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Successful living donor liver transplantation of the left lateral sector with simultaneous atypical resection of the S2 for focal nodular hyperplasia

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The purpose of presenting a clinical observation. Description of a clinical observation of a successful atypical resection of focal nodular hyperplasia of the segment II of the liver to a relative donor, followed by transplantation of the left lateral sector of the liver to a one-year-old child with cirrhosis of the liver in the outcome of biliary atresia.
General provisions. Patient Z., 29 years old, was examined as donor for related liver sector transplantation. The oval structure with fairly smooth contours, hyperechoic in the central parts and hypoechoic in the periphery, was found in the segment II during the ultrasound examination; assessment of blood flow was difficult due to acoustic interference. According to computed tomography, it was a weak hypervascular formation with fuzzy contours, accumulating contrast medium unevenly in the arterial phase, with visualisation of a small hypodense area around, also accumulating contrast medium in the venous phase. According to magnetic resonance imaging, there is a zone of increased MR signal, with contrast enhancement uniformly accumulating contrast in the arterial phase. It was assumed that the patient had focal nodular liver hyperplasia. The patient underwent surgery, during which the left lateral sector of the liver was removed and a single-step atypical resection of the segment II was performed. Histological examination of the surgical material confirmed the presence of FNH. After excision and suturing of vascular structures in conditions of continued blood circulation, the left lateral sector was removed from the wound and implanted into the recipient in an orthotopic position. The postoperative period passed without complications. The donor and recipient were discharged from the hospital in a satisfactory condition on the planned date.
Conclusion. This observation demonstrates the possibility of using a liver fragment as a transplant after resection of focal nodular hyperplasia.
Keywords: liver transplantation, focal nodular hyperplasia, liver cirrhosis, ultrasound diagnostics, computed tomography, magnetic resonance imaging
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Успешная родственная трансплантация левого латерального сектора печени с одномоментной атипичной резекцией 2-го сегмента по поводу фокально-нодулярной гиперплазии

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Цель: Описать наблюдение успешно проведенной родственному донору атипичной резекции фокально-нодулярной гиперплазии 2-го сегмента печени с последующей трансплантацией левого латерального сектора печени ребенку в возрасте одного года с циррозом печени в исходе билиарной атрезии.
Основные положения. У пациентки Х., 29 лет, в рамках обследования родственных доноров печени во 2-м сегменте печени при ультразвуковом исследовании было выявлено овальное образование с достаточно

ровными контурами, гиперэхогенное в центральных отделах и гипоэхогенное по периферии; оценка кровотока была затруднена из-за акустических помех. По данным компьютерной томографии: слабогиперваскулярное образование с нечеткими контурами, в артериальную фазу неравномерно накапливающее контрастное вещество, с визуализацией небольшой гиподенсной зоны вокруг, также накапливающей контрастное вещество в венозную фазу. По данным магнитно-резонансной томографии: зона повышенного МР сигнала, при контрастном усилении равномерно накапливающая контраст в артериальную фазу. Было предположено наличие у пациентки фокально-нодулярной гиперплазии (ФНГ) печени. Проведено оперативное вмешательство, в ходе которого был изъят левый латеральный сектор печени и проведена одномоментная атипичная резекция 2-го сегмента. Данные гистологического исследования операционного материала подтвердили наличие ФНГ. После иссечения и ушивания сосудистых структур в условиях сохраненного кровообращения левый латеральный сектор был изъят из раны и имплантирован реципиенту в ортотопическую позицию. Послеоперационный период проходил без осложнений. Донор и реципиент были выписаны из стационара в удовлетворительном состоянии в планируемый срок.

Заключение. Данное наблюдение демонстрирует возможность использования фрагмента печени в качестве трансплантата после резекции фокально-нодулярной гиперплазии.

Ключевые слова: трансплантация печени, фокально-нодулярная гиперплазия, цирроз печени, ультразвуковая диагностика, компьютерная томография, магнитно-резонансная томография

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To date, the only radical method of treatment of end-stage liver failure is liver transplantation [1–4]. In a number of foreign countries, this problem has been practically solved thanks to the use of cadaveric organs transplantation of a donor with confirmed brain death. However, nowadays on the territory of the Russian Federation there is a shortage of cadaveric organs due to ethical, religious and legal problems in society regarding organ donation. At the same time, over the period from 2012 to 2020, there has been a 4.6 times increase in the number of patients on the waiting list for liver transplantation [4]. Given the current situation, the only way out in this case is to expand the donor pool through the use of liver fragments from living related donors [4–7].

As part of the preoperative examination of related donors, the generally accepted instrumental diagnostic methods are ultrasonography (ultrasound), magnetic resonance imaging (MRI) and multispiral computed tomography (MSCT) using contrast medium, which allows obtaining reliable information about the state of the liver parenchyma, assessing its vascular and biliary anatomy, measuring the volume of both the entire liver and its fragments [1, 8].

This paper presents a clinical example of a successfully performed atypical resection of focal nodular hyperplasia (FNH) of the segment II of the liver in a related donor with subsequent transplantation of the left lateral sector of the liver to a one-year-old child suffering from cirrhosis of the liver in the outcome of biliary atresia.

Description of clinical observation

Patient Z., 29 years old, was admitted to the Russian Scientific Center of Surgery named after Academician Petrovsky as a related donor of a part of the liver for her daughter, who had been diagnosed with cirrhosis of the liver in the outcome of biliary atresia.

At the preoperative stage, the patient underwent a full range of clinical, laboratory and instrumental examination methods according to the developed protocol for the examination of related liver donors, presented in the national clinical guidelines for living-donor liver transplantation [9].

Ultrasound diagnostics of the liver condition within the framework of this protocol includes a qualitative assessment of the parenchyma (uniformity, echogenicity, anterior-posterior liver size) and an assessment of hepatic blood flow (the state of the main branches of the hepatic artery, portal and hepatic veins).

According to ultrasound data, the patient had clear, even contours, normal dimensions, and a homogeneous structure of the liver. The echogenicity of parenchyma was insignificantly diffusely increased. In the segment II of the liver, an oval formation sized 1.6 × 2.4 cm with fairly even contours, hyperechoic in the central parts and hypoechoic in the periphery, was revealed (Figure 1). During colour Doppler imaging (CDI), the vessels in this formation were not clearly defined due to acoustic interference arising from heart contractions. Ultrasound data did not allow differentiating this formation.

According to the protocol, the patient underwent MSCT using an intravenous contrast medium.

During MSCT of the abdominal cavity and retroperitoneal space, performed by volumetric scanning with a thickness of 3 mm and 1 mm slices during and after intravenous bolus administration of the contrast medium Visipaque 320, the liver

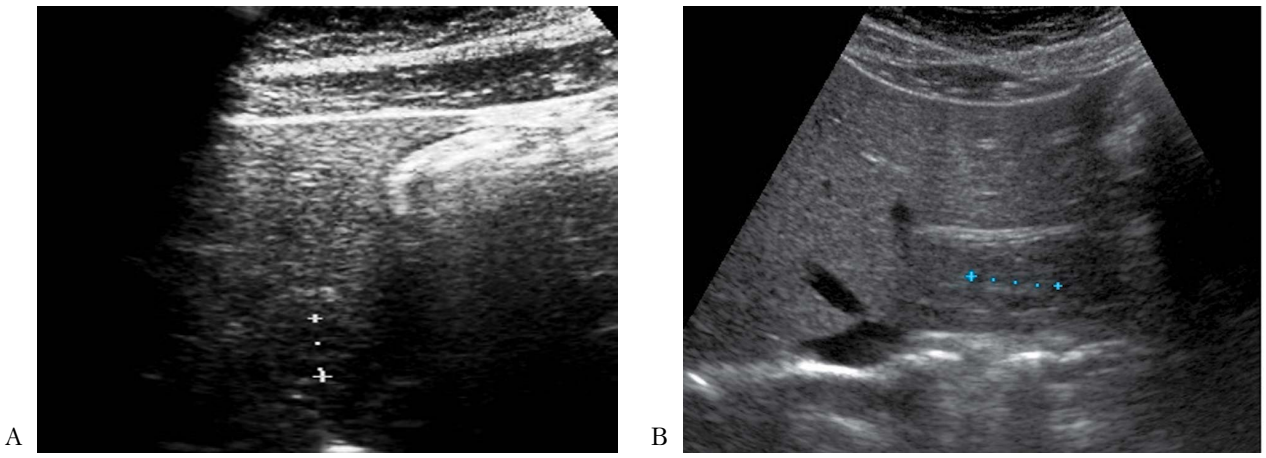


Fig. 1. A — ultrasound picture of the FNH of the segment II of the liver during longitudinal scanning along the midline of the abdomen; B — during transverse scanning

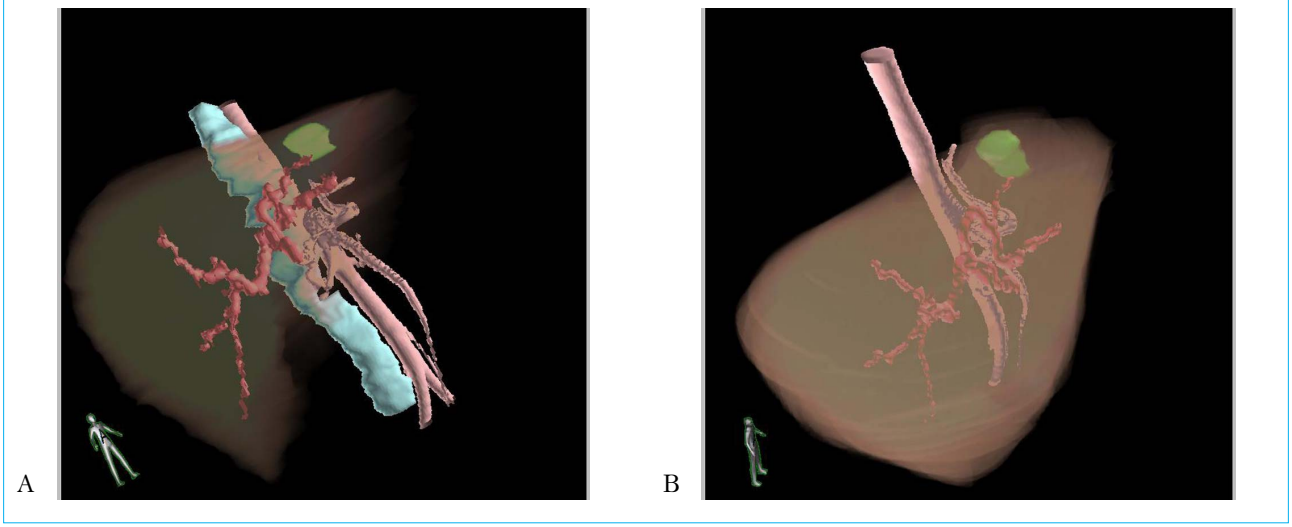


Fig. 2. Picture of FNH of the segment II of the liver, three-dimensional image

dimensions were not changed. Liver parenchyma is of normal densitometric density of 65–68 HU units. In the segment II of the liver, during the arterial phase of contrast, a weak hypervascular formation with fuzzy contours, sized 1.7 × 1.9 cm, accumulating contrast medium unevenly (with visualization of a small hypodense area around) was revealed. An arterial vessel (segment II artery) was attached to this formation (Fig. 2). In the venous phase, this formation continued to accumulate a contrast medium. In the native phase, this formation was isodense to the liver parenchyma, and therefore it was necessary to differentiate this formation between FNH and hemangioma.

To clarify the nature of the detected formation, the decision was taken to conduct magnetic resonance imaging (MRI) using hepatotropic contrast agents.

During MRI of the abdominal cavity and retroperitoneal space, the zone of increased MR signal on the diffusion-weighted image (DWI) B-factor 50, 400 was differentiated subdiaphragmally in the segment II of the liver. With contrast enhancement, there was a uniform accumulation of contrast by this zone in the arterial phase. In the venous phase and in the hepatospecific phase, the MR signal was identical to the liver parenchyma. The visible size of the formation was 1.7 × 1.5 cm. According to MR diagnostics, this formation was more consistent with FNH by the nature of contrast accumulation (Fig. 3).

Given the progressively deteriorating condition of the child and considering that the identified formation was benign, as well as taking into account the absence of a cadaveric organ and other potential related liver donors, it was decided to perform a related liver transplant from the mother. The estimated scope of the surgery included the removal of the left lateral sector of the liver with single-step atypical resection of its segment II.

The patient underwent surgical intervention according to the estimated volume. Intraoperative examination revealed a liver of the usual shape, colour and consistency, with a sharp edge. In the lateral part of the segment II, a volumetric formation of a tightly elastic consistency, sized 2.5 × 3.5 cm, was palpated. An additional determination of this formation was performed by intraoperative ultrasound. The boundaries of this formation were marked on the surface of the liver using an electrocoagulator. The left lateral sector of a triangular pyramid shape, sized 11.0 × 9.0 × 6.0 cm, was mobilised from the diaphragm by the intersection of the left triangular ligament. A ligature was placed under the left hepatic vein. With an indentation of 1.0 cm from the pre-marking line, the patient underwent atypical resection of the segment II of the liver. Further, by electrocoagulation, the liver parenchyma was separated from the porta hepatis parallel to the edge of the falciform ligament towards the right wall of the left hepatic vein. After excision and suturing of vascular structures in conditions of continued blood circulation, the left lateral sector was removed from the wound and implanted into the recipient in an orthotopic position.

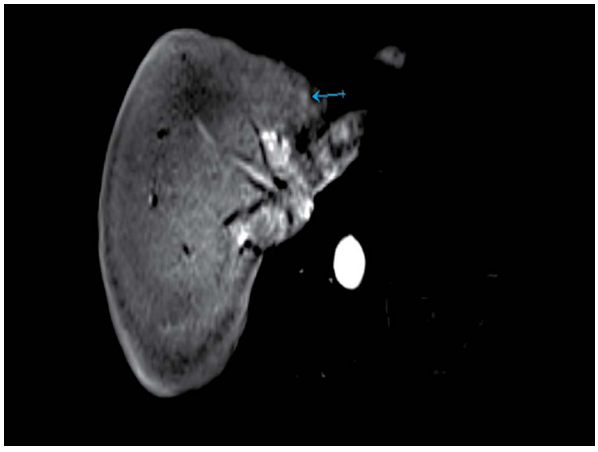


Fig. 3. MR image of the FNH of the segment II of the liver, an axial image in the arterial phase of the scan

As a result of a histopathology of the surgical material, a node was macroscopically identified, represented by a hyperplastic liver parenchyma divided into nodules by fibrous septa, in the center of which there was a stellate scar with a large arterial vessel (Fig. 4).

On microscopic examination (Fig. 5), the focus was represented by hepatocytes with a large amount of glycogen and focal steatosis forming trabeculae. There were no portal tracts. Groups of trabeculae formed nodules separated by fibrous septa of varying thickness with moderate lymphocytic infiltration, in the layer of which proliferating bile ducts were determined.

Such studies corresponded to the FNH and allowed us to confirm the presence of this benign formation in the patient.

The postoperative period passed without complications. The donor and recipient were discharged from the hospital in a satisfactory condition on the planned date.

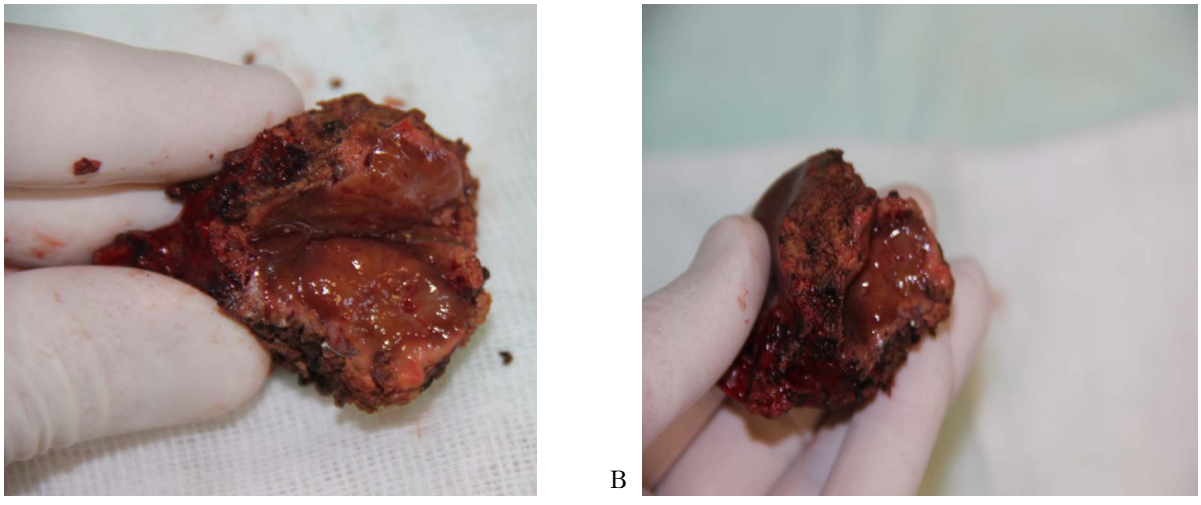
Discussion

It is assumed that FNH is formed due to hyperplastic growth of normal hepatocytes with a deformed drainage system of the biliary tract, which could potentially be a response to a pre-existing arteriovenous malformation [10, 11]. Arterial blood supply comes from the hepatic artery, while venous outflow is carried out into the hepatic veins.

There are two main types of FNH [11]:

1. Typical, which accounts for 80 % of cases;
2. Atypical, accounting for 20 % of cases.

Macroscopically, a typical FNH is represented by large areas with fairly well-defined edges, with



A B

Fig. 4. Macroscopic picture of the drug, after atypical resection of the segment II of the liver

an unexpressed capsule. A characteristic feature is a noticeable central scar with radially divergent fibrous septa, which is present in less than 50 % of cases [12]. The central artery is determined with characteristic features of the "spokes of the wheel" type [11].

Histologically, the formation consists of an abnormal nodular area, deformed vessels and proliferation of cholangioles. Almost normal hepatocytes are arranged in the form of plates, with a thickness of one to two cells. Bile ducts are usually located on the line between hepatocytes and fibrous cords [10]. Kupffer cells are determined. There are no signs of malignancy in the FNH [11].

In atypical forms of FNH, there is no central scar and central artery. These areas of the liver are difficult to distinguish from other lesions using medical imaging methods [11].

Atypical signs also include a pseudocapsule, heterogeneity of the lesion (more often observed in adenoma), lack of strengthening of the central scar and intralesional fat [12].

Nodules can grow and disappear, and new nodules can appear even after resection [13].

According to the international classification of liver and intrahepatic bile duct tumors, FNH is not a true neoplasm, but a secondary hyperplastic regenerative response of hepatocytes to local vascular disorders [14].

Based on the frequency of occurrence among all benign liver formations, FNH ranks second after hemangiomas, accounting for up to 3–8 % of primary volumetric organ pathologies [15]. The disease is more common in women than in men (up to 9:1 cases), and is usually detected in people aged 30–40 years. In 80–95 % of cases, FNH is a single section of altered tissue, most often less than 5 centimeters in size. Less than 20 % of cases occur in multiple sites of FNH. In addition, in 5–10 % of cases, FNH is combined with hemangioma.

FNH has no clinical symptoms and is usually an accidental revelation during instrumental examination methods (ultrasound, MSCT, MRI) applied for other diseases. According to foreign studies, the expression of estrogen receptors may be observed in the FNH tissue, but at present it is not known for certain whether estrogen participates in the development of FNH [16].

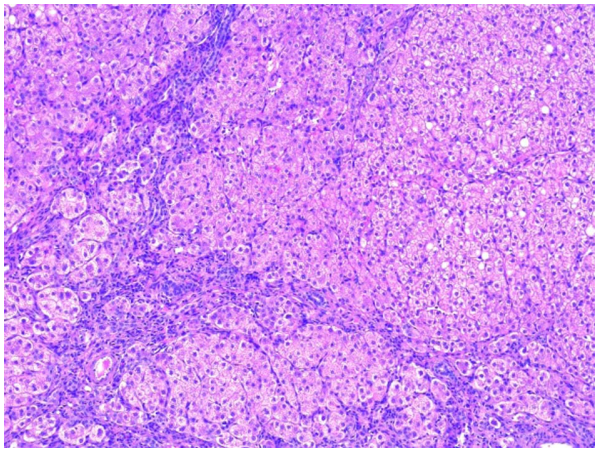


Fig. 5. Histological examination of the removed tissue sample, x100 magnification

In the literature, there are a small number of descriptions of clinical cases of liver transplantation from a donor with benign formations. The article by Tan M. et al., 2001, described the first case of transplantation of a donor cadaveric liver with FNH detected in it, which was resected during the donor stage of the operation and then transplanted to the recipient [17]. Publications on liver transplantation from a living donor who was diagnosed with FNH were not previously presented. However, in 2015 Onishi Y. and co-authors demonstrated for the first time a case of successful transplantation of a donor liver from a living donor with hemangiomas in the segments II and IV [18], showing that the presence of benign liver formations is not a contraindication to liver transplantation.

Conclusions

The presented clinical observation objectively demonstrates the possibility of successful clinical application of related transplantation of a liver fragment with FNH with its single-step resection at the donor stage.

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