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WILMS' TUMOR IN HORSESHOE KIDNEY: POSSIBILITIES OF ORGAN-PRESERVING TREATMENT (A CLINICAL CASE)

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ABSTRACT

Relevance: *Horseshoe kidney is the most typical kidney fusion anomaly among children (0.25%) and is associated with various urological and non-urological abnormalities. Wilms' tumor is the most common malignant neoplasm of the kidneys and the third most common solid malignant neoplasm in pediatrics.*

The study aimed to *present a clinical case of Wilms' tumor detected in a horseshoe kidney, describing the diagnostic and treatment methods.*

Methods: *The article describes a clinical case of Wilms' tumor in the horseshoe kidney of a 4-year-old girl treated at the Moscow Regional Oncological Dispensary (Balashikha, Russia).*

Results: *A combined treatment was performed, including neoadjuvant and adjuvant chemotherapy and surgical resection of the left half of the horseshoe kidney at the isthmus level. At the time of writing, there were no manifestations of the disease.*

Conclusion: *The clinical case highlights the importance of a timely diagnosis of Wilms' tumor and the initiation of treatment, which significantly contributes to a favorable outcome. Early diagnosis and treatment allowed a particular patient to evaluate all possible outcomes and determine further tactics. This made it possible to remove Wilms' tumor localized in the horseshoe kidney with minimal loss of renal and urinary system function.*

Keywords: *pediatrics, surgical treatment, nephroblastoma, Wilms' tumor, horseshoe kidney, chemotherapy.*

Introduction: Wilms' tumor, also known as nephroblastoma, is the most common kidney tumor in children [1-3]. Kidney tumors account for approximately 5% of malignant neoplasms in children under 15 years old and 3.6% of malignant neoplasms in children under 18 years old. Among the 9,731 patients enrolled in the National Renal Tumor Research Group (NWTSG) (1969–2002), nephroblastoma accounted for the vast majority of childhood kidney tumors (92%), followed by clear cell renal sarcoma (3.4%), congenital mesoblastic nephroma (1.7%), malignant rhabdoid tumor (1.6%), and rare neoplasms, including primitive neuroectodermal tumor, synovial sarcoma, neuroblastoma, and cystic nephroma (1.1%). Although renal cell carcinoma has not historically been included in NWTSG studies, it accounts for 8% of kidney tumors in children from birth to 19 years old, according to the Surveillance, Epidemiology, and Outcomes (SEER) Program [2]. Horseshoe kidney is the most common abnormality of kidney formation [4].

The primary kidney migrates from the pelvic cavity to the level of the upper lumbar vertebrae during the formation and development of the renal system, accompanied by additional rotation and fixation of the organ in its typical position [5]. Renal fusion anomalies can occur during rotation and kidney elevation during the 9th week of ontogenesis [6]. The isthmus of a horseshoe kidney may contain a functioning renal parenchyma or a fibrous band [7]. In up to 80% of horseshoe kidney cases, the isthmus contains functional renal parenchyma tissue, and in 90% of cases, fusion occurs at the lower pole [8]. Horseshoe kidneys are often asymptomatic and are usually discovered by chance, often due to symptoms or secondary disorders of the genitourinary system, such as genitourinary infections or obstructions [8]. It is believed that these patients are at increased risk of developing malignant neoplasms, such as Wilms' tumor [6]. Nephroblastoma is the most common malignant kidney tumor detected in childhood [7]. The risk of developing Wilms' tumor in children with horseshoe kidney is 2-6 times greater than in children in general [7]. Approximately 50% of Wilms' tumors in horseshoe kidneys develop from the isthmus, probably due to abnormal proliferation of the metanephric blastema [6]. The same abnormality that causes the development of a horseshoe kidney can also lead to the development of Wilms' tumor [8]. Nephroblastoma is asymptomatic; approximately 10% are detected accidentally after injury, while 25% are found to have microhematuria or hypertension, which occurs against the background of hyperproduction of renin [1].

Ultrasound is used to diagnose a horseshoe kidney, whereas computed tomography (CT) and magnetic reso-

nance imaging (MRI) are used in staging the process [3]. On ultrasound, the tumor appears as a large mass that can be solid or cystic, characterized by large hypoechoic areas resulting from central necrosis and cyst formation [1]. The areas are characterized by fat deposits, calcifications, or hemorrhages [1]. CT scans show tumors with a lower density and are less visualized than the normal renal parenchyma [4]. Tumors are often characterized by heterogeneous enhancement and may have inclusions in the form of accentuated calcifications [4]. In magnetic resonance imaging, tumors exhibit low signal intensity in T1-weighted images, varying signal intensity in T2-weighted images, and limited diffusion in diffusion-weighted images [5]. CT is also used to detect lung metastasis or local recurrence [5].

Wilms' tumor may contain inclusions of embryonic renal elements, including blastema, epithelium, and stroma [4]. Wilms' tumor can be divided into 2 types based on the prognosis: favorable (more than 90%) and unfavorable (6–10%) [5]. Histopathological analysis is the modern gold standard for diagnosing Wilms' tumor.

Surgery, chemotherapy, and radiation therapy are used to treat Wilms' tumor [6]. The National Wilms' Tumor Study Group (NWTSG)/Children's Oncology Group (COG) and the International Society of Pediatric Oncology-Renal Tumor Study Group (SIOP) have established main guidelines for the management of patients with Wilms' tumor [8]. SIOP recommends the use of preoperative chemotherapy to reduce tumor size and prevent intraoperative complications due to tumor rupture [7]. In contrast, the NWTSG/COG recommends the use of primary surgery before any conservative therapy [2]. The overall survival rate for children with Wilms' tumor in the horseshoe kidneys is similar to that for children with Wilms' tumor in normal kidneys: NWTS-4 stage I-IV (event-free survival – 80.6%-94.9%, overall survival – 93%-98.7%) [6].

The study aimed to present a clinical case of Wilms' tumor detected in a horseshoe kidney, describing the diagnostic and treatment methods.

Description of the clinical case.

Patient's data: A 4-year-old girl, 4 months from the 8th pregnancy, second childbirth. The weight at birth was 2980 g. No chronic diseases. There were no surgeries or injuries. The ultrasound results revealed the formation of the abdominal cavity during a routine examination at the place of residence. The child was hospitalized in the Moscow Regional Oncology Dispensary (Balashikha, Russia).

Diagnostics: During the examination, palpation revealed a volumetric formation of tightly elastic consistency, painless and immobile, on the right side. General urinalysis revealed microhematuria. Blood pressure was higher than the norm for the age. A CT scan of the abdominal cavity with intravenous contrast enhancement was performed on 04.09.24: CT scan of a horseshoe kidney, a tumor mainly of the right half of the kidney with a size of 6.6×8.3×8 cm, heterogeneous structure. Renal veins contrast homogeneously (Figure 1).

The diagnosis was established based on instrumental research methods: "Nephroblastoma of the horseshoe kidney on the right."

The child was admitted to the Moscow Regional Oncology Dispensary to determine further treatment tactics and receive specific treatment.

Treatment: From 06.09.2024, after the diagnosis, the child began to receive therapy according to the Umbrella SIOP 2016 protocol, AV block:

Week 1 (06.09.2024) – Vincristine 1.5 mg/m^2 IV bolus (single dose of 1.0 mg), Actinomycin D 45 µg/kg IV bolus (single dose of 0.72 mg).

Week 2 (13.09.2024) – Vincristine 1.5 mg/m^2 IV bolus (single dose of 1.0 mg).

Week 3 (20.09.2024) – Vincristine 1.5 mg/m^2 IV bolus (single dose of 1.0 mg), Actinomycin D 45 µg/kg IV bolus (single dose of 0.72 mg).

Week 4 (27.09.2024) – Vincristine 1.5 mg/m^2 IV bolus (single dose of 1.0 mg).

Due to technical difficulties during surgery at Week 5, an additional injection of vincristine was administered following the recommendations of the RF Ministry of Health.

Week 5 (04.10.2024) – Vincristine 1.5 mg/m^2 IV bolus (single dose of 1.0 mg).

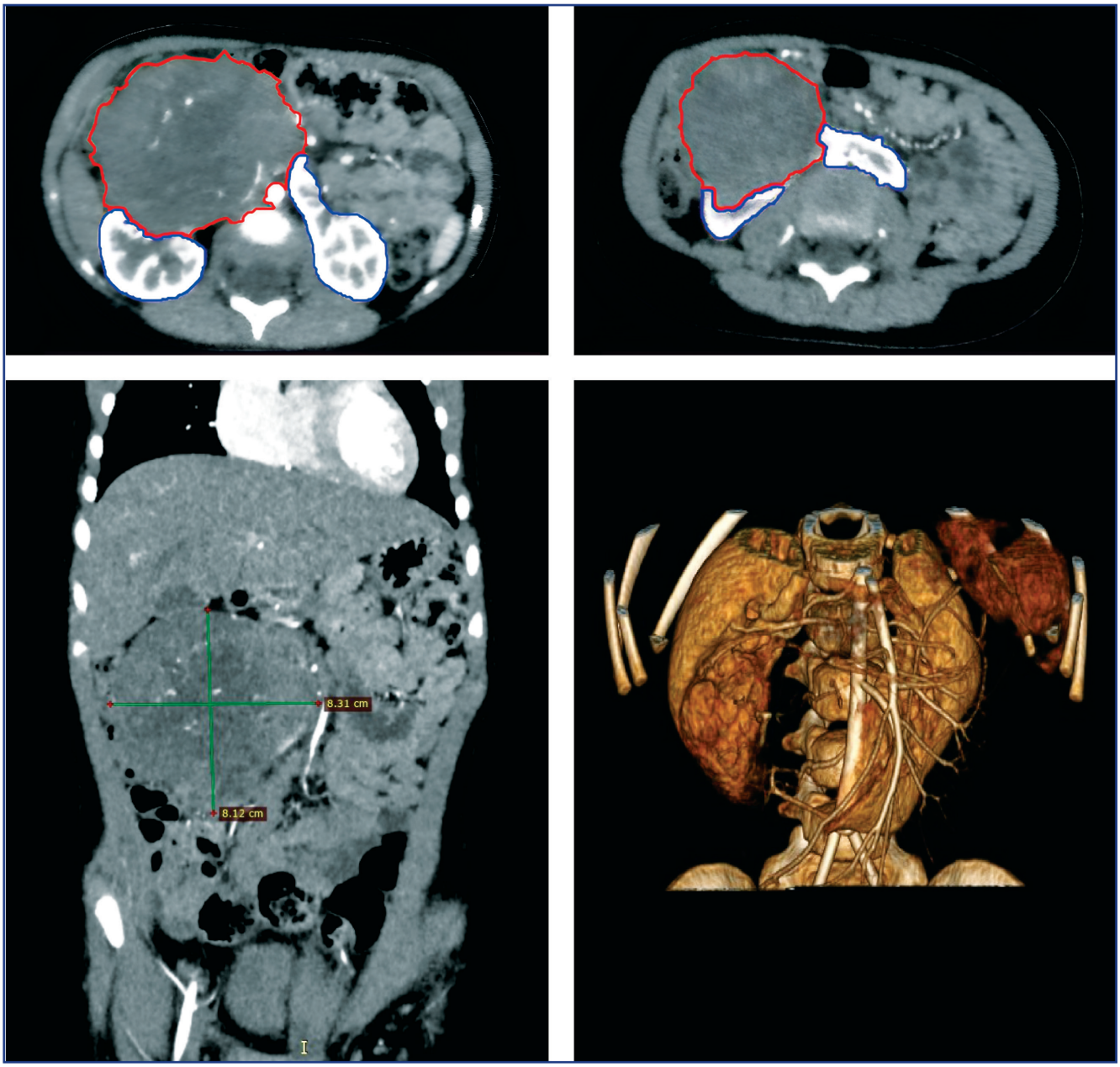
Against the background of preoperative chemotherapy, a CT scan of the abdominal cavity and retroperitoneal space with intravenous contrast showed a reduction in the size of the tumor node from 6.6×8.3×8 cm (230 cm^3) to 5.1×5.7×5.7 cm (86 cm^3). The tumor has decreased by 62.6% of its initial volume (Figure 2).

It was decided to perform surgery after neoadjuvant maintenance chemotherapy within the framework of the Umbrella SIOP 2016 protocol.

The surgical intervention was performed on October 14, 2024. The access was via median laparotomy. The revision revealed no pathology in the abdominal organs. A rounded tumor of 6×6×5 cm was visualized in the retroperitoneal space in the centre and to the right at the level of the lower poles of the kidneys. It emanated from the isthmus of the horseshoe kidney and spread to a greater extent to the lower parts of the right half of the horseshoe kidney. The right ureter passed along the anterior edge of the tumor node; the inferior vena cava (compressed by the tumor) was posterior to the tumor, and there was the bifurcation of the aorta. The right lateral canal was opened, and the right half of the horseshoe kidney, including the tumor and the isthmus to the level of the left part, was mobilized. The right ureter was isolated and mobilized to the right pelvis (Figure 3).

The inferior vena cava was abruptly separated from the tumor node, and the aorta was mobilized at the bifurcation level. A feeding vessel extending from the aorta to the isthmus of the horseshoe kidney was found, li-

gated, and dissected. The left ureter was checked. Acute resection of the left half of the horseshoe kidney was performed at the level of the isthmus within healthy tissues. Suturing of the lower group of calyces of the left half of the horseshoe kidney was performed with Prolene 4-0 thread. The encircling stitch was made with Vicryl 0 thread on the lower pole of the left kidney half. Acute resection of the right half of the horseshoe kidney was performed at the level of the lower pole within healthy tissues. Suturing of the lower group of calyces of the right half of the horseshoe kidney was performed with Prolene 4-0 thread. The encircling stitch was made with Vicryl 0 thread on the lower pole of the right kidney half (Figure 4).



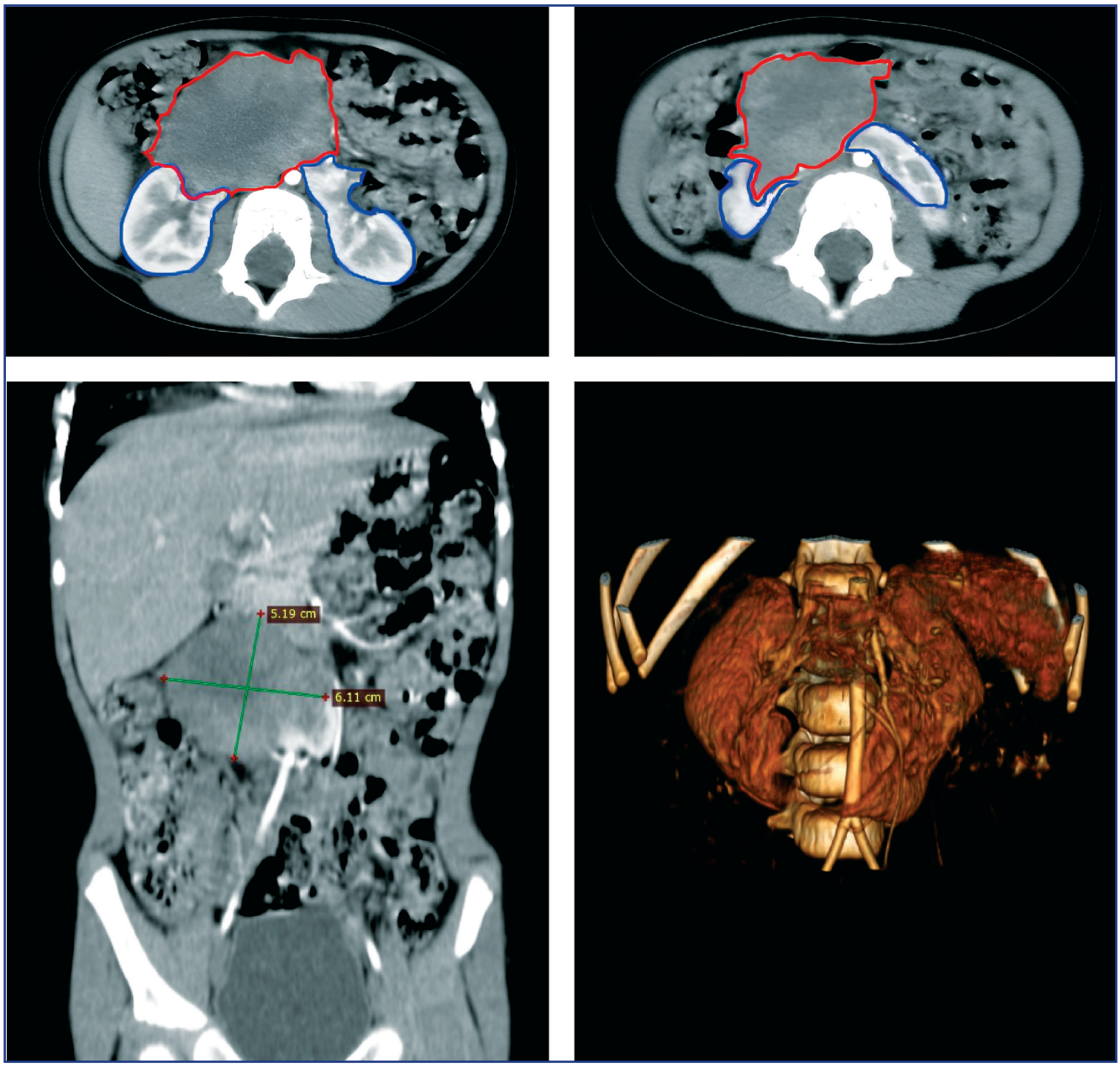
Blue line – Boundaries of normal renal tissue; Red line – Boundaries of the neoplasm
Figure 1 – Computed tomography of the abdominal cavity and retroperitoneal space with intravenous contrast dated 04.09.2024, arterial phase

Hemostasis during the operation – dry. The revision revealed no pathological findings; the sutures were consistent, with no leaks. The following procedures were performed: drainage of the pelvis with a silicone tube led through a counter-aperture on the right; plastic surgery of the right lateral canal; layer-by-layer suturing of the postoperative wound; cosmetic skin suture. Urine was drained through a Foley catheter inserted into the urethra.

The material obtained during the operation was sent for pathomorphological examination (Figure 5).

The child in stable condition was transferred from the intensive care unit to the children's oncology department on the 2nd day after the operation. Blood pressure was

within the normal range for the age. Urine outflow through the urethral catheter was adequate, without pathological inclusions or discoloration. Discharge through the drainage was below 100 ml.



Blue line – Boundaries of normal renal tissue; Red line – Boundaries of the neoplasm

Figure 2 – Computed tomography of the abdominal cavity and retroperitoneal space with intravenous contrast dated 02.10.2024

The drainage was removed along with the urethral catheter on the 7th day after surgery.

Histochemical study dated 16.10.2024: Nephroblastoma, epithelial type, intermediate risk group, R0-resection. pT2N0M0.

The final clinical diagnosis was established: Nephroblastoma on the right, epithelial type, intermediate risk group, local stage 1.

The child began to receive adjuvant chemotherapy according to the Umbrella SIOP 2016 protocol for the intermediate histological risk group block AV1 from 24.10.2024, after surgical treatment:

Week 1 (24.10.2024) – Vincristine 1.5 mg/m^2 IV bolus (single dose of 1.0 mg).

Week 2 (01.11.2024) – Vincristine 1.5 mg/m^2 IV bolus (single dose of 0.9 mg), Actinomycin D 45 µg/kg IV bolus (single dose= 0.65 mg).

Week 3 (08.11.2024) – Vincristine 1.5 mg/m^2 IV bolus (single dose of 0.9 mg).

Week 4 (15.11.2024) – Vincristine 1.5 mg/m^2 IV bolus (single dose of 0.9 mg).

Control computed tomography of the abdominal organs and retroperitoneal space with intravenous contrast enhancement was performed on 21.11.2024: functionally sound

right and left kidneys were visualized, with dimensions of 34×37×76 mm on the right, and 52×27×90mm on the left. The pelvic system of both kidneys was not deformed or dilated, and radiopaque concrements were not found (Figure 6).

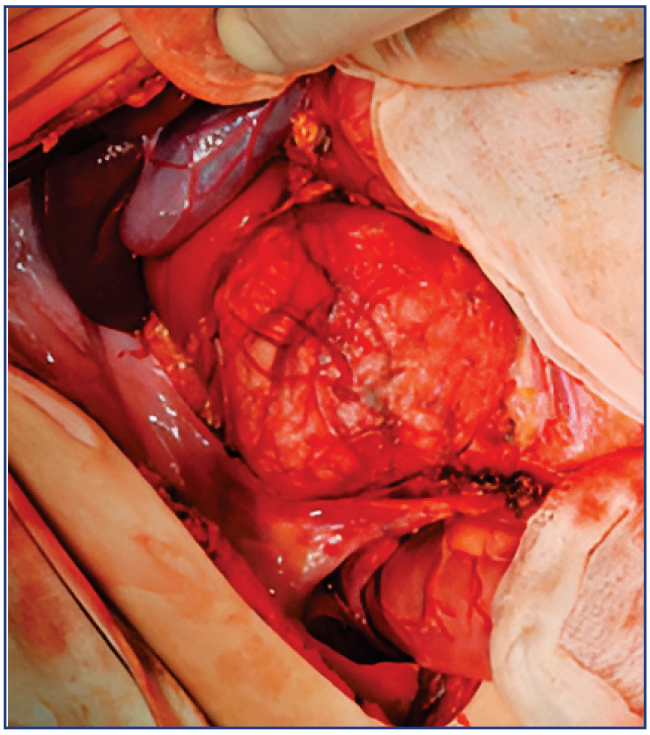


Figure 3 – Horseshoe kidney mobilization and formation

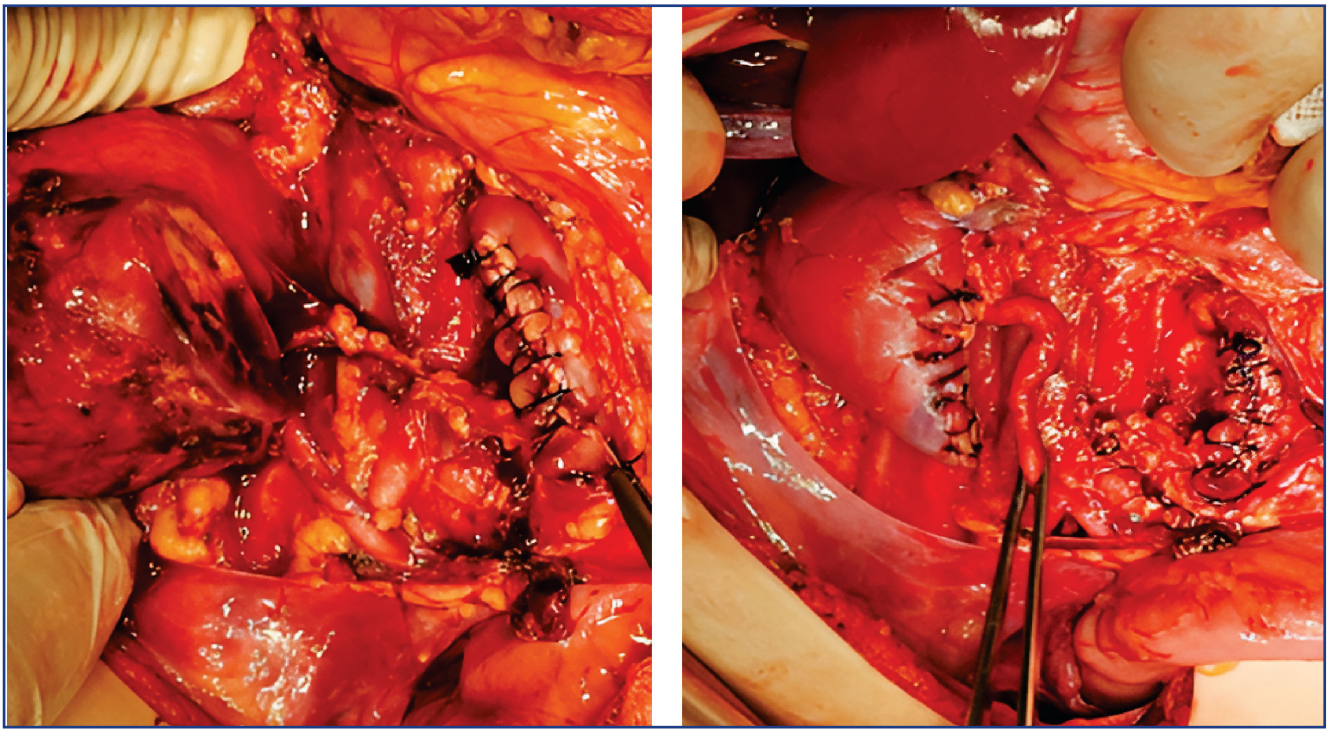


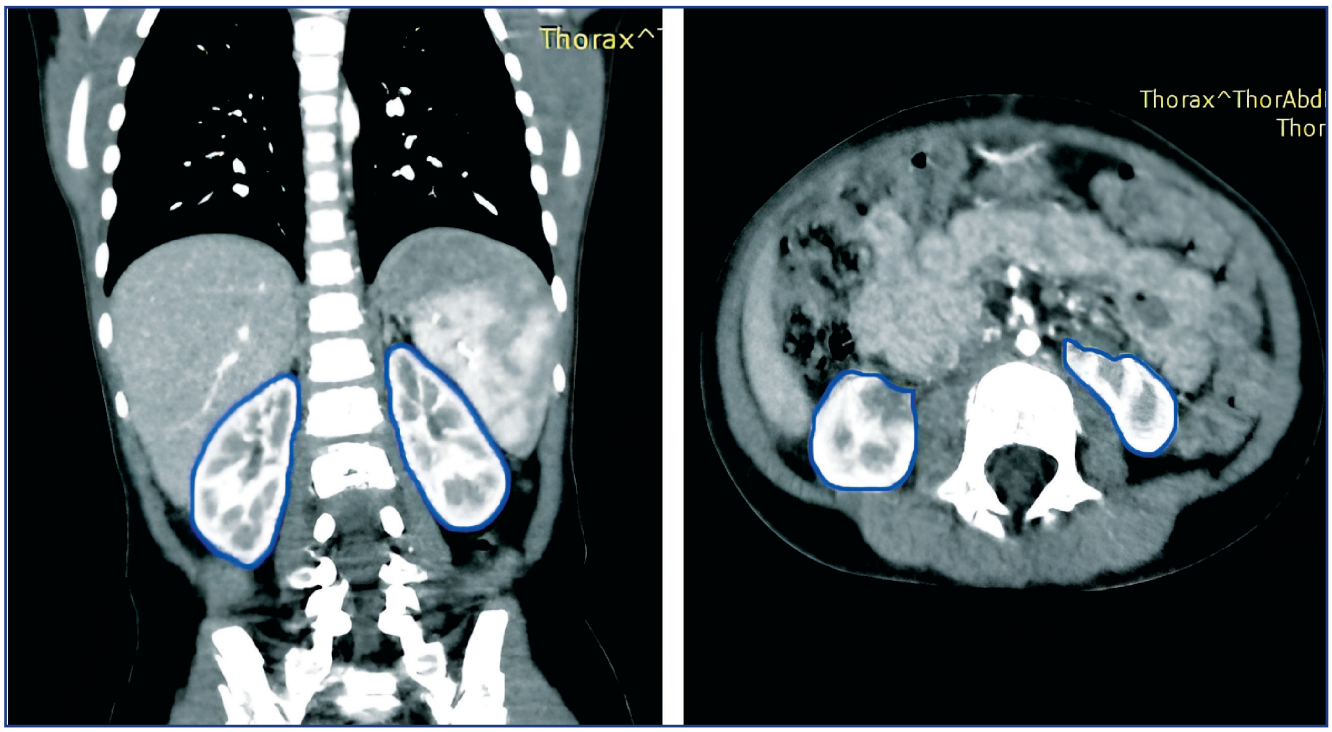
Figure 4 – Suturing of the lower poles of the right and left kidneys

Results: A combined treatment was performed, including neoadjuvant and adjuvant chemotherapy and a resection of the left half of the horseshoe kidney at the isthmus. The primary indicators, including blood pressure and a general urinalysis, were monitored during treatment. These indicators demonstrated the positive dynamics of the patient, from the moment of admission to the moment of withdrawal, as erythrocytes ceased to be detected in the general urinalysis, and BP values stabilized within the age norm. In dynamics, radiation di-

agnostic methods showed a positive trend in response to chemotherapy. A follow-up examination at the time of withdrawal from treatment made it possible to verify the efficiency of the therapy and the integrity of the urogenital system after reconstructive plasty. There were no manifestations of the disease at the time of writing the article.

The timeline of the clinical case is presented in Figure 7.

Figure 5 – Taken gross specimen



Blue line – Borders of normal renal tissue

Figure 6 – Computed tomography of the abdominal cavity and retroperitoneal space with intravenous contrast dated 21.11.2024

Discussion: Wilms' tumor is the most common kidney malignant neoplasm in children and the fifth most common malignant neoplasm in children in general [1]. Horseshoe kidney is a kidney fusion abnormality characterized by the fusion of the kidneys through the isthmus at the lower pole in approximately 90% of cases. This isthmus is predominantly composed of functional renal tissue, although it can sometimes appear as a fibrous band [1, 3]. Horseshoe kidney is a risk factor for kidney malignant neoplasms [8]. Despite the increased risk compared to the general population, horseshoe kidney is not currently recommended as a condition requiring Wilms' tumor screening [6]. Since horseshoe kidney is usually asymptomatic [3, 4], most cases described in the literature were diagnosed at the same time as the tumor itself [8].

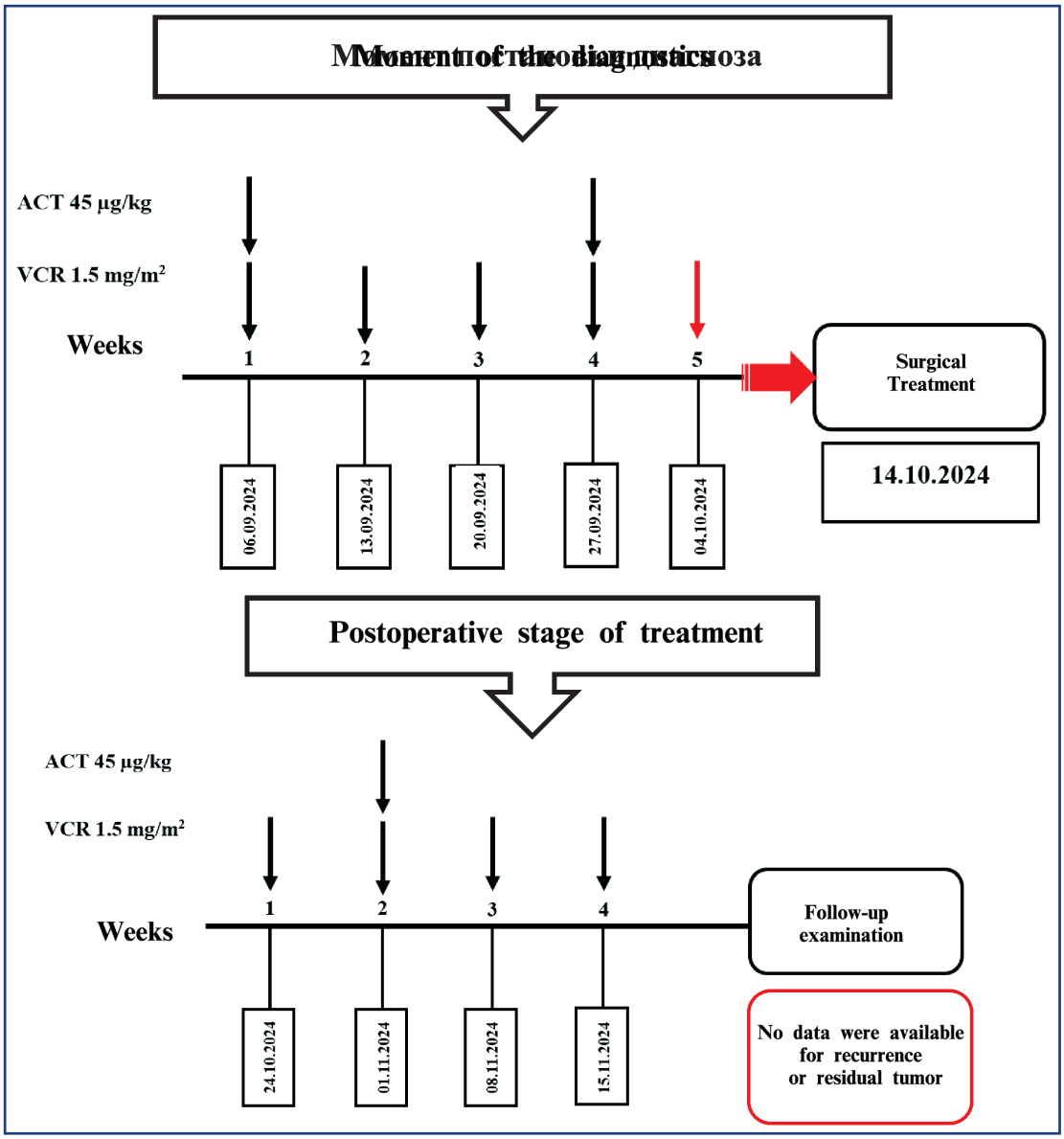


Figure 7 – Timeline of the clinical case of Wilms' tumor in a horseshoe kidney in a 4-year-old girl

Radical nephroureterectomy with lymph node removal using a wide transverse, transperitoneal approach is recommended for adequate tumor resection [8]. Complete removal of the affected kidney, along with the isthmus and tumor, is recommended in case of a unilateral tumor in the horseshoe kidney. When the tumor is located in the isthmus, an organ-preserving treatment method is possible [7, 8].

In the presented case, the tumor node was removed along with the isthmus, with the simultaneous formation of two separate kidneys.

Conclusion: Wilms' tumor is the most common malignant neoplasm of the kidneys among the pediatric population. This nosology can be associated with various congenital anomalies, such as sporadic aniridia, hemihypertrophy, and genitourinary anomalies, which in some cases make it possible to verify the diagnosis at an early stage, during the process of studying background conditions. But in most cases, nephroblastoma is diagnosed at the stage of visual changes in the child's body, when parents begin to see body asymmetry and can palpate the formation. It of-

ten leads to several complications, such as rupture of the tumor capsule, which in turn is an unfavorable prognosis in terms of contamination of the surrounding tissues and organs located in the abdominal cavity and retroperitoneal space with tumor cells.

Early diagnostics of concomitant renal pathologies makes it possible to adequately route patients and plan management tactics.

Preoperative chemotherapy allows for the avoidance of several possible complications during surgery.

A combined anomaly of the structure of the kidneys and neoplasms requires a highly qualified surgical team to perform one-stage removal of the tumor and use a reconstructive method.

The rational performance of computed tomography of the abdominal cavity and retroperitoneal space enables the adequate assessment of the effect of each stage of treatment, as well as the comparison of dynamics relative to the initial data obtained during the detection of the formation.

If it is impossible to perform surgery at the appointed time, it is worth considering the option to add a course of chemotherapy in order to maintain the therapeutic effect of the block until the moment when surgery becomes possible.

This clinical case highlights the importance of timely diagnosis of Wilms' tumor and the initiation of treatment, which contributes to a favorable outcome. Early diagnostics and treatment made it possible to assess all possible outcomes in a particular patient and determine further tactics; as a result, Wilms' tumor, localized in the horseshoe kidney, could be corrected with minimal loss of kidney function and urinary system function.

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АҢДАТПА

АТТАСҚАН БҮЙРЕКТЕ ДАМЫҒАН ВИЛЬМС ІСІГІ: АҒЗАЛАРДЫ САҚТАУ ЕМІНІҢ МҮМКІНДІКТЕРІ (КЛИНИКАЛЫҚ ЖАҒДАЙ)

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Өзектілігі: *Аттасқан (таға тәрізді) бүйрек – балаларда жиі кездесетін бүйрек қосылу аномалиясы (0,25%) болып табылады және ол әртүрлі урологиялық және бейурологиялық ауытқулармен байланысты. Вильмс ісігі – балалардағы ең жиі кездесетін қатерлі бүйрек ісігі және үшінші жиі кездесетін қатты тіндік қатерлі ісік болып саналады.*

Зерттеу мақсаты: *Аттасқан бүйректе анықталған Вильмс ісігінің клиникалық жағдайын сипаттау, диагностика және емдеу әдістерін баяндау.*

Әдістер: *Бұл мақалада 4 жастағы қыз баланың аттасқан бүйрегінде анықталған Вильмс ісігінің клиникалық жағдайы сипатталады. Емдеу Мәскеу облыстық онкологиялық диспансерінде (Балашиха, Ресей) жүргізілген.*

Нәтижелері: *Неоадъювантты және адъювантты химиотерапияны және аттасқан бүйректің сол бөлігін мойын тұсында резекциялау көлемінде жасалған операцияны қамтитын кешенді ем жүргізілді. Мақала жазу кезінде аурудың көріністері тіркелмеген*

Қорытынды: *Бұл клиникалық жағдай Вильмс ісігін дер кезінде анықтап, емдеудің маңыздылығын көрсетеді, бұл қолайлы нәтижеге қол жеткізуге мүмкіндік берді. Ерте диагностика мен ем нақты науқастың барлық мүмкін нәтижелерін бағалап, әрі қарайғы ем тактикасын анықтауға септігін тигізді. Соның арқасында аттасқан бүйректе орналасқан Вильмс ісігін бүйрек пен несеп шығару жүйесінің функциясына барынша аз зиян келтіре отырып емдеуге мүмкіндік болды.*

Түйінді сөздер: *педиатрия, хирургиялық ем, нефробластома, Вильмс ісігі, аттасқан бүйрек, химиотерапия.*

ABSTRACT

WILMS' TUMOR IN HORSESHOE KIDNEY: POSSIBILITIES OF ORGAN-PRESERVING TREATMENT (A CLINICAL CASE)

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Relevance: *Horseshoe kidney is the most typical kidney fusion anomaly among children (0.25%) and is associated with various urological and non-urological abnormalities. Wilms' tumor is the most common malignant neoplasm of the kidneys and the third most common solid malignant neoplasm in pediatrics.*

The study aimed to *present a clinical case of Wilms' tumor detected in a horseshoe kidney to describe diagnostic and treatment methods.*

Methods: *The article describes a clinical case of Wilms' tumor in the horseshoe kidney of a 4-year-old girl treated at the Moscow Regional Oncological Dispensary (Balashikha, Russia).*

Results: *A combined treatment was performed, including neoadjuvant and adjuvant chemotherapy and surgical resection of the left half of the horseshoe kidney at the isthmus level. At the time of writing, there are no manifestations of the disease.*

Conclusion: *The clinical case reflects the need for a timely diagnosis of Wilms' tumor and initiation of treatment, which contributes to a favorable outcome. Early diagnosis and treatment allowed a particular patient to evaluate all possible outcomes and determine further tactics. This made it possible to remove Wilms' tumor localized in the horseshoe kidney with minimal loss of renal and urinary system function.*

Keywords: *pediatrics, surgical treatment, nephroblastoma, Wilms' tumor, horseshoe kidney, chemotherapy.*

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