## Endoscopic Papillotomy in Infancy Due to Biliary Cutaneous Fistula

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### Özet: BİLİYOKÜTANÖZ FİSTÜL NEDENİYLE BEBEKTE ENDOSKOPİK PAPİLLOTOMİ

Onaltı aylık kız çocuğunda, hepatoblastoma nedeniyle yapılan amealiyattan hemen sonra safra fistülü gelişti. Fistül debisi günde ortalama 250 ml idi. Endoskopik papillotomi, post operatif 29. günde yapıldı. Fistül, endoskopuk papillotomiyi takiben 8. günde kesildi. Papillotomi sonrası komplikasyon görülmedi. Hasta kısa zamanda düzeldi ve kemoterapi uygulandı. Safra sızıntısı 6 aylık takibimiz sırasında nüksetmedi. Endoskopik papillotomi bebeklerde de yapılabilir, ve özellikle seçilmiş vakalarda konvensiyonel cerrahi tedaviye alternatif yaklaşım olarak kabul edilebilir.

Anahtar Kelimeler: Endoskopik papillotomi fistül, bebek.

**B**illiary cutaneous fistula is one of the major complications of biliary tract surgery. Obstructions of the biliary ducts due to a tumor, stricture, or stone may provocate this unfavorable condition.

Injury to the bile ducts after surgery is usually manifested by pain in the upper right quadrant, jaundice, sepsis, or external drainage of bile from the wound or drain sites in the immediate postoperative period. Surgical reexploration of such patients who are usually critically ill, is often difficult due to infection, edema, and fibrosis in the periportal area.

GATA Hospital, Department of General Surgery. Ahmet Örs Hospital, Department of Internal Medicine. **Summary:** A sixteen months old girl was operated on having a hepatoblastoma. Bile leakage appeared immediately after the surgery with a rate of 250 ml per day.

An endoscopic papillotomy (EP) was performed on 29th day post operatively. The fistula ceased draining after eight days of EP. No complications occured after the EP. She recovered immediately and received chemotherapy. Bile leakege did not recur during the 6 month follow-up period.

EP can be performed in infancy and should be accepted as an alternative approach to conventional surgical treatment in carefully selected cases.

Key Words: Endoscopic papillotomy, fistula, infancy.

EP has been accepted with a wide spectrum of indications mainly in the adult population of patients since it was first described by Classen and Kawai in 1974, however experience with this procedure in the paediatric patient population is absent1,2. Herein, we present a 16 months old child with biliary cutaneous fistula due to resection of hepatoblastoma of the liver who was then succesfully treated by EP.

#### CASE REPORT

A sixteen months old female child was brought to Sosyal Sigortalar Hospital in Ankara, due to abdominal tenderness and vomiting during the last one month. On physical examination, she weighed 7.8 kg, she was pale and dehydrated. Oral temperature was 37 degrees Celsius, pulse rate was 128 beats per minute. Blood pressure was 100/60 mmHg. Chest, heart and neurological examination were all normal.

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Figure 1: Yellowish tumor mass located in the left lobe of the liver is seen on the fresh resected specimen

On abdominal examination the liver was palpable 15 cm below the right costal margin. A firm, tender mass of 15x15 cm in diameter was palpated in the liver site. Laboratory investigation revealed a hemoglobin level of 4.9 gr/dL, a hematocrit of 19.2 %, WBC count of 18.600/mm<sup>3</sup>, RBC count of 3,620,000/mm<sup>3</sup>, serum total and direct bilirübin levels were 2 and 1.5 mg/dL respectively. Alkaline phosphatase was measured to be 16 units of Bodansky. Blood urea, creatinin, ALT, AST, blood electrolytes and calcium levels were all within normal limits.

Ultrasonography revealed a hypoechoic lesion located in the left lobe of the liver. Liver biopsy revealed hepatoblastoma of the liver. The tumor was removed by a left lobectomy of the liver (Figure 1). The weight of the tumor mass was 1.3 kg. There was bile oozing 250-300 mL per day from the drain placed under the liver site during the all 19 days of post operative follow up (Figure 2). She was consulted to the department of Gastroenterology, Ankara University Medical School. Endoscopic Papillotomy was performed without complication. Oozing of the bile decreased day by day and it stopped completely on the 8th day. The drain was removed. She quickly recovered and gained 1.1 kg after one month of



Figure 2: Bile leakage seen from the drain inserted under the liver.

EP (Figure 3). She received chemotherapy and there was no recurrence of the tumor and fistula after 6 months follow-up period.

#### DISCUSSION

Biliary cutaneous fistula still remains as a major complication of biliary tract surgery. Glemm and Mannix reported 40 biliary-digestive fistulas in 4500 bile tract operations (3). Surgical reexploration of such patients is often difficult due to infection, edema, and fibrosis in the periportal area. A relatively high morbidity and mortality rate is also associated with surgical repair4. Recently non-surgical techniques, such as percutaneous transhepatic biliary decompression (PTBD), endoscopic papillotomy, naso-biliary drainage, endoscopic or percutaneous endoprosthesis were employed to treat the mentioned complications (4-10).

The aim of these non surgical techniques for biliary fistula is to reduce the biliary,duodenal pressure gradient. The basal tonic pressure of the sphincter of Oddi is about 4 mm Hg being higher than that in the common bile duct, and about 16 mm Hg higher than in the duodenum; it reaches contraction amplitudes of 50-120 mm Hg11,12.

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Figure 3: Bile leakage completely ceased after endoscopic papillotomy.

After EP, the bile is expected to be redirected towards the site of smallest resistance by way of the papilla of Vater. As a result of redirection of the bile, the fistula is managed to cease. The percutenous way of decompression provides delineating the anatomy of the bile ducts, the site and cause of the stricture and fistula. PTBD may lead to healing of the fistula by decompressing the biliary system and hence diverting bile flow from the defect into the bile ducts.

#### KAYNAKLAR

- 1. Classen, M., Demling, L.: Endoscopische Sphinterotomie der papilla vateri and stein extraktion aus dem ductus choledochus. M.M.W. 99: 496, 1974.
- Kawai, K., Akasaka, Y., Mura Kami, K., Tada, M., Kohli, M.Y.: Endoscopic sphincterotomy of ampulla of vater. Gastrointestinal endoscopy 20: 148, 1974.
- Jorge, A., Diaz, M., Lorenzo, J., Jorge, O.: Choledochoduodenal fistulas. Endoscopy 23: 76-78, 1991.
- Smith, C.A., Schapiro, H.R., Kelsey, B.P., Warshaw, L.A.: Successful treatment of nonhealing biliary-cutaneous fistulas with biliary stents. Gastroenterology 90: 764-769, 1986.
- Feretis, C., Kekis, B., Bliouras, N., Shaheen, G.N., Daras, D., Golematis, B.: Postoperative external and internal biliary fistulas, unassociated with distal bile duct obstruction: Endoscopic treatment. Endoscopy 22: 211-213, 1990.
- Goldin, E., Libson, E., Rachmilewitz, D.: Endoscopic insertion of endoprostheses in the treatment of postoperative cutenaous biliary fistula. Surgery 102 (1): 88-90, 1987.
- Gökçora, H.I., Örmeci, N., Dökmeci, A., Barlas, M.: Treatment of biliary fistulas and cholelithiasis: Is endoscopic sphincterotomy acceptable in the paediatric age group? Int Surg 74: 51-54, 1989.

PTBD lost favour due to complications, uncomfortable puncture sites, sometimes difficulty in keeping the catheter in very narrow biliary tracts13. Allthough percutaneous or endoscopic endoprosthesis has complications like bleeding, cholangitis, bile leakage, pneumothorax, and retroperitoneal perforation, it can be used in selective cases with large fistulas. But the EP, which has less complications than the PTBD or endoprosthesis, can be successfully used in cases with fistulas. Biliary fistulas may also be treated in the paediatric age group by the EP.

Endoscopic Retrograde Cholangiopancreaticograply is succesfull with a rate of 92-96% in childhood (3,9,10,13).

It is of great importance that we have not come across any series involving infant patients requiring endoscopic sphincterotomies.

The EP can be succesfully used in selected cases such as fistula and/or common bile duct stones as an alternative method to surgery infant age group.

- Hsu, D.: Endoscopic sphincterotomy in the management of biliary cutaneous fistula. The American journal of gastroenterology 82 (10): 1078-1080, 1987.
- Kaufman, L.S., Kadir, S., Mitchell, E.S., Chang, R., Kinnison, L.M., Cameron, L.J., Jr. White, I.R.: Percutaneous transhepatic biliary drainage for bile leaks and fistulas. AJR 144: 1055-1058, 1985.
- Steenberger, V.W., Haemers, A., Pelemans, W., Ponette E., Vanwing, J., Verbeken, E., Fevery, J.: Postoperative biliocutaneous fistula: Successful treatment by insertion of an endoprosthesis. Endoscopy 19: 34-36, 1987.
- 11. Allescher, D.H.: Papilla of vater: Structure and function. Endoscopy 21: 321-329, 1989.
- Tanaka, M., Ikeda, S., Nakayama, F.: Change in bile duct pressure responses after cholecystectomy: Loss of gallbladder as a pressure reservoir. Gastroenterology 87: 1154-1159, 1984.
- Yee, N.C.A.: Complications of percutaneous biliary drainage: Benign vs malignant diseases. AJR 148: 1207-1209, 1987.
- Allendorph, M., Werlin, L.S., Geenen, E.J., Hogan, J.W., Venu, P.R., Stewart, T.E., Blank, L.E.: Endoscopic retrograde cholangio pancreatography in children. The journal of pediatrics. 110(2): 206-211, 1987.
- Cotton, B.P., Laage, J.N.: Endoscopic retrograde cholangio pancreatography in children. Archives of disease in childhood 57: 131-136, 1982.