

ORIGINAL PAPER

## Multicystic dysplastic kidney with severe abdominal obstruction signs – case reports and management of children

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### ABSTRACT

**Introduction:** Multicystic dysplastic kidney (MCDK) is a relatively common developmental abnormality of the urinary tract. In most cases, it is an asymptomatic disease and rarely leads to problems emerging from the pressing effect of large MCDK kidneys on the adjacent organs. Nowadays, surgical intervention has mostly been replaced by a non-invasive approach with long-term follow-up.

**Aim of the study:** Analysis of the diagnostic process, patients' clinical condition, treatment, and follow-up in neonatal patients with extreme MCDK.

**Material and methods:** Retrospective analysis of medical records of four infants with extreme MCDK, who were hospitalised in the Department of Intensive Therapy and Neonatal Pathology in Independent Public Clinical Hospital No. 1 in Zabrze in 2014–2019. The study also includes data from an out-patient nephrological unit, where infants are checked up by paediatric nephrologists every 6 to 12 months.

**Results:** All four patients involved in this study were diagnosed during the prenatal period. After birth, the initial diagnosis was confirmed by ultrasound, X-ray, computed tomography (CT), magnetic resonance imaging (MRI), and/or scintigraphy. All patients in their neonatal period presented the symptoms caused by the pressing effect of the large size of MCDK kidneys, such as abdominal or flank discomfort, digestive disturbances, or respiratory distress. The life-threatening clinical condition of two of our patients led to surgical intervention, which significantly improved their life functions. The stable clinical condition of another two infants enabled a non-invasive approach. All four infants still remain under long-term follow-up. They are not afflicted with any problems emerging from the urinary tract and they do not require any pharmacological treatment.

**Conclusions:** Despite the currently proposed non-invasive approach to MCDK, there are cases when a nephrectomy should be considered, especially regarding patients with extreme-sized kidneys.

### KEY WORDS:

nephrectomy, multicystic dysplastic kidney, urinary tract malformations, renal imaging, non-invasive approach.

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## INTRODUCTION

Multicystic dysplastic kidney (MCDK) is one of the most commonly identified urinary tract anomalies, the overall incidence of which is approximately 1 in 4300 live births. It is one of the most frequent causes of a palpable abdominal mass in the neonatal period and is the most common cystic malformation of kidneys in infancy [1–3].

There are two leading theories explaining the aetiology of MCDK. The first one suggests that MCDK results from a failure of differentiation of the mesenchymal metanephros and epithelial cells of the ureteral bud, although this view has been questioned because sometimes kidney in MCDK contains some functional renal tissue. An alternative theory suggests that the alteration of the nephrogenesis arises from an impaired foetal urine flow in the early development, which is confirmed by the common finding that each MCDK is generally attached to a non-functioning or atretic ureter [4–6].

The picture of MCDK is characterised by a grapelike cluster of cysts, usually with no identifiable normal renal parenchyma. Currently, it is one of the most commonly detected anomalies on prenatal ultrasound, and its diagnosis has increased in the past 20 years due to the widespread use of antenatal and postnatal ultrasonography. MCDK usually occurs unilaterally, predominantly in males, and coexists with other urinary tract malformations such as ureteral dilation, ureterovesical stenosis, ureterocele, and ureteral valves. Thus, a more detailed postnatal follow-up is required as a consequence of prenatal detection of MCDK [3, 7, 8].

In the natural course of unilateral MCDK, involution of the afflicted kidney is accompanied by compensatory contralateral hypertrophy. Most infants (about 90%) develop contralateral hypertrophy at the age of three years, which is considered as a good indicator of further kidney function. Unilateral MCDK without contralateral compensatory hypertrophy is regarded as a risk factor for future renal insufficiency and requires imaging follow-up [9, 10].

Renal ultrasound is a natural imaging choice for the follow-up of MCDK patients. Although there are some reports about using fetal magnetic resonance imaging (MRI) for the diagnosis of urinary tract anomalies, there is no confirmation about its clinical value [8]. An alternative approach includes renal scintigraphy, which provides additional information about the current renal function and can be used to confirm a diagnosis of MCDK and accompanying urinary tract malformations. Some authors believe that constant improvement of the renal ultrasound technique is sufficient and there is no need for further confirmation of the MCDK diagnosis by nuclear medicine scan in patients with normal bladder ultrasound image [11]. The first-line imaging for the diagnosis of contralateral vesicoureteral reflux (VUR), which is the most frequent abnormality, occurring in 5 to 43% of patients with unilateral MCDK, is voiding cystourethro-

graphy (VCUG). However, the necessity of routine VCUG is still debated in the literature [12–14].

Until recently, MCDK was generally managed by nephrectomy to avoid complications, such as urinary tract infections, hypertension, or malignant transformation. Currently, the non-invasive protocol has been appointed due to recent studies showing high rates of spontaneous partial or complete involution, low risk of hypertension, and neoplasia. Long-term follow-up of children with MCDK by ultrasound monitoring seems to be the first-line management [15–18].

The most common means of identification of MCDK is ultrasonography or another imaging method. However, there are rare cases when surgical intervention is required, and the diagnosis is made during the surgery itself [19].

This study describes the case studies of four infant patients, in whom the process of diagnosis, treatment, and follow-up was difficult to manage due to the extraordinary dimensions of the affected kidneys.

## MATERIAL AND METHODS

We reviewed retrospectively the medical charts of four patients (two males and two females) with extreme MCDK hospitalised in the neonatal period in the Department of Intensive Therapy and Neonatal Pathology in Zabrze in the years 2014–2019. All patients were followed-up by experienced paediatric nephrologists.

The diagnosis of MCDK was given after prenatal ultrasound findings in all four patients, based on the detected features of multiplied various-sized cysts located in the kidney with no visible parenchymal tissue. In order to extend the diagnosis, additional imaging was performed after birth, including abdominal ultrasound (US) (all patients), abdominal X-ray (three patients), CT scan (one patient), MRI scan (one patient), renal scintigraphy (three patients), and voiding cystourethrogram (two patients). Alongside the postnatal imaging findings we simultaneously evaluated the following clinical parameters: gender, gestational age, condition after birth, APGAR score, renal function tests (serum creatinine and urea level 72 h after birth), cancer risk factors, MCDK side, occurrence of other urological and non-urological abnormalities and dysfunctions, modelling effect on another organs, deviations in physical examination, hypertension, and recurrent urinary tract infections.

Senior staff consisting of experienced neonatologists, paediatric nephrologists, urologists, and paediatric surgeons decided that two of our patients required surgical interventions in the neonatal period. The remaining two patients were treated with a conservative approach.

Since being discharged from the hospital all four patients are being checked up for general development, weight, height, concomitant illnesses, or alterations during the control visits every 6–12 months. Abdominal

ultrasound examination and arterial blood pressure are also verified the same as serum creatinine and urea level, acid-base balance parameters, blood morphology, and urine analysis.

## RESULTS

We reviewed the process of diagnosis, treatment, and follow-up in neonates with unilateral MCDK, in whom affected kidneys were extremely large, thus modelling other adjacent organs (left kidneys in three cases and right kidney in one case). The characteristics of enrolled patients are presented in Table 1. The diagnosis was made in the prenatal period using the maternal sonography for foetal screening in all four cases.

Two infants were born in the full term. One of them was in good condition, while the other required oxygen therapy for a couple of hours after birth. Two more infants were born preterm, and both required additional support.

Palpable masses were discovered during the physical examination of the abdomen in all four infants, although they were clearly palpable in only three of them. All infants presented digestive tract symptoms, such as disturbance of passage, stomach alimentary residuals, decreased appetite, and weakened suction reflex.

A differential diagnosis was performed to prove a connection between cystic structure and the urinary tract. Even though this imaging confirmed the large-size polycystic character of masses and their contribution to relocating other organs, it did not reveal clearly their origin in all of our cases.

The first patient's renal scintigraphy revealed no function of the left kidney and normal function of the right one. The performed VCUG was normal. Because of the disturbance in the functioning of the alimentary tract and bilateral pneumothorax in parbasal parts of lungs, surgical intervention was necessary. The initial procedure was decompression of the cysts by using a nephrostomy catheter. Even though the sterile fluid was evacuated, the US imaging revealed no dimensional change of the cysts. Because of the clinical condition, the patient was qualified for the left-sided nephrectomy by laparoscopic method, which finally confirmed the MCDK diagnosis based on the macroscopic image and histopathological examination (Fig. 1).

The second patient's MRI scan revealed compensatory hypertrophy of the right kidney and multicystic forms mainly on the right side of the abdomen, which was in favour of the MCDK diagnosis with crossed ectopia. Due to the bad clinical condition of the patient, including decreased appetite, distended stomach, and weakened suction reflect, the decision to perform left-sided nephrectomy was undertaken. This procedure led to the improvement of the patient's clinical condition and confirmed the diagnosis of MCDK.

The third patient had early syndromes of digestive tract disability in her early days; however, they gradually disappeared, leading to a conservative approach instead of surgical intervention. At the age of one year, a performed VCUG did not reveal any abnormalities in the lower urinary tract. Five months later renal scintigraphy was performed revealing absent function in the affected right kidney and normal renal excretion on the left side. Until now, only one episode of the urinary tract infection was noted in this child. Control abdominal ultrasound revealed the stationary size of the affected kidney.

The fourth patient was diagnosed with MCDK based on an abdominal ultrasound. Initial problems with the disturbed passage of the digestive tract could be due either to infection or patent ductus arteriosus (PDA). After the successful pharmacological closure of PDA and the reduction of the inflammatory parameters, there were no alarming symptoms from the urinary tract. The decision to use a non-surgical approach was made. During the follow-up, no complications arising from left-sided MCDK were detected. The ultrasound image revealed that the afflicted kidney started to involute. Renal scintigraphy is planned in the future.

Follow-up data presented in Table 2 were collected from all patients, who are now at the age between 5 and 58 months. During follow-up, the first patient was hospitalised because of performed adenotomy, the third patient required surgical intervention due to ovarian torsion on the opposite side to MCDK kidney, and the fourth patient was hospitalised due to respiratory tract infections. All children have normal general development, they do not suffer from any problems from the urinary tract, and they do not require any permanent drug treatment.

## DISCUSSION

Before the widespread use of maternal sonography for foetal screening, most of the diagnosed MCDK cases were large palpable masses identified during the physical examination of a newborn. At that time detecting these abnormalities demanded nephrectomy as a primary treatment, which was considered as the safest therapy to avoid further complications, such as recurrent urinary tract infections, hypertension, and neoplasia [15, 20]. Several studies showed that the prevalence of malignant transformation and hypertension among MCDK patients reflects the occurrence in the general paediatric population [2, 17, 18, 21–24]. Moreover, a prospective study published in 2004 by Rabelo *et al.* demonstrated that complete or partial involution is the most frequently observed course of MCDK. In that study 43 children with MCDK diagnosed by ultrasound scans were involved. The average length of the MCDK was 62 mm (range 18–148 mm). Follow-up lasted for 12–156 months and showed partial involution of the MCDK in 70% of cases, complete involution in 19%, and stable size in 11% [22]. A number of studies have found

TABLE 1. Characteristics of patients with multicystic dysplastic kidney (MCDK)

Characteristic	Patient 1	Patient 2	Patient 3	Patient 4
Gender	Male	Male	Female	Female
Gestational age	Full-term	Pre-term (33 weeks)	Pre-term (35 weeks)	Full-term
Condition after birth	Normal	Average, required ventilator support	Average, required external stimulation	Average, required oxygen therapy for few hours
Antenatal diagnosis	Yes	Yes	Yes	Yes
MCDK side	Left	Left	Right	Left
Physical examination of abdomen	Palpable masses and quiet peristalsis on the left side of abdomen	Palpable masses in both sides of abdomen	Poorly detectable masses on the right side of abdomen	Palpable masses
Urinary tract infections	Once in neonatal period	No	Once in third month of life	No
Other urological affections	No	No	Small cyst in left kidney	No
Syndromes from digestive tract	Alimentary residuals, blood appearing in the stools	Decreased appetite, weakened suction reflex	Decreased appetite, weakened suction reflex	Disturbed passage of digestive tract
Non-urological dysfunctions	No	No	No	Patent ductus arteriosus, dysmorphic facial features
Size of the cysts cluster	92 × 66 × 100 mm (CT)	57 × 97 × 100 mm (MRI)	67 × 38 mm (US)	80 × 55 mm (US)
Modelling effects on another organs	Revealed in CT scan relocation of intestines to the opposite side, modelling the pancreas, stomach, spleen, mesenteric arteries, urinary bladder. Bilateral pneumothorax in parbasal parts	Revealed in MRI modelling neighbouring structures, especially the pancreas, liver, spleen, stomach, intestines, right kidney	No	No
Scintigraphy	No detected function of left kidney. Normal function of right kidney	–	No detected function of right kidney. Normal function of left kidney	Planned
Renal function tests 72 h after birth				
Creatinine serum level (μmol/l)	64	68	51	87
eGFR (ml/min/1.73 m <sup>2</sup> )	36.1	19.3	28.0	22.9
Urea serum level (mmol/l)	6.3	11.3	–	–
Cancer risk factors				
AFP (ng/ml)	740.3	488.6	–	–
CEA (ng/ml)	6.98	–	–	–
β-hCG (mIU/ml)	0.458	–	–	–
Chirurgical intervention				
Laparoscopic nephrectomy	In neonatal period – firstly decompression of cysts by using nephrostomy tube. Because of lack of contenting effect nephrectomy was needed	In neonatal period	–	–
Histopathological examination	Validated diagnosis of MCDK	Validated diagnosis of MCDK	–	–

CT – computed tomography, MRI – magnetic resonance imaging, US – ultrasound, eGFR – estimated glomerular filtration rate, according to new Schwartz formula, AFP – alpha-fetoprotein, CEA – carcinoembryonic antigen

that initially smaller MCDKs are more likely to involute at an earlier age in comparison to larger MCDKs [25, 26].

MCDK is usually asymptomatic; however, large size of abnormal kidneys can cause a pressure effect on adjacent organs, manifested by abdominal or flank discomfort, disorders in the digestive tract, or respiratory distress. These symptoms of abdominal compartment syndrome could be explained by the constricting effect of large-size cysts situated in dysplastic kidneys, which can also lead to respiratory problems caused by consecutive diaphragmatic elevation. When those disturbances are distinctly connected with the pressing effect of MCDK kidney, the abnormal kidney should be removed [1, 27, 28]. In our study, among four patients with enormous size of the affected kidney, two infants required early nephrectomy during their hospitalisation in the Department of Neonatal Pathology. This procedure enabled clinical stabilisation of newborns and improved functioning of the urinary tract and other organs. On the other hand, in these two patients, the diagnosis of MCDK was confirmed after surgical intervention, based on the macroscopic and microscopic view. Previously accomplished imaging did not give assured answers about the origin of cystic masses or its connection with the urinary tract, and it could not exclude malignant proliferation.

Long-term follow-up appears to be widespread, proceeding in both conservative and surgical approaches; however, no universally accepted management protocol has been elaborated. Abdominal ultrasound is a widely used imaging method for monitoring of growth or involution of both cystic and contralateral kidneys, as well as identifying other urinary tract anomalies. Other investigations widely used during follow-up are blood pressure control, urine analysis, and measurement of creatinine, urea, and electrolytes serum levels. More diversified imaging techniques containing VCUG or renal scintigraphy are considered individually.

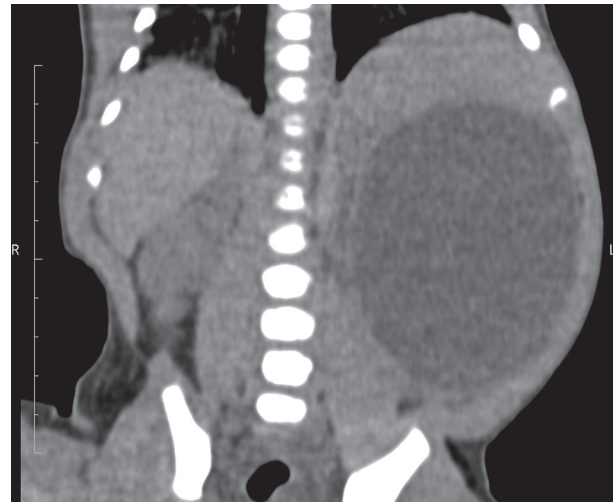


FIGURE 1. First patient's computed tomography scan

In recent years the majority of studies have found a non-invasive conservative approach with long-term ultrasound follow-up as the most appropriate treatment [17, 18, 21, 24, 25, 29]. The retrospective cohort study performed by Brown *et al.* revealed a decreasing trend in using an invasive approach for MCDK patients between January 2006 and September 2015. Meanwhile, the percentage of minimally invasive nephrectomies (laparoscopic non-robotic and robotic) among all performed nephrectomies in this period increased significantly (from 8% in 2006 to 29% in 2015) [30, 31]. No universal indications have been established for nephrectomy in unilateral MCDK patients, except for a few clinical indications, such as: uncontrolled blood pressure despite the use of pharmacological treatment, ultrasound image suggesting the presence of malignant proliferation, and an enlarging renal mass pressing adjacent organs [2, 28]. The last one was the reason why it was decided to perform surgery on our patients. The size of the affected kidney that should determine the need for a nephrectomy is discussed. Some

TABLE 2. Follow-up data

Characteristic	Patient 1	Patient 2	Patient 3	Patient 4
General development	Normal	Normal	Normal	Normal
Actual age	5 years 3 months	2 years 3 months	2 years 4 months	9 months
Weight	> 97 pc	10–25 pc	< 3 pc	75 pc
Height	> 97 pc	3 pc	< 3 pc	75 pc
Hospitalisations	Adenotomy at the age of 4	No	At the age of 3 months urinary tract infection; at the age of 4 months left-sided ovarian torsion – surgical intervention	Respiratory system infections
Pharmacological medication	No	No	No	No
Hypertension	First 5 months after birth	No	First 2 weeks after birth	No
Urinary tract condition	Normal	Compensatory hypertrophy of the remaining kidney	Normal, stable size of the affected kidney	Normal, ultrasound present involution of affected kidney



clinics present the approach that nephrectomy is the best treatment for children if the size of a cluster is  $\geq 5$  cm in the first year of life or  $< 5$  cm with no signs of regression during follow-up [19]. The currently offered laparoscopic surgery instead of open surgery is considered as a safe solution in justified cases. In addition, there are many benefits of performing the procedure, such as exemption from regular check-up visits [30]. An additional advantage is the complete removal of residual renal tissue, while a conservative approach has been proven to reveal the presence of atrophic renal tissue despite the apparent total evolution in the US image [32]. Yamataka *et al.* presented a retrospective study comparing the costs of leading patients with unilateral MCDDK after using different therapeutic options. It was concluded that if the observation of the patient lasted longer than five years due to lack of regression, the costs of necessary control visits and imaging were higher than the costs of laparoscopic nephrectomy [33].

In conclusion, although nowadays routine nephrectomy in MCDDK is regarded as an inappropriate procedure, surgical intervention is always an option to consider in cases with extreme symptomatic dimensions of MCDDK.

## DISCLOSURE

The authors declare no conflict of interest.

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