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Hepatogonadal fusion (HGF), a rare cause of undescended testis.

7th case in the Literature

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Hepatogonadal fusion (HGF), a rare cause of undescended testis. 7th case in the Literature

BACKGROUND: *Hepatogonadal fusion (HGF). It is a rare congenital anomaly characterized by the fusion of the liver and gonads in the intrauterine period. We report the 7th hepatogonadal fusion case in the literature and its treatment.*
CASE PRESENTATION: *In the physical examination of a 4-month-old male patient who applied with the complaint of recurrent swelling in the right inguinal region, a right inguinal hernia was detected and the right testis could not be palpated. The patient underwent laparoscopy. It was observed that the right testis was in the abdomen at the level of the inner ring of the inguinal canal and adhered to the level of the liver segment 6 with a thick band. Hepatogonadal fusion was separated, then hernia repair and orchidopexy were performed. The patient was discharged on the 1st post-operative day. Both testes of the patient were palpable in the scrotum at the 6th month postoperative follow-up.*
CONCLUSIONS: *In conclusion, HGF may cause undescended testis with intra-abdominal localization. The use of laparoscopy in intrabdominal testis cases is a very accurate choice in the diagnosis and treatment of rare cases such as HGF.*

KEY WORDS: Hepatogonadal fusion, Intra-abdominal testis, Laparoscopy, Orchidopexy, Undescended testis

Introduction

Fusion of gonadal structures with internal organs is extremely rare and has been associated with the development of inguinal hernia and undescended testis [1,2]. The organs reported to be fusion with gonads in the literature are the surrenal, kidney, spleen, and liver [3-6]. The presence of surrenal remnants within the para-testicular tissue is the most common form of organogonadal fusion [7]. Splenogonadal fusion (SGF) is the second most common. Hepatogonadal fusion (HGF) is the rarest known form of organogonadal fusion. Only 6 cases of HGF have been reported in the literature. Our case will be the seventh case brought to the literature.

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Case Presentation

It was learned that the antenatal history of a 28-year-old male baby who was born by normal spontaneous vaginal delivery from the first pregnancy of his mother was uneventful, and the mother had no history of hormone use during or before pregnancy. It was learned that the patient who applied to us at the age of 4 months had a complaint of recurrent swelling in the right groin for the last month. In the physical examination of the patient, there was a reducible inguinal hernia on the right side and a silk sign on the left side. While the left testis was in place, the right testis could not be palpated in the scrotum and inguinal canal. Other physical examination findings of the case were normal. He had no additional disease. Scrotal and abdominal ultrasonography was performed on the patient. On ultrasonography, testicular tissue could not be seen in the right inguinal canal and inside the abdomen. It was decided to perform a laparoscopy on the patient.
In laparoscopy, the left inguinal canal was open and the vas deferens and cord were passing through it (Fig. 1).

Fig. 1: Left inguinal hernia on laparoscopy.

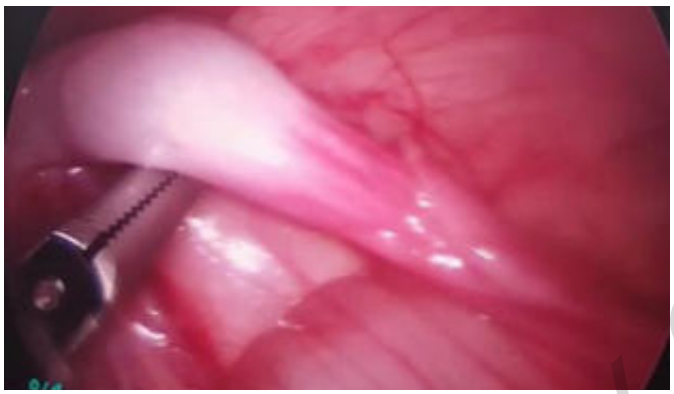

Fig. 2: Hepatic Gonadal fusion laparoscopic appearance (testicular side).

Fig. 3: Hepatic Gonadal fusion laparoscopic appearance (liver side).

"Percutaneous Internal Ring Suturing" technique left inguinal hernia repair was performed. When the exploration was continued, it was observed that the right testis was at the level of the inner ring of the inguinal canal in the abdomen and adhered to the liver at segment 6 with a thick fibrotic band (Figs. 2, 3). The tape was cut using ligasure. The testis was dissected from the surrounding tissues. It was observed that the testis was the same size as the other testis and there was no epididymal anomaly. The testis was placed in the inguinal canal and inguinal exploration was started. The testis was found by passing the layers with the right lower inguinal incision. Hernia repair was performed. The testis was placed in the dartos pouch created in the scrotum. The patient was discharged on the first postoperative day. In the postoperative third-month follow-up of the patient, it was observed that both testicles were in the scrotum and were in size and compatible with the patient's age.

Discussion

During intrauterine development, the testis is contained within the peritoneal sheaths known as the mesonephric sheaths, which ensures that the testis is in close proximity to the internal organs [8]. In 5-8 weeks of pregnancy, this closeness between mesenchymal cell clusters and testicles during organogenesis in weeks of gestation may cause SGF or HGF [9]. SGF was first described by Bostroem in 1883 and is relatively more common among these fusion anomalies [10]. The number of SGF cases brought to the literature is slightly more than 200 [11]. On the other hand, HGF is very rare and only 6 cases have been reported before [10,12-16]. SGF is 16 times more common in males and only 8 female cases have been reported [2]. All of the reported HGF cases are male patients. The incidence of SGF and HGF in girls is not clear, since female gonads are in the abdomen and fusion does not cause any problems for the patients.

SGF and HGF appear in two types continuous and discontinuous. In discontinuous fusion, there is no continuous connection between the testis and the organ, there are tissues of the relevant organ around the testis, and the testis may be in its normal position. In the continuous type, the testis is almost always intrabdominal and there are fibrous bands connecting the testis and the related organ [10]. Cases of SGF and HGF may present as testicular pain, epididymitis, and testicular mass, or are found incidentally during inguinal hernia repair. Undescended testis is the most common form of application [17].

The first case of HGF was reported by Fabio Ferro et al. in 1996 [12]. This case is a 4-month-old male patient with normal testes and nodular tissue was noticed in the upper pole of the testis during inguinal hernia repair. It was observed that this tissue extended as a thin fibrous band up to the liver. Nodular tissue and the fibrous band were excised. In his pathological examination, only nodular liver tissue was found around the testis.

The second case described was a 3.5-year-old patient who underwent laparoscopy for nonpalpable testis [13]. Unlike the first case, in this patient, atrophic testicular tissue was detected adhering to the right lobe inferior to the liver, and orchiectomy was applied to the patient.

Another case in the literature was a patient who was operated on for a 3-month-old right inguinal hernia and undescended testis [14]. During inguinal exploration, it was found that the testis was at the level of the inner inguinal

TABLE I - *The demographic and clinical characteristics of HGF cases*

		Ferro, Fabio, et al. (1996)	Lund, J.M., et al. (2001)	Fan, Rong, et al. (2012)	Al-Saied, Gamal, et al. (2016)	El-koutby, Montasser, and Mahmoud Eljalby (2018)	Durmus, Gül, et al. (2022)	Total
Age(month)		4	42	3	14	10	18	15.2±5.8
Type	Continuous		+	+	+	+	+	5/6 (83%)
	Discontinuous	+						1/6 (17%)
Surgery procedure	Laparoscopy		+			+	+	3/6 (50%)
	Inguinal incision	+		+	+			3/6 (50%)
Undescended testis	Yes		+	+	+	+	+	5/6 (83%)
	No	+						1/6 (17%)
Orchidopexy			+		+	+	3/6 (50%)	
Orchiectomy		+		+			2/6 (33%)	

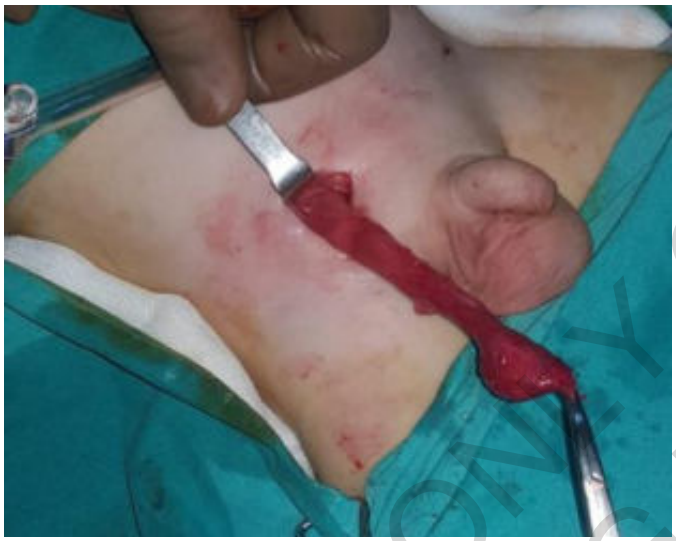

Fig. 4: View of testis after inguinal exploration and dissection.

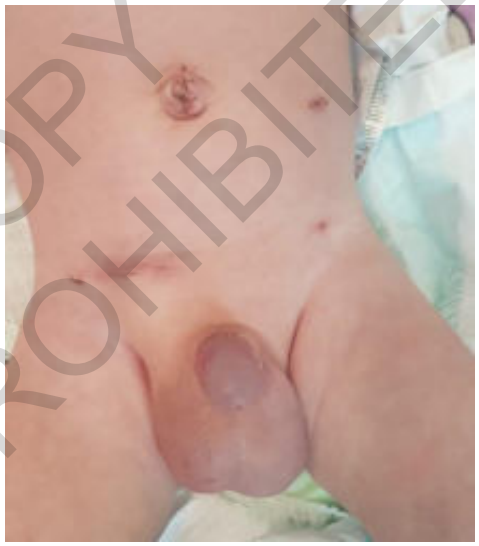

Fig. 5: Postoperative view of the patient.

ring and there was a thin fibrous band extending from the testis to the inferior of the liver. After the band was completely excised, orchidopexy was applied to the patient. Liver cells were detected in the pathological examination of the excised fibrous tissue.

Inguinal exploration was performed in the fourth case in the literature due to the right nonpalpable testis [16]. In this case, atrophic testicular tissue was seen adjacent to the liver and adhered to the inferior of the liver by a fibrous band, and an orchiectomy was performed.

In another HGF case who underwent laparoscopy for a 10-month-old nonpalpable testis, the authors observed that the right testis was attached to the liver adjacent to and inferior to the right lobe of the liver by a fibrous band, and they successfully applied orchidopexy [15].

The authors claimed that their case was the first laparoscopic HGF case in the literature, but as mentioned above, J.M. Lund et al. used laparoscopy for HGF before these [13].

The last case in the literature differed from both other cases and our case. In this case, HGF and SGF were detected simultaneously in the laparoscopy performed for bilateral nonpalpable testis [10]. Bilateral orchidopexy was performed in the case due to normal testicular development. The authors claim that they have not come across any other similar case in the literature.

Demographic characteristics and clinical data of previously published HGF cases are given in Table 1. The mean age of the cases was 15.2±5.8. One of the cases was discontinuous and the other five were continuous. It was observed that laparoscopy was performed in three cases and inguinal exploration was performed in the other three cases. While five patients had undescended testicles, one patient had intact testicles. HGF was detected incidentally while this patient was being operated on for inguinal hernia. Orchiectomy was performed in two cases due to atrophic testis, and orchidopexy was performed in three cases. In our case, as in most cases in the literature, there was a continuous type of HGF and laparoscopic orchidopexy was successfully applied to the case. No malignancy associated with HGF has yet been reported. It is not necessary to excise the fibrous struc-

tures in the paratesticular area, as there is a possibility of damage to the testis during the excision of the fibrous tissues and there is no risk of malignancy. If there is viable testicular tissue in HGF cases, orchidopexy should be applied and unnecessary orchiectomies should be avoided. Orchidectomy can only be considered for atrophic testis cases.

Conclusion

In conclusion, the use of laparoscopy in cases of intraabdominal testis facilitates the diagnosis and treatment of rare cases such as HGF. In the presence of viable testicular tissue, single-stage laparoscopic orchidopexy can be successfully applied to HGF cases.

Riassunto

FUSIONE EPATOGONADICA: È una rara anomalia congenita caratterizzata dalla fusione del fegato e delle gonadi nel periodo intrauterino. Riportiamo il 7° caso di fusione epatogonadica in letteratura e il suo trattamento.
Nell'esame obiettivo di un paziente di sesso maschile di 4 mesi che si è presentato con la denuncia di tumefazione ricorrente nella regione inguinale destra, è stata rilevata un'ernia inguinale destra e non è stato possibile palpare il testicolo destro. Il paziente è stato sottoposto a laparoscopia. Si è osservato che il testicolo destro si trovava nell'addome a livello dell'anello interno del canale inguinale ed aderiva al livello del segmento 6 del fegato con una spessa fascia. La fusione epatogonadica è stata separata, quindi sono state eseguite la riparazione dell'ernia e l'orchidopessia. Il paziente è stato dimesso in 1a giornata postoperatoria. Entrambi i testicoli del paziente erano palpabili nello scroto al sesto mese di follow-up postoperatorio.
CONCLUSIONE: Fusione epatogonadica può causare ritenzione testicolare con localizzazione intra-addominale. L'uso della laparoscopia nei casi di testicolo intraddominale è una scelta molto accurata nella diagnosi e nel trattamento di casi rari come Fusione epatogonadica.

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