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Case Study

Pediatric renovascular hypertension due to fibromuscular dysplasia presenting with refractory hypertension and status epilepticus. A case study.

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Abstract

Introduction

Renovascular hypertension is an uncommon but important cause of secondary hypertension in children and may result in severe neurological complications if diagnosis is delayed. Fibromuscular dysplasia is a recognized cause of renal artery stenosis in the pediatric population.

Case Presentation

A 5-year-old girl presented with recurrent episodes of status epilepticus and refractory hypertension despite treatment with three antihypertensive medications. Magnetic resonance angiography demonstrated left renal artery stenosis with juxta-ostial occlusion and distal recanalization. A radionuclide scan revealed a non-functioning hypoplastic left kidney contributing only 0.9% of total renal function. Balloon angioplasty was attempted but was unsuccessful. The patient subsequently underwent laparoscopic left nephrectomy. Histopathological examination demonstrated fibromuscular dysplasia involving the renal artery. The postoperative course was uneventful, with gradual improvement in blood pressure control and reduction in antihypertensive requirements during follow-up.

Conclusion

This case highlights the importance of investigating secondary causes of hypertension in children presenting with refractory hypertension and neurological manifestations. Laparoscopic nephrectomy may be an effective treatment option in selected patients with a non-functioning kidney due to renovascular disease.

Take-away Lessons

Early recognition of renovascular hypertension in children is essential to prevent complications. Comprehensive imaging and functional assessment aid diagnosis, while timely surgical intervention can achieve favorable outcomes in appropriately selected patients.

Keywords: *Refractoy hypertension, status epilepticus, fibromuscular dysplasia, renovascular hypertension.*
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Introduction

Hypertension is a common chronic condition increasingly encountered in pediatric clinical practice [1]. Its global prevalence among children is rising, with reported rates ranging from 1.6% to 3.5%. In addition, approximately 2.2% to 9.4% of children have blood pressure levels above the normal range [2].

Hypertension in children is increasingly recognized as a significant clinical concern [1], with secondary causes accounting for the majority of cases, particularly in younger age groups. Among these, renovascular hypertension due to renal artery stenosis is a well-established etiology [3]. Delayed diagnosis can result in

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complications such as hypertensive encephalopathy, seizures, and end-organ damage [3,4].

The present case emphasizes a rare presentation of refractory hypertension associated with unilateral renal artery stenosis and a non-functioning kidney, successfully managed with surgical intervention.

Case Presentation

A 5-year-old girl, born at term with a low birth weight of 1.6 kg, was asymptomatic for the first two years of life. Subsequently, she developed recurrent episodes of status epilepticus, prompting further evaluation. During the course of investigations, she was found to have early-onset hypertension, which remained uncontrolled despite treatment with three antihypertensive medications.

Due to refractory hypertension, the patient was referred to a specialized cardiovascular institute for further evaluation. Magnetic resonance angiography revealed left renal artery stenosis. MR aortography showed no evidence of wall thickening, enhancement, or calcification of the thoracoabdominal aorta (Figure 1A); however, there was juxta-ostial occlusion of the left renal artery with distal recanalization (Figure 1B).

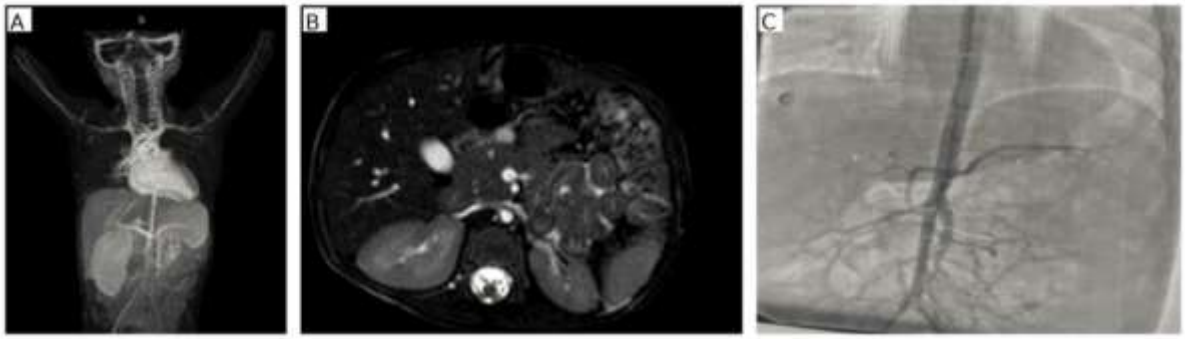

Figure 1: Radiological Evaluation of Renovascular Abnormality

(a) MR Aortogram: No wall thickening or enhancement or calcification of thoracoabdominal aorta

(b) MR Aortogram: Juxta ostial occlusion of left MRA with distal recanalisation

(c) Aortic angiogram

The patient underwent an aortic angiogram (Figure 1C), which revealed an occluded left renal artery; balloon angioplasty was attempted but was unsuccessful.

Despite optimal medical therapy, the patient's blood pressure remained persistently elevated. Ultrasonography demonstrated a hypoplastic left kidney with a normal right kidney. A DMSA scan confirmed a non-functioning left kidney contributing only 0.9% of total renal function, while the right kidney showed compensatory hypertrophy with 99.1% function.

On examination, the patient was alert, oriented, and afebrile. Her pulse rate was 90/min, and blood pressure was 130/96 mmHg. Systemic examination revealed no additional abnormalities.

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Table 1: Laboratory Investigations at Presentation

Parameter	Patient Value	Reference Range
Hemoglobin	12.5 g/dL	12–16 g/dL
Total leukocyte count	15,880/mm³	4,000–11,000/mm³
Platelet count	4.28 lakh/mm³	1.5–4.5 lakh/mm³
Blood urea	19 mg/dL	7–20 mg/dL
Serum creatinine	0.41 mg/dL	0.5–1.2 mg/dL
Serum sodium	140 mEq/L	135–145 mEq/L
Serum potassium	4.0 mEq/L	3.5–5.0 mEq/L
Serum chloride	106 mEq/L	98–106 mEq/L
Serological tests	Non-reactive	Non-reactive
Blood group	O positive	—

Note. Laboratory investigations obtained during initial evaluation.

Renal dimercaptosuccinic acid (DMSA) scan confirmed a non-functioning left kidney with compensatory hypertrophy of the right kidney.

After thorough evaluation, the patient was planned for surgical intervention. Necessary precautions were taken to optimize blood pressure, and the high-risk nature of the procedure was explained to the parents. The patient subsequently underwent left laparoscopic nephrectomy. Intraoperative findings were consistent with a hypoplastic, poorly functioning kidney (figure 2A&B).

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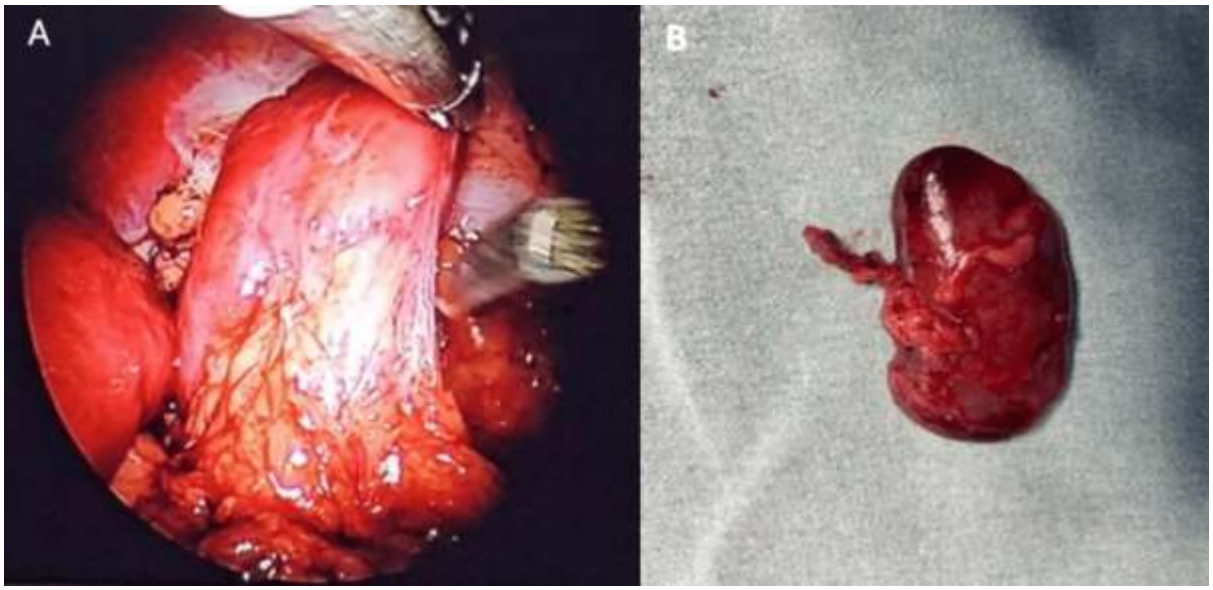

Figure 2: Intra Operative and Post Operative images

(A) Intra Operative Image of laparoscopic nephrectomy

(B) Post operative Image of removed kidney

A medium-sized biopsy specimen from the left kidney was obtained in a case clinically diagnosed as left renal artery stenosis with persistent hypertension. The specimen, received as a left nephrectomy, measured 4.5 × 3.5 × 2 cm, and the cut surface showed preserved corticomedullary differentiation. The renal artery measured 1.5 cm in length. Microscopic examination revealed focally crowded glomeruli with evidence of both global and segmental glomerulosclerosis. The tubulointerstitial compartment showed tubular atrophy, interstitial fibrosis, and chronic inflammatory infiltrates composed predominantly of lymphocytes and plasma cells, with no evidence of interstitial granuloma. Examination of the vessels demonstrated adventitial fibroplasia highlighted on MTS staining, with extension into the tunica media at certain areas. No features of vasculitis were identified in the sections examined. Overall, the histopathological findings were consistent with fibromuscular dysplasia, leading to the final diagnosis of fibromuscular dysplasia of the renal artery in the left kidney (figure 3).

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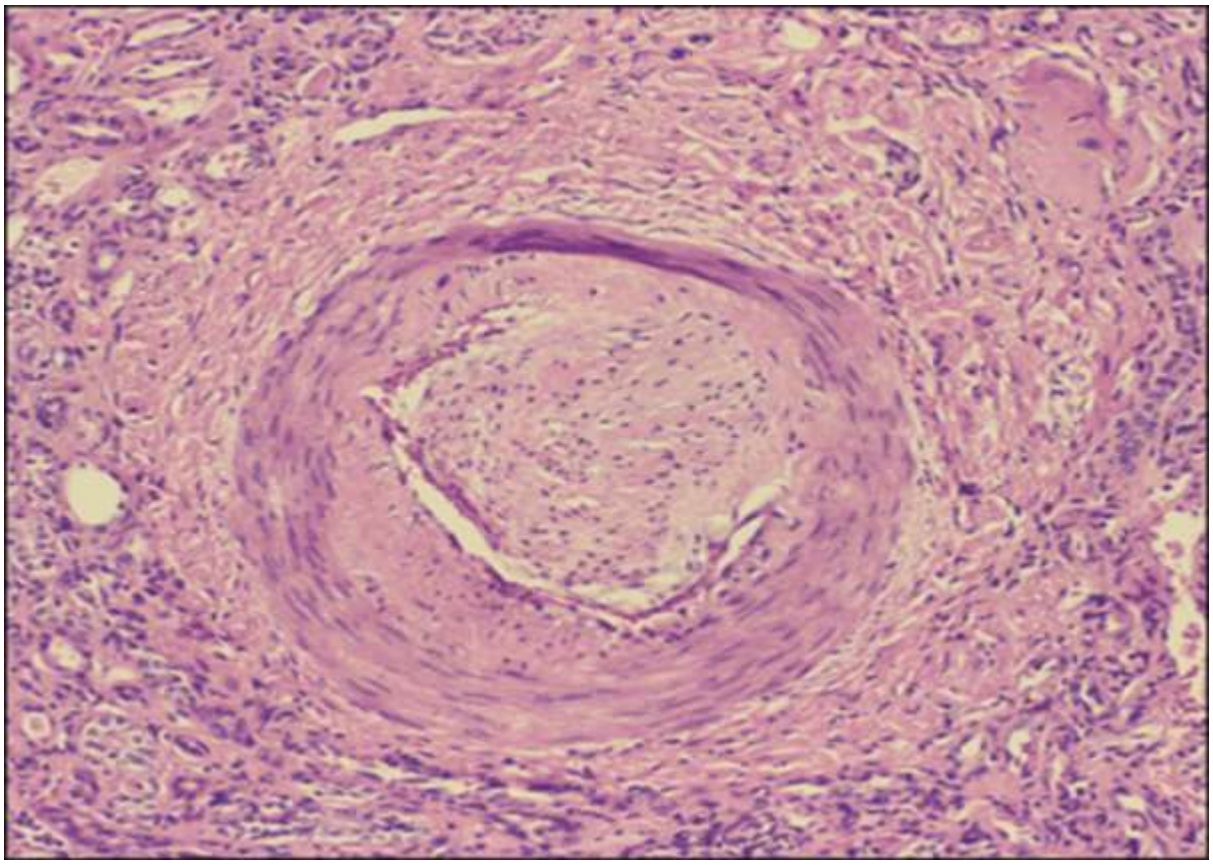

Figure 3: Histopathological picture depicting the arterial wall was composed of fibrous and muscular tissue, showing a characteristic whorled appearance, particularly within the media, along with surrounding renal parenchyma. The kidney section demonstrated a centrally located artery in which the media was expanded by eosinophilic fibrous tissue, interspersed with spindle-shaped smooth muscle cells and fibroblasts.

The postoperative period was uneventful, except for persistent hypertension in the immediate postoperative phase. The surgical wound remained healthy, and the Foley catheter was removed on postoperative day 1. No abdominal drain was required. The patient was discharged on postoperative day 4 in stable condition.

During follow-up, a gradual and consistent reduction in blood pressure was observed over time. The preoperative blood pressure was recorded at 130/96 mmHg. By postoperative day 4 (at discharge), it decreased slightly to 126/92 mmHg. At the 2-week follow-up, the blood pressure further declined to 124/88 mmHg, and by 4 weeks (1 month), it reached 120/84 mmHg. Continued improvement was noted at 6 weeks with a reading of 118/82 mmHg. At 8 weeks (2 months), the blood pressure reduced to 114/78 mmHg, and by 12 weeks (3 months), it stabilized at 110/76 mmHg, indicating a progressive normalization over the follow-up period.

Thus, on follow-up, there was gradual improvement in blood pressure control, allowing tapering of antihypertensive medications.

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Discussion

Blood pressure regulation is governed by the complex interplay between cardiac output, peripheral vascular resistance, arterial compliance, and neurohormonal mechanisms. In pediatric populations, hypertension is more often secondary in origin, particularly in younger children, and frequently attributable to renal or renovascular causes. In contrast to primary hypertension, which lacks an identifiable etiology, secondary hypertension is typically associated with underlying structural or functional abnormalities and often presents with severe or refractory clinical features. [5] In the present case, the early onset of hypertension, resistance to triple antihypertensive therapy, and associated neurological manifestations in the form of recurrent status epilepticus strongly suggested a secondary etiology, warranting detailed evaluation.

Renovascular hypertension (RVH), though uncommon in children, remains a critical and potentially reversible cause of severe hypertension. It is most commonly caused by renal artery stenosis, which leads to reduced renal perfusion and subsequent activation of the renin-angiotensin-aldosterone system, resulting in persistent elevation of blood pressure. While atherosclerosis is the predominant cause of RVH in adults, fibromuscular dysplasia is more commonly implicated in younger patients and shows a marked female preponderance, affecting approximately 80-90% of cases [6]. The present case aligns with this demographic tendency, as the patient was a young female with unilateral renal artery involvement.

The clinical presentation in this case is notable for its severity and atypical features. Unlike many reported pediatric cases where hypertension may be detected incidentally, in the present case, the child initially presented with recurrent status epilepticus, likely secondary to hypertensive encephalopathy, highlighting a neurological manifestation of severe hypertension. In contrast, the case described by Ding GG et al presented with gastrointestinal symptoms such as vomiting, along with features suggestive of fluid-electrolyte imbalance, including polydipsia and polyuria.

Imaging plays a central role in the diagnosis of renovascular hypertension, as laboratory investigations are often nonspecific. Modalities such as Doppler ultrasonography, magnetic resonance angiography (MRA), and computed tomography angiography (CTA) are commonly utilized [7]. In the present case, MRA effectively demonstrated left renal artery stenosis with juxta-ostial occlusion and distal recanalization, while MR aortography excluded large vessel vasculitis by showing no wall thickening or enhancement [8]. This is consistent with previous reports emphasizing the utility of MRA in pediatric patients due to its noninvasive nature and high diagnostic accuracy.

Functional assessment using radionuclide imaging further strengthened the diagnosis. Dimercaptosuccinic acid (DMSA) scanning revealed a non-functioning left kidney contributing only 0.9% of total renal function, with compensatory hypertrophy of the contralateral kidney. DMSA scanning is particularly advantageous in pediatric populations as it does not require bowel preparation, has minimal radiation exposure, and avoids the use of contrast agents, thereby eliminating the risk of hypersensitivity reactions. These features make it a preferred modality for evaluating renal cortical function in children.

Management of renovascular hypertension includes medical therapy, revascularization procedures such as percutaneous transluminal renal angioplasty (PTRA), and surgical interventions. Percutaneous transluminal angioplasty is generally the first-line intervention, although its success is limited in cases of complete occlusion. The choice of treatment depends on factors such as the extent of vascular involvement, renal function, and response to pharmacological therapy. In patients with a non-functioning kidney contributing to hypertension, nephrectomy is an effective therapeutic option [9]. The present case fulfilled these criteria, as the patient had a poorly functioning kidney with severe arterial stenosis and uncontrolled hypertension despite optimal medical therapy. Thus, in the present case, laparoscopic nephrectomy resulted in reduction of antihypertensive requirements. The presence of associated infrarenal aortic stenosis represents an additional contributor to hypertension and is being addressed in a planned, stepwise manner. Such an approach is consistent with current understanding that multilevel vascular disease may require sequential interventions for optimal outcomes.

Similarly, Shimoda M et al [10] described a case of a 49-year-old woman with renovascular hypertension due to renal artery stenosis who underwent laparoscopic nephrectomy after failed medical management, resulting in improved blood pressure control and reduced dependence on antihypertensive medications. Although

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their patient was an adult with bilateral disease, the therapeutic response parallels the present case, highlighting the effectiveness of nephrectomy in selected patients.

Comparable outcomes have been reported in the literature. In the case presented by Ding GG et al, [11] due to the severity of left renal artery stenosis, patient was not a candidate for angiographic intervention, and decision was made to proceed with left nephrectomy. Together, the case presented by Ding GG et al, [11] and the present case emphasize the heterogeneity in clinical manifestations of pediatric renovascular hypertension and the need for individualized diagnostic and therapeutic approaches. Further these cases underscore the importance of recognizing renovascular causes of pediatric hypertension and the role of nephrectomy in selected cases, the present case uniquely highlights neurological presentation and relatively preserved biochemical parameters.

The postoperative course in the present case was largely uneventful, with gradual improvement in blood pressure control and successful tapering of antihypertensive medications during follow-up. This favorable outcome underscores the importance of early diagnosis and timely intervention. However, a potential limitation of nephrectomy is the long-term risk associated with a solitary functioning kidney, necessitating careful monitoring for renal function and blood pressure control.

In comparison with previously reported cases, the present case is unique due to the combination of early-onset refractory hypertension, seizure presentation, unilateral renal artery stenosis, and a non-functioning hypoplastic kidney in a pediatric patient. It reinforces the need for high clinical suspicion of secondary causes in children presenting with severe or resistant hypertension.

Overall, this case highlights the critical role of comprehensive evaluation, including advanced imaging and functional studies, in diagnosing renovascular hypertension. It also emphasizes that timely surgical intervention, particularly laparoscopic nephrectomy in appropriately selected cases, can lead to significant clinical improvement and reduction in long-term morbidity.

Generalizability

Although this report describes a single patient, the findings may be applicable to pediatric patients presenting with severe or resistant hypertension, particularly when associated with neurological manifestations. The case emphasizes the importance of considering renovascular causes in children with refractory hypertension and supports the role of advanced imaging and functional assessment in diagnosis. While treatment decisions should be individualized, this report contributes to the growing evidence supporting nephrectomy in selected cases involving a non-functioning kidney secondary to renal artery stenosis.

Take-away Lessons

- Severe hypertension in children warrants evaluation for secondary causes.
- Seizures may be a manifestation of hypertensive encephalopathy.
- MRA and DMSA scanning are valuable diagnostic tools.
- Nephrectomy remains an effective treatment option in selected patients with a non-functioning kidney.
- Early intervention can reduce long-term morbidity.

Conclusions

This case underscores the need to consider renovascular etiologies in pediatric hypertension, particularly when resistant to medical therapy. Comprehensive evaluation using imaging and functional studies is essential for diagnosis.

When associated vascular abnormalities are present, a staged multidisciplinary approach with close follow-up is crucial for optimal outcomes. Laparoscopic nephrectomy may be an effective treatment option in selected patients with a non-functioning kidney secondary to renal artery stenosis.

Documentation of such cases contributes to improved clinical awareness and supports evidence-based management strategies.

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List of Abbreviations

CTA – Computed Tomography Angiography
DMSA – Dimercaptosuccinic Acid
FMD – Fibromuscular Dysplasia
MRA – Magnetic Resonance Angiography
MR – Magnetic Resonance
PTRA – Percutaneous Transluminal Renal Angioplasty
RAAS – Renin–Angiotensin–Aldosterone System
RVH – Renovascular Hypertension

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The authors received no financial support for the research, authorship, or publication of this study.

Conflict of Interest

The authors declare no conflicts of interest.

Data Availability

The data supporting the findings of this study are available from the corresponding author upon reasonable request. Patient-identifying information has been omitted to maintain confidentiality.

Author Contributions

Conceptualization: Gowda, Amruthraj G
Patient management: R, Vijayakumar; Dharwadkar, Sachin
Data collection: Ansari, Adeel; A, Vivek
Literature review: Gowda, Amruthraj G; R, Vijayakumar
Manuscript drafting: Gowda, Amruthraj G ; Ansari, Adeel
Critical revision and final approval : All authors

Author Biography

The authors are clinicians and researchers affiliated with tertiary care teaching institutions with interests in pediatric urology, minimally invasive surgery, renovascular diseases, and clinical research.

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