PATENT VITELLINE DUCT IN AN ADULT DECEPTIVELY APPEARED TO BE ACQUIRED UMBILICAL URACHAL SINUS: A CASE REPORT

Masahiko Takano
From the Division of Urology, Ohmura Municipal Hospital
Shuhji Yamashita, Shuh Inuzuka and Hiroshi Suzu
From the Division of Urology, Nagasaki University School of Medicine

Here is presented a surprisingly rare case in a 40-year-old male who had patent vitelline duct by nature. However, his congenital disease appeared deceptively to be an acquired umbilical urachal sinus on the diagnostic evaluations including fistulography before surgery. The diagnosis was definitely confirmed after the successful surgical procedure. The principal reason why these diseases were indistinguishable was reviewed. The incidence of each disease and incidence of association with umbilical fistula in each disease were discussed. With regard to these incidences, we compared urachal anomalies with vitelline duct anomalies through reference of several literatures. This is the most unique event we have ever clinically experienced.
(Acta Urol. Jpn. 41: 613-616, 1995)

Key words: Patent vitelline duct, Urachal sinus, Umbilical fistula, False exhibition, Adult

INTRODUCTION

Neither disease of enteroumbilical duct remnant nor that of urachal remnant is commonly encountered Here is reported an extremely rare case in a 40-year-old male who has retained patent vitelline duct, falsely exhibited as acquired umbilical urachal sinus on preoperative diagnostic appreciation.

CASE REPORT

About 10 years ago, this 40-year-old male began to notice an intermittent wetting around his navel. For the last several months prior to admission, he has often experienced a purulent discharge from there. On April 20, 1992, he was admitted to hospital suspected of having an urachal anomaly with wet umbilicus. In the center of his umbilical fossa a fistulous orifice of pin-hole in size was visible, and a small amount of serous fluid was continuously oozing out. However, no evidence of infection was seen around it. Excretory urography revealed normal findings. Cystoscopy showed a normal bladder dome. Abdominal CT-scan revealed a cystic lesion just behind the anterior abdominal wall (Fig. 1). Fistulography revealed a long cystic filling shadow about 5 cm in length and 2 cm in width lying downward toward the pubic symphysis from the umbilicus along the midline of the lower abdominal wall (Fig. 2). Judging from these findings, the diagnosis was made as an acquired umbilical urachal sinus, and the

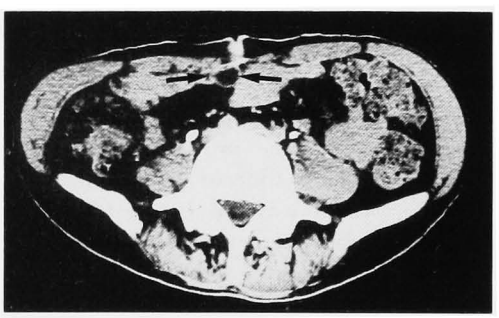

Fig. 1. Computed tomography shows cystic tubular lesion (arrows) in the abdomen.

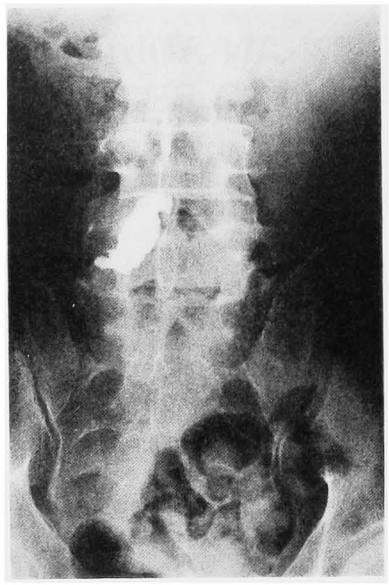

Fig. 2. Fistulography in frontal view shows a long cystically filled shadow.

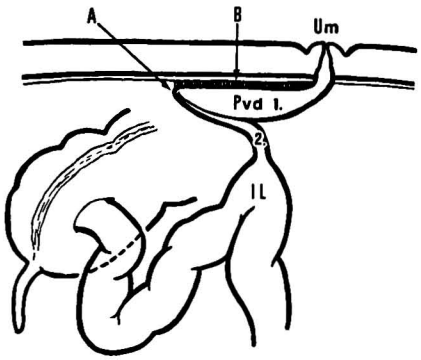

Fig. 3. Schema of intraperitoneal circumstances at the time of operation.
Um : Umbilicus
Pvd: Patent vitelline duct
1. Cystically dilated portion
2. Slenderized portien by tension
IL : Ileum
A : Point of crooking of Pvd
B : Adherence area of Pvd

operation was decided to remove en masse from umbilicus to bladder vertex.

Figure 3 shows the intra-abdominal circumstances at the time of operation. The structure, which was predicted to be an acquired umbilical urachal sinus before operation, was found to be lying within the peritoneal cavity and was adhered closely to behind the anterior abdominal wall. Its end was attached to the wall just behind

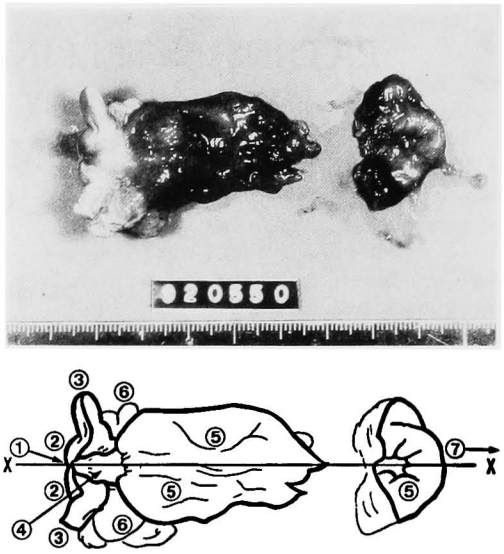

Fig. 4. I. (Upper) Macroscopic appearance of resected patent vitelline duct (mucosal surface). (Tne duct was divided into two parts of necessity during loosening of hard complex adhesions witn many surrounding enteric loops.)
II. (Lower) Schema shows each part of the upper resected tissue.
① Fistulous opening in the center of navel fossa ② Eminence of umbilicus within the fossa ③ Skin flap around the umbilicus ④ Fistulous lumen within umbilicus ⑤ Mucosal surface of patent vitelline duct ⑥ Fat tissue ⑦ Entry into ileum X. Central axis of the duct

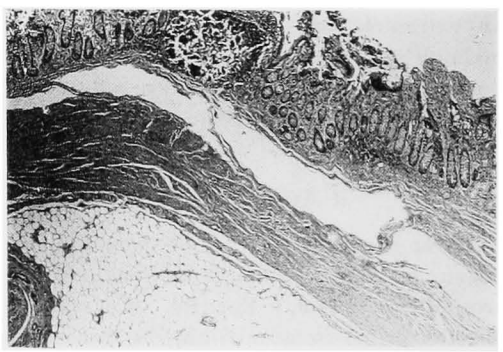

Fig. 5. Microscopic appearance of the patent vitelline duct with construction of ileum wall, consisting of layers arrangement of villi epithelium with lymph follicle, submucosa, smooth muscle and serosa. (HE-stain, × 100)

the umbilicus. Another end was suddenly crooked with an angle of 180 degrees, and was extended inward toward the cavity.

Finally it was attached to the opposite side of mesentery attachment of terminal ileum at about 50 cm oral from ileum end. It was associated with complex inflammatory adhesions with surrounding many enteric loops. After loosening of the severe adherence, this structure was resected en bloc from the umbilicus to the entry into ileum side. Macroscopically it showed just the appearance of intestine itself and made a complete tubular communication between umbilicus and ileum (Fig. 4). Microscopically it showed the construction of ileum wall consisting of layer arrangement of villi epithelium, submucosa, flat muscle and serosa without other ectopic tissues (Fig. 5). Consequently, the final diagnosis was patent vitelline duct.

DISCUSSION

Patent vitelline duct is an entire persistence of entero-umbilical duct, which makes a complete conduit between umbilicus and intestine. This is heralded by the excretion of enteric contents from the umbilicus. This is commonly seen in the neonate and should be treated promptly because of its severity. The incidence of Meckel's diverticle ranged from 1 to 2% of the population[1,2]. According to Mackey and coworkers[2], of a total of 402 patients with Meckel's diverticle, 4 (0.99%) had omphalomesenteric duct, consisting of 3 with patent omphaloenteric fistula and one with miscellaneous symptom. Of these 4 patients, three were children but the remaining one was an adult. Vane and associates[3] described that of a total of 217 children with vitelline duct anomaly, 4 patients (1.8%) had patent vitelline duct with bilious drainage from the umbilicus. Accordingly, the patent vitelline duct alone had draining umbilicus among the vitelline duct anomalies, and the incidence is only 1~2% in all patients with Meckel's diverticle. Therefore, as for patent vitelline duct, the incidence of population is estimated from 0.01 to 0.04%.

Acquired umbilical urachal sinus is a partial persistence of urachus, in which umbilical end opens but another end closes. At any age this condition manifests symptoms with infected excretion from umbilicus. Contemporary common classification on urachal diseases has been reported by Blichert-Toft and Nielsen[4]. From their report, of a total of 315 cases of urachal disease, 150 (47.6%) were congenital and 165 (52.4%) were acquired. The 165 acquired cases consisted of 58 (18.4%) of umbilical sinus, 10 (3.2%) of vesicourachal diverticle and 97 (30.8%) of urachal cyst including 4 (1.3%) of alternating sinus. Of these types, congenital patent urachus, acquired umbilical sinus and alternating sinus have a chance of associating with umbilical fistula. Therefore the incidence of associating with the fistula in urachal diseases is calculated as a total of 67.3%. This is indeed 33~67 fold rate in entero-umbilical duct leavings.

Mackey and Vane collected 402 and 217 cases of disease of entero-umbilical duct leavings during 50 and 16 years, only within their medical center, respectively[2,3]. On the other hand, Blichert-Toft collected 315 cases of disease of urachal leavings from the world literature during indeed 420 years[4]. In the latter, the time-length and the space-width of the collected cases were far longer and far wider, respectively, than in the former. Consequently, the occurrence rate of urachal anomalies is thought to be far less than that of vitelline duct anomalies. In reality, as to the rate, Blichert-Toft informed also that among 40,000 patients who underwent abdominal surgery at their institution, 8 cases (0.02%) were found to have acquired urachal disease[5].

Conventionally, fistulography is of the utmost importance and of indispensability to differentiate urachal anomaly from vitelline duct one before surgery[6]. However, in our case this test practically resulted in uselessness. The greatest cause was that this inherent illness has latently retained a strange crooking en route the conduit. Furthermore, an angular corner induced by this kinking has played the role of a stopcock against both inflow of contrast agents into the intestine on fistulography and discharge of enteric contents from umbilicus until the time of operation, and has deceived us into confusion in the

result.

In conclusion, an exceptionally rare case of patent vitelline duct in adults falsely exhibited as umbilical urachal fistula, was reported. While it is necessary to obtain correct findings through helpful diagnostic evaluations about urachal disorders before operation, it is also necessary to suspect the existence of such a case.

ACKNOWLEDGMENT

We thank Dr Masao Tomita, former Professor of the first division of surgery, Medical Department of Nagasaki University, for the well-meaning assistance of the literature citing about the disease of vitelline duct remnant, allowing this paper to be published.

We wish also to acknowledge the invaluable help of surgical staff in this hospital, Dr Katsutoshi Hashiguchi and Dr Takashi Ohshima without whose technical assistance surgical treatment of this patient would not have been possible.

REFERENCES

1) Griffen WO Jr: Meckel's diverticulum. In: Textbook of Surgery: The Biological Basis of Modern Surgical Practice. Edited by Sabiston DC Jr. 14th ed., pp. 866-868, WB Saunders, Philadelphia, 1991
2) Mackey WC and Dineen P: A fifty year experience with Meckel's diverticulum. Surg Gynecol Obstet **156**: 56-64, 1983
3) Vane DW, West KW and Grosfeld JL: Vitelline duct anomalies - Experience with 217 childhood cases. Arch Surg **122**: 542-547, 1987
4) Blichert-Toft M and Nielsen OV: Diseases of the urachus simulating intra-abdominal disorders. Am J Surg **122**: 123-128, 1971
5) Blichert-Toft M, Koch F and Nielsen OV: Anatomic variants of the urachus related to clinical appearance and surgical treatment of urachal lesions. Surg Gynecol Obstet **137**: 51-54, 1973
6) Gearhalt JP and Jeffs RD: Urachal abnormalities (in the section of Exstrophy of the bladder, epispadias, and other bladder anomalies). In: Campbell's Urology. Edited by Walsh PC, Retic AB, Stamey TA, et al. 6th ed., pp. 1815-1818, WB Saunders, Philadelphia, 1992

(Received on February 28, 1995)
(Accepted on May 2, 1995)

(和文抄録)

偽性後天性尿膜管臍瘻を呈した卵黄腸管開存症の1成人例

大村市立病院泌尿器科
高野 真彦
長崎大学医学部泌尿器科学教室(主任:斉藤 泰教授)
山下 修史, 犬塚 周, 鈴 博司

きわめて稀有な症例を経験したので,報告する.症例は40歳男性,約10年前より臍からの液性分泌物に気付いていた.術前,瘻孔造影を含めて諸検査により,臍瘻を伴った後天性尿膜管洞の診断のもとに手術に着手したが,開腹してみると卵黄腸管開存症であった.詳細に検索すると,この卵黄腸管は途中で180度の屈曲を有しており,この特異的な屈曲がその通過に対して留栓として作用していたため,造影剤の腸管内流入と生来から腸管内容物の臍からの排出が妨げられ,鑑別診断が不可能という結果に終わった.組織学的に絨毛上皮,粘膜下固有層,平滑筋層および漿膜からなる,完全な小腸壁の構造が認められた.ほかに,卵黄腸管と尿膜管の遺残の発生頻度,臍瘻の合併頻度等につき,文献より両者を比較してみた.

(泌尿紀要 **41**: 613-616, 1995)