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Diaphragmatic Flutter in Adolescent Male: A Rare Case Report

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Abstract
Diaphragmatic flutter is an infrequent disorder characterized by high-frequency rhythmic involuntary contractions of the diaphragm and other respiratory muscles In this communication, we are reporting a rare case of diaphragmatic flutter in 20-year-old male -thoracic , abdominal and para-spinal muscles showed a myoclonic pattern of involuntary contractions in thoracic level (T4 to T12). Since there was a suspicion of diaphragmatic flutter, we evaluated movements of diaphragm with fluoroscopy. It showed a repetitive rhythmic movements of the diaphragm (112 movements per minute) and normal diaphragm mobility with respiratory cycle both in inspiration and expiration. Thus the patient was diagnosed to have diaphragmatic flutter. He was advised on carbamazepine 400 mg thrice daily with other supportive measures. Patient was symptomatically improved and discharged Later we got the opinion of cardiothoracic surgeon who suggested VATS guided Clipping/Crushing of phrenic nerve at C4 level if there was poor clinical response /recurrence.

INTRODUCTION

Case Report

History: 20-year-old male was presented to our casualty with a history of sudden onset of pain in the right side of chest and lumbar region and radiated to back with breathlessness for 10 days. His pain was associated with rhythmic involuntary movements of the trunk. He also complained of burning sensation in the right side of the face, neck and chest combined with nausea /vomiting.

The patient had previous consultations with various specialities in many hospitals. He was advised for physiotherapy and analgesics but his symptoms did not improve. He was admitted in our intensive care unit and subjected for evaluation.

Examination: On examination patient was thin-built, looked pale and afebrile. We collected the following vitals: BP-120/70mmHg, HR-86bpm, RR-19breaths/min, SPO 2 room air 96%.

Systemic Emanation:

- **RS:** Normal, vesicular breath sounds heard in equal intensity
- **CNS:** Alert, Higher mental functions –Intact, cranial nerves-unaffected
- His deep tendon reflexes graded 3+/4+, with preserved muscle strength, no evidence of Babinski sign.

During our examination, he complained of unusual burning sensation on the right side of face, neck and the chest. Involuntary rhythmic movements were also noted in right side of chest and abdomen, which were not exacerbated by touch. Psychiatrist opinion was sought and advised to rule out organic cause and found no psychiatric interventions.

Investigations: Complete blood count was normal except for iron deficiency anaemia. Liver function tests, renal function tests, serum electrolytes and thyroid function tests were advised and found within normal ranges. Anti-nuclear antibody, RA factor were found negative. ECG was normal.

Electromyography and nerve conduction studies of cervico-thoracic, abdominal and para-spinal muscles showed a myoclonic pattern of involuntary contractions in thoracic level (T4 to T12). MRI brain and spine were normal.

Since there is a suspicion of diaphragmatic flutter, we evaluated movements of diaphragm with fluoroscopy. It showed an unique repetitive rhythmic movements of the diaphragm (112 movements per minute) and normal diaphragm mobility with respiratory cycle both in inspiration and expiration.

Treatment: Thus the patient was diagnosed to have diaphragmatic flutter based on the foregoing investigations. He was started on carbamazepine 400 mg thrice daily with other supportive measures. The patient was symptomatically improved and discharged. Cardiothoracic surgeon opinion was obtained for VATS guided Clipping/Crushing of phrenic nerve at C4 level if there was any poor clinical response /recurrence.

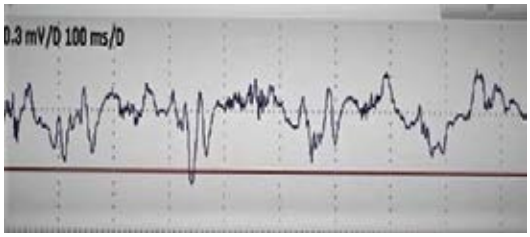

Fig 1: Right Diaphragm surface EMG showing rhythmic discharges

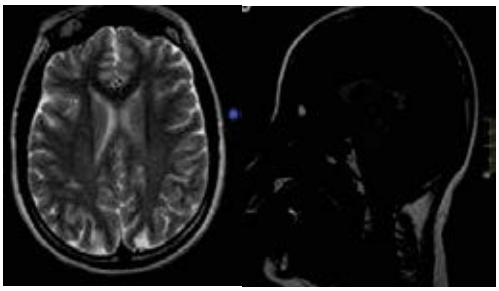

Fig. 2: MRI BRAIN

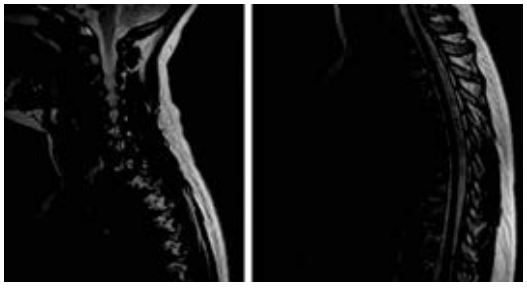

Fig. 3: MRI SPINE

Diaphragmatic flutter is an infrequent disorder characterised by high-frequency rhythmic involuntary contractions of the diaphragm and other respiratory muscles innervated by cervical nerve roots[1]. A.V. Leeuwenhoek gave his first ever description about this condition after being affected by the same. Even though his physician suggested that it was due to cardiac cause, van Leeuwenhoek realised that his heart rate did not change when symptoms appeared and thus concluded that it was the diaphragm and not the heart that caused the palpitations[2,3] Diaphragmatic flutter has also been called by various names as Leeuwenhoek disease, diaphragmatic myoclonus, respiratory myoclonus, belly dancer's syndrome and belly dancer's dyskinesia[1,4].

In the case of diaphragmatic flutter, clinical

symptoms are highly variable and the syndrome is very rare, which results in late diagnosis. It may present with pain, but pain cannot be used to locate damage since it is perceived in the thorax, epigastrium and lumbar region. It mimics Ischaemic heart disease due to the pain location if it involves left hemi diaphragm. In these cases, pain radiates to the left arm and patients experience dyspnoea.

It was described in conjunction with CNS, peripheral nerve system disorders, pleurisy, mediastinal diseases, peritonitis, rheumatic fever, post thoracic surgeries.

Diagnosis can be based on fluoroscopy to evaluate its amplitude and rate[3]. Needle electromyography is accurate in diagnosis but its more invasive which needs the needle insertion to diaphragm. Surface electro physiological studies are less accurate since thoracic movements may interfere.

Because of nonspecific and vague symptoms, diagnosis is often delayed. Patients are usually diagnosed with suspected psychiatric disorder and therefore been treated with multiple drugs including valproic acid, haloperidol, pimozide and clonidine[5]. Treatment options include carbamazepine, phenytoin, phrenic nerve block with bupivacaine/methyl prednisolone and VATS guided phrenic nerve crush/clipping[5].

Michael Chiou *et al* successfully used manual resuscitator and subsequently, mouthpiece and nasal non-invasive ventilator support (NVS) instantaneously halted the flutter for 3 months and almost for another 6 months for a case with diaphragmatic flutter. Further, they demonstrated that resting the diaphragm could give positive results in flutter episodes[6].

CONCLUSION

Thus “Diaphragmatic flutter” is an imfrequent disorder with nonspecific symptoms which needs high index of clinical suspicion for early diagnosis with help of fluoroscopy and EMG.

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