OPEN ACCESS

Scholars International Journal of Anatomy and Physiology

Abbreviated Key Title: Sch Int J Anat Physiol
ISSN 2616-8618 (Print) |ISSN 2617-345X (Online)
Scholars Middle East Publishers, Dubai, United Arab Emirates
Journal homepage: https://saudijournals.com

Case Report

Association of Morgagni-Larrey Hernia and Diaphragmatic Eventration: A Case Report

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DOI: https://doi.org/10.36348/sijap.2025.v08i06.003 | **Received:** 05.10.2025 | **Accepted:** 26.11.2025 | **Published:** 03.12.2025

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Abstract

Introduction: Larrey's cleft hernia is defined by the permanent or intermittent passage of part of the abdominal contents through the costo-xyphoid hiatus of the diaphragm. Its occurrence in trisomy 21 is classic, but its association with diaphragmatic eventration is rare. We report the case of a 03-year-old patient with Down syndrome who presented an anterior diaphragmatic hernia associated with diaphragmatic eventration. ***Observation:*** A.H, aged 03 with Down syndrome, was admitted to the Thoracic Surgery Department of Fann Hospital for the management of a hernia of the Larrey's cleft. On admission, she presented a sub occlusive syndrome, without vomiting or rectal bleeding, the abdomen was supple, depressible, with no palpable mass. Thoracic-abdominal-pelvic computed tomography showed a midline anterior diaphragmatic hernia (Larrey). The barium enema showed an uninhabited right iliac fossa with intra thoracic digestive gas in front of the heart, an attachment anomaly affecting the cecum, with sigmoid dollish-colon. Larrey's hernia was repaired by supra umbilical midline laparotomy. After 3 weeks, the child again presented an occlusive syndrome, and the CT scan was strongly suggestive of a recurrence of the hernia. Revision by right thoracotomy supplemented by a mini subcostal approach made it possible to rectify the diagnosis, highlighting diaphragmatic eventration, which was treated by plication. The suites were simple. ***Conclusion:*** The association of diaphragmatic eventration and hernia of Morgagni is possible and surgical treatment gives good results.

Keywords: Larrey's cleft hernia, diaphragmatic eventration, Trisomy 21, case report.

INTRODUCTION

Congenital diaphragmatic eventration (D.E.) corresponds to the ascent of a hemi-cupola consecutive to amyotrophic or muscular agenesis without solution of continuity [1]. The latter constitutes, with congenital diaphragmatic hernia (CDH), two rare pathological entities, with respective rates of 1 in 10,000 births (E.D) and 1 in 2,000 to 5,000 births (CDH) [2, 3].

Diaphragmatic eventration is one of the diagnosis major differentials of congenital diaphragmatic hernia, in most cases the correct diagnosis can only be made intraoperative [4]. However, their association has been rarely described. We report the case of a 3-year-old patient with Down syndrome who presented an anterior diaphragmatic hernia associated with diaphragmatic eventration.

OBSERVATION

Patient information:

A.H, aged 03 with Down syndrome, was admitted to the Thoracic Surgery Department of Fann Hospital for treatment of a Larrey's cleft hernia. On admission, she presented with paroxysmal abdominal pain, ceasing on the emission of stools associated with chronic constipation without vomiting or rectal bleeding. Her history included an ischemic stroke. She is the result of a well-monitored pregnancy carried to term, delivery by cesarean section, without the notion of resuscitation at birth. Her vaccination record was up to date according to the expanded vaccination plan in force.

Clinical Findings

the general examination objectified the followings: a good general state, a clear conscience, norm colored mucous membranes, a trisomic facies; there were no signs of dehydration, malnutrition, or

edema of the lower limbs; the calves were supple. Her constants were as follows: blood pressure=143/82mmHg, pulse=94bpm, Respiratory rate=20bpm, saturation=98%, temperature=37.2°C. On physical examination, the chest expansion was good, the chest was slightly bulging, the lung fields were free, the heart sounds were muffled at all foci, the abdomen, supple, depressible, without a palpable mass, the fossa right iliac was uninhabited, the hydro-aeric noises were absent. Neurologically, the patient presented with sequel right hemiparesis.

Timeline:

we then asked for a Thoracic-abdominal-pelvic Computed Tomography followed by a barium enema to confirm our diagnosis.

Diagnostic Assessment

Thoracic-abdominal-pelvic computed tomography objectified a median anterior diaphragmatic hernia (Larrey) of 102.9x27.4mm for a neck of 38.4mm, with small intestine content and exerting a mass effect on the heart, a bilateral peri-hilar alveolar syndrome (Figure 1).

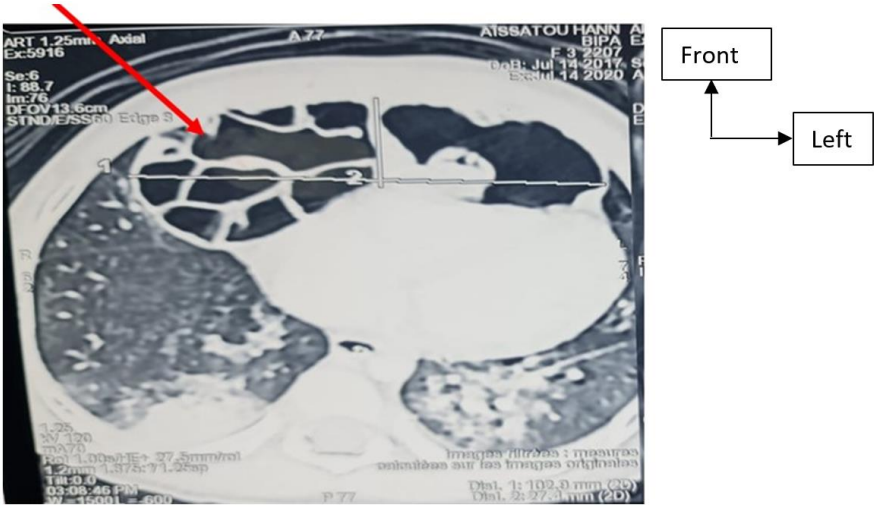

Figure 1: Median anterior diaphragmatic hernia (Red arrow) of 102.9x27.4mm with a neck of 38.4mm, with small intestine content and exerting a mass effect on the heart chambers

The barium enema showed an uninhabited right iliac fossa with intra thoracic digestive gas in front of the heart, an attachment anomaly affecting the cecum which is in an ectopic position (intra thoracic) as well as the ileo cecal junction and the last ileal loop. The sigmoid colon is abnormally long and describes several loops.

In conclusion it was therefore a hernia of the Larrey's cleft with intra thoracic cecum (due to lack of attachment or mal rotation) and sigmoid dolicho-colon (Figure 2).

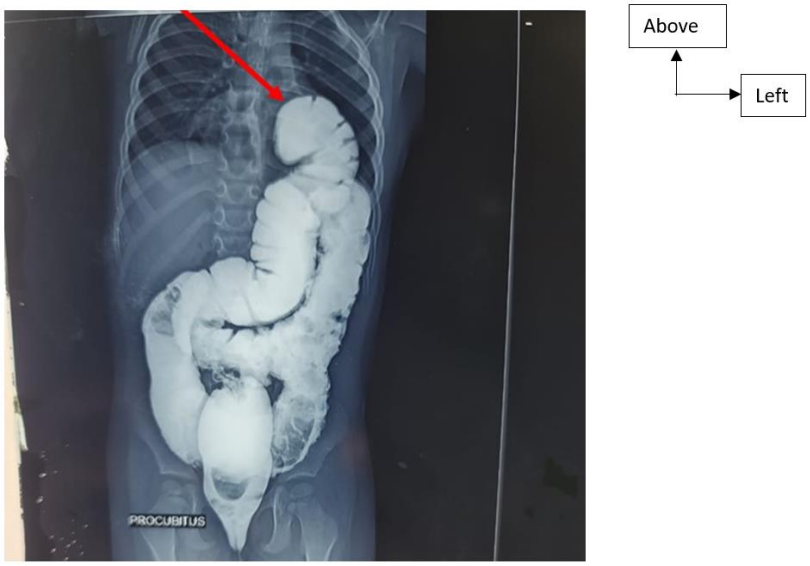

Figure 2: Larrey's cleft hernia with intra thoracic cecum (red arrow) and sigmoid dolico-colon

Therapeutic Interventions:

The surgical indication was posed. After a supra-umbilical midline laparotomy, exploration found a left anterior diaphragmatic hernial orifice, oval, approximately 3 cm in long axis, providing passage to the hernial sac containing the transverse colon and the intestinal loops which were viable (figure 3).

Figure 3: Left anterior diaphragmatic hernial orifice, oval, about 3 cm long axis (red arrow), providing passage to the hernial sac containing the transverse colon and the intestinal loops which were viable

After reintroduction of the hernial contents, the hernial orifice was closed with 5 simple stitches with polyester 0**,** after placement of a thoracic drain.

Follow-up and outcomes:

The postoperative course was simple with a clear regression of pain. The chest X-ray performed on D5 postoperatively showed persistence of the pre-cardiac air pocket with an intra-thoracic membrane. (Figure 4) We had recommended monitoring, because the patient was asymptomatic. The removal of the drain was performed on postoperative day 3, then the patient was discharging home. We had recommended monitoring, because the patient was asymptomatic.

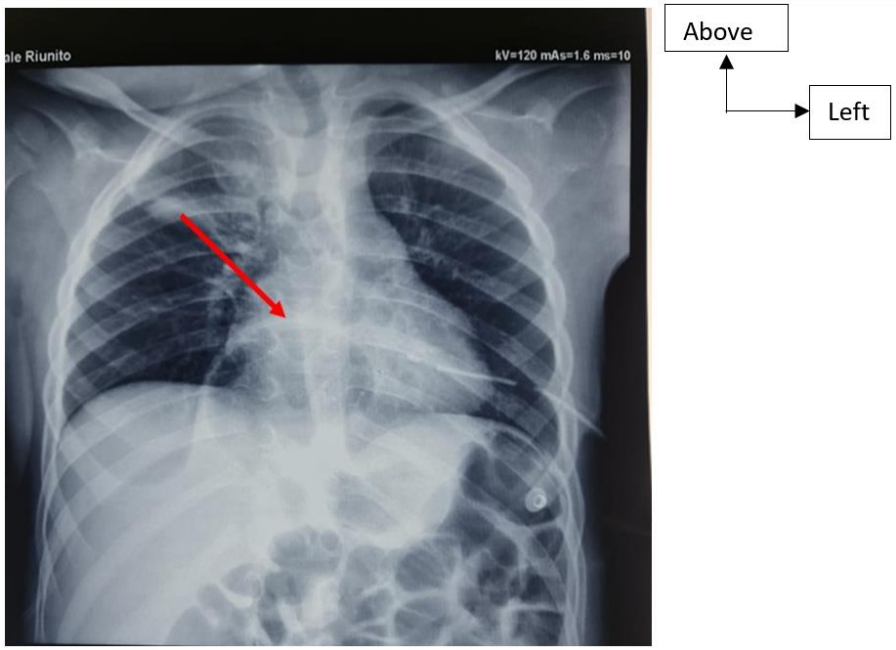

Figure 4: Persistence of the pre-cardiac air pocket with an intra-thoracic membrane (Red arrow)

She was readmitted 21 days after the intervention, for a sub-occlusive syndrome (early postprandial food vomiting with stopping of materials without stopping of gases). On examination, we noted: a nasogastric tube in place, dehydration, malnutrition, a midline laparotomy scar above the umbilical. The abdomen was supple, depressible, without meteorism or palpable mass, and silent on auscultation. Heart sounds were audible, regular without superimposed sounds; bilateral rhonchi were noted.

Computed tomography revealed a retro-xyphoid hernia with passage of part of the gastric body into the anterior mediastinum in contact with the right heart chambers. (Figure 5) This strongly suggestive picture of a recurrence of Larrey's cleft hernia required revision by left postero lateral thoracotomy. On opening, the digestive organs were covered by congestive tissue thicker than the normal diaphragmatic peritoneum and thinner than the diaphragm, on which no macroscopic structure was recognized, allowing us to differentiate the hernia and the disembowelment.

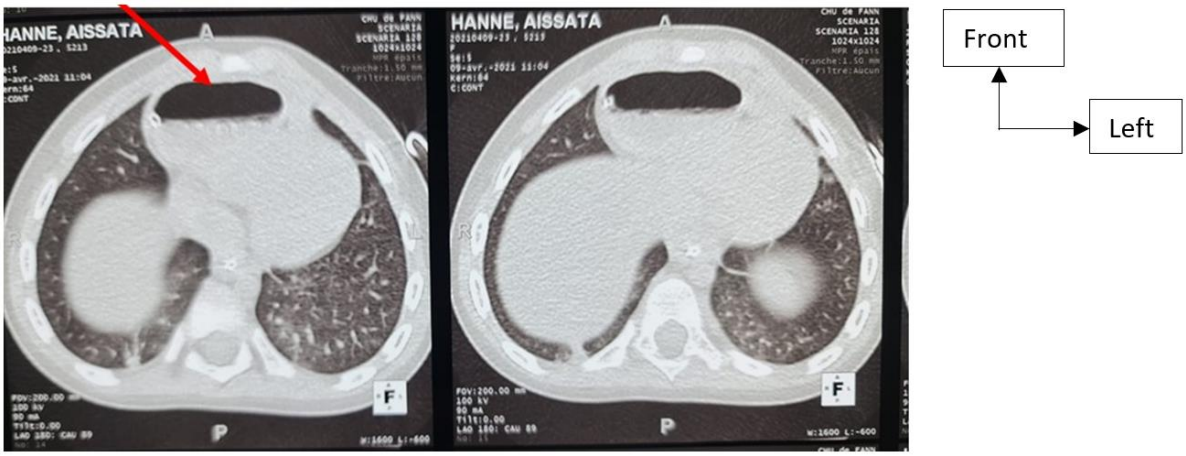

Figure 5: Retro-xyphoid hernia with passage of a part of the gastric body in the anterior mediastinum in contact with the right cardiac chambers (Red arrow)

This doubt forced us to perform a left subcostal transverse approach, which showed a sealed diaphragm, without defect with a zone of weakness opposite its left sterno-costal portion. The Larrey's cleft suture was in place and tight, the stomach in the intra-abdominal position.

Diaphragmatic eventration was affirmed and treated by plication on the left diaphragmatic dome intra thoracically with a Blalock overlock with number 0 polyester. The thorax was closed on a drain and the subcostal approach in several planes. The post-operative follow-up to this recovery was simple and the patient was executed on D5 post-operative. The chest X-ray taken on D2 post-recovery showed no elevation of the left diaphragmatic cupola with complete disappearance of the pre-cardiac air sac (figure 6).

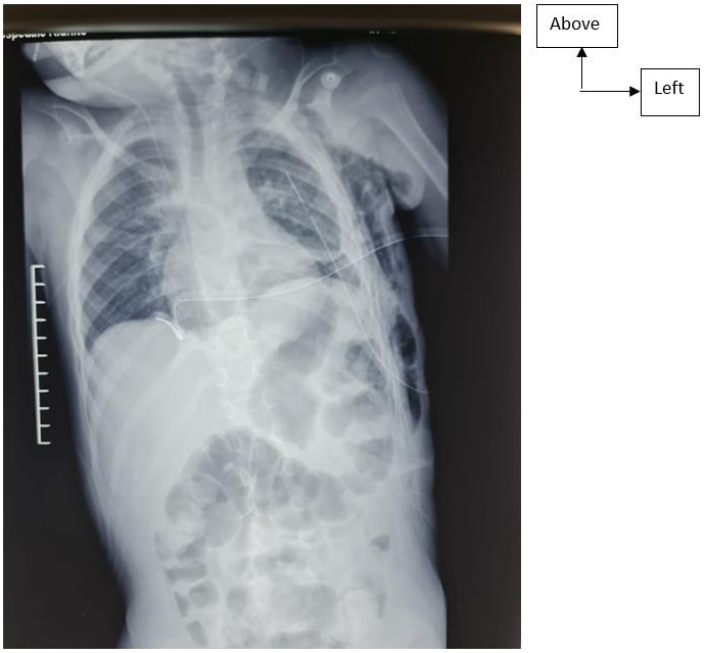

Figure 6: Complete disappearance of the pre-cardiac air pocket and the intra-thoracic membrane after diaphragmatic plication

Informed consent: The patient's parents, considered as legal guardians, gave their informed consent.

DISCUSSION

Morgagni-Larrey hernia is a rare condition, representing less than 5% of all congenital diaphragmatic hernias [3]. The diaphragm is made up of a multitude of digastric muscles, each of these muscles attaching to the periphery of the rib cage: forward on the posterior aspect of the xiphoid appendage, laterally on the medial aspect of the last six ribs and posteriorly on the antero-lateral part of the 2nd and 3rd lumbar vertebrae. Morgagni hernias (MH) occur due to a defect between the sternal and costal fibers of the diaphragm, both inserting at the tendinous center of the diaphragm. The majority (90%) of anterior diaphragmatic hernias occur on the right side, 2% on the left side and 8% are bilateral [5]. The rarity of the latter, is caused by the fixity of the pericardium to the diaphragm which is more important on the left side, which gives support and protection to this side [3].

Morgagni-Larrey hernias are usually diagnosed in adulthood and are most often asymptomatic, unlike in the pediatric population where they are often symptomatic [6]. The average age at diagnosis is 22.2 months (1-120 months) with a sex ratio of 2,5 [5]. In 71.2% of cases, Larrey cleft hernias are associated with abnormalities dominated by congenital heart disease, followed by Down syndrome, inguinal and umbilical hernias, then hydrocephalus [5]. In our patient, congenital heart disease was not found on echocardiography but she presented as an anomaly associated with Down syndrome. The hernia-trisomy 21 a association was reported by Cigdem et al [7], Kubiak et al [8], and a Saudi study involving 22 patients [9] at respective rates of 31.25%, 34.8% an 50 %.

In adults, the Morgagni-Larrey hernia is most often asymptomatic, unlike the pediatric population where the symptomatology is much more marked and varied. This can be respiratory (Broncho-pulmonary infection, respiratory distress, cough, dyspnea) or digestive (post-prandial vomiting, rectal bleeding). The discovery can also be fortuitous [4].

The chest X-ray allows the diagnosis to be made in 71% of cases. The barium enema shows part of the digestive tract intra-thoracically, more often the colon or the stomach. Thoracic computed tomography is the reference examination for the diagnosis of Morgagni-Larrey hernia, as it gives anatomical details of the hernia, the contents of the sac and the complications [10]. The most dreaded complication of MH remains the volvulus of the colon or stomach [10]. In our patient, no complications were found.

The treatment of Morgagni-Larrey hernia is always surgical, even when it is asymptomatic, because of the risk of strangulation or volvulus of the hernial contents [10,11]. The approach can be done by open surgery (laparotomy, thoracotomy, median sternotomy) or minimally invasive (thoracoscopy or laparoscopy), each approach having its advantages and disadvantages. The most commonly used approach is the midline laparotomy [10], which is the case in our patient. The latter is used when the Morgagni hernia is bilateral or complicated, in the event of a hostile abdomen or when the diagnosis of Larrey cleft hernia is uncertain.

The disadvantages are the recovery time, the complications of the operative wound and the aesthetic aspect [10]. The minimally invasive surgery approach is better, especially when it comes to a simple hernia, because it has fewer complications (5%) and has the advantages of a shorter hospital stay and recovery time. This approach presents also a faster return to activities and food, with a better aesthetic result [10].

In our patient the content of the hernial sac is the transverse colon as we have seen in a retrospective, multicenter study carried out in Iran involving 36 patients, where in 52.8% of cases, the hernial content was the omentum and the transverse colon [11]. The hernia sac can be resected or left in place. The repair of the hernia can be made by direct suturing or by prosthesis. The latter is recommended in hernias greater than 20 to 30 mm. In this Iranian study, the 36 patients underwent repair by direct suture and none of them presented a recurrence [11].

The association between Morgagni-Larrey hernia and diaphragmatic eventration has rarely been described [4]. Distinguishing congenital diaphragmatic eventration from diaphragmatic hernia before surgery can be difficult. Imaging interpreted by experienced radiologists and surgeons may miss the diagnosis of diaphragmatic eventration [4]. In our patient, this association was not seen either at laparotomy during initial surgery, or at thoracotomy. The subcostal approach complementary to the thoracotomy, combined with the palpation, made it possible to certify that it was hypo plastic diaphragm. From there we were able to cure the disembowelment. The best treatment for diaphragmatic eventration remains diaphragmatic plication [4]. As this is a very rare case, it is difficult for us to generalize our results. The association of diaphragmatic eventration and hernia of Morgagni is possible and your study confirms that the treatment of choice is a reintroduction of the hernial contents, closure of the hernial orifice and diaphragmatic plication.

Patient perspective:

The parents were satisfied with the successful outcome of the surgery despite the fact that their child has been operated twice.

CONCLUSION

Larrey's cleft hernia and diaphragmatic eventration are rare conditions, especially in the pediatric population. Their association is much rarer, difficult to diagnose, all these two entities requiring surgical treatment. Message to remember: think about the association of diaphragmatic eventration and hernia of Morgagni.

Conflicts of Interest

The authors declare no conflict of interest.

Authors' contributions

Patient care: Ba Papa Ousmane, Camara Mory, Diatta Souleymane, Diop Moussa Seck, Ndiaye Assane. Manuscript drafting: Ba Papa Ousmane, Camara Mory, Diatta Souleymane. Manuscript revision: Ndiaye Assane, Diatta Souleymane, Ba Papa Ousmane. All authors have reviewed and corrected the final version of this document.

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