CONGENITAL LUMBAR HERNIA: A CASE SERIES STUDY

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ABSTRACT

Background

Congenital lumbar hernia is a rare anomaly with only 45 cases reported in the English-language literature, usually presents since birth either isolated or, less commonly, in association with other congenital malformations.

Objectives

This study done to record the prevalence rate of this very rare congenital anomaly at Sulaimani province, with reviewing the important clinical and anatomical aspects, along with the current surgical management of this malformation.

Methods

A case series study on just four cases presented over period of 10 years (May 2007 to June 2017), data were collected regarding the age at presentation, sex, associated anomalies, surgical management, and outcome, with measuring the prevalence rate.

Results

During the study period only four patients were recorded to have congenital lumber hernia. Male to female ratio was 1:1. All of them presented during infancy, with unilateral, and right sided CLH. One female presented with recurrence after a previous surgical correction. Lumbocostvertiberal syndrome was an associated anomaly in a female patient; bilateral undescended testes in a male patient. Three cases were managed with open surgical repair; without post operative morbidity and mortality. The recurrent case's father declined further surgery.

Conclusion

CLH is a rare condition. The diagnosis is usually clinical. Due to the high frequency of associated congenital anomalies, a proper physical examination supplemented by plain X-ray and US study is important in every case. The treatment is surgical repair as soon as Possible. There is a high chance of recurrence if big defect not corrected with meshplasty.

Keywords: *Congenital Lumbar Hernia (CLH), Grynfeltt Hernia (GH), Petit Hernia (PH), Superior Lumbar Hernia (SLH), Inferior Lumbar Hernia (ILH).*

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INTRODUCTION

Congenital lumbar hernia is a rare anomaly with only 45 cases reported in the English-language literature, usually present in the first six months of infant's life, either isolated, or associated with other congenital malformations [1, 2]. The causes of congenital hernias have not been completely defined: according to Touloukian, a somatic mutation in the first weeks of embryogenesis caused by transitory anoxia leading to an alteration of muscles and aponeuroses of the lumbar region may be the underlying cause [3]. This theory is supported by the fact that lumbar hernias are associated with the lumbar-costo-vertebral syndrome in 75% of cases [3].

Like all LHs in general, CLH occurs through one of two areas of potential weakness in the posterior abdominal wall; the Superior Lumbar Triangle (SLT) of "Grynfeltt" (GH) or the Inferior Lumbar Triangle (ILT) of "Petit"(PH) [4].

The classical clinical picture is a unilateral flank mass that usually present since birth and usually carrying the features of a reducible hernia; an increase in size on crying with easy manual, and sometimes spontaneous, reduction [5,6]. Most of CLHs reported are of the GH variety and occur unilaterally although Bilateral CLH had also been reported [5-7].

The content of the hernial sac is usually small bowel, less commonly the colon, and unlike the acquired or adult variety of LH; herniation of the retroperitoneal fat, kidney and spleen are very rare events [8, 9]. In contrast with the acquired variety; obstruction, incarceration and strangulation of a CLH are rare events, although it had been reported when a CLH occurred in juxtaposition to an associated anomaly resulting in incarceration and delayed diagnosis [9, 10].This study done to record the prevalence rate of this very rare congenital anomaly at Sulaimani province, with reviewing the important clinical and anatomical aspects, along with the current surgical management of this malformation.

PATIENTS AND METHODS

A case series study on 4 infant, had been admitted in Pediatric Surgery Department/Sulaimani Teaching Hospital, over a period of 10 years from May 2007 to June 2017. Data were collected regarding the age at presentation, sex, associated anomalies, surgical management and outcome. All of the patients were diagnosed clinically with reducible swelling in lumbar region present since birth, which increased in size on crying or coughing .X-rays of the chest and lumbosacral spine were taken for all, Echocardiography done to rule out congenital heart diseases, Ultrasound of the abdomen to exclude visceral abnormality and CT scan done for one patient with small CLH to exclude other pathologies.

After routine investigations, three cases were managed with open surgical repair. Surgery was done under general anesthesia. Skin incision was done along the sac and the defect was identified. Sac was opened; contents were identified and reduced. Defect was repaired using local healthy fasciomuscular tissue. One case had large defect (about 7×7 cm), where meshplasty was done with drainage .According to European Hernia Society classification for primary abdominal wall hernias, small hernia is < 2cm, intermediate hernia ≥ 2-4cm, and large hernia ≥ 4cm. In postoperative period, patients were started orally after 1 day and discharged after day 3. Stitches were removed on day 10. In case of meshplasty, drain was removed after 72 hours. All the operated cases were doing well on follow-up with no evidence of recurrence of hernia.

RESULTS

During the study period, 4 patients presented with CLH during infancy, it was unilateral, and right sided Petit Hernial (PH) type .Male to female ratio was 1:1 (2 male and 2 female). This study record two large defects , one intermediate defect and one small defect, a 1-month old male (Fig. 1), 2-month old male (Fig. 2), 5-month-old female with recurrent CLH (Fig. 3), and the fourth patient was one year-old female (Fig. 4,5). All of the patients were diagnosed clinically with reducible swelling in lumbar region present since birth; X-rays of the chest and lumbosacral spine were abnormal in two patients, showing Lumbocostovertebral syndrome (Figure 5), bilateral undescended testes was found in one patient. The management was open surgical repair, except for the recurrent case, the father declined further surgery. No post operative recurrence or other complications were recorded. Post operative mortality was nil. The prevalence rate measured and it was 1.5 per 100,000 births.

Figure 1. Case no.1 (1 month old male patient with CLH).

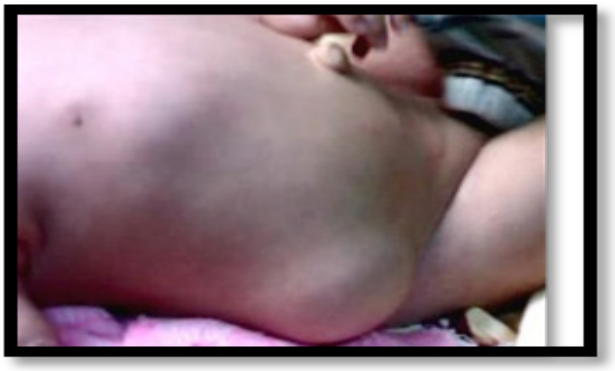


Figure 2. Case no.2 (2 month old female patient with CLH).

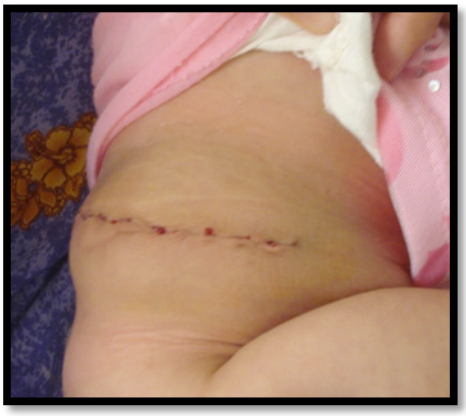


Figure 3. Case no.3 (5-month old female).

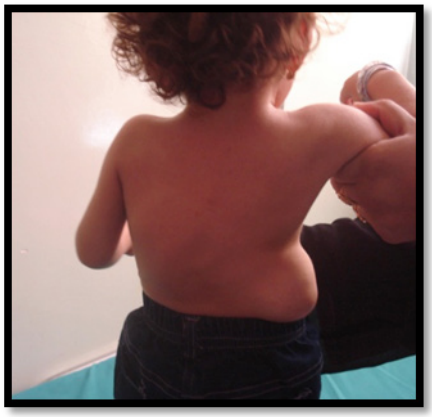


Figure 4. Case no .4 (Posterior view, shows left scoliosis).

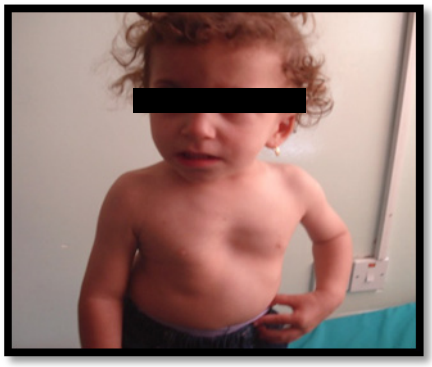
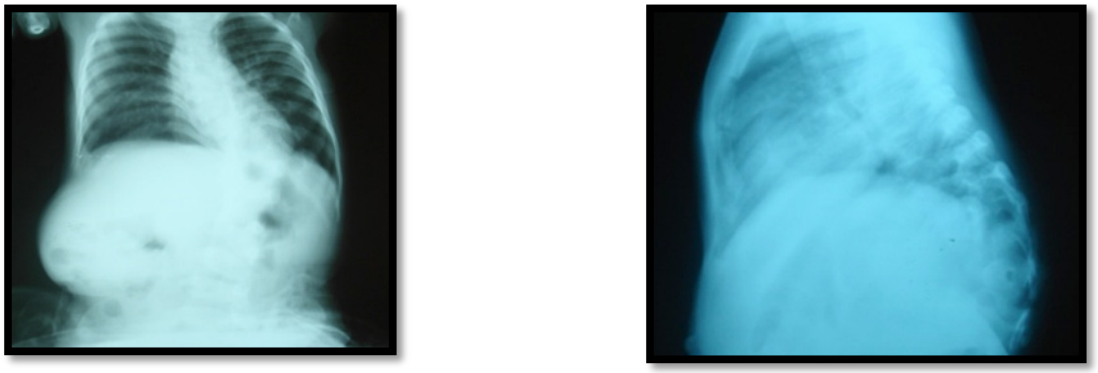


Figure 5. Case no.4 (Anterior view, shows Pectus Excavatum)



A B

Figure 6. (Case no 4). A: AP View chest and Abdomen. B: Lateral View

DISCUSSION

During this case series study that extended for ten years, four cases had been recorded, the English literatures reported that CLH is rare[1], Male to female ratio was 1:1 (2male and 2 female),close to a result from a study done in India extended for 20 years [8]. Majority of the cases were being diagnosed in the first 6 months of life and typically they presented with abnormal protrusions in the lumbar region [5], same was noticed in this study.

Most of CLHs reported are of the GH variety and occured unilaterally. Bilateral CLH had also been reported [9,10]. Unlike this study,although were unilateral but all of them with PH type.

Associated anomalies were diagnosed in all of them, unlike what found in other studies that, less than half of them were associated with other congenital anomalies [4]. Many congenital anomalies had been known to associate CLH; most commonly LCV Syndrome [11,12] ,and it was recorded in two patient out of four in this study.

Unusual association with Hydrometrocolpos, anorectal malformations, undescended testis, uretero pelvic junction obstruction, sciatic hernia, Carpus Equino Varus and bilateral renal agenesis, had been reported [13,14,15,16].Undescended testis diagnosed in one patient in this study.

The diagnosis of CLH is usually clinical in most of the cases. Complementary studies are needed to confirm the diagnosis and to reveal the absence or presence of associated anomalies [11, 12, 17], same what we did during this study.

Unlike the acquired type of LH; CT scan, MRI and barium studies are not usually indicated except when clinical diagnosis is difficult or when incarceration or obstruction is suspected [18], as in this study, it was done just for one patient.

Recent studies on cadavers revealed that the anatomy of the Superior lumber triangle and inferior lumber triangle is variable, and on the basis of the surface area, either of which had been classified in to Small (50%), intermediate (25%) and large (10%) [19, 20],in this study two of the cases had large defects , the third had intermediate and the fourth had small defect.

CLH must be treated surgically as soon as the diagnosis is made and immediate operation, after correction of any life threatening conditions, is always indicated. With a small and moderate size defects, repair with local tissue is preferable and both simple tension-free approximation of the muscle, or the use of muscular flaps, is applicable. For larger defects or for recurrent large hernia, it is preferable to use prosthetic materials like Prolein Mesh [21]. Same management approach was followed in this study, and a mesh was used in one of the cases with large defect History of recurrence was recorded in another patient with large recurrent hernia, not repaired with mesh.

Recently; laparoscopic techniques have been successful in repairing CLH using a variety of synthetic mesh [22]. It is not tried yet in Sulaimani.

The prevalence rate was 1.5 per 100,000 births, it was not measured before, but 18 patients were recorded over 20 years in a single center study done in India [8]. In conclusion, CLH is rare and the diagnosis is usually clinical. Due to the high frequency of associated congenital anomalies, a proper physical examination, supplemented by plain X-ray and US study is important in every case. The treatment is surgical repair as soon as possible, there is a chance of recurrence if a big defect not corrected with meshplasty.

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