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CLINICAL SERIE

Adrenal tumors in pediatric patients treated with minimally invasive surgery

Tumores suprarrenales en pacientes pediátricos tratados mediante técnica quirúrgica mínimamente invasiva

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What do we know about the subject matter of this study?

Adrenal tumors in the pediatric population are mainly malignant neoplasms, with surgical resection as the suggested management. Recent reports suggest that laparoscopic adrenalectomy is a safe approach and should be the first line of treatment.

What does this study contribute to what is already known?

We report the surgical outcomes of a minimally invasive approach in the management of adrenal tumors in the pediatric population, through an observational, retrospective, and descriptive study of the interventions performed between 2012 and 2023 in a teaching hospital. In our series, the laparoscopic approach proved to be a safe technique, associated with short hospital stays, and the absence of perioperative morbidity and mortality.

Abstract

Adrenal tumors in children are frequently neoplastic and malignant, and surgical resection is the first management option. Minimally invasive surgery (MIS) has proven to be a safe management alternative and is suggested as a preferred alternative approach. **Objective:** To report the surgical outcomes of patients with adrenal tumors treated by MIS. **Patients and Method:** Retrospective descriptive study of a cohort of pediatric patients with adrenal gland tumors undergoing MIS in the period 2012 - 2023. **Results:** 15 MIS of the adrenal gland were performed in 14 patients, 60% were male, mean age 1.8 years (range: 5 days - 7 years). Histological diagnoses included neuroblastoma (n = 7), pheochromocytoma (n = 3), ganglioneuroma (n = 2), extrapulmonary sequestration (n = 1), mature teratoma (n = 1), and adrenal cortical neoplasm of uncertain malignant potential (n = 1). 14 adrenalectomies and one biopsy were performed, all via laparoscopic surgery, with no need for

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conversion to open surgery. No perioperative complications greater than Clavien-Dindo grade II were recorded; only 2 patients required red blood cell transfusions and there was no perioperative mortality. The mean tumor diameter was 3.7 cm. The median hospital stay was 1 day. The mean follow-up period was 29 months. **Conclusions**: The laparoscopic technique has proven to be a safe approach in pediatric patients with adrenal tumors, offering advantages in terms of hospital stay and perioperative complications.

Introduction

The adrenal gland is made up of cortex and medulla, structures with different embryological origins, each of which can give rise to different types of tumors[1]. Some may have clinical manifestations, but most frequently they are asymptomatic and detected incidentally in imaging studies. Adrenal masses in the pediatric population can develop spontaneously or in the context of a genetic syndrome[2]. Their treatment depends on clinical, radiological, and pathological factors, which determine the need for an initial biopsy, eventual pre- or postoperative chemotherapy, and the type of surgery to be performed[3].

Unlike in adults, adrenal tumors in the pediatric population are more frequently malignant neoplasms, and therefore, their surgical resection is recommended in most cases[2,4]. The most frequent in children is neuroblastoma of the adrenal medulla, while adrenocortical tumors such as carcinoma and adenoma are less frequent[4].

Currently, the minimally invasive approach is the first line of treatment for adrenal tumors in adults, having demonstrated benefits compared to the open technique such as faster recovery, shorter hospital stays, and better cosmetic outcomes[5], even in adrenal tumors larger than 6 cm[6].

However, given the lower incidence of adrenal tumors in the pediatric population, the use of laparoscopic surgery as a surgical approach in the resection and/or biopsy of adrenal tumors in children has not yet been widely disseminated. Recent reports regarding laparoscopic adrenalectomy suggest that this approach is safe and that, with well-established selection criteria, minimally invasive surgery should be the first line in the resection of small adrenal tumors[7].

The objective of this manuscript is to report the surgical outcomes of the minimally invasive approach in the diagnosis and management of adrenal tumors in the pediatric population.

Patients and Method

Observational, retrospective, and descriptive study. Data were collected from the institutional clinical records of 14 pediatric patients diagnosed with adrenal tumors, in whom a minimally invasive surgical intervention was performed, either for biopsy or resection of the lesion, between September 2012 and January 2023. Patients undergoing open surgery were excluded. Table 1 shows the demographic and clinical data of the series. This review was authorized by the institutional Ethics Committee.

The clinical, radiological, and pathological information on the tumors was collected from the clinical records, imaging reports, and anatomopathological results of the surgical specimens. The surgical approach, perioperative complications, length of hospital stay, and patient discharge status were collected from the corresponding surgical protocol and clinical summary.

The diagnosis of adrenal mass was made by radiological imaging, including ultrasound, CT scan, and MRI. Laterality, etiology-oriented characteristics, and unfavorable aspects of surgery were evaluated, being essential to rule out extensive invasion into vascular structures (figures 1 and 2). Depending on the case, some patients were evaluated by multidisciplinary teams, including oncology and/or pediatric endocrinology.

In all patients, a transperitoneal laparoscopic approach was performed, with the patient in a lateral decubitus position according to the location of the lesion, with a 30° elevation of the affected side, under general anesthesia, and orotracheal intubation. The camera was positioned through a periumbilical incision, along with two or three auxiliary trocars depending on the case. Monopolar and bipolar coagulation, as well as advanced hemostasis instruments, were used as required. The vascular structures were carefully identified and preserved during dissection. The surgical specimen was removed through a midline incision, either by direct extraction or using a retrieval bag, requiring incision enlargement in some cases.

Results

14 pediatric patients underwent 15 surgeries with a preoperative diagnosis of adrenal tumor. All underwent laparoscopic surgery for biopsy or resection of the lesion. The patient who required two procedures

Table 1. Demographic and clinical data of patients with adrenal tumors who underwent laparoscopic surgery

N°	Age at intervention (months)	Gender	Side	Surgery	Histopathology	Largest diameter (cm)	Perioperative complications	Length of stay (days)	Follow up (months)
1	50.8	M	Right	Resection	Pheochromocytoma	4.2	RC Transfusión	28	122.0
2	81. 5	M	Left	Resection	Pheochromocytoma	3	No	6	91.3
3	3.4	F	Right	Resection	Neuroblastoma	3.8	No	1	14.6
4	81.1	M	Left	Resection	Ganglioneuroma	5.7	No	1	17.0
5	30.4	F	Right	Resection	Adrenal cortical neoplasm	7	No	3	69.7
6	2.2	F	Left	Resection	Lung tissue with adenomatoid malformation	2.7	No	2	1.6
7	6.7	F	Right	Resection	Neuroblastoma	3.1	No	3	35.6
8	52.6	M	Right	Resection	Neuroblastoma	2.8	No	1	0.9
9	0.1	M	Right	Resection	Pheochromocytoma	4.3	No	1	32.9
10	5.75	M	Right	Resection	Neuroblastoma	2.6	No	1	32.7
11	11.3	M	Right	Resection	Ganglioneuroma	4.4	No	2	1.2
12	15.5	F	Left	Resection	Neuroblastoma	3	RC Transfusión	5	14.6
13	14.7	F	Right	Resection	Mature teratoma	4.9	No	1	0.7
14	6.4	M	Left	Biopsy	Neuroblastoma	4.3	No	1	6.1
15	3.4	M	Left	Resection	Neuroblastoma	4	No	1	4.2

M: male; F: female; RC: Red blood cells.

had a diagnosis of Von Hippel-Lindau syndrome and developed tumors in both adrenal glands.

The age of the patients at the time of surgery was on average 21 months, ranging from 0 to 82 months, and 60% were male.

Regarding the diagnosis of the adrenal lesion, in 53% of the cases, it was made in the context of clinical manifestations, including abdominal pain (n =4), fatigue (n = 1), early pubertal development (n = 1), and study of hypertension (n = 1). In the rest of the patients, the diagnosis was made as a finding after an imaging study because of another cause.

The estimated preoperative size of the tumor was measured by imaging, with a mean largest diameter of 3.7 cm, with a range of 2.7 - 5.7 cm. Regarding the laterality of the adrenal mass, 60% were on the right side, while the remaining were on the left.

Patients with functioning tumors were evaluated by multidisciplinary teams that included an endocrinologist, nephrologist, and anesthesiologist. Preoperative preparation with beta-blockade and/or alpha-blockade was performed in all cases. During the intraoperative period, only one patient required the use of a nitroprusside pump for blood pressure management.

Of the total series studied, 14 resections and one biopsy were performed. No perioperative complications greater than grade II on the Clavien-Dindo scale were recorded and only 2 patients required red blood cell transfusion. There was no mortality associated with the surgical technique.

The anatomopathological reports on the etiology of the adrenal tumor showed that most corresponded to neuroblastomas (n = 7) and pheochromocytomas (n = 3), followed by ganglioneuromas (n = 2), extrapulmonary sequestration (n = 1), mature teratoma (n = 1), and adrenocortical neoplasm of uncertain malignant potential (n = 1).

The median hospital stay was 1 day, ranging from 1 to 28 days. The patient with the longer hospital stay had a viral respiratory complication that prolonged his need for care. No perioperative mortality was recorded. The median follow-up was 29 months and no local recurrences have been detected at the date of this data review.

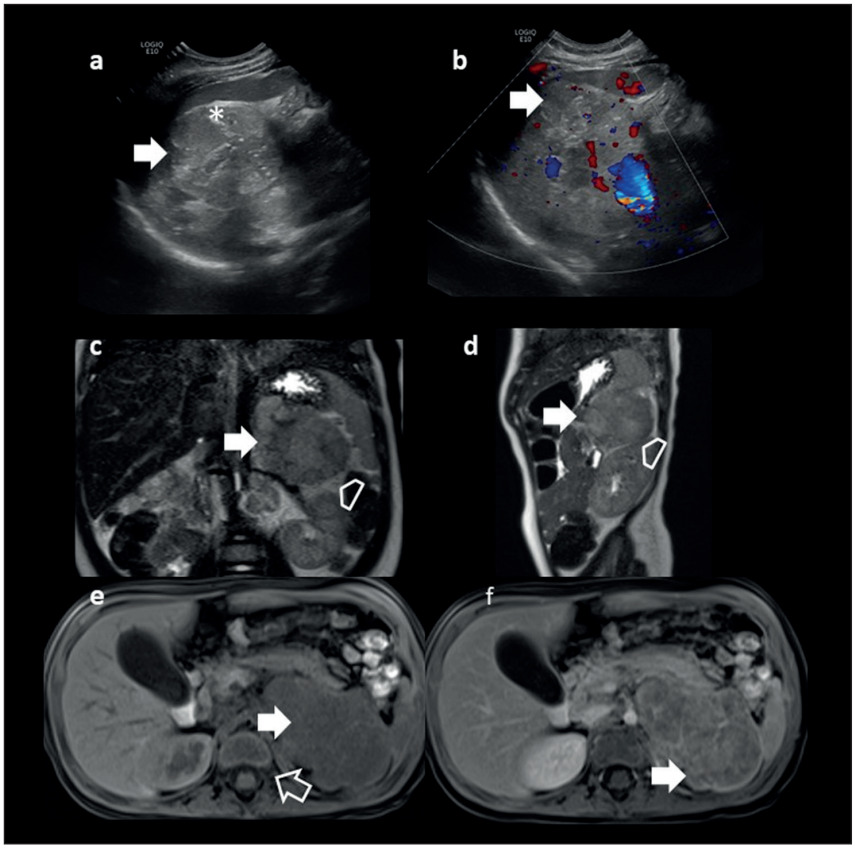

Figure 1. 1a y 1b. Abdominal ultrasound showing a solid lesion located in the left adrenal region (solid arrow) that presents hyperechoic foci inside compatible with calcifications (asterisks) and vascular flow in the Doppler study. 1c, 1d, 1e, 1f. Magnetic resonance image in coronal (c) and sagittal (d) T2-weighted sequences, and axial (e and f) T1-weighted pre- and post-contrast sequences respectively; a solid lesion (solid arrow) is observed hyperintense with the muscle in T2-weighted sequences and hypointense in T1-weighted sequences with progressive enhancement after the use of paramagnetic contrast (unfilled arrow), without evidence of invasion of the neuroforamen and with a cleavage plane with the kidney (arrowhead). These images are compatible with a neuroblastoma.

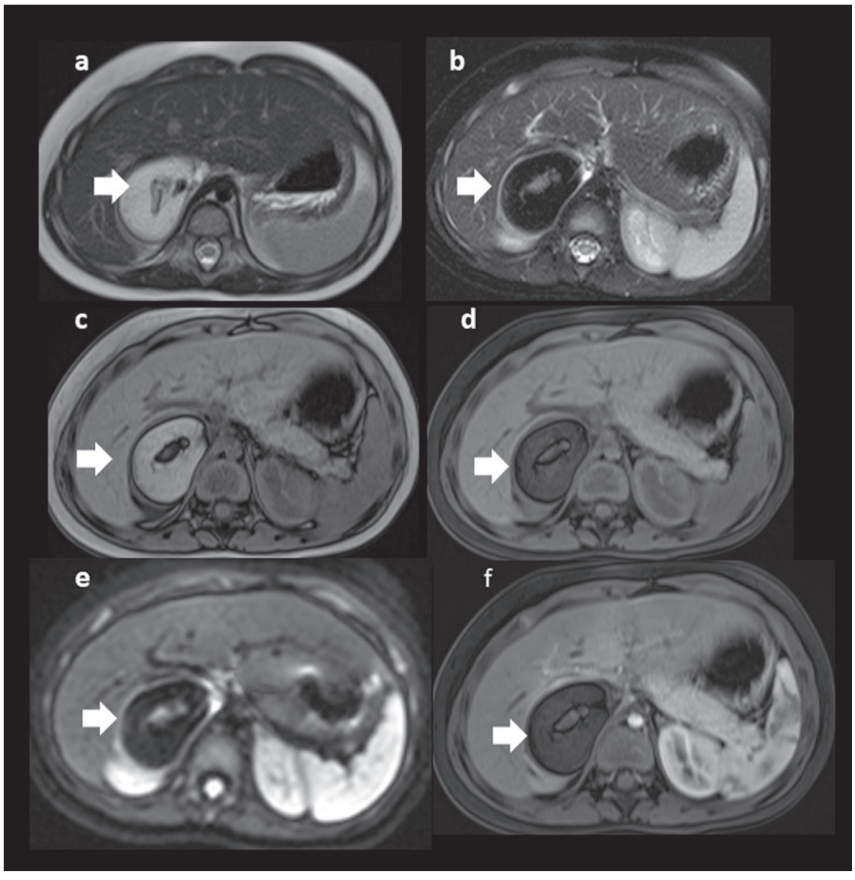

Figure 2. Abdominal MRI showing a solid lesion located in the right adrenal region that is hyperintense (solid arrow) on T2-weighted (a) and T1-weighted sequences (d) and presents a drop in signal on the suppressed sequences. fat (b and c), which translates the presence of macroscopic fat. It does not present diffusion restriction (e) or significant impregnation in the post-contrast sequences (f). Histopathology confirmed a mature teratoma.

Discussion

There is growing evidence in pediatric surgery supporting the benefits of the minimally invasive approach compared to open surgery. Recently, the working group of Yao et al. compared the long-term prognosis in 37 children with adrenal neuroblastoma who underwent either laparoscopic adrenalectomy or open surgery. They observed equivalent recurrence and mortality rates in both series, suggesting that for small tumors without vascular invasion, laparoscopic surgery is considered a safe option and the first-line management of adrenal neuroblastoma when performed by experienced surgeons[8]. This experience is complemented by a case review published in 2015 by Rodriguez et al. which already indicated favorable laparoscopic surgery outcomes, highlighting low morbidity and mortality rates, quick recovery, shorter hospital stays, and satisfactory cosmetic outcomes[9]. In this study, no morbidity or mortality associated with the surgical technique was observed, while the hospital stay was short.

In reports of uncommon adrenal tumors, such as pulmonary sequestration, a conservative approach has been proposed, involving patient observation, primarily based on the low complication rate associated with this condition. However, due to the low morbidity associated with minimally invasive surgery and the importance of establishing an accurate etiological diagnosis, the option of performing early resection of such lesions instead of adopting an expectant approach has been proposed[10].

The age of presentation in patients with neuroblastoma is between 1 and 5 years in 90% of cases, occurring during the first months of life in up to 50%[1], while children diagnosed with pheochromocytoma have a later presentation. The age composition in our series is consistent with that described in the literature, with an average age at intervention of 1.4 years in patients with neuroblastoma and 3.6 years in pheochromocytoma.

Traditionally, the management of adrenal tumors in pediatrics has been a surgical intervention. In this regard, it has been suggested that the determining factors that influence the choice of approach include the etiology of the tumor, its size, and its relationship with nearby vascular structures[4]. In the case of neuroblastoma, there is no consensus on the eligibility criteria for the indication of minimally invasive surgery, however, it is suggested that it could be adequate for small tumors (diameter < 5 cm) and without image-defined risk factors (IDRF), such as tumor invasion of blood vessels. However, in a multicenter European study where more than 70 pediatric patients were included, no correlation was observed between tumor size and unfavorable surgical outcomes, while the invasion of adjacent structures prolonged surgical time and increased the risk of rupture and bleeding, showing no relation to the need for conversion to open surgery, post-surgical complications, or recurrences[11]. On the other hand, Traynor et al. suggested that in children with non-neuroblastoma tumors with a low probability of malignancy, the use of laparoscopic adrenalectomy can be performed with low morbidity, that is, no intraoperative complications, less blood loss, and shorter hospital stay when compared to open surgery[12].

Given the teaching hospital context where this work was carried out, it is interesting to report on a multicenter study published in 2021 that compared laparoscopic surgical resection of adrenal neuroblastomas performed by pediatric surgical residents under the supervision of expert surgeons with those performed exclusively by the latter, revealing no significant differences in terms of tumor size, surgical time, and amount of bleeding between the two groups[13].

The case series analyzed in this study focuses exclusively on the surgical laparoscopic approach, with favorable results in terms of surgical morbidity and mortality. The main limitation lies in the retrospective design of this work and with it the limited availability of data collected from medical records.

The experience of the use of the minimally invasive approach in children is mainly based on the report of small retrospective series, in contrast to the extensive evidence reported in adults[14]. In addition, given that the current preference for the laparoscopic approach in pediatric surgeons in Latin America is unknown and few reports have been published in this continent[7], the contribution of this publication lies in the dissemination of experience and outcomes in minimally invasive techniques in the management of adrenal tumors in pediatric patients in a teaching hospital in Chile.

In conclusion, in this series of patients diagnosed with adrenal tumors, the minimally invasive surgical approach, either for biopsy or resection in selected cases, proved to be a safe technique in the pediatric population. This surgical modality has evolved in the last decades, consolidating as an alternative to open surgery, offering benefits in terms of hospital stay, morbidity, mortality, and cosmetic outcomes.

Although laparoscopic surgery has proven to be effective and safe in the treatment of pediatric adrenal tumors, it is essential to remember that each case must be evaluated individually and discussed by a multidisciplinary medical team. The surgeon's experience and appropriate case selection, based on tumor size, etiology, and possible complications, are crucial to the success of this surgical approach.

The contribution of clinical reports from different health institutions is essential to enrich the knowledge of the benefits and complications associated with the

laparoscopic technique in the treatment of adrenal tumors in the pediatric population, as it contributes to the strengthening of medical evidence and will allow further progress in the field of minimally invasive surgery, ensuring better outcomes for patients presenting with these pathologies.

Ethical Responsibilities

Human Beings and animals protection: Disclosure the authors state that the procedures were followed according to the Declaration of Helsinki and the World Medical Association regarding human experimentation developed for the medical community.

Data confidentiality: The authors state that they have followed the protocols of their Center and Local regulations on the publication of patient data.

Rights to privacy and informed consent: This study was approved by the respective Research Ethics Committee, which, according to the study's characteristics, has accepted the non-use of Informed Consent.

Conflicts of Interest

Authors declare no conflict of interest regarding the present study.

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References

1. Balassy C, Navarro OM, Daneman A. Adrenal masses in children. Radiologic Clinics of North America. 2011;49(4):711-27. doi:10.1016/j.rcl.2011.05.001.
2. Kastenberg ZJ, Scaife ER. Adrenocortical tumors in children. Seminars in Pediatric Surgery. 2020;29(3):150927. doi:10.1016/j.sempedsurg.2020.150927.
3. Mihai R. Rare adrenal tumors in children. Seminars in Pediatric Surgery. 2014;23(2):71-5. doi:10.1053/j.sempedsurg.2014.03.004.
4. Emre ş, Özcan R, Bakır AC, et al. Adrenal masses in children: Imaging, surgical treatment and outcome. Asian Journal of Surgery. 2020;43(1):207-12. doi:10.1016/j.asjsur.2019.03.012.
5. Öz B, Akcan A, Emek E, et al. Laparoscopic surgery in functional and nonfunctional adrenal tumors: A single-center experience. Asian Journal of Surgery. 2016;39(3):137-43. doi:10.1016/j.asjsur.2015.04.009.
6. Öz B, Cücük Ö, Gök M, et al. Laparoscopic transperitoneal adrenalectomy for adrenal tumours of 6 cm or greater. Journal of Minimal Access Surgery. 2023;Publish Ahead of Print. doi:10.4103/jmas.jmas_217_22.
7. Oesterreich R, Varela MF, Moldes J, et al. Laparoscopic approach of pediatric adrenal tumors. Pediatric Surgery International. 2022;38(10):1435-44. doi:10.1007/s00383-022-05188-x.
8. Yao W, Dong K, Li K, et al. Comparison of long-term prognosis of laparoscopic and open adrenalectomy for local adrenal neuroblastoma in children. Pediatric Surgery International. 2018;34(8):851-6. doi:10.1007/s00383-018-4294-5.
9. Rodríguez PI, Gómez-Chacón JV, Rodríguez LC, et al. Laparoscopic adrenalectomy: advantages of the minimally invasive approach. Cir Pediatr. 2016;29(4):162-5.
10. Lemus Marcenaro M, Campos Varas JM, Vuletin Solís JF, et al. Diagnóstico diferencial y Cirugía Mínimamente invasiva de un tumor suprarrenal de Hallazgo Antenatal. Revista Chilena de Pediatría. 2019;90(3):321. doi:10.32641/rchped.v90i3.823.
11. Fascetti-Leon F, Scotton G, Pio L, et al. Minimally invasive resection of adrenal masses in infants and children: Results of a European multi-center survey. Surgical Endoscopy. 2017;31(11):4505-12. doi:10.1007/s00464-017-5506-0.
12. Traynor MD, Sada A, Thompson GB, et al. Adrenalectomy for non-neuroblastic pathology in children. Pediatric Surgery International. 2019;36(2):129-35. doi:10.1007/s00383-019-04589-9.
13. Zenitani M, Tsukada R, Masahata K, et al. Safety and feasibility of laparoscopic resection of neuroblastoma without image-defined risk factors performed by pediatric surgical trainees: A multicenter comparison study. Journal of Laparoendoscopic & Advanced Surgical Techniques. 2021;31(8):954-8. doi:10.1089/lap.2020.0957.
14. Dokumcu Z, Divarci E, Ertan Y, et al. Laparoscopic adrenalectomy in children: A 25-case series and review of the literature. Journal of Pediatric Surgery. 2018;53(9):1800-5. doi:10.1016/j.jpedsurg.2017.11.055.