Association of capillary haemangioma with bilateral hydronephrosis in an infant

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Introduction
Capillary (or strawberry) haemangiomas are benign localized tumours of blood vessels, usually occurring in the head and neck region1. They have a malformational, hamartomatous basis and spontaneously disappear within the first few years of life2. They occur in 1-2% of all neonates and have a female: male ratio of 3:11. They typically arise early in life, grow rapidly during a proliferative phase and then slowly regress in an involutional phase1. We present a case of capillary haemangioma of face, neck and upper part of chest associated with bilateral hydronephrosis.

Case report
A 7 month old boy came to the outpatient clinic with his parents. He was conscious and playful, immunized according to age, with normal development. His systemic examination was unremarkable. His mother had developed gestational diabetes mellitus and she gave a history of swollen kidneys in the antenatal ultrasound examination of fetus. Infant had developed a reddish skin rash (which later became hypertrophied) on lower face including left parotid region, some anterior part of neck and extending to upper part of chest from the 18th day of life, which was diagnosed as a capillary haemangioma (Figures 1A and 1B).

He was treated with an oral β-blocker, propranolol 1.5 mg/kg and Timolol drops for local application on affected site. No side effects were recorded. On further evaluation, his ultrasound scan of abdomen revealed a right kidney 5.4×3.0 cm in size and a left kidney 6.1×4.1 cm in size at 4 months. Ultrasound examination did not reveal any intra-abdominal mass. Hence, to minimize radiation hazard to the infant, CT scan or MRI of the abdomen was not performed to rule out the possibility of intra-abdominal mass lesion. Later, bilateral hydronephrosis was confirmed by intravenous pyelography, which suggested left sided vesicoureteral junction (VUJ) obstruction and right sided pelvicalyceal ureteric junction (PUJ) obstruction (Figure 1C). Patient was surgically managed with ureteric reimplantation on left side and pyeloplasty was performed on the right side. The post-surgical course of infant was uneventful with normal renal function.
Discussion

Most haemangiomas are solitary; when multiple (with or without associated lesions in internal organs) or affecting a large segment of body, condition is known as multifocal angiomatosis1. Kasabach and Merritt reported a case of haemangioma involving skin and deep soft tissues of thigh in 19403. Haemangiomas may present as small isolated lesions or as large masses that cause systemic symptomatology, impair vital or sensory functions or cause disfigurement, despite their self-limited course1. Their pathogenesis and optimal management remain unknown1.

Schwalb D et al reported a case of left capillary renal haemangioma which caused massive hematuria in a 27-year-old male with left hydronephrosis and nephrolithiasis without facial involvement4. Isolated cavernous haemangioma was reported in the right kidney of a newborn, who presented with moderate abdominal distension and a large abdominal mass reaching to right iliac fossa5. Rastogi R et al reported a vesical haemangioma at VUJ in a patient with abdominal pain secondary to hydroureteronephrosis6. Our patient had bilateral hydronephrosis with capillary haemangioma involving lower face, left parotid region, anterior part of neck and upper part of chest, an unusual co-occurrence. There was no external compression observed due to any mass producing (PUJ) obstruction and VUJ) obstruction in right and left kidney respectively in this patient. Corticosteroids are the first line of treatment for infantile capillary haemangiomas. Other options include laser therapy, interferon alfa-3, vincristine and propranolol which can inhibit the growth of these haemangiomas7.

References


