

Laparoscopic adrenalectomy for benign adrenal tumors in pediatric surgical practice

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ABSTRACT

Objective. To estimate the results of minimally invasive adrenalectomy in children and compare our data with worldwide results.

Material and methods. There were 65 patients aged from 3 months to 17 years with different organic diseases of adrenal glands. Children have undergone surgery for the period since 2003 to 2018.

Results. Incidentalomas accounted 33.8% of all resected tumors. Bilateral lesion was diagnosed in 12% of patients. The largest neoplasm (12 cm) was diagnosed in a 9-year-old child. Intraoperative bleeding occurred in 2 patients. Endoscopic surgery did not require conversion in any case.

Conclusion. We accumulated unique experience of laparoscopic resection of benign lesions of adrenal glands. Our results are satisfactory and comparable with data of other national and foreign colleagues. Only close collaboration of pediatric surgeons and endocrinologists could lead to successful and safe treatment of these patients. Patients with suspected malignancies should receive treatment at special hospitals with oncological service.

Keywords: adrenalectomy, laparoscopic, children, incidentaloma, benign adrenal lesions.

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Introduction

History of adrenal surgery dates back a little over 100 years. The first minimally invasive endoscopic adrenalectomy was performed by the team of prominent Canadian surgeon M. Gagner in 1992 in the patient with Cushing's syndrome and pheochromocytoma [1]. Maistrenko et al. reported that the first laparoscopic adrenalectomy in Russia was performed by military surgeon Yu.N. Sukhopara in November 1995 at the Fedorov Abdominal Surgery Clinic (St. Petersburg). Robot-assisted resection of adrenal glands (Da Vinci) has been reported since 2003 [2].

Pediatric minimally invasive surgery develops after an adult one. Endoscopic adrenalectomy became preferable in pediatric patients as soon as this approach took its place in adult surgery from the second half of the 90s of the last century [3, 4].

Benign adrenal neoplasm is a rare disease in pediatric patients. However, this diagnosis has become more common in recent years that is associated, inter alia, with improved primary diagnostic imaging. However, sample

size does not usually exceed 20 cases in reports devoted to adrenal pediatric surgery [5]. Minimally invasive resection of adrenal glands for benign neoplasms in pediatric surgical practice is still poorly described in national literature despite more than 20-year international experience.

Material and methods

A retrospective cohort study was conducted.

Inclusion criteria: patients under 18 years of age with adrenal gland tumors consulted by an endocrinologist except for neoplasms with high probability of malignancy in accordance with preliminary preoperative examination. Tumor dimension, surgery time, complications and need for conversion were evaluated.

We have performed 67 minimally invasive adrenalectomies in 65 patients with various adrenal gland lesions since 2003. In our opinion, surgical treatment of this disease requires a special consideration while pediatric surgeon may encounter this diagnosis in a non-oncological hospital. There were 34 girls and 31 boys (52.3% and

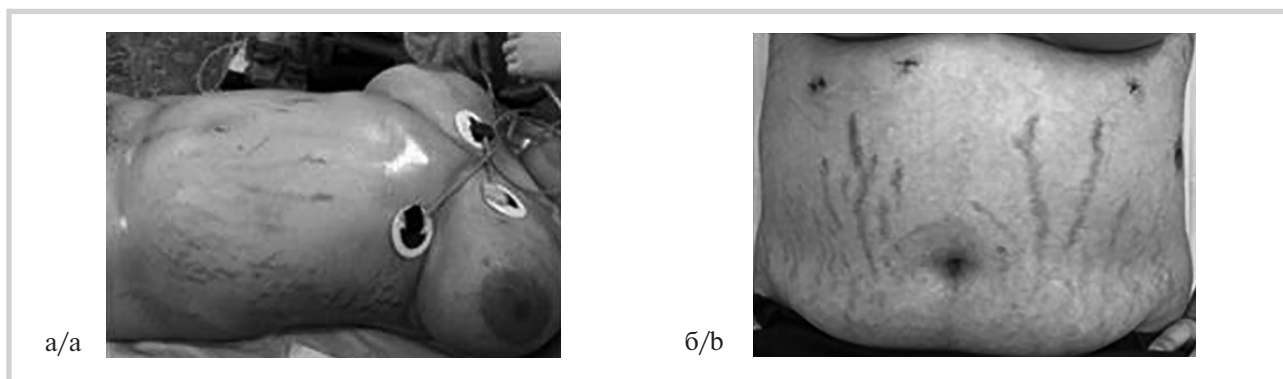


Fig. 1a. Patient 16 y.o., Carney complex with bilateral nodular adrenal disease, severe symptomatic hypercorticism (1a – preoperative view; 1b – postoperative view); 1b. Number of trocars – 6. Surgery time – 190 min (the most protracted operation in our investigation)

47.7%, respectively). Age of patients ranged from 3 months to 17 years. Median age was 90.6 months.

We used laparoscopic lateral transperitoneal approach in all cases. Three trocars were deployed for left-sided lesions, four trocars (additional trocar for hepatic retractor) – for right-sided lesions, five trocars – for bilateral lesions. Right-sided adrenalectomy was followed by left-sided resection and change of patient’s position on the operating table [6]. There were 27 right-sided solid adrenal neoplasms and 24 left-sided tumors. Cystectomy with preservation of adrenal parenchyma was performed in 6 patients with adrenal cysts. Bilateral lesion was diagnosed

in 8 patients. According to the recommendations of attending endocrinologists, two patients with bilateral lesions underwent staged right– and left-sided adrenalectomy. One patient required contralateral adrenalectomy due to persistent Cushing’s syndrome in 1 year after primary surgery. Nodular lesion of excised adrenal gland was histologically confirmed in the other case. Therefore, endocrinologists determined necessity of resection of the second adrenal gland. Resection of extraadrenal chromaffinoma was made in 1 case.

It is important to note that surgical indications were determined only in conjunction with endocrinologists in all cases. In our research, minimally invasive surgical approach was contraindicated in patients with potentially malignant neoplasms in accordance with preoperative examination data. Nevertheless, postoperative histological examination revealed adrenocortical cancer in 4 cases. Adrenalectomy was carried out in R0 fashion in all cases. En-bloc resection of adrenal gland with tumor was followed by its extraction from the abdominal cavity in endoscopic bag. Children were followed-up by attending endocrinologists. There were 2 cases of recurrence-free 5-year period. Moreover, adrenal neuroblastoma was confirmed in 3 patients after postoperative histological examination. These children were referred to oncologists for subsequent follow-up.

Results and discussion

Adrenal gland neoplasm over 6-7 cm was a contraindication for endoscopic resection for a long time [7, 8, 9]. In 2008, Chilean surgeons analyzed a sample of patients with various adrenal gland neoplasms. They divided these patients into 3 groups depending on tumor dimension. The first group included patients with tumors < 6 cm, the second group – 6 – 8 cm, the third group – over 8 cm. Surgery time in the third group was 20 min longer than in the first one (80 vs. 60 min), blood loss was more by 50 ml (100 vs. 50 ml), hospital-stay was higher by 1 days (3 vs. 2 days). However, incidence of complications was similar

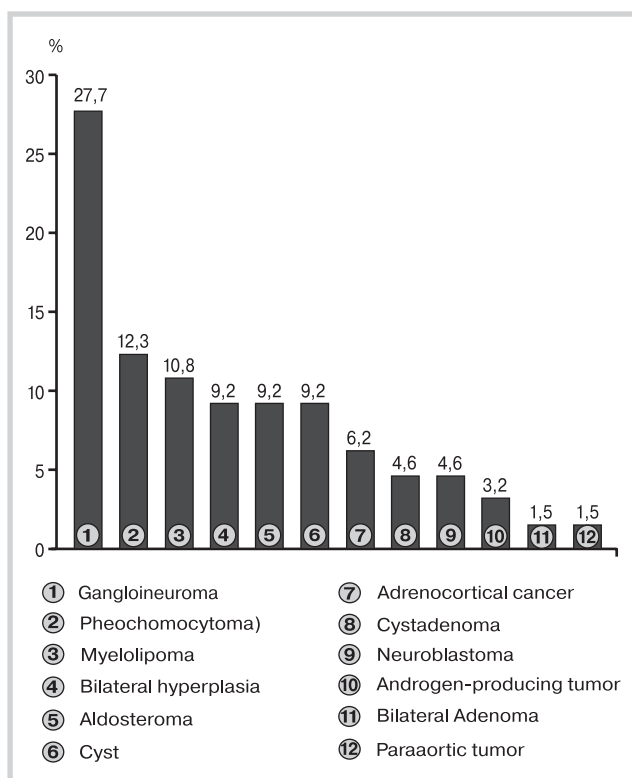


Diagram 1. Morphology of lesions

Table 1. Comparison of our findings with worldwide data

	Number of surgeries	Benign tumors, %	Mean age, years	Side of lesion (right/left)	Bilateral lesion	Tumor dimension, cm	Surgery time, min	Conversion, %
Miller et al, 2002 [16]	17	10 (58,8%)	9,8	5/12	0	4,8	120	5,8
Castilho et al, 2002 [17]	14	11 (78,6%)	6,3	8/4	1	4,1	107	15,4
Skarsgard et al, 2005 [18]	21	12 (57,1%)	6,4	6/13	1	6 (3,5 – 8,5)	-	4,8
Al-Shanafey et al, 2008 [19]	32	12 (37,5%)	3	11/15	3	2,8 – 7	-	9,4
Lopes et al, 2011 [20]	19	13 (52,6%)	3,9	10/5	2	4,3 (2 – 6,5)	-	0
Nerli et al, 2011 [21]	18	8 (44,4%)	5,8	6/12	0	-	-	0
St Peter et al, 2011 (multicenter) [22]	145	78 (53,8%)	8,8	59/76	5	4,3 (1-14)	-	8,97
Kelleher CM, et al, 2013 [23]	18	0 (0%)	2,4	-	-	2,5	-	2
Kagantsov I.M. et al., 2017 [12]	28	0 (0%)	2,4	14/14	0	-	107,5	2 (7,1%)
Andreev E.S. et al., 2017 [24]	39	0 (0%)	11 months	21/17	1	3,5	135	2 (5%)
Z Dokumcu et al, 2018 [5]	26	9 (34,6%)	5,5	14/10	1	3,9	-	0
Poddubniy I.V. et al., 2018	67	60 (89,6%)	7,5	31/26	8	4,65 ± 1,29	90,00 ± 36,82	0

in all groups. Thus, minimally invasive adrenalectomy should not be contraindicated in patients with adrenal gland tumor over 8 cm [10]. It is currently agreed that tumor dimension cannot be a contraindication for endoscopic approach in well-experienced surgeons [11].

In 2017, the results of laparoscopic adrenalectomy in children with adrenal neuroblastoma were analyzed in national literature. Considering the data of this multiple-center study, the authors concluded that minimally invasive resection of adrenal neuroblastoma is advisable if tumor volume does not exceed 120 cm³ and other contraindications are absent [12].

In our study, mean dimension of adrenal tumors was 4.65 ± 1.29 cm. The largest neoplasm (12 cm) was found in a 9-year-old patient with pheochromocytoma of the left adrenal gland.

Some authors consider retroperitoneoscopic approach to adrenal glands as an alternative to the most common transperitoneal approach [13, 14]. However, we used only laparoscopic lateral transperitoneal approach in our work. In our opinion, this method is the most rational, valuable for wide and free manipulation in the abdominal cavity, early vascular occlusion for prevention of probable release of hormones associated with dissection of the adrenal gland. Abdominal approach is valuable for examination of abdominal cavity, bilateral surgery via the same approaches and simultaneous procedures in patients with concomitant surgical diseases (hernia, paraovarian cysts). In our study, left-sided paraovarian cyst with a diameter of 2.0 cm was observed in a 12-year-old girl with left adrenal gland tumor (adenoma) during intraoperative examination of the abdominal cavity and small pelvis. Simultaneous resection was carried out without installation of additional trocars.

According to the world data, bleeding is the most common complication of laparoscopic adrenalectomy. Blood transfusions are performed in 1.9% of cases [5]. In

our study, bleeding associated with central adrenal vein stump failure occurred twice (on the right and on the left) that accounted 3% of complications among all operations. In both cases, additional clipping was required without conversion to open surgery [15]. Bleeding was moderate and did not require blood transfusions.

The advantages of minimally invasive approaches have long ceased to be a matter of debate. However, they acquire special significance compared to open surgery in patients with adrenal gland diseases especially in those with hypercorticism because advanced coagulation of subcutaneous fat is associated with increased risk of local infectious complications. For example, there are literature data on the incidence of this complication of 31.1% in the cases of abdominal approach, 26.7% in lumbar approaches and 9.7% in thoracic approaches [8]. It is worth noting that these events were absent in our sample.

Histological examination was carried out in 100% of cases. There were endocrine-active and inactive incidentalomas (22 tumors) diagnosed accidentally during clinical examinations or target diagnostics for non-endocrinological complaints. Resection of 8 pheochromocytomas (including 1 patient with bilateral pheochromocytoma) was performed in presence of endocrinologist in the operating theatre who carried out preoperative, intraoperative and early postoperative correction of hormonal therapy. Curation of patients with bilateral nodular adrenal lesion ($n=8$) was performed in a similar fashion.

Conclusion

Analysis of national and foreign reports devoted to minimally invasive surgical treatment of various adrenal gland neoplasms showed that our experience is quite unique regarding the number of endoscopic adrenalectomies in children with benign adrenal tumors. Moreover, our results are not inferior to the data of foreign authors.

Only 22 trials with sample size over 5 have been published in the foreign literature over the past 20 years. Only four of these reports included over 20 patients and two studies were multiple-center [5]. Moreover, most of patients in these studies were children with neuroblastoma.

We can summarize and emphasize all above-mentioned statements once again. Minimally invasive procedures in adult and pediatric surgery reduce duration of interventions, intra- and postoperative morbidity and length of hospital-stay. Moreover, endoscopic approach often becomes even more comfortable in patients undergoing resection of be-

nign adrenal gland tumors as anatomically deep structures. These procedures are valuable to avoid resection of small tumor through a relatively large incision [7]. Simultaneous surgical treatment of bilateral adrenal lesions and various simultaneous procedures are possible and performed via installation of additional trocar.

It must be emphasized once again that only team work of endocrinologist and surgeon at all surgical stages ensures effective care in these patients.

No conflict of interests to declare.

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