A 35-year-old Swiss woman with AIDS experienced fever, jaundice and cough. Laboratory evaluation revealed signs of an infection and cholestasis. The examination by ultrasound showed thickening of the intra- and extrahepatic bile ducts and gallbladder wall, without dilatation or stones. Endoscopic retrograde cholangiography demonstrated diffuse sclerosing cholangitis like lesions in the biliary tract and confirmed the diagnosis of a HIV related cholangiopathy. The cause was a cytomegalovirus infection as shown by liver biopsy with detection of cytomegalovirus early antigen. The treatment with ganciclovir was of some efficacy with improvement of jaundice.