CYSTIC HYGROMA A RARE CAUSE FOR OBSTRUCTED LABOUR AND A UROLOGICAL CHALLENGE.

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Introduction

Cystic hygroma is a benign congenital malformation of the lymphatic system that has its genesis in the lack of development of communication between the lymphatic and venous systems. The cyst may be unilocular or multilocular and its incidence is approximately 1/6000 live births. 70–80% of cystic hygromas occur in the neck, usually in the posterior cervical triangle . The remainder 20–30% occurs in the axilla, superior mediastinum, chest wall, mesentery, retro-peritoneal region, pelvis and lower limbs. Cystic hygroma is known to present at birth in about 50% of the affected newborns and 90% present by age 2 years[(1)]. Prognosis of fetal cystic hygroma detected during the first trimester is poor. Outcome of pregnancy is unfavourable with increased risk of miscarriage, elective termination and serious structural abnormalities in 77.7% of cases[(2)]. Identification of the defect is based on the visualization of a cystic structure located in the occipitocervical region[(3)].

CASE SUMMARY

32 years old female, product of non-consanguineous marriage presented to hospital with unduly Enlarged abdominal girth, edema of both feet's and lower limbs with breathlessness on exertion. There was history of single live full term vaginal delivery 3years back with uneventful post-Operative period. On general physical examination there was a pulse of 92 bpm, blood pressure of 150/100 mmHg, respiratory rate of 28 breaths per minute and pitting oedema of feet, legs and sacral region. Chest and cardiovascular examination was normal. Abdominal examination showed fundal height of 38 weeks not corresponding to period of amenorrhoea, foetal lie could not be made out because of hydraamnios, foetal heart sound were present, no visible veins were on abdominal wall, no other intra-abdominal swelling was palpable. vaginal examination revealed soft cervix with Os closed, presenting part of foetus could not be made out, pelvis was gynaecoid. Investigations showed haemoglobin of 12.4grams%, total leucocyte count of 6.5×10^3 /mm^3, blood urea of 48mg/dl, serum creatinine of 3.2mg/dl, blood sugar of 90 mg/dl, serum electrolyte sodium and potassium was 145meq/l and 4.5 meq/l respectively. Ultasonography abdomen showed 36 weeks pregnancy with fetal hydrocephalous and large hydrammnios. Both kidneys of mother showed grade second hydronephrosis with maintained corticomedullary junction.

Biophysical profile showed; Fetal heart rate score of 2, Fetal breathing movements score of 2, Fetal activity score 2,Fetal muscle tone score 2,AFI score 2. Total score was 10. No other imaging like X-rays, CT abdomen and pelvis was done as these are contraindicated although MRI could have been done but patient didn't gave consent. In view of HTN, deranged KFT, grade II hydronephrosis patient was planned for bilateral ureteric stenting.

Taking all aseptic precautions cystoscopy was done and both ureters were stented under ultrasound guidance with the cover of antibiotic of inj. Ceftriaxone and sulbectami/v stat followed by oral antibiotics .within two weeks time kidney function test normalised and Ultrasound revealed 38 weeks of gestation cephalic presentation with hydrocephalous and large hydramnios and no foetal ascites,hydronephrosis or any other congenital abnormalities . Both maternal kidneys were normal with no hydronehrosis with both ureteric stents inside.In view of above mentioned scenario termination of pregnancy was suggested for which patient gave written consent. So the patient was admitted in labour room and monitored.

Labour was induced by giving enemas, intra vaginal prostaglandins were inserted for induction of labour. Patient was watched for any undesirable side effect , after two hours examination was repeated revealing distant fetal heart sound with initiation of labour pains and PV revealing soft cervix Os admitting one finger, membranes present, presenting part not made out and pelvis was adequate. After this one unit of oxytocin was started in 5% of dextrose for augmentation resulting in intensification of labour and patient complaint of decreased fetal movements. After two hours of post induction period of labour patient revealed pulse of 100/m, B.P 120/80mmhg, presenting part could not be made out, FHS 160/minute which slows in between on Doppler , Pervaginal examination revealed Os 8cms, cervical rim felt,

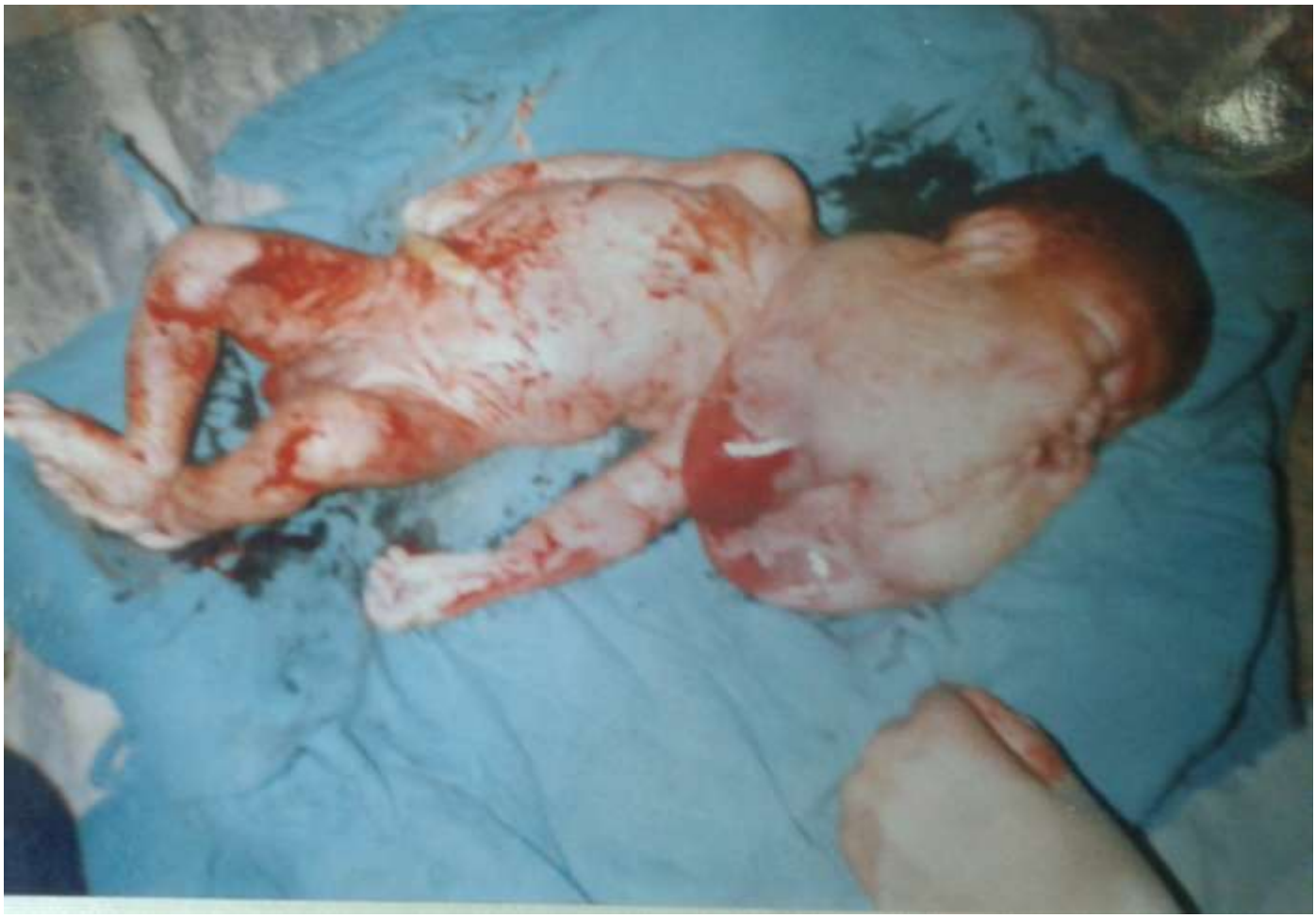
Membranes not ruptured, features of hydrocephalous present, meconium tinge was present. patient started getting exhausted with complaints of decreased fetal movements. Per abdomen examination done revealing no fetal heart sound with ultrasound didn't show any FHS and diagnosis of IUD was made. Patient was shifted to operation and was given epidural anaesthesia. Right mediolaterial episiotomy was done in lithotomy position under all aseptic precautions. per vaginal examination was done again revealing cervix fully effaced fully dilated Os, soft cystic swelling felt per vaginally and part of cranium also felt. Anterior fontanellle was punctured under ultrasound guidance resulting in drainage of very little CSF, which didn't helped in termination of pregnancy and so vacuum assisted delivery was suggested. After application of vacuum cup to scalp of foetus taking care of surrounding structures around the cup and patient was encouraged to bear down, sustained vacuum pull with assistant help in supporting perineum , a full term dead male baby was delivered.

Examination of fetus revealed huge cystic swelling occupying whole of neck. there was no fetal hydrocephalus or any other visible abnormality present. Biopsy of swelling revealed cystic hygroma .Autopsy of the foetus was not allowed by the parents. Patient was observed overnight and was discharged after two days in healthy condition and was advised to report back for removal JJ stent after two weeks. Follow up of patients after two weeks showed normal kidney function and no hydronephrosis on ultrasound and hence ureteric stents were removed.

Discussion

Lymphangiomas are benign hamartomatous tumours which occur in head and neck in75% of cases. Rare cases have been reported where these lesions have been a cause for obstructed labour [8]. A baby with prenatally diagnosed cystic hygroma should be delivered in a major centre equipped to deal with

neonatal complications. An obstetrician usually decides the method of delivery [9]. In this case a rare congenital anomaly, cystic hygroma neck was associated with polyhydroamnios and caused a bilateral maternal hydronephrosis and obstructed labour. Hence cystic hygroma with polyhydroamnios should be considered as a cause although very rare for maternal bilateral hydronephrosis.



Fetus with Large Cystic Hygroma

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