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Original Article

Clinical effect evaluation of interventional embolization combined with pingyangmycin injection in the treatment of trunk and limbs Kaposi type hemangioendothelioma

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Abstract: Objective: To evaluate the clinical efficacy of interventional embolization combined with Pingyangmycin injection in children with Kasai-type hemangioendothelioma (KHE). Methods: A retrospective analysis was conducted on 60 children with KHE admitted between January 2015 and December 2024. All patients were treated with interventional embolization combined with Pingyangmycin injection. Demographic and clinical data of patients were collected. The platelet count, coagulation function, therapeutic effect, and health-related quality of life (HRQoL) scores were compared before and after treatment. Survival outcomes were analyzed using Kaplan-Meier method. Factors affecting HRQoL were analyzed using univariate and multivariate regression analysis. Results: The total effective rate was 80%. The 1-year and 3-year survival rates were 93.3% and 66.67%, respectively. After treatment, the platelet count was significantly increased, and the coagulation function was significantly improved ($P<0.05$). In addition, the HRQoL scores of each dimension were significantly improved after treatment ($P<0.01$). Univariate and multivariate analysis identified activity disorder, platelet count, and parents' education level as independent factors affecting HRQoL (all $P<0.05$). Conclusion: Interventional embolization combined with Pingyangmycin injection is an effective method for the treatment of KHE involving the trunk and limbs in children. It demonstrates good effects on correcting thrombocytopenia and improving the quality of life of affected children.

Keywords: Kaposiform hemangioendothelioma, trunk and limbs, interventional therapy, pingyangmycin, efficacy evaluation

Introduction

Kaposiform hemangioendothelioma (KHE) is an aggressive vascular endothelial tumor characterized by purple to dark purple skin or mucosal plaques and nodules [1, 2]. It predominantly affects infants and young children due to their immature immune system, often causing serious health complications [3, 4]. The potential onset of Kasabach-Merritt phenomenon (KMP) further challenges its management. KMP is a severe consumptive coagulopathy characterized by thrombocytopenia, hypofibrinogenemia, and coagulation factor depletion [5, 6]. This complication significantly increases mortality, severely impairs quality of life and poses a major clinical challenge in pediatric care.

Systemic drug therapy remains the standard for KHE complicated with KMP, despite limited evidence for many drugs. There has been a paradigm shift in first-line treatment: since the effect of glucocorticoids is not ideal, international guidelines now recommend sirolimus due to its high efficiency (>90%) [7]. However, some patients either fail to respond to or tolerate sirolimus. Other alternative methods, such as Pingyangmycin combined with interventional embolization, have shown satisfactory results, but evidence remains limited to small-scale

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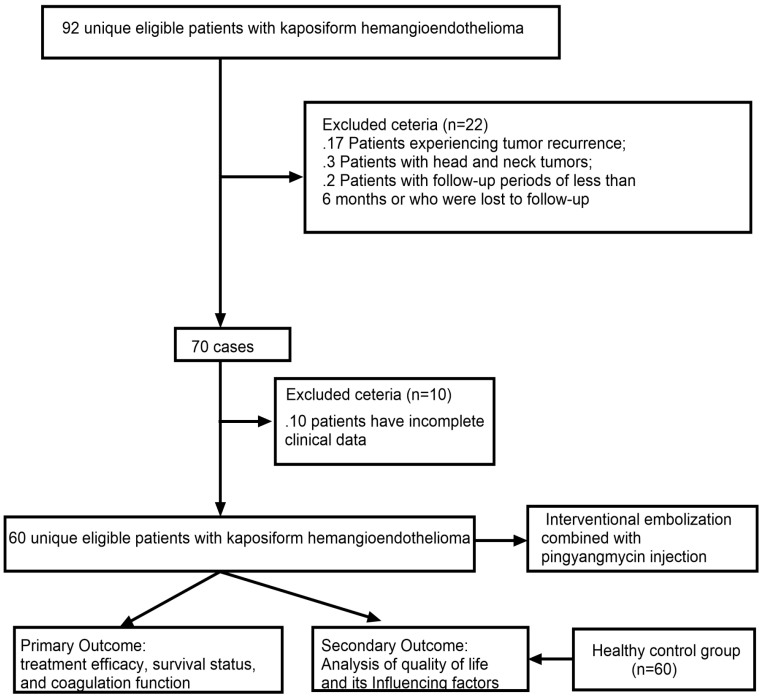

Figure 1. Research flowchart.

studies, limiting wider applicability. In addition, the incidence of pediatric KHE varies significantly in different regions and populations, indicating a complex interaction between genetic, environmental, and socio-economic factors [8, 9]. This regional difference highlights the necessity of localized research. Therefore, to supplement current evidence, this study retrospectively analyzed the clinical data of children with KHE in Guangzhou region.

General information and methods

Study population

This retrospective study analyzed data from 60 pediatric patients with KHE who underwent interventional embolization combined with pingyangmycin injection at our institution between January 2015 and December 2024.

Inclusion criteria: (1) diagnosis of KHE confirmed by characteristic clinical presentation, supportive magnetic resonance imaging (MRI) findings, and histopathological examination [10]; (2) availability of complete clinical data; (3) receipt of initial combination therapy with interventional embolization and pingyangmycin; (4) tumor located at the limbs or trunk, exhibiting ill-defined borders and a purplish-red appearance; (5) imaging evidence of a solid, locally invasive tumor; and (6) age between 2 months and 12 years. Exclusion criteria: (1) incomplete clinical data; (2) other concurrent tumors or hematologic diseases; (3) treatment-related disputes; (4) follow-up duration less than 6 months or loss to follow-up; (5) lesion located in the head or neck region; and (6) history of prior treatment or disease recurrence. The detailed screening process is illustrated in **Figure 1**.

The study protocol was approved by the Ethics Committee of Guangdong Provincial Maternal and Child Health Hospital (Approval number: 2025-10641). Since all data were collected from the hospital's electronic medical record system and only involved anonymized clinical and demographic data without patient intervention, the requirement for informed consent was waived.

Treatment protocol

All patients received a preoperative intravenous injection of methylprednisolone (Medrol®, Pfizer, Batch No. MP12345) (1.6 mg/kg) prior to emergency interventional embolization under general anesthesia. First, the femoral artery (left or right) was punctured as the access route; for upper-limb lesions, the brachial artery or radial artery was selected. Under ultrasound guidance, the femoral artery was punctured using a Seldinger technique, and a 4 F (1 F= 0.33 mm) arterial sheath was introduced. The catheter tip was carefully positioned without crossing the origin of the internal iliac artery. Diagnostic angiography was then performed using a 4 F Cobra catheter, usually displaying 2-3 main feeding arteries with multiple small branches. The parenchymal phase showed characteristic 'cloud-like' staining, and no arteriovenous fistula was observed. Subsequently, a 1.9 F microcatheter was superselectively advanced into the feeding artery, and superselective angiography was repeated to evaluate potential anastomosis with normal blood ves-

sels. Embolization was performed by slowly injecting pirarubicin iodized oil emulsion mixture (containing 2 mL iodized oil, 2 mg pingyangmycin (Bleomycin A5, Nippon Kayaku, Batch No. PY67890), 2 mg dexamethasone (Decadron®, Merck, Batch No. DXZ13579), and 2 mL iodol) or polyvinyl alcohol (PVA) particles (350-560 μm). The dose of pingyangmycin was 0.2-0.3 mg/kg each time, and the cumulative dose of each course was no more than 2.0 mg/kg. The drug was dissolved in 3-5 mL normal saline with 1-2 mg dexamethasone before injection. Embolization was repeated every 2 weeks until satisfactory results were achieved (defined as a 50% reduction in volume, stable lesions, or visible pale skin) or a maximum cumulative dose was achieved.

For residual lesions, 0.1 mL of 0.1 g/L diluted pingyangmycin solution was injected at an interval of 5 mm under ultrasound guidance. After operation, self-adhesive elastic bandage was used to stop bleeding at the puncture site, and the lower limbs were fixed for 8 hours. Intravenous methylprednisolone (1.6 mg/kg/day) was maintained, and the whole blood cell count was monitored every 24 hours. Once the platelet remained above 100×10^9/L, glucocorticoids were gradually decreased and discontinued. Weekly blood test was performed in the first month after operation. If the platelet counts remained stable within the normal range, the monitoring frequency was then reduced to once per month.

Data collection

Patient data were retrospectively collected from the Hospital Information System (HIS), including outpatient and inpatient records. The dataset covers patient demographics, disease characteristics, diagnostic results, treatment details, clinical outcomes, recurrence rates, and mortality. Health-related quality of life (HRQoL) was assessed 6 months after discharge.

To evaluate therapeutic efficacy, platelet counts and coagulation parameters (activated partial thromboplastin time [APTT], prothrombin time [PT], and thrombin time [TT]) were measured one week after surgery to monitor early coagulation and surgical safety. Clinical symptoms and lesion changes were monitored during the 6-month follow-up.

Therapeutic efficacy was evaluated according to following criteria: Effective: (1) clinical symptom relief or improvement; (2) the lesion ≥20% reduction in lesion volume or complete resolution; (3) improvement of complications (e.g. alleviation of functional impairment, chronic pain, or compression symptoms); (4) normalization of hematological and coagulation parameters (e.g. fibrinogen level and clotting time). Any of these findings were considered effective.

Laboratory index detection method: fasting venous blood was collected at the time of admission. Whole blood cell counts were performed using an automated hematology analyzer (Model: Sysmex XN-9000) to determine white blood cell (WBC) counts. Liver and kidney function and blood lipid levels were assessed by a biochemical analyzer (instrument model: Roche Cobas c702). Specific indicators include: alanine aminotransferase (ALT), aspartate aminotransferase (AST), assessment of liver function; serum creatinine (SCr) and blood urea nitrogen (BUN) were measured to evaluate renal function. All detection operations strictly followed the standard operating procedures provided by the reagent manufacturer.

Ineffective: (1) no significant change in lesion size or <20% volume reduction; (2) no significant improvement in clinical symptoms or complications; (3) no obvious normalization of hematological parameters; (4) death due to KHE or its related complications. Any of these findings were considered ineffective.

Assessment of health-related quality of life (HRQoL): HRQoL was assessed using the Pediatric Quality of Life Scale (PedsQL TM) version 4.0, a validated modular instrument for multidimensional HRQoL measurements in children and adolescents. The Chinese version of the scale has demonstrated good reliability and validity. The PedsQL TM 4.0 suite used in this study included the Family Information Form (FIF), the General Core Scale (GCS), and the Chinese version of the PedsQL TM 4.0 Infant Scale.

Follow-up

Patients were systematically followed up through medical records review and telephone interviews every three months, with an aim to determine the survival status of all enrolled children. The follow-up period ended on December 31,2024.

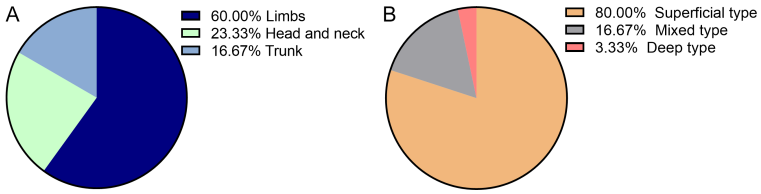

Figure 2. The location of the lesions and clinical classification in group.

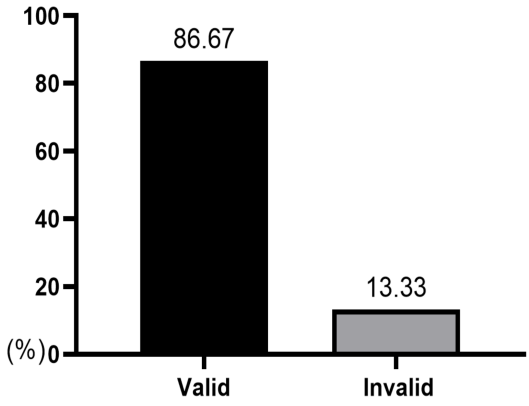

Figure 3. Treatment efficacy in patient.

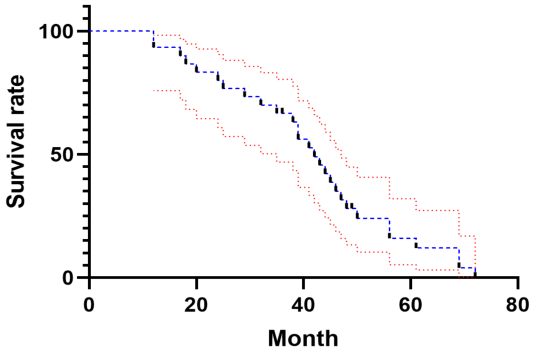

Figure 4. Post-treatment survival curve of the study cohort.

Statistics methods

Statistical analysis was performed using SPSS software (version 26.0). Continuous variables were expressed as mean ± standard deviation or median (interquartile range) according the data normality (assessed by Shapiro-Wilk test) and homogeneity of variance (assessed by Levene test). For between-group comparisons, data with a normal distribution was compared using independent-samples t-test, otherwise, using Mann-Whitney U test. Categorical variables were expressed as numbers (percentages) and compared using the chi-square test. A binary logistic regression model was established to explore the factors related to the improvement of quality of life. Variable selection was performed using the reverse stepwise method. The threshold for entry was $P<0.05$, and the threshold for removal was $P<0.10$. Cox regression analysis was used to analyze the factors affecting patient survival. A two-tailed $P<0.05$ was considered statistically significant.

Result

Baseline characteristics

The cohort comprised 32 males (53.33%) and 28 females (46.67%). The age at onset ranged from 3 days to 48 months, with a median of 9 months and a mean of 13.30±9.9 months. The mean lesion size was 3.68±2.18 cm. Critically, 25 patients (41.67%) presented with KMP at diagnosis. The lesion site and clinical classification are detailed in **Figure 2**.

Treatment outcomes

Amont the 60 patients, 52 (86.67%) achieved an effective response following interventional embolization combined with Pingyangmycin injection. During a mean follow-up period of 18.5±7.8 months, the 1-year and 3-year survival rates were 93.33% and 66.67%, respectively (**Figures 3**-**5**).

Comparison of liver and kidney function before and after treatment

No significant difference were observed in liver (ALT & AST) or kidney function (Scr & BUN) markers before and after the treatment, as detailed in **Table 1**.

Changes in coagulation function before and after treatment

The platelet counts and fibrinogen levels were significantly elevated after treatment, accompanied by significantly improved coagulation parameters (APTT, PT, and TT) (all $P<0.05$) (**Table 2** and **Figure 6**).

Comparison of PedsQL™4.0 scores before and after treatment in KHE patients

As shown in **Table 3**, after treatment, the scores of physical function, physical symptom func-

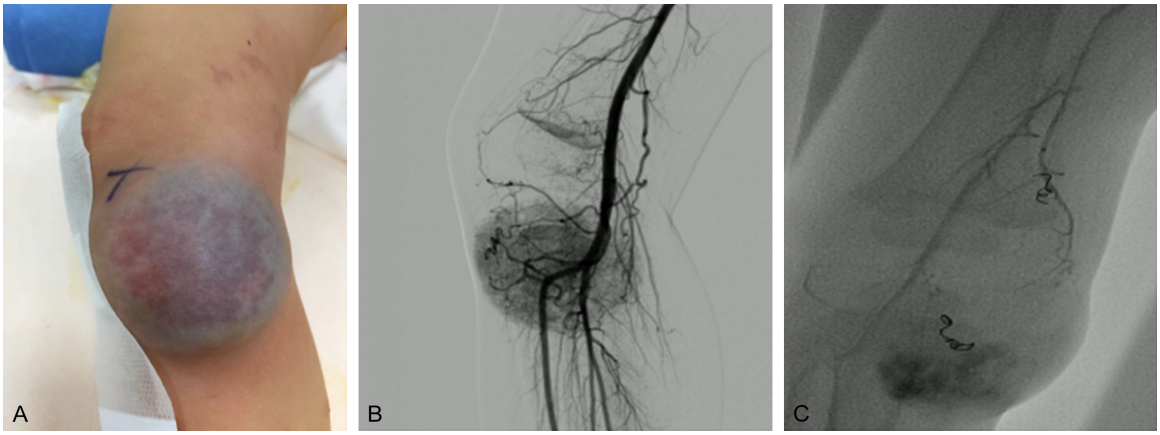

Figure 5. Clinical examples. A: Preoperative; B: Preoperative angiography image; C: Postoperative angiography image.

Table 1. Comparison of liver and kidney function before and after treatment in pediatric KHE patients

Time	AST (U/L)	ALT (U/L)	Scr (μmol/L)	BUN (mg/dl)
Before treatment	25.6±3.8	26.0±4.2	56.4±8.8	5.9±0.4
After treatment	25.2±3.5	26.3±4.3	56.2±8.7	5.8±0.5
t value	0.424	0.273	0.089	0.855
P value	0.673	0.786	0.930	0.396

AST: Aspartate Aminotransferase; ALT: Alanine Aminotransferase; Scr: Serum Creatinine; BUN: Blood Urea Nitrogen.

Table 2. Comparison of coagulation parameters before and after treatment in pediatric KHE patients

Time	APTT (S)	PT (S)	TT (S)
Before treatment	32.2±3.8	13.7±0.9	15.2±0.7
After treatment	26.6±4.1	12.0±0.8	13.4±0.5
t value	5.487	7.733	11.461
P value	<0.001	<0.001	<0.001

APTT: Activated Partial Thromboplastin Time; PT: Prothrombin Time; TT: Thrombin Time.

tion, emotional function, social function, and cognitive function in the KHE children significantly increased compared to before treatment (all P<0.001).

Binary logistic regression analysis of quality-of-life scores in children with KHE

All domain and total scores from the PedsQL™ 4.0 were linearly transformed and standardized to a 0-100 scale. Using one standard deviation below the mean total score (83.2) as the cutoff for *impaired quality of life*, a total of 33 children (55.55%) were identified as having impaired quality of life. Subsequently, Univariate analysis revealed that children with activity impairment, lower platelet counts, or parents with lower education attainment had significantly lower scores across all HRQoL domains compared to their counterparts without these risk factors (all *P*<0.05). Subsequent multivariate analysis further identified the presence of an activity disorder, reduced platelet count, and lower parental education as independent factors adversely affecting HRQoL in children with KHE (**Tables 4**-**6**).

Multivariate COX regression analysis of survival in KHE patients

Univariate Cox proportional hazards regression analysis identified age, platelet count, coagulation function, and tumor size as significant risk factors affecting the survival of children after treatment. Multivariate analysis further confirmed that age and platelet count were independent prognostic predictors of survival duration in children (both P<0.05) (**Tables 5** and **7**).

Discussion

The pathological feature of KHE is the formation of tumor-like vascular structures resulting from the proliferation of endothelial cells [11, 12]. The lesions are not confined to the skin but also involve internal organs such as the lungs

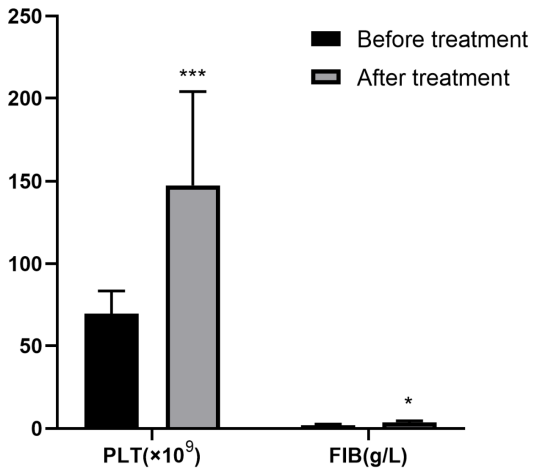

Figure 6. The levels of platelet (PLT) and fibrinogen (FIB) in the two groups of patients.

and liver, severely compromising the quality of life of affected patients [13, 14]. A severe complication associated with KHE is the Kasabach-Merritt phenomenon (KMP), which is characterized by severe thrombocytopenia and coagulation dysfunction. This condition results from platelet capture and activation within tumor-feeding blood vessels. Clinically, affected patients often present with bleeding tendency, including petechiae, nosebleed, or gingival bleeding; in severe cases, internal bleeding may occur, posing threat to life. Its pathophysiology is closely related to the biological behavior of tumors: the proliferation of endothelial cells not only drives tumor growth, but also causes hemodynamic changes, leading to platelet sequestration and the release of procoagulant factors, thereby aggravating coagulation disorders [15, 16].

A variety of treatments have been proposed for KHE. Although surgical resection can achieve radical removal of local lesions, it may cause severe aesthetic and functional damage to aesthetically or functionally sensitive parts [17, 18]. This study showed that interventional embolization combined with pingyangmycin injection provided favorable therapeutic outcomes, as evidenced by significantly increased platelet count after treatment. The therapeutic mechanism can be explained as follows: interventional embolization, a minimally invasive technique, delivers embolic material to the tumor-feeding arteries, thereby blocking blood supply and inducing ischemic shrinkage of the lesion. This process not only reduces tumor volume but also relieves symptoms and improves the quality of life. At the same time, pingyangmycin is an anti-tumor antibiotic with cytotoxic effects that inhibit tumor cell proliferation and angiogenesis. The combination of these two methods creates a synergistic effect - physically blocking tumor perfusion while chemically targeting tumor cells - thereby establishing a multidimensional treatment paradigm consistent with previous reports [19, 20].

Quality of life is closely related to the therapeutic effect. Our study demonstrated that this combination therapy significantly improved the quality of life by reducing the tumor burden and conferring psychosocial benefits [21, 22]. The regression of lesions enhances the mental health of children and helps to restore normal activities [23, 24]. Further analysis found that activity disorders, low platelet count, and low parental education were significant negative predictors of patients' quality of life. Due to fear of bleeding, thrombocytopenia often limits physical activity, which fosters a disease-centered lifestyle, contributing to psychological distress. Low parental education levels may hinder access to health information and support resources, weakening adaptive coping. For example, children from such environment are more vulnerable, with aggravated helplessness. These factors together contribute to impaired emotional and social functions. These findings collectively indicate that the physical activity, psychological well-being, and social support of KHE patients are particularly fragile domains of QoL [25, 26].

Limitations

This study has several limitations. First, although the sample size is informative, it is still limited; future multicenter studies with larger cohorts are warranted to verify these findings. Second, as a single-arm retrospective study, this study has inherent limitations, mainly due to the lack of a control group. Future work should involve rigorously designed prospective randomized controlled trials to directly compare this treatment plan with current first-line treatment (such as sirolimus), thereby providing a more accurate assessment of its clinical efficacy. In addition, the use of standardized laboratory reference ranges rather than age-stratified pediatric standards may affect the interpretation of hematological parameters,

Table 3. Comparison of quality of life before and after treatment in pediatric KHE patients

PedsQL™4.0	Body functions	Physical Symptoms Function	Emotional Function	Social features
Before treatment	81.5±14.3	75.3±14.1	76.8±13.2	82.1±11.2
After treatment	89.4±12.9	82.5±13.6	85.4±12.9	93.4±12.6
t value	2.247	2.013	2.552	3.671
P value	0.028	0.049	0.013	<0.001

Table 4. Univariate analysis of factors affecting health-related quality of life in pediatric KHE patients

PedsQL™4.0	Body functions	Physical symptom function	Emotional Function	Social Features
Presence of functional impairment in mobility				
Yes	82.5±15.5	85.4±16.3	77.9±17.2	93.4±12.6
None	70.4±14.9	72.3±15.7	67.3±14.2	82.1±11.2
P value	0.005	<0.001	<0.001	<0.001
Platelet count ($\times 10^9$)				
< mean value	86.2±10.4	87.8±12.7	81.7±13.5	96.5±3.7
≥ mean value	66.6±17.1	76.4±15.5	63.5±15.4	80.4±14.6
P value	<0.001	<0.001	<0.001	<0.001
Lesion location				
Limbs	72.3±17.6	78.7±13.3	67.5±16.1	83.6±15.2
Torso	76.5±18.6	83.5±13.6	74.2±18.6	88.7±12.3
P value	0.675	0.836	0.492	0.676
Mother's education level				
Secondary education or lower	77.8±6.5	85.6±6.0	80.1±6.4	83.4±6.5
Tertiary education	89.5±5.4	89.1±4.9	87.3±7.8	91.3±8.0
P value	<0.001	<0.001	<0.001	<0.001
Father's education level				
Secondary education or lower	82.3±6.1	81.5±5.4	80.1±7.1	82.3±6.1
Tertiary education	89.0±6.4	88.6±4.9	89.2±5.7	90.1±6.3
P value	<0.001	<0.001	<0.001	<0.001

Table 5. Assignment table

Variable	Assignment
Age	≤9 month =0; >9 month =1
Coagulation dysfunction	Normal =0; Abnormal =1
Platelet count	Below Median =0; Above Median =1
Tumor diameter size	Below Median =0; Above Median =1
Accompanied by limb dysfunction	Normal =0; Abnormal =1
Mother's education level	Secondary education or lower =0; Tertiary education =1
Father's education level	Secondary education or lower =0; Tertiary education =1

Table 6. Multivariate Analysis of health-related quality of life in pediatric KHE patients

Variable	β	OR value	95% CI	*P* value
Accompanied by limb dysfunction	0.549	1.731	1.632-4.001	0.036
Platelet count	0.417	1.518	1.217-3.428	0.029
Mother's education level	0.534	1.706	1.033-3.364	0.030
Father's education level	0.594	1.813	1.210-3.227	0.017

Table 7. COX regression analysis of survival in pediatric KHE patients

Variable	Univariate COX regression analysis			Multivariate COX regression analysis		
	HR	95% CI	P	HR	95% CI	P
Age	1.080	1.038-1.120	<0.001	2.358	1.076-3.011	<0.001
Coagulation dysfunction	1.580	1.477-15.958	0.009	-	-	-
Platelet count	1.267	1.104-1.453	<0.001	3.044	2.112-3.992	<0.001
Tumor diameter size	1.032	1.067-1.589	0.038	-	-	-

and future studies should consider age-adjusted ranges for more accurate assessment.

Conclusion

Interventional embolization combined with pingyangmycin injection achieves favorable clinical outcomes and improves the quality of life of children with KHE. Quality of life is influenced by disease severity and parental education, emphasizing the need for integrated clinical management and psychosocial support in comprehensive care for these patients.

Disclosure of conflict of interest

None.

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