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Congenital alveolar proteinosis, a simple diagnosis but often missed: A case report

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This near term, 36⁺⁴ weeks, 2.8 Kg birth weight, AGA, non con-sanguinous product, male infant was admitted to Surya Child Care Neonatal Intensive Care Unit (SCH-NICU) on day 23 of life with respiratory distress since birth. Infant was born to a 32 years G2P1L1 mother. Antenatal period was uneventful. It was an LSCS delivery for non-reassuring fetal status. Baby was vigorous at birth and resuscitation was not required. However, infant was noticed to have respiratory distress soon after, for which infant was admitted to an NICU at Indore, India. At admission infant was given one dose of surfactant for suspected respiratory distress syndrome. Infant was ventilated up to day 8 of life, given one more dose of surfactant at day 8 of life, extubated and put on oxygen supplementation through nasal prongs. Infant was reintubated at day 11 of life for increasing respiratory distress. Chest X-rays showed persistent bilateral haziness. Infant was given multiple antibiotics with suspicion of congenital pneumonia. However there were no significant antenatal risk factors for sepsis and infant's sepsis work-up was unremarkable. CT chest showed patchy areas of air space consolidation bilaterally. As respiratory distress was persistent, infant was transferred on 23 day of life to Surya Child Care NICU, Mumbai for further care. On admission, infant was put under ventilation support for significant respiratory distress. Differential diagnoses was considered which included; unresolved pneumonia; congenital heart disease (TAPVC); GERD; H type tracheo-esophageal fistula; immunodeficiency; cystic fibrosis; alpha 1 antitrypsin deficiency; congenital pulmonary alveolar proteinosis; congenital lymphangiectasia; primary ciliary dyskinesia and CMV pneumonitis. Respiratory distress persisted throughout the admission. Chest X-rays performed periodically showed persistent haziness. Ventilation assistance was required throughout the admission. Serial sepsis screens and blood cultures didn't show any evidence of sepsis. CSF study, tracheal secretion cultures, CMV antibody titers and CMV DNA-PCR were all non-remarkable. Work-up for immunodeficiency including flow cytometric lymphocyte sub-set analysis was unremarkable. 2D echocardiogram of the heart, cranial ultrasound and USG of abdomen and pelvis were normal. Milk scan revealed presence of high grade (IV) gastro esophageal reflux. Repeat milk scan after fundoplication showed no evidence of GER. Infant's HRCT revealed diffuse ground glass opacities bilaterally suggestive of alveolar edema of uncertain cause with normal tracheo-bronchial tree. Work-up for cystic fibrosis (delta 508 mutation) and alpha 1 antitrypsin deficiency were unremarkable. Electron microscopy of nasal scraping for cilia morphology was unremarkable. Bronchoalveolar lavage revealed lipid laden macrophages but little PAS positive staining. Histopathological examination of the excisional lung biopsy revealed PAS positive material in alveolar spaces with preserved alveolar architecture. Electron microscopy examination was suggestive of congenital alveolar proteinosis. For financial constraints further evaluation could not be performed. Despite giving optimal supportive care, performing partial broncho-alveolar lavage (once), administering IVIG, methylprednisolone, and granulocyte colony stimulating factor, no significant response was observed. On request, infant was transferred back to Indore and died peacefully in an NICU after 3 days of transfer.

Biography

Shagufta Yousuf is working as Assistant Professor at Adesh University, India. After obtaining her Bachelor's degree in 2007, she obtained her Post-graduate Diploma in Maternal Child Health from IGNOU in 2011 and MD in OBG from University of Kashmir in 2016. In the same year she joined Adesh University as Assistant Professor, OBG. She has several publications in national and international journals. Recently she has been invited to deliver a speech at International Congress of Gynecology and Obstetrics, Taiyuan, China (Nov 2017)

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