Case Report

An unusual case of villous adenoma of the ampulla Vateri in a renal allograft recipient

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Introduction

Ampullary epithelial neoplasms include benign (5%) and malignant tumours (95%) centred in the region of the papilla of Vater [1–3]. The clinical manifestations for all types of lesions are similar. Carcinoma and benign adenomas are the most important ampullary tumours, and there is evidence that adenomas are premalignant [1–3]. After a complete survey of the literature, no cases of ampullary adenoma in renal transplant recipients were found.

Case report

A 64-year-old man was diagnosed with chronic renal failure secondary to IgA nephropathy in 1984, and was included in our haemodialysis programme in January 1985. Mild hypertension was easily controlled with oral medication. Ten months later the patient underwent a cadaveric renal transplantation. Immunosuppressive therapy during these 10 years included prednisone (0.5 mg/kg/day tapered to 0.1 after 1 year) and azathioprine 2.5 mg/kg/day. One year after grafting he developed posttransplant hyperglycaemia, which was controlled by dietary measures.

During a scheduled follow-up examination he complained of polydypsia, polyuria, nicturia, and polyphagia, with a 5-kg weight loss during the previous 6 months. Physical examination was unremarkable; he appeared healthy, with normal skin and painless abdominal palpation. Blood pressure was 150/80 mmHg. Leukocyte count was 6580 cells/mm³, haemoglobin 13.6 g/dl, haematocrit 40%, and platelet count was 221 000/mm³. Serum chemistry showed urea 43 mg/dl, creatinine 1.4 mg/dl, sodium 134 mEq/l, potassium 4 mEq/l, chloride 100 mEq/l, total CO₂ 29.5 mEq/l, glucose 270 mg/dl, uric acid 4.6 mg/dl, calcium 9.4 mg/dl, phosphorus 3.8 mg/dl, triglyceride 99 mg/dl, total cholesterol 231 mg/dl, total protein 7.3 g/l, albumin 45 g/l, total bilirubin 1.8 mg/dl, aspartate aminotransferase (ASAT) 44 IU/l, alanine aminotransferase (ALAT) 30 IU/l, γ-glutamyltranspeptidase (GGT) 1339 IU/l, lactic dehydrogenase 275 IU/l, alkaline phosphatase 785 IU/l, IgG 1328 mg/dl, IgA 504 mg/dl, IgM 101 mg/dl, Carcinoembryonic antigen level was 9.4% (normal 5.4–7.6), CA.19.9 was higher than 200 U (normal less than 10). Coagulation studies were normal. Hepatitis B surface antigen was negative, anti-core and anti-surface hepatitis B antibodies were positive. Hepatitis A and C serologies were negative. Cytomegalovirus, herpes, Epstein–Barr virus, and toxoplasma serologies showed past but not recent infection. A culture of the urine was sterile. Antinuclear, antinucleolar, antimitochondrial, anti-LKM, and antithyroglobulin antibodies were all negative. Further studies included a 24-h urine collection; the volume was 4.8 litres, glycosuria 7.4 g/l, proteinuria 0.2 g/l, sodium 9 mEq/l, and fractional excretion of sodium 0.3%. Creatinine clearance was 71 ml/min.

Ultrasonography examination revealed an important dilatation of intra- and extrahepatic bile ducts, down to the intrapancreatic common bile duct (diameter 20 mm), and no clear image of a mass. CT scan showed the dilated common bile duct and the pancreatic duct, but there was no definite mass (Figure 1). Endoscopic retrograde cholangiopancreatography (ERCP) revealed an excentric mass, 3 cm in diameter, with an irregular surface. A histological diagnosis of villous adenoma was made. The patient underwent ampullectomy with sphincteroplasty after cholecystectomy. Extensive study of the surgical specimen showed typical findings of tubular–villous adenoma with moderate superficial dysplasia (Figure 2).
crine tumours. The villous adenoma is the most common one [4]. Rosenberg et al. [5] found six examples of such tumours among one million surgical pathology specimens. They range in size from 4 to 7 cm, and cause biliary obstruction in approximately 70% of the cases [4,5]. A high percentage of the tumours are carcinomas: in 27% of cases reviewed by Sobol and Cooperman [4], the tumours were malignant.

As an initial approach, ECRP should be performed after a diagnosis of obstructive bile duct disease is made with ultrasonography. The biopsy obtained with this technique gave us essential diagnostic information which guided our conservative surgical procedure. Indications for ampullectomy in the management of ampullary tumours are controversial, but the technique has been advocated for patients with benign or villous adenomas [4–7]. Our transplant patient was treated with wide local resection of the ampulla, because the previous biopsy and frozen sections showed no malignancy. Cholecystectomy was performed to avoid future gall-bladder disease and to help the identification of the common bile duct at the ampulla. Sobol and Cooperman [4] observed that over 85% of the patients who were conservatively managed were without evidence of recurrence for varying periods of follow-up. Therefore, ampullectomy could be the procedure of choice for management of benign or even premalignant processes of the ampullary region. Indeed, the Whipple procedure (pancreatoduodenal resection) is associated with high morbidity and mortality [6], and the withdrawal of immunosuppressive therapy could result in loss of the renal graft.

Discussion
A wide variety of benign tumours may occur at the ampulla of Vater, including benign adenoma (tubular and villous), lipoma, fibroma, neurofibroma, leiomyoma, lymphangioma, haemangioma, and neuroendo-

Fig. 1. CT scan showing dilatation of both common bile and pancreatic ducts, as well as the gall-bladder.

Fig. 2. Histological section of a typical villous adenoma without significant dysplasia. There is no infiltration of the adjacent submucosa. (H & E ×40)

The postoperative course was unremarkable. Five days after surgery, renal function was normal, and serum determinations were: glucose 178 mg/dl, total bilirubin 0.67 mg/dl, ASAT 21 IU/l, ALAT 19 IU/l, GGT 198 IU/l, and alkaline phosphatase 171 IU/l. One year after surgery, ultrasonography revealed normal bile ducts, and serum chemistry was unremarkable. No glycosuria was noted and the patient is asymptomatic.

References

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