Case Report

COLPOCEPHALY WITH SEIZURE DISORDER IN AN ADULT- REPORT OF A RARE CASE

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ABSTRACT

Colpocephaly is a congenital abnormality in the ventricular system of the brain. It is a rare anatomic finding in the brain characterized by occipital horns that are disproportionately enlarged compared to other parts of the lateral ventricles. The radiological diagnosis is usually made in the perinatal period and early childhood. The patient usually suffers from mental retardation, seizures and delayed motor milestones in early childhood and presents other congenital anomalies like meningomyelocele. Adult cases of colpocephaly have rarely been reported. This case report describes a case of a 21 year old man who presented with epileptic seizures occurring for the last three years. Nothing significant was found on the physical and mental state examination. His EEG was unremarkable. His CT Scan revealed Colpocephaly with dysgenetic corpus callosum with intracerebral lipoma. The patient was put on antiepileptics and his epileptic seizures resolved.

Keywords: colpocephaly, congenital.

INTRODUCTION

Colpocephaly represents a congenital disproportionate enlargement of the occipital horns of the lateral ventricles of the brain, often associated with agenesis/dysgenesis of the corpus callosum.[1] It may be misdiagnosed as normal pressure hydrocephalus. It is a rare nonspecific finding and is usually associated with Chiari malformation and microcephaly.

A review of the literature shows that various aetiologies may be associated with colpocephaly, including intrauterine infection, intrauterine growth retardation, perinatal anoxic ischaemic encephalopathy, maternal drug ingestion (corticosteroids, salbutamol, theophylline). There is evidence of genetic transmission, either X-linked or autosomal dominant.[2]

Another possible explanation for the development of disproportionately enlarged occipital horns of the lateral ventricles is periventricular leukomalacia due to the destruction of the white matter of the occipital lobe.[3,4] We present a case of colpocephaly with dysgenetic corpus callosum in a patient with seizures manifestation.

CASE REPORT

A 21-year-old man with a chief complaint of periodic abnormal behaviour for the last three years came to the psychiatry outpatient department for evaluation. Three years back, he had sudden attacks of abnormal behavior characterized by walking briskly in circles for about 2-3 mins, followed by standing still with wide open eyes and appearing confused. On being called by the attendants, he did not respond and became all right after half an hour. He reported no recollection of the episode. He remained asymptomatic for another year. Over the next two years, the frequency of these episodes had increased to 2-3 per month. When the patient presented to the

Access the article online:
https://kjponline.com/index.php/kjp/article/view/304
DOI: https://doi.org/10.30834/KJP.35.1.2022.304
Received: 02/12/2021. Accepted: 27/02/2022.
Web publication: 03/03/2022

QR Code

Please cite the article as: Arora P, Mahesh K. Colpocephaly with seizure disorder in an adult- report of a rare case. Kerala Journal of Psychiatry. 2022; 35(1): 68-69

outpatient department, he was experiencing similar kinds of episodes. However, there was no tonic clonic movements of any part of the body, no nystagmus, no loss of consciousness, no frothing from the mouth, and no bladder or bowel incontinence. No episodes at night time or in sleep were reported. The patient reported no history of trauma, substance abuse, or hallucinations during or after the episode. There was no history of suspiciousness, manic or depressive episode, any apparent stressor of any kind, any chronic medical illness or long term medication use. The patient's past history was non-contributory. No family history of a similar illness was reported.

In personal history, developmental milestones were normal, and he was educated up to the 12th standard. He was good in his studies but left school due to poor socio-economic status. His physical examination did not reveal any abnormality. Mental state examination did not reveal any abnormality. His MMSE score was 29/30.

Baseline investigations were normal. No papilloedema was seen on fundoscopy, and Intraocular pressure was not raised. CT Scan revealed dysgenetic corpus callosum with colpocephaly and midline intracerebral lipoma. EEG did not reveal any interictal abnormality. A neurology and neurosurgical opinion was sought and suggested conservative management. So, the patient was started on tablets of carbamazepine 300 mg ~~daily~~ twice a day. He was compliant with treatment. After a month of regular treatment, he was seen again in the outpatient department. He had experienced no further episodes of seizures. He was advised regular follow up and medication~~.~~

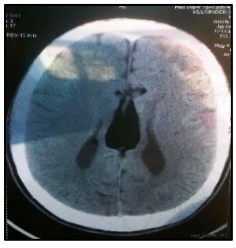

Figure 1- Brain CT scan in the axial plane showing massive dilation of the occipital horns and the lateral ventricles

DISCUSSION

Colpocephaly is an abnormal enlargement of the occipital horn of the lateral ventricle, also described as persistence of the fetal configuration of the lateral ventricles. It has been found associated with several abnormalities of the brain. Apparently, a specific form of fetal ventricular configuration persists into postnatal life. The occipital horns are disproportionately large and dilated. Colpocephaly with or without corpus callosum agenesis is a rare neuro migrational disorder during the first trimester. It mainly presents as neurological and neurodevelopmental disorders in infancy. Discovery in adulthood is remarkably uncommon. The identification of colpocephaly in adulthood with a seizure disorder is a rare and uncommon phenomenon

Reviewing the literature showed two case reports of colpocephaly presenting in previously normal adults. A thirty years old adult male presented with new onset complex partial seizures, and his brain MRI showed colpocephaly. This was reported by Wunderlich G[5] et al. and is similar to our case report. The second case was reported by Esenwa[6] et al. from Columbia University Medical Center. It was about a 60 years old lady who presented in the emergency department after a series of falls. Her non-contrast CT of the head showed massive dilation of the lateral and third ventricles, thus diagnosed with congenital colpocephaly.

Colpocephaly discovered incidentally in asymptomatic adults requires no specific treatment. If the patient is symptomatic, then treatment is based on symptoms. For seizure disorder, antiepileptics are beneficial, as in our case.

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