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Delayed Rise of Serum Thyroid Stimulating Hormone in a Micro-preemie With Congenital Hypothyroidism

Preterm neonates are at higher risk for deranged thyroid function test (TFT). Repeated thyroid screening tests by measuring thyroid stimulating hormone (TSH) and free thyroxine (FT4) levels are recommended in preterm and very low birth weight (VLBW) neonates [1]. Delayed TSH elevation, either alone or in association with low thyroxine level are more common in preterms as compared to term babies. We managed a preterm micro-preemie with congenital hypothyroidism (CH) diagnosed at 10 weeks of age.

A third order preterm (28+4 weeks) girl with birth weight of 630 g (IUGR), was delivered by caesarean section for abnormal doppler with fetal distress. Product of non-consanguineous marriage, was a spontaneous conception with antenatal history of two previous first trimester abortions and no drug or radiation exposure. Mother was diagnosed with hypothyroidism during second trimester, remained euthyroid on oral thyroxine 25 µg/day. Anticipating preterm delivery, antenatal steroids were given. Baby was limp at birth requiring intubation in delivery room with APGAR 4/10 and 5/10 at 1 min. and 5 min, respectively. Baby had respiratory distress syndrome (RDS), sepsis with shock (received dopamine infusion for initial 3 postnatal days), hemo-dynamically significant PDA, neonatal jaundice and was managed conservatively as per NICU protocol. Screening cranial ultra-sound revealed Grade I intraventricular hemorrhage. Baby was weaned off to nasal CPAP on day 13 and to room air on day 37 of life. Trophic feeding was started on day 2 along with total parenteral nutrition (TPN), on increasing feeds baby developed feed intolerance with NEC like features on day 6 and therefore, was continued on TPN only. Feeding was restarted on day 13, gradually increased to reach full feeds (120 mL/kg/day) by day 20. Baby received four units of packed red blood cell (PRBC) transfusions during hospital stay. Pale stool with high coloured urine and yellowish discoloration of body was noticed in third week of life and liver function test suggested neonatal cholestasis. Sepsis evaluation including urine culture, metabolic screening, TFT, TORCH profile, urine CMV PCR and eye examination were done for cholestasis evaluation. She was managed conservatively for TPN induced cholestasis and it improved gradually over next ten weeks. She had fully vascularized retina and normal cranial ultrasound at 40 weeks corrected gestational age (CGA). Initial thyroid profile on day 5 of life showed FT4 - 1.1 ng/dL, TSH - 4.6 µIU/mL; on day 27 FT4 - 1.6ng/dL, TSH 9.58 µIU/mL and on day 48 FT4 - 1.2 ng/dL, TSH - 14.8 µIU/mL. Considering rising trend of TSH and multiple prematurity related illness, repeat TFT was done at 10 weeks showing FT4 - 0.2 ng/dL and TSH >100 µIU/mL. Neck ultrasound showed normal sized thyroid gland with isthmus and *X* ray bilateral knee had presence of femoral and tibial epiphyses. A diagnosis of atypical CH was made at 2 months CGA and baby was started with oral levothyroxine at 15 µg/kg and dose adjusted as per serial TFT [2]. Currently baby weighs 7.75 kg at corrected age of 9 months, in euthyroid state on levothyroxine at a dose of 16 µg/day and planned to be followed up every three monthly till three years of age.

The thyroid profile on day 5 of our baby was not absolutely normal (low FT4 and normal TSH), which could be attributed to sick euthyroid syndrome, prematurity or dopamine infusion. Subsequent rising TSH with normal FT4 at 4 and 7 weeks is suggestive of either improvement from sick euthyroid syndrome or progressive maturity of hypothalamic pituitary adrenal (HPA) axis. In sick euthyroid syndrome, neonates may have lower T3/FT3, normal or low T4/FT4 with normal TSH during stress (RDS, IUGR) and usually manifest with rising trends of TSH during recovery [3]. Few ELBW neonates develop hypothy-roxinemia with delayed TSH rise during recovery from sick euthyroid syndrome labelled as atypical CH [4]. This should be distinguished from delayed rise of TSH, which warrants starting lower dose of levothyroxine (8µg/kg/day). The dose of levothy-roxine supplementation for maintenance of normal TFT with advancing age may help to differentiate transient CH from permanent CH. In this neonate as need of levothyroxine dose is declining with age it might be a case of transient CH. So brief trial of stoppage of medication may be considered after 3 years of age.

The incidence of delayed TSH elevation is inversely related to gestational age and birth weight; neonates born at gestational age 23-24 weeks or birth weight <800 gm are at higher risk [5]. In a cohort of preterm neonates caesarean section, mechanical ventilation, PDA, pneumothorax, PRBC transfusion and some specific medications (antibiotics, dopamine, postnatal steroids) found to be risk factors of delayed TSH elevation [6]. Therefore, this case

report recommends serial repetition of TFT in ELBW neonates, particularly those having initial stormy course during hospitalisation.

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Adenomyomatosis of Gallbladder in a Neonate

Adenomyomatosis of gallbladder is defined as hypertrophy of gallbladder mucosal epithelium that invaginate into thickened muscularis propria, leading to formation of intramural diverti-cula. It is mainly seen in adults and the incidences increases with increasing age, as most of the documented cases are above 50 years of age, and many were detected in cholecystectomy specimens. Adenomyomatosis of gallbladder is very rare in the paediatric population. We herein report a case of adenomyo-matosis of gallbladder in a newborn infant.

A term infant was delivered to a primigravida mother at 37 weeks of gestation at our hospital. Post-natal transition of the baby was smooth. We noted that one of the antenatal ultrasonographic scan of the mother was suggestive of umbilical varices in the fetus. To detect any associated abnormality in the neonate due to the umbilical arices, a postnatal ultrasound was done. The scan showed a partially distended gallbladder with diffuse wall thickening and multiple tiny echogenic foci in anterior and posterior walls involving entire gallbladder showing comet tail artefacts (**Fig.1**). The imaging findings were consistent with the adenomyomatosis of gallbladder. The baby continued to do well clinically and discharged with the advice to repeat the abdominal USG later.

The exact pathophysiology of adenomyomatosis of gallbladder is not clearly known. One theory suggests that change in intra-cystic pressure because of disruption of gallbladder function may lead to proliferation of cells in the gallbladder mucosa and hyperplasia of the muscle layer. The epithelial layer can then invaginate into the muscular layer, leading to the formation of Rokitensky Ashoff Sinuses (RAS) [1]. This is also seen on histopathology, which generally shows hyperplasia of the epithelium and mucosal out pouching through the muscular layer [2].

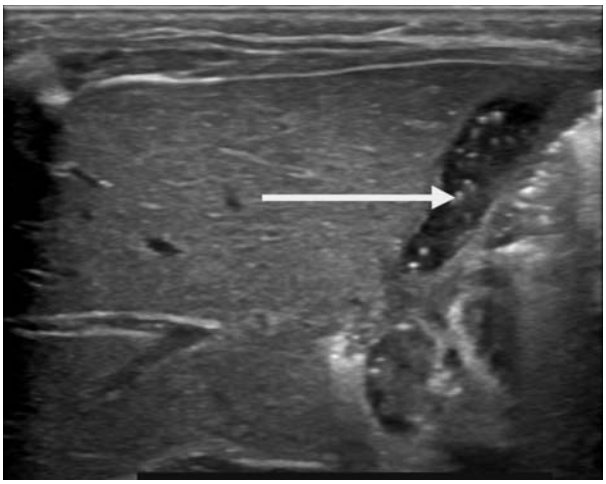

Fig. 1 Ultrasonography of abdomen showing adenomyomatosis of gall bladder.

Most of the cases in adults were diagnosed when they developed cholelithiasis or any other gall bladder condition. Although, there are some reports suggesting an association between adenomyomatosis and gallbladder cancer, there is no concrete evidence to prove this association [3].

A review of the literature showed only nine reported cases of paediatric gallbladder adenomyomatosis, with ages ranging from 1 day to 14 years. The most common symptom was abdominal pain, which was seen in 7 of the patients. Cholecystectomy was performed in seven paediatric cases [4]. One patient was a 1-day old newborn baby with suspected heterotaxy syndrome and complete atrioventricular canal. Patients generally did not have other significant comorbid conditions [5].

In children, adenomyomatosis of gall bladder remains an incidental finding on USG scan for some other reason as there are no specific clinical symptoms. Computed tomography and magnetic resonance imaging have also been used in the diagnosis of this