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Diaphragmatic eventration in a 15-month-old girl presented with recurrent hiccups only

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Abstract: The hiccups are involuntary, spasmodic contractions of the diaphragm and intercostal muscles that results in sudden inspiration and ends with abrupt closure of the glottis. As shown in the table below (table 1) it is either of central or peripheral reasons and even under the peripheral reasons most of the paediatrics medical references did not mentioned the diaphragmatic eventrations per say as a leading cause of hiccups. i report here a case of 15 months old girl with bouts of hiccups since birth and with further work up found to be having a diaphragmatic eventration after an incidental chest x-ray performance. After the eventration correction her hiccup disappeared completely so this raised an important hidden question for all cases of long-term hiccups in children's, shall we consider the possibility of DE in the differential diagnosis in each paediatrics hiccup presentation like what is in the adult hiccup differential diagnosis.

Keywords: Diaphragmatic eventration, Eventration correction, Recurrent hiccups.

Abbreviation:

- DH: Diaphragmatic hernia.
- DE: Diaphragmatic eventration.
- CXR: Chest x-ray.
- BA: Bronchial asthma.
- FU: Follow up.
- KAMC: King Abdulaziz medical City.
- POP: Post-operative.
- PHC: Primary health Center.
- US: Ultrasound.

1. INTRODUCTION

There are numerous causes of hiccups in children showed in (table 1) and still the gastric elements are the commonest cause in the paediatrics group.

The diaphragmatic eventration as a surgical disorder that mostly congenital in origin result from a defect of inadequate development of the diaphragmatic muscles or absence of the phrenic nerve, few cases of the diaphragmatic eventration can be acquired in origin like that result from injury of the phrenic nerve occurring during the birth trauma or the thoracic surgeries.



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(Table 1: Causes of hiccups in children):

Causes of persistent and intractable hiccups

Central nervous system disorders	
Vascular	
Ischemic/hemorrhagic stroke*, arteriovenous malformations (AVMs), temporal arteritis	
Infections	
Encephalitis*, meningitis, brain abscess, neurosyphillis, subphrenic abscess	
Structural	
Head trauma*, intracranial neoplasms, brainstem neoplasms, multiple sclerosis, syringomyelia, hydrocephalus	
Vagus and phrenic nerve irritation	
Goiter*, pharyngitis*, laryngitis, hair or foreign body irritation of tympanic membrane, neck cyst or other tumor	
Gastrointestinal disorders	
Gastric distention, gastritis, peptic ulcer disease, pancreatitis, pancreatic cancer, gastric carcinoma, abdominal abscesses disease, inflammatory bowel disease, hepatitis, aerophagia, esophagael distention, esophagitis, bowel obstruction	, gallbladder
Thoracic disorders	
Enlarged lymph nodes secondary to infection or neoplasm [®] , pneumonia, empyema, bronchitis, asthma, pleuritis, aortic a mediastinitis, mediastinal tumors, chest trauma, pulmonary embolism	neurysm,
Cardiovascular disorders	
Myocardial infarction, pericarditis	
Toxic-metabolic	
Alcohol*	
Diabetes mellitus	
Herpes zoster	
Hypocalcemia	
Hypocapnia	
Hyponatremia	
Influenza	
Malaria	
Tuberculosis	
Uremia	
Postoperative	
General anesthesia	
Intubation (stimulation of glottis)	
Neck extension (stretching phrenic nerve roots)	
Gastric distention	
Traction on viscera	
Drugs	
Alpha methyldopa	
Short-acting barbituates	
Chemotherapeutic agents (eg, carboplatin)	
Dexamethasone	
Diazepam	
Psychogenic	
Anorexia nervosa	
Conversion reaction	
Excitement	
Malingering	
Schizophrenia	
Stress	

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Though of that as you see the in preceding table and as I researched in most of the paediatrics literatures and publishments, I did not find anybody frankly impacted the "DE" as a direct cause of the hiccups in children's per say in contrast to what occurring in adults despite the seldom reported frank cases in them pointed to the "DH" as a causative agent for adults' hiccups.

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Case Presentation

A 15-month-old girl who presented to my clinic with an episode of diarrhoea, cough and mild shortness of breath since of the 'BA'' episode she is known to have and among the inspection I noticed a few bouts of hiccup between the coughs, further history taken from the mother about that bout of hiccups that was seeming not of value to the mother. That history questions directed about presence of any eventful pre-natal or peri-natal history like birth trauma or maternal or neonatal perinatal infections, head traumas, neck lumps, recurrent vomiting with abdominal distension, regurgitations, long term fever, chest traumas, contact with tuberculosis patient, also about the vaccination status which were came all with negative history from the mother side. Mother completed all prenatal clinical visits without any medical problems and mentioned normal results of all pre-natal gestation US images.

From the detailed history about this hiccup the mother mentioned it started to observe it after first attack of child's bronchial asthma at seven months old, it comes almost ten to thirteen times per day mainly at the bedtime extending almost 3-5 minutes in each, exaggerated mainly by the bronchial asthma cough, and laying supine position but no certain reliving factors.

That hiccups was not bothering the child's daily activities like interplaying or eating but sometimes interferes with her sleeping.

The mother despite of that did not seek a medical advice about that hiccups and most of the child's visit to the clinics were since of the "BA" attacks. Mother mentioned only one time among the clinical visits to their family physician in "PHC" she asked about that hiccups and he reassured her as it is something transient and will self-limit with time and advice to burp the child frequently.

Clinically the patient was entirely normal and CXR done for her showed a marked elevation of the right diaphragm mostly diaphragmatic eventration (Figure 1) with a further request from the radiologist to rule out the presence of diaphragmatic hernia (Figure 2).



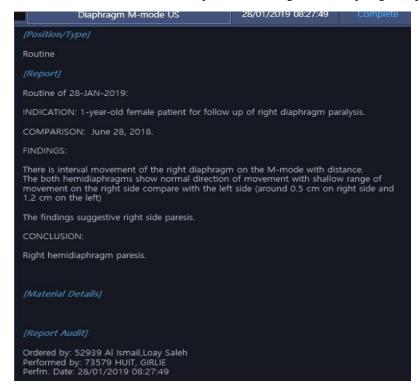
(Figure 1: The initial chest x-ray of the patient showing a right sided diaphragmatic eventration).

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Chest X-ray	27/06/2018 08:41:48	Complete
[Position/Type]		
AP, Lateral		
[Report]		
AP of 27-JUN-2018:		
Lateral of 27-JUN-2018:		
Lateral of 27-JUN-2018:		
The cardiothymic contour is normal. The lungs showed background features of reactive parenchymal abnormality seen. T	airway disease changes.	No focal
here is marked elevation of the right hemidiaphrag in keeping with diaphragmatic eventration. Howe cannot be ruled out.	gm. The radiographic feat ver, the less likely possibili	ures are more ty of hernia
Further evaluation and characterization by ultrason advised.	und of the abdomen and c	liaphragm is
[Material Details]		
[Report Audit]		
Ordered by: 30563 Aseery , Ahmed Moosa Performed by: 47106 Nabus, Noemi Perfm. Date: 27/06/2018 08:41:48		

(Figure 2: The radiology report of the initial x-ray).

Further diaphragm M-mode ultrasound done and showed a paresis on the right hemi-diaphragm (Figure 3).



(Figure 3: The radiology report of the diaphragm M-mode ultrasound).



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Review of the patient's outpatient medical records showed a regular visit to the well-baby clinic for vaccinations (without any concern about this hiccup from the parents or the physician who check the child) and one visit to my clinic with complaint of diarrhoea and cough the visit I focused on the hiccups presentation (Figure 4).

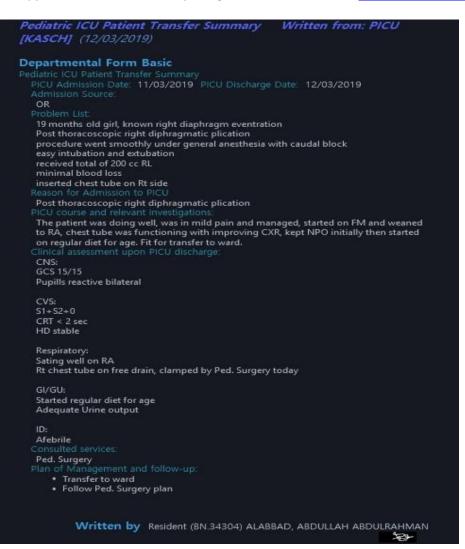
This is TT months old boy who came with:	
S)_ Diarrhea 3 days.	
_LBM six times per days,Watery,Non-bloody and no mucous or tenismus. _There is no vomitting. _The mother mentioned a history of on-off hiccups at the bed time since she was 6 months old, takes almost 10 minutes and sometimes bothers her sleepin _There is no history of trauma of chest. _There was also anorexia and poor daily actevity. _Fever only for 3 days,Abdomenal pain or history of food poisioning. _Non-eventfull perinatal history. _No chronic medical illnesses. _No clear allergies. _No surgical procedures. _No family history of any medical illnesses. _The mother is house wife.	g.
 O)_(GENERAL):Conscious alert and hydfrated .No palor, cyanosis or tingue of jau appearing with fair bodybuid. no abnormal movements(GROWTH INDEX):Within normal SD(VS):Within normal(CNS):Normal craneal nerves,Normal tone,power and reflexes with normal ga _(ENT):Normal(CHEST):Clear without added sounds thoug the air entery is diminisheed on t middle and lowrer zones of the right hemi-thorax(CVS):S1+S2+O(ABDOMEN):Soft and lax and no organomegally(GENIT-URINAR SYSTEM):Within normal(LYMPHATIC SYSTEM):Normal(LYMPHATIC SYSTEM):Shoty balbable cervical LN(SKIN):Normal(EYES):Normal. 	it.
Assessment & Plan A)_Normal child who came with GE. P)_Reassurance . _Dietery instructions. _Symptomatic treatment. _Chest x-ray. _RTC In NA appointment.	
NB:Chest x-ray showed an abnormal right lung pathology so referred to the ER.	

(Figure 4: The first presentation visits of the patient to my clinic).

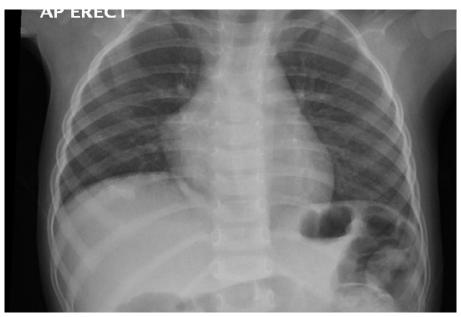
Apart from the 'CXR' and the diaphragm M-mode ultrasounds no further laboratory and radiological work up done for the patient to rule out the non-thoracic causes of this hiccup.

A paediatrics surgery consultation done, and the patient seen by paediatrics surgeon after five months and diaphragmatic repair done for the patient (Figure 5) with a subsequent clinical and radiological follow up which showed a significant "CXR" findings improvement (Figure 6) with complete absence of the hiccup in the out-patient follow up with my clinic (Figure 7).

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(Figure 5: The first day post-operative report in the PICU for the patient).



(Figure 6: The post-operative chest-x-ray done for the patient in the PICU).

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(Figure 7: The first follow-up visits of the patient post-operatively with my out-patients clinic).

The patient given a subsequent follow up appointments that came without any further complications from the surgery or reoccurrence of the symptoms.

2. DISCUSSION

The prevalence hiccup in the children's is still not well estimated (though few studies have been done in the adults only with hiccups caused by advanced cancers) and despite of that most of the paediatrics references mentioned that the most common cause of hiccups is the gastric distension whatever is the cause of that distension.

No reference or literature mentioned the direct relation between the hiccups in children with any coexisting congenital thoracic anomalies like "DE" except few literatures I have collected one of them published on the *Journal of Family Medicine and Primary Care (2014 Apr – Jun; 3(2):* 161-163) which did not pointed directly by the name of the "DE" as direct cause of hiccups on children's, but in this current special case and after focussing on the hiccup history with subsequent radiological study findings of the diaphragmatic defect and with ultimate hiccup resolution with the correction of that eventration we concluded a distinct and incised cause of the hiccups in paediatrics that nobody mentioned previously in a clear cut that there is a definite relation between the hiccup and the "DE".

So, is it rationale to do 'CXR' with every child with prolonged intermittent hiccup as a role or not? And the next related leading question why in this case specially such defect of the 'DE' not diagnosed prenatally?

3. CONCLUSION

From the cinereous of the mentioned case as a paediatrician (or even general practitioner doctor) don't turn a blind eye any long term frequent hiccups episodes in children's and don't hesitate in doing a radiological assessment for that to roll out any missed serious cardio-thoracic anomalies.



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Rationale for consent of the patient's family to publish the case:

The patient's father talked about all the rationales behind publishing the case in this medical journal and he agreed on that and signed on a paper that kept secretly in our institution's medical records to respect the patient's privacy and also for any unsuspected further medico-legal claims from the family side.

REFERENCES AND SOURCES

- [1] All attached figures taken from our main official operating computer system screens in KAMC (National Guard Hospital).
- [2] UpToDate (Wolters Kluwer) website, Prince G, Sergel M. Persistent hiccups as an atypical presenting complaint of COVID-19. Am J Emerg Med 2020; 38:1546.
- [3] The Journal of Family Medicine and Primary Care (2014 Apr Jun; 3(2): 161-163).