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Baylor Regional Transplant Institute, Dallas, TX. 2 Dallas Transplant Institute, Dallas, TX, USA.

**BACKGROUND:** Polycystic liver disease (PLD) is a rare disorder frequently associated with polycystic kidney disease (PKD). Transplantation is a treatment option for these patients. Because of preservation of hepatic function in these patients, liver transplantation is not routinely utilized. We report a large series of PLD patients and their outcomes following liver and kidney transplantation. **METHODS:** Fourteen patients underwent orthotopic liver transplantation (OLTx) for PLD between 1987 and 2003. Twelve patients had PKD combined with PLD. Nine patients received only liver transplantation. Five patients had combined liver and kidney transplantation. Thirteen patients (93%) survived for at least one year following liver transplantation. Two out of eight patients who received solitary liver transplantation later required kidney transplantation. **RESULTS:** Pretransplant glomerular filtration rate (GFR) in patients with PKD was 75.8+/-25.4 ml/min/1.73 m. One year later, GFR was 37.2+/-8.3 ml/min/1.73 m. Kaplan-Meier analysis showed that one- and two-year graft survival for combined liver and kidney transplantation was 80% (n=5), whereas graft survival for solitary liver transplantation was 100% (n=9). Mean survival of patients who had combined liver and kidney transplantation was 46.7+/-54.2 months (n=5), whereas the mean survival for solitary liver transplant patients was 80.4+/-68.6 months (n=9) (P=0.36). **CONCLUSION:** Transplantation is an excellent option for PLD with dramatic improvement in quality of life and acceptable morbidity. For combined liver and kidney transplantation one- and two-year patient survival rates were similar to combined transplantation for other indications. For patients with acceptable renal function at time of transplantation, solitary liver transplantation has an excellent outcome.

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