Case Report

Splenogonadal Fusion Presented With Cryptorchidism

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INTRODUCTION

Splenogonadal fusion is a rare congenital condition which was first reported in 1883 by Boestrom.⁽¹⁾ Then in 1956, Putschar and Manion published a review of 30 cases and classified the condition into 2 types of continuous and discontinuous fusions.⁽²⁾ We report a case of splenogonadal fusion presented with undescended left testis.

CASE REPORT

A 2-week-old boy was referred to our hospital for evaluation of left cryptorchidism. He was a result of a full-term normal pregnancy. The infant was well developed proportional to his age. On genital examination, the right testis was in normal size and located in the scrotum, but the left testis was not palpable in the scrotum. No other physical abnormalities were detected.



Intra-operative gross photograph of the spleen attached to the left testis in a 1-year old boy with splenogonadal fusion.

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Outpatient follow-up till the age of 1 year revealed no change in his clinical picture. Thus, left orchiopexy was planned. The operation was performed through a left lower-quadrant incision. After exploration, the left testis was found in the abdominal cavity adjacent to the lower pole of the left kidney. A reddish-brown structure appeared posteriorly. The incision was laterally extended and a cord of dark-red structure was found, which was in close relation to the spermatic cord and the vessels. The structure was traced to the lower pole of the spleen and excised completely (Figure). Then, the spermatic cord vessels were ligated, and finally after 3 months, a left orchiopexy-on-vas was performed. Histopathological examination of the structure revealed normal splenic tissue.

DISCUSSION

Splenogonadal fusion is most commonly an incidental finding during groin exploration for an undescended testis or hernia. The diagnosis is rarely suspected preoperatively.⁽³⁾ It is usually present on the left side (98%) and in males (95%) with a male-female ratio of about 16:1. However, the reported incidence may not reflect its true incidence in females because of the inaccessibility of the ovary for examination.^(3,4) In a previous study, 37% of the patients with

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splenogonadal fusion underwent an unnecessary orchiectomy for suspicion of a primary testicular neoplasm. Another presentation is acute painful scrotal swelling secondary to affection of the ectopic splenic tissue by various processes.⁽⁵⁾ Falmann and Settle reported cases of splenogonadal fusion presenting with painful scrotal swelling secondary to malaria.^(6,7) Three cases were preoperatively diagnosed by ^{99m}Tcsulphur colloid liver-spleen scintigraphy.⁽⁸⁾ Many congenital abnormalities have been described to be accompanied by splenogonadal fusion, especially the continuous type, including limb defects and micrognathia.^(3,9-11)

Continuous and unconscious types of splenogonadal fusion occur in equal frequencies.^(2,3) In the continuous type, there is a direct anatomical connection between the main spleen and the gonad by a cord which is totally splenic or is composed of fibrous tissue. There is no anatomic connection between the ectopic and main spleens in the discontinuous type. Discontinuous splenogonadal fusion may be very rarely associated with the same congenital abnormalities as the continuous type is.⁽³⁾ The etiology of malformation is not determined yet. Most probably, it arises during the 5th through 8th week of the embryonic life when the developing spleen is close to the mesonephricgonadal anlage.^(9,12) However, there is no evidence suggesting that discontinuous splenogonadal fusion has a similar etiology.⁽¹⁰⁾ Additionally, right splenogonadal fusions cannot be explained with the above theory. It is therefore proposed that the discontinuous type may present a rare variant of an accessory spleen.^(2,9)

In summary, splenogonadal fusion is a rare congenital abnormality that is rarely suspected

preoperatively. Careful medical history recording and high suspicion for the condition may prevent the patient from unnecessary orchiectomy.

CONFLICT OF INTEREST None declared.

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