



# Laparoscopic treatment of adrenal masses in children: Report of two cases and review of literature

Antonio Orofino, Cosetta Maggipinto, MariaPaola Lanzillotto, Michele D'Amato, Massimo Ronzini, Guglielmo Paradies

## ABSTRACT

Laparoscopic adrenalectomy has become a common alternative to open surgery for the resection of adrenal lesions in adults: The advantages are to provide better exposure of the adrenal gland, diminish soft tissue dissection, decrease morbidity and postoperative pain; however, reporting on the laparoscopic adrenalectomy in paediatric patients has been limited. We present two cases of laparoscopic adrenalectomy performed at our institution in two children, for left adrenal neuroblastoma in a first patient with opsomyoclonus syndrome, and for a right incidentaloma in the second case. According to recent literature, our experience has demonstrated that the laparoscopic adrenalectomy is a feasible procedure in children with small, well-circumscribed adrenal masses: It can be used a safety to treat suspected benign and malignant adrenal masses in children, with minimal morbidity and short hospital stay. The lateral trans-peritoneal approach offers optimal visualisation and good outcomes in terms of minimal discomfort, rapid recovery and excellent cosmesis. However, in the paediatric field, the number of patients is limited, making the learning curve longer.

**Key words:** Adrenal tumour, laparoscopic adrenalectomy, minimally invasive surgery, paediatrics

## INTRODUCTION

Recently, minimally invasive surgery (MIS) has evolved as a standard technique for the management of numerous abdominal and thoracic pathologies in

children. The advantages are an overall decrease in pain and need for post-operative narcotics, reduced morbidity, shorter hospital stay and better cosmesis. Since Gagner *et al.* described the first laparoscopic adrenalectomy in 1991,<sup>[1]</sup> laparoscopic approach has become the standard of care for most benign adrenal lesions in adults, instead the role of laparoscopic adrenalectomy in children is still debated. However, reporting on the laparoscopic for adrenal disease in children has been limited. A retrospective analysis was performed involving two patients with radiologically confirmed adrenal masses who underwent laparoscopic resection in our institution in last 2 years.

The aim of this report is to evaluate the role of MIS in the surgical management of paediatric adrenal masses, reporting our experience, discussing the technical aspects and surgical outcomes and review the current literature focusing some questions still debated.

## CASE REPORTS

### First case

An 8-year-old female was referred by neurologist colleagues to our institute with a diagnosis of left adrenal neuroblastomas in a patient with the opsomoclonic syndrome. This is a rare paraneoplastic paediatric syndrome, also known by the name of “dancing eyes syndrome” characterised by neurological disorders, such

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**Cite this article as:** Orofino A, Maggipinto C, Lanzillotto M, D'Amato M, Ronzini M, Paradies G. Laparoscopic treatment of adrenal masses in children: Report of two cases and review of literature. Afr J Paediatr Surg 2016;13:98-102.

Department of Science and Pediatric Surgery, Operative Unit of Pediatric Surgery, Policlinico-Giovanni XXIII University Hospital, Bari, Italy

#### Address for correspondence:

Dr. Michele D'Amato, Department of Science and Pediatric Surgery, Operative Unit of Pediatric Surgery, Giovanni XXIII University Hospital, V. Amendola, 207, 70125, Bari, Italy.  
E-mail: mikdam@virgilio.it

as opsoclonus, a rapid eyes movements in all planes of space, ataxia, myoclonus, irritability and psychomotor regression. Another characteristic of the pathology is that the peripheral neuroblastic tumour associated with this syndrome generally is poorly aggressive, with favourable histology and biology and high rate of 5-years survival. The abdominal computerised axial tomography computed tomography (CT) scan demonstrated that the left adrenal gland presented a solid lesion of diameter of 30 mm with progressive and dishomogeneous enhancement [Figure 1]; the metaiodobenzylguanidine (MIBG) scintigraphy also showed a pathological fixation of MIBG in the left adrenal gland corresponding to the formation detected by CTS.

A laparoscopic left adrenal resection with lateral transperitoneal approach was performed.

The patient was placed in a lateral decubitus position, over a right flank lift, so as to maximally expose the space between the costal margin and the iliac crest. The procedure started inserting with open technique a 10 mm port for 0° camera, along the anterior axillary lines, 2 cm below the costal arch; the abdominal cavity was insufflated with carbon dioxide to a pressure of 10 mm Hg, so under vision one 12 mm port was placed along the posterior axillary lines below the costal arch and additional two 5 mm trocars in epigastrio and along the left midclavicular line. The splenic flexure of the colon, the spleen and the tail of the pancreas were mobilised and retracted medially *en bloc*, incising lateral peritoneal attachments. The dissection of the retroperitoneal space, the mobilisation of the adrenal gland and the control of the blood supply were accomplished using 5 mm bipolar forceps. The left adrenal vein, once mobilised, was clipped and divided.

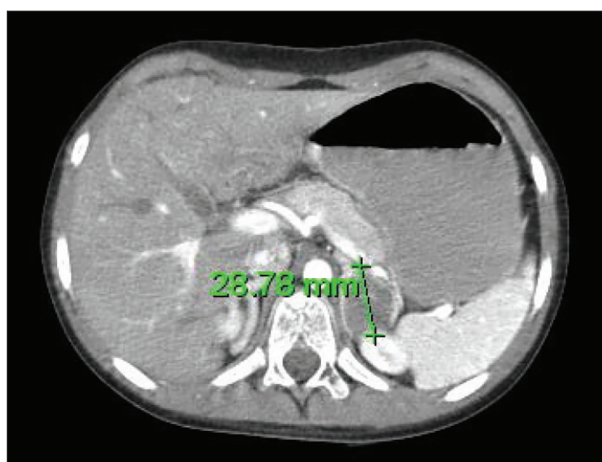


Figure 1: Contrast-enhanced abdominal computed tomography scan shows in the left adrenal gland a solid lesion of diameter of 30 mm, with progressive and dishomogeneous enhancement

The adrenal gland completely free from surrounding tissues was placed into a specimen retrieval bag and removed from the abdominal cavity through the 12 mm port access. The retroperitoneal space was examined for any evidence of bleeding or lymph node swelling; a drain was placed into the retroperitoneum to evacuate any fluid or blood then removed 24 h postoperatively. The operative time was 130 min, there were no intra-operative or post-operative complications [Figure 2].

Microscopic examination revealed an adrenal neuroblastoma, without *N-Myc* amplification.

The hospital stay was uneventful, and the patient was discharged home on the 4<sup>th</sup> post-operative day and started to neurological care.

### Second case

A 15-year-old male presented to our Institute with a diagnosis of right adrenal incidentaloma; by definition “Incidental adrenal masses” are those that are found when testing is being performed for other reasons and a mass is discovered in the adrenal gland. In this case, a magnetic resonance imaging (MRI) performed 1-year before, in the follow-up for immature teratoma of the left testis, surgically treated at 6-month-old, had demonstrated a right adrenal lesion with a diameter of 33 mm. The MRI control performed after 1-year showed a volumetric increase of the lesion which had a diameter of 4, 3 cm [Figure 3].

The fundus oculi examination was normal, tumour markers, plasma and urinary catecholamines were in the normal range, excluding suspect that was a hormonally active mass.

The clinical and instrumental investigations confirmed the biological nature of the lesion directing our decision-making towards a laparoscopic approach; indeed the lesion presented: 4 cm of size, regular margins and form, homogeneous appearance, the

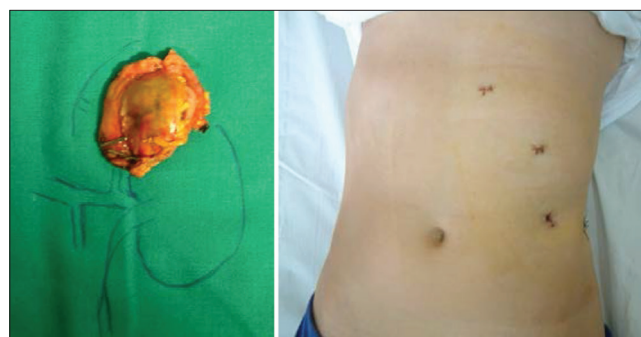


Figure 2: The excised left adrenal gland (left image). Location of access points (right image)

absence of calcifications, no infiltrative disease.<sup>[2]</sup> Laparoscopic resection with lateral trans-peritoneal approach was performed through four ports, two of 12 mm and two of 5 mm, positioned below the costal margin along the anterior and posterior axillary lines and in epigastrio [Figure 4]. The posterior peritoneum was incised below the liver lifted above to expose the adrenal gland and the inferior vena cava. Dissection was done along the medial border of the adrenal gland to identify the main adrenal vein that was sectioned between clips.

Small vessels were cauterised and sectioned. The gland was retrieved intact from abdominal cavity after placement in an endo-bag, through one 12 mm port access. The operative time was 150 min. There were no intra-operative or post-operative complications. Intra-operative blood loss was negligible. Post-operative course was uneventful, and the patient was discharged home 72 h after the procedure. Pathological examination revealed an adrenal with extensive bleeding site long time [Figure 5].

## DISCUSSION

Since Gagner *et al.* described the first laparoscopic adrenalectomy in 1991 this approach has become the gold standard, and a common alternative to open surgery for the resection of most benign adrenal masses in adults, while limited literature exists on the laparoscopic adrenalectomy in paediatric patients. Minimally invasive technique to perform adrenalectomy includes trans-peritoneal approach (lateral or anterior) and retroperitoneoscopic approach (RPA) (lateral, posterior or anterior).

At present, lateral trans-peritoneal adrenalectomy (LTA) is the most widely utilised procedure that offers the advantage of a wider and efficient working space with optimal visualisation and operator's familiarity with the anatomy. The RPAs offer direct access to the adrenal gland avoiding mobilisation of the bowel, the spleen or of the liver, but provide a significantly smaller working space with poor exposure of the vasculature, especially on the right side, with increased risk of bleeding and open conversion; moreover, the reversed orientation of the kidney and hilum make this approach difficult to master.<sup>[3]</sup>

In the paediatric population, RPA rarely has been described; recently Eassa *et al.* presented an anterior approach to RPA in two children with incidental right adrenal masses, confirming the advantageous exposure

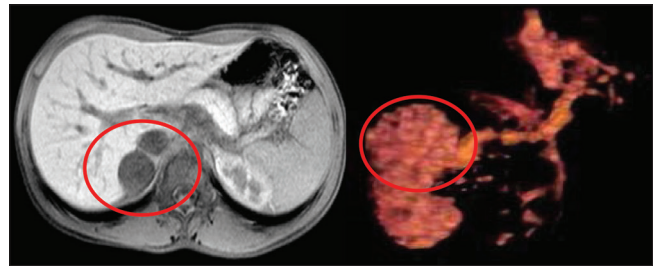


Figure 3: Magnetic resonance imaging: The adrenal lesion and processing three-dimensional of images

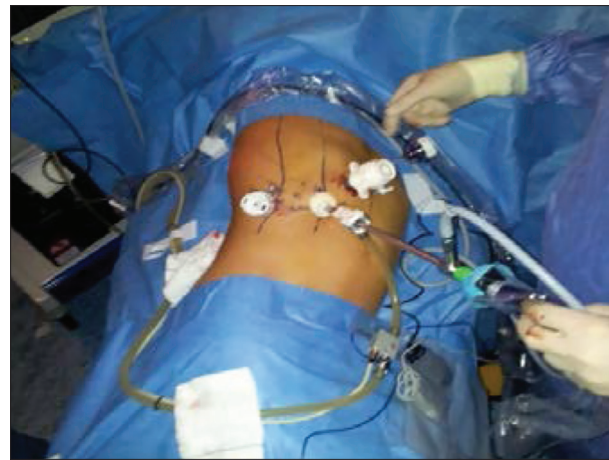


Figure 4: Port placement sites



Figure 5: The excised right adrenal gland

provided by this technique advocate in adults by Zhang, and its applicability in children with large masses even on the right side.<sup>[4]</sup>

As already mentioned, the majority of the literature on laparoscopic adrenalectomy is based on adults cohort, and this is easily explained considering the relative rarity of benign adrenal disease in children since the adrenal gland is the principal site of abdominal localisation of neuroblastoma, the most solid tumour in infancy.



Articles and abstracts published in English from 1992 to 2013 were searched using Cochrane controlled trials register and Medline: 201 publications were found using the key words “Laparoscopic adrenalectomy in children” and “Minimally invasive adrenalectomy in children.”

The analysis of those publications showed that during a first period between 1992 and 2003, there were 82 publications about laparoscopic adrenalectomy exclusively in adults, while there was no evidence of paediatric studies in literature. From 2003 to 2013, there was a significant increase of publications (119) that evaluate the role of MIS adrenalectomy also in children with malignancies. Indeed, 52 of 119 publications were related to the paediatric age: Numerous of these reports have described successful laparoscopic removal of the adrenal gland in children, and it is actually an accepted procedure to treat children with adrenal tumours; however, most of these studies included retrospective cohort studies, only a few prospective cohort studies were performed, missing randomised controlled trials or controlled clinical trials and long-term follow-up, so the advantages and disadvantages of laparoscopic in paediatric abdominal neuroblastoma still need to be defined, and the role of MIS in paediatric cancer is a matter of debate, and its use is limited to lesions that are small, well-circumscribed, without local infiltration or metastasis.<sup>[5]</sup>

Iwanaka *et al.* have shown that laparoscopic resection of solid tumours, such as neuroblastomas, is feasible.<sup>[6]</sup> The study by Romano *et al.* comparing open to laparoscopic adrenalectomy in children but is not randomised.<sup>[7]</sup>

A prospective study conducted by Metzelder *et al.*, including 276 children with abdominal/retroperitoneal and thoracic cancer showed how MIS was successfully employed in 77% of operations with a conversion rate <5%.<sup>[8]</sup>

In selected patients, laparoscopic surgery is a valid technique for resection of solid paediatric malignancies, but the numbers reported are still limited: Warmann *et al.* operated on nine patients and converted in 5;<sup>[9]</sup> Iwanaka *et al.* On 6 with 2 conversions,<sup>[10]</sup> De Lagausie *et al.* resected 9 adrenal neuroblastoma and converted in 1, because of adhesions to renal vessels.<sup>[11]</sup>

Kelleher *et al.* in a retrospective review published in 2013 comparing long-term oncologic outcomes in 79 patients with adrenal neuroblastoma undergoing open versus MIS adrenalectomy, confirms that laparoscopic resection of adrenal neuroblastoma is feasible and offers

equivalent recurrence E-mortality rates in patients who meet selection criteria.<sup>[12]</sup> The feasibility of laparoscopic approach for the management of adrenal incidentalomas is validated by various retrospective studies. In our second case, as it was an incidental finding of adrenal lesion, the indication for ablative surgery was performed according to current literature. In literature reviewed, the diagnostic and therapeutic approaches of adrenal incidentalomas are controversial, but, according to the most of authors, laparoscopic approach is the preferred option for incidentalomas with size equal to or >4 cm, without suspected malignancy.<sup>[13,14]</sup> A Chinese study, including 143 patients, has shown as adrenal incidentalomas clearly benign (with capsula, regular margins and form, in the absence of calcifications, without locoregional invasion and metastasis) are successfully treated with MIS.<sup>[15]</sup>

Our experience showed that more experience gained at laparoscopic has allowed to remove adrenal masses with MIS. Because laparoscopic adrenalectomy is considered as an advanced technique, it is necessary a long learning curve, understood as the average number of procedures that surgeon has to perform without risks. In addition to in the paediatric field, the number of patients is limited, making the learning curve longer.<sup>[16,17]</sup>

## CONCLUSION

The advances in MIS techniques have provided an alternative to open procedure in infants and children, resulting in decreased surgical stress and improved post-operative morbidity. The development of laparoscopic surgery has extended its uses to include adrenalectomy in children.<sup>[7]</sup> The advantages of laparoscopy are to provide better exposure of the adrenal gland, diminish soft tissue dissection, decrease morbidity and post-operative pain, resulting in earlier ambulation, shorter hospital stay and faster return to normal activity. Our experience confirms, according to the literature, that laparoscopic adrenalectomy is a safe, effective technique in children with small, well-circumscribed adrenal masses. Both laparoscopic and RPA are feasible, with the choice being dictated by the experience of the surgeon; the most common approach is the LTA that offers an efficient working space with optimal visualisation optimal and excellent outcomes in terms of minimal discomfort, rapid recovery and good cosmesis.<sup>[18,19]</sup>

Laparoscopic adrenalectomy can be used if the selection of cases is rigorous and the operations are performed by well-trained laparoscopic surgeons, with care to maintain the principles of cancer surgery. Therefore,

more experience and longer follow-up are necessary to validate this procedure.<sup>[20]</sup>

### Financial support and sponsorship

Nil.

### Conflicts of interest

There are no conflicts of interest.

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