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A bolt from the blue: A rare case of agenesis of corpus callosum with colpocephaly in a 32-weeks pregnant woman presenting as eclampsia

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Abstract
Agenesis of Corpus Callosum (ACC) is a developmental anomaly that occurs as an isolated abnormality of the central nervous system or as a component of syndromes with other structural abnormalities. This report briefs about a 26-year-old G2P1L1 with 32 weeks of gestation admitted at the institution with complaint of having continuous seizures at home for about 20 minutes. Patient was taken up for Emergency LSCS under general anaesthesia in view of eclamptic seizures with an unfavourable cervix and delivered a female premature baby of weight 1.4 kg. This case is reported since, it presented as a diagnostic dilemma either to be classified as an eclamptic seizure or seizures due to neuro-developmental abnormality.

Keywords: Key words-proximal tibia fracture, MIPPO, knee stiffness, wound dehiscence

Introduction
Agenesis of Corpus Callosum (ACC) is a developmental anomaly that occurs as an isolated abnormality of the central nervous system or as a component of syndromes with other structural abnormalities. Studies report a higher incidence in males in comparison to females [1]. National Organisation for disorders of Corpus Callosum, California reports the frequency of this condition to be between 0.5 and 70/10,000 of general population [1].
Isolated Corpus Callosal Agenesis in adults may present most commonly with an episode of seizure. Children with ACC may have neuro-developmental delay.

Case report
Mrs X, a 26-year-old G2P1L1 with 32 weeks of gestation was referred from a nearby sub-district hospital in an intubated state. She was admitted there with complaint of having continuous seizures at home for about 20 minutes as witnessed by her family members. At the sub-district hospital patient was intubated in view of continuous seizure episodes. She was a booked case and had been on regular antenatal visits without any comorbidities or any history of elevated BP recordings.
On admission, patient was on mechanical ventilation with GCS of E1VTM1, BP 150/90mmHg, PR:148/mt, RR:16/mt, SpO2 99% on mechanical ventilation, Temperature 98.6 F. On clinical examination, patient had mild pallor, no pedal edema, uterus uniformly distended to 32 weeks with FHR 178bpm. Per vaginal examination revealed a tubular cervix with closed os without any bleeding/draining.
Emergency blood investigations were carried out with the results tabulated in table 1. Fundus examination had no evidence of hypertensive retinopathy. The ECHO study revealed normal findings. As the facilities for MRI was not available at our centre, and immediate decision had to be taken to prevent maternal morbidity and mortality, CT was done with abdominal lead shield, which revealed Corpus Callosal agenesis with Colpocephaly.
Patient was treated with IV Levetiracetam 1gm, Inj Dexamethasone 6 mg IM, Inj Labetalol 20 mg IV Stat and IV antibiotics. Patient was taken up for Emergency LSCS under general anesthesia in view of eclamptic seizures with an unfavorable cervix and delivered a female premature baby of weight 1.4 kg. Intraoperatively patient had a BP drop to 80/40 mmHg and started on Inj Noradrenaline infusion at the rate of 8 drops per minute and 2 units of PRBC

transfused. Patient was transferred to the intensive care unit for post op monitoring. Postoperatively patient was extubated on day 1, nor adrenaline drip tapered and stopped. Patient was stepped down to postpartum ward on post operative day 3 and was put on tab Levetiracetam from post operative day 5.

Clinical history elicited normal psychosocial development. Patient was left-handed. Her previous pregnancy was uneventful. The NT-NB scan done at 11 weeks and 6 days for the delivered baby found no defects. The anomaly scan done at 20 weeks reported no anomalies.

Physical examination done postoperatively for ruling out any syndromic association with agenesis of corpus callosum like facial dysmorphism/ bitemporal narrowing/ low anterior hairline, which were absent other than hypertelorism. Optic examination revealed no abnormality. Hearing tests like Pure tone audiometry revealed near normal hearing. Motor strength was normal. Coordination was intact.

Patient was discharged on 10th post op day with oral antiepileptic (Leviteracetam 500 mg) and advised follow-up after 3 weeks, and review at neurology OPD every 3-6 months.

Table 1: Results of Blood Investigations

Sl. No	Tests	Results
1	Hemoglobin	8.6g/dl
2	White blood cells	22,700cells/mm3
3	Platelets	2.13L/ mm3
4	Serum creatinine	0.7mg/dl
5	Blood urea	16mg/dl
6	AST	60IU/L
7	ALT	79IU/L
8	Alkaline phosphatase	119IU/L
9	Total bilirubin	0.9mg/dl
10	Direct bilirubin	0.2mg/dl
11	Random blood sugar	60mg/dl
12	PT /INR	15sec/1.08
13	Serum sodium	138mmol
14	Serum potassium	4.3mmol
15	Urine albumin sugar deposits	Nil Nil 1-2 pus cells

ECHO: Normal study.

EEG: Intermittent sharp waves seen suggestive of seizure activity.

CT brain plain: Corpus Callosum - not visualised. Bilateral occipital horn of lateral ventricle appears prominent and pointed. Impression: Complete Agenesis of Corpus Callosum with Colpocephaly.

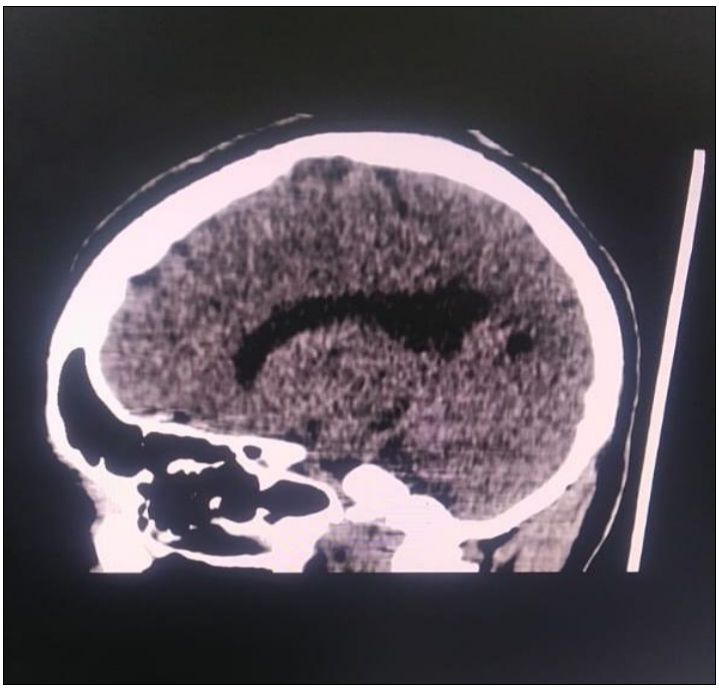

Fig 1: CT brain plain -sagital image showing agensis Corpus Callosum.

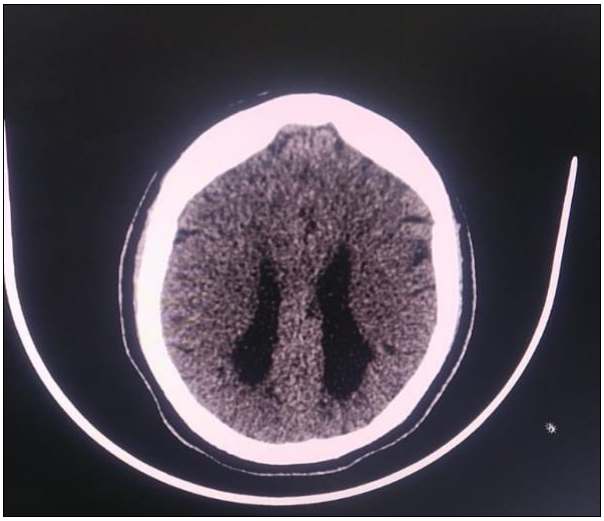

Fig 2: CT- brain plain- coronal image.

Discussion

Corpus Callosum is a thick bundle of nerve fibres that connects the two cerebral hemispheres allowing them to communicate [1]. Agenesis of Corpus Callosum is a cerebral developmental anomaly that may be classified as partial or complete based on the missing segments of Corpus Callosum. The potential cause may be genetic, infection, vascular or toxin (alcohol), etc.

Most of the cases of agenesis of Corpus Callosum with Colpocephaly may be diagnosed antenatally as a finding in ultrasonograpy done for anomaly screening [2]. In childhood they may present with developmental delay or cognitive impairment. Adults with isolated Agenesis of Corpus Callosum may present with seizures.

CCA may present as an isolated entity or in association with other central nervous system abnormalities. Syndromes that include this agenesis as an associated feature are Apert syndrome/ basal cell nevus syndrome/ Joubert syndrome/ Lyon syndrome/ Aicardi syndrome and X linked aqueduct stenosis [2]. Studies quote Chromosomal abnormalities like trisomy 18 and 13 to be associated with CCA in about 20% of cases [2].

The clinical manifestations range from asymptomatic to severe neuro-developmental delay, the degree of which depends on the part of Corpus Callosum that has not been developed. Cases may present with subnormal intelligence, visual problems, speech impairment, seizures, socio behavioral problems, problem solving skills, etc. [3]. People who are asymptomatic or only with mild neurological problems like in this case report, may be able to lead a normal daily life.

Evaluation includes MRI which is the investigation of choice. The following are the signs described in Agenesis of Corpus Callosum: racing car sign, moose head appearance, sunray appearance, colpocephaly (dilatation of the trigonal and occipital horns of the lateral ventricle) [3].

There is a paucity of literature suggesting the pregnancy and obstetric management in patients with agenesis of corpus callosum. Zamurovic & Andjelic, in their study, reported the absence of specific medical treatments for callosal disorders but suggested the beneficial effects of a multi-disciplinary approach for additional support and services [4]. Treatments are mainly symptomatic and supportive with antiepileptic medication when needed. Special education, psychological and psychiatric therapy, genetic counselling, visual/ speech rehabilitation as deemed appropriate may be advised [5].

Conclusion

This case is reported since it was presented as a diagnostic dilemma either to be classified as an eclamptic seizure or seizures due to neuro-developmental abnormality. In order to rule out the possibilities of other differential diagnoses of seizures during pregnancy, a CT scan was performed identifying the presence of complete agenesis of corpus callosum with colpocephaly. The possibility of other causes of seizures in pregnancy should be considered in clinical settings for the better management of the patient.

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