. Kaplan et al[6] summed up 47 cases and only seven cases were positive; moreover it is believed that retroperitoneal lymph node dissection on children in stage I did not improve the prognosis. Hu CG, et al[7] confirmed that 2 of 3 cases were more than 2 years old. Therefore, for children under 1 year age in clinical stage I, without recurrence and metastasis, retroperitoneal lymph node dissection was unnecessary either for therapeutic, prevention or staging purposes. Besides, the retroperitoneal lymph node dissection can cause major trauma and many short-term complications such as enteroparalysis, lymphatic fistula and atelectasis[7]. We followed 6 cases (under 1 year age) in this group, of which five cases underwent only high ligation of internal spermatic cord vein and orchiectomy, and were finally cured with an average survival time of 5.6 years (ranging 2 to 10 years).

Several reports showed that among the pediatric testicular yolk sac tumors diagnosed, 80% of the lesions were confined to the testis, and that the survival rate of patients was not influenced by chemotherapy. Flamant et al[8] compared 24 cases of children with Stage I, there was no significant difference in survival rates between the 12 cases treated by systemic chemotherapy and the 12 cases underwent high ligation of internal spermatic cord vein and orchiectomy. It is generally accepted that the prognosis of pediatric testicular yolk sac tumor is closely related to patient age, i.e., the tumor has slow growth and low degree of malignancy with decreasing age. This report showed that the survival rate of children under 1-year-age without recurrence was 80%, while this rate dropped to 25% for the 2-year-old patients. Leonard et al[9] reported that the survival rate of children under 1 year of age who underwent high ligation of internal spermatic cord vein and orchiectomy can reach up to 96%, but this dropped to 29% if the age of the patient was more than 2 years. Huang et al[10] also think that the prognosis of children under 1 year age is superior to the older children. All patients in this group were not treated by postoperative chemotherapy; among the nine survival cases followed up, six cases were under 1 year of age. In this study, 3 dead patients died and included two cases in stage II and another case older than 2-years age in stage I, who underwent postoperative AFP examination for three times with positive results. In view of the lack of improved survival rate by the post-operative chemotherapy and also because of significant side effects and complications of chemotherapy, the authors of this study[10] suggested that the children under 1 year age in stage I, without clinical signs of recurrence and metastasis or AFP decreasing to normal levels, don't need to be treated by systematic postoperative chemotherapy.

The vast majority of pediatric testicular tumors are malignant. When not fully diagnosed before surgery, high ligation of internal spermatic cord vein and orchiectomy can be implemented as the first measure of therapy, prohibiting preoperative tumor puncture and avoiding hematogenous or lymphatic metastasis. If clinical staging is not completely determined, the surgery may be performed first, and the decision to implement the retroperitoneal lymph node dissection and systemic chemotherapy can be made after postoperative pathological findings and AFP examination. The patients with pediatric testicular yolk sac tumors, especially the children in stage I implemented with high ligation of internal spermatic cord vein and orchiectomy, should be closely followed up after surgery. It is known that the disseminated metastasis of tumors mostly occurs during the first 14 months[7] and, therefore, should be followed up for at least 2 years. If the postoperative AFP does not drop and still higher than normal and/or significantly increased in a short term, which suggests the existence of residual tumor or recurrence, the retroperitoneal lymph node dissection and metastasectomy should be implemented in these patients followed by chemotherapy with the first selected regimen of cisplatin, vincristine and bleomycin.

Conflict of Interest

The Authors declare that there are no conflicts of interest.

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