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Unilateral Combined Radial and Ulnar Dysplasia: A Rare Paediatric Case

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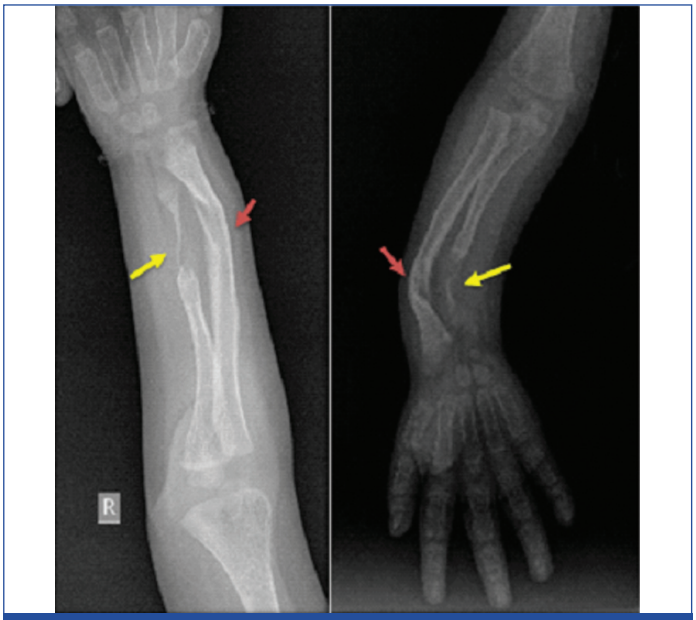
Congenital longitudinal deficiencies of the forearm involving both the radius and ulna are exceedingly rare. Early diagnosis is critical for planning surgical correction and optimising outcomes.

A two-year-old male presented with a four-month history of progressive shortening and abnormal curvature of the right forearm, as noted by his parents. There was no history of trauma, infection, or any preceding illness that could explain the deformity. The family's primary concern was cosmetic, prompting them to seek medical evaluation for possible corrective surgery. The child was born full-term via lower segment caesarean section, weighing 3 kg, and exhibited a vigorous cry at birth. Developmental milestones were appropriate for his age. There were no available antenatal ultrasound reports, and the deformity had not been detected prenatally. The child was the third in the family, with no history of congenital anomalies in his siblings or relatives. The mother's prenatal history was significant for dengue haemorrhagic fever during the first trimester, during which her platelet count dropped to 40,000/µL. She was managed symptomatically, and there were no other infections, medications, or known teratogenic exposures during pregnancy.

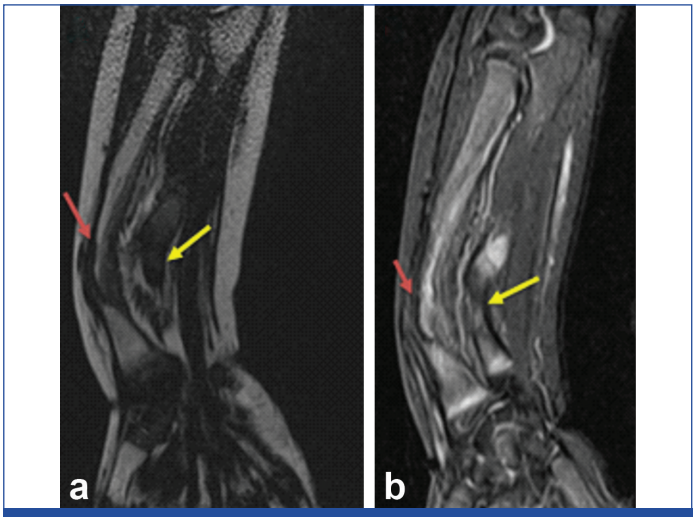
On physical examination, the right forearm appeared visibly shortened with lateral bowing. The elbow joint was clinically mobile and structurally normal. The hand was well-developed with five digits and exhibited a good grip. No neurovascular deficits were detected in the affected limb. No dysmorphic features or anomalies of other limbs were noted. Systemic examination revealed a normal spine and lower limbs, with no abnormalities elsewhere. The child demonstrated functional use of the hand, although the reach was limited due to forearm shortening. Fine motor development appeared normal, with no delays noted.

The provisional diagnosis included congenital longitudinal deficiencies involving the radius and ulna. Differential diagnoses considered included isolated radial dysplasia, isolated ulnar dysplasia, and other congenital limb deficiencies. Radiographs of the right forearm [Table/Fig-1] demonstrated significant dysplasia of the distal third of the ulna with lateral bowing and replacement by fibrous tissue, while the radius showed sharp anterolateral angulation and lateral bowing. Magnetic Resonance Imaging (MRI) [Table/Fig-2a,b] further confirmed these findings by revealing a fibrous bar extending from the distal radius to the wrist, consistent with radial dysplasia. The distal third of the ulna showed fibrous replacement medially throughout its length, indicative of ulnar longitudinal deficiency type 2. The elbow and wrist joints were preserved and structurally intact. The combined involvement of both the radius and ulna supported the final diagnosis of unilateral combined radial and ulnar dysplasia.

The patient was placed under evaluation for limb lengthening surgery, planned to be performed after sufficient development of the ossification centers of the radius and ulna. Counselling regarding the prognosis and the staged surgical correction was initiated with the family. The present case underscores the rarity of combined radial and ulnar dysplasia and highlights the importance of early imaging and genetic evaluation for accurate diagnosis and management.



[Table/Fig-1]: X-ray- Anteroposterior (AP) view of right forearm shows ulnar bowing with convexity towards the lateral side, and the distal third of the ulna shows a radiolucent defect (yellow arrow). There is also bowing of the radius, with convexity towards the lateral side, and the distal third of the radius shows a radiolucent defect (red arrow). Normal hand, five digits, and normal elbow joint were noted.



[Table/Fig-2]: a) T2-W1 coronal and b) T1-WI coronal images reveal abnormal signal intensity along the medial aspect of the radius, most prominent in the distal third (red arrows), with areas appearing iso-hypointense on T2-WI and hypointense on T1-WI, suggesting fibrous tissue or anlage. The ulna shows sharp angulation and convexity anterolaterally, with fibrous tissue replacing the bone, visible as hypointense areas on all sequences and irregular cortical margins (yellow arrows).

The maternal history of dengue haemorrhagic fever during early pregnancy raises the possibility of an intrauterine insult contributing to the developmental anomaly, although this remains speculative [1]. Previous reports have documented similar combined deficiencies, though bilateral or contralateral presentations are more common [2,3]. Surgical intervention is essential to correct the structural

deformity and length discrepancy, with timing carefully coordinated to optimise functional outcomes [4,5]. The present case adds valuable insight to the limited literature on such rare congenital upper limb anomalies and emphasises the need for multidisciplinary care and careful long-term follow-up.

Combined congenital dysplasia of both the radius and ulna in a unilateral presentation is an exceedingly rare limb anomaly, with few cases documented in the medical literature. Most congenital longitudinal deficiencies affect either the radius or ulna independently, making this combined involvement particularly challenging in diagnosis and management. Classification systems such as Bayne LG and Klug MS for radial dysplasia and Al-Qattan's et al., classification for ulnar ray deficiencies provide frameworks for isolated deformities but are less applicable for combined forms [4,5]. In present case, MRI was instrumental in delineating the extent of fibrous tissue replacing the distal radius and ulna, indicating a developmental disruption during early limb morphogenesis. Potential aetiologies include genetic mutations, vascular insults, or intrauterine infections. The history of maternal dengue haemorrhagic fever during the first trimester suggests a possible vascular insult, though this remains speculative [5].

Surgical management must be carefully timed with the child's skeletal maturation to address the forearm's structural imbalance and length discrepancy, aiming to improve function while minimising complications [2-5]. Multidisciplinary care, including orthopaedic surgeons, radiologists, and geneticists, is essential for optimal diagnosis, treatment planning, and follow-up.

In conclusion, the present case of unilateral combined radial and ulnar dysplasia in a two-year-old boy represents a rare congenital anomaly likely resulting from early developmental disruption. Detailed imaging, particularly MRI, was crucial in defining the pathology and guiding management. Surgical correction is planned following the maturation of the ossification centers, with the goal of improving limb function and cosmetic appearance. Early detection, thorough diagnostic evaluation, and multidisciplinary planning remain vital in optimising outcomes for patients with such complex congenital limb deformities. The present report contributes to the scarce literature on combined upper limb dysplasias and highlights the importance of ongoing clinical and radiological follow-up.

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