

Paraplegia after thoracotomy: a single center experience with pediatric patients and a review of the literature



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AIM: Paraplegia is an infrequent although fearsome complication of anesthesia and surgical procedures, such as epidural anesthesia and thoracotomy. It may occur in both adults and children and a medullary lesion may be confirmed by magnetic resonance imaging, rather than computed tomography. The aim of this study is to describe the experience of two pediatric tertiary centers, contextualizing it with the other cases reported in literature.

MATERIAL AND METHODS: We reported three pediatric cases of post-operative paraplegia in oncological patients, focusing on the potential causes and underlying the possible strategies to prevent this complication.

RESULTS: From our study, two principal features emerged: 1) Epidural anesthesia may expose children to a greater risk of spinal cord permanent damage due to the execution of the procedure under general anesthesia, which deprives the anesthesiologist of an important feedback about the position of the device; 2) In thoracotomy, the risk of paraplegia tends to increase along with the proximity to the costo-vertebral angle, especially if electrocautery or hemostatic materials are used.

CONCLUSIONS: A prompt post-surgical neurological status routine assessment in pediatric patients undergoing epidural anesthesia or thoracotomy should be mandatory and, associated with the correct imaging study, may lead to the most appropriate therapeutic pathway and to a better prognosis.

KEY WORDS: Anesthesia, Children, Epidural, Thoracotomy, Paraplegia

Introduction

Paraplegia is an infrequent and unfortunate complication of thoracic surgery but also of epidural anesthesia that can occur in both adults and children. The literature emphasizes the association between paraplegia and epidural anesthesia, although underlining its rarity with an estimated incidence of 0,003%^{1,2}.

It can be caused by various mechanisms: epidural hematoma, direct spinal cord trauma, spinal cord infarction, neurotoxicity by accidental subarachnoid injection³.

Furthermore, paraplegia after thoracotomy has a reported incidence of 0.08% in patients not receiving thoracic epidural anesthesia, suggesting the presence of other underlying causes⁴.

The gold-standard for the diagnosis of this undesirable complication is magnetic resonance imaging (MRI), rather than Computed Tomography (CT)⁵.

In our study, we describe our experience in two different pediatric tertiary centers and we report a brief review of the literature, underlining the importance of prevention and early detection of this undesired complication.

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Case Reports

We present two cases of thoracotomy for mediastinal neuroblastoma (NBL) and one case of rib excision for

TABLE I - Summary table of the cases reported in our study

Case Number	1	2	3
Year of Diagnosis	1999	1999	2019
Age at diagnosis(years)	1	3	9
Sex	F	M	F
Diagnosis	Thoracic left neuroblastoma	Thoracic left ganglioneuroblastoma	Osteosarcoma
Cause of paraplegia	Thoracotomy	Decompression laminotomy	Epidural anesthesia
Surgery	Tumor excision	Excision of the intraspinal portion of the tumor	Sixth right rib surgical margins excision
Neurological level loss	T9-T10	T2	T5-T6
Long-term outcome	Partial improvement	Improvement	No improvement
Notes			Osteosarcoma induced by radiation therapy for rhabdomyosarcoma

osteosarcoma. In the first two cases paraplegia was caused by a lesion of the arteria magna of Adamkiewicz; for the third one, a certain underlying pathogenetic mechanism has not been proven so far, although we strongly suspect a correlation with epidural anesthesia (Table I).

CASE N. 1

A 1-and-half-year-old girl with walking difficulty was referred to Bambino Gesù Children's Hospital, in Rome. A total-body CT-scan showed a large left mediastinal mass with intraspinal extension (as a dumbbell lesion) from T8 to T11, causing an spinal cord compression leading to the clinical manifestation. The histological diagnosis, obtained by open biopsy, was neuroblastoma.

After four-cycles of D-CECaT chemotherapy (desferoxamine 150 mg/Kg, carboplatin 1.000 mg/m², etoposide 300 mg/m², cyclophosphamide 1.200 mg/m², thiotepa 30 mg/m²) and a 30% reduction of the mass, the infant underwent surgery and the left-sided tumor was excised en bloc along with four large spinal roots and four intercostal arteries that were stretched and infiltrated by the tumor. In order to achieve a nearly total excision of the mass, a fifth right spinal root was transected.

By the second postoperative day, the child showed loss of sensibility up to the xiphoidal level. After 15 days, the patient was submitted to another intervention to remove the intraspinal part of the tumor; a 1-cm-long segment of the spinal cord at the level of T10 appeared pale and grayish. After surgery, four more cycles of D-CECaT chemotherapy were administered.

After the postoperative treatment (at 24-months follow-up), the girl was actually free of disease with a good leg muscles tone and preserved anal sphincter function, although she retained urine and had to go through manual bladder voiding.

CASE N. 2

A 3-years-old boy was referred to the Instituto Portugues de Oncologia Francisco Gentil, in Lisbon, with a history of bronchitis and fever. Thoracic CT-scan showed a large tumor of the left costo-vertebral gutter, with several calcifications and invasion of the epidural space, without widening of the intervertebral foramina. The alleged diagnosis, based on MIBG-scans and catecholamine dosage, was neuroblastoma and a decompression laminotomy was performed, with removal of all the intraspinal portion of the tumor. Histopathology showed a ganglioneuroblastoma.

On the second postoperative day, the child showed urinary and fecal incontinence, with painful analgesia at middorsal level, lower limbs hypotonia and withdrawal reflexes after painful feet stimulation. Suspecting an acute high dorsal pyramidal lesion, the child underwent immediate revision of the laminotomy (which turned into a laminectomy); during the procedure, a hemostatic pledget (positioned during the previous surgical procedure) was removed. Nevertheless, postoperative, the neurological examination did not show significant improvement, indicating persistent spinal cord damage at the level of T2. One month later, despite the use of high-dose corticosteroids, the clinical situation was unchanged.

Neurosurgical procedures were followed by three cycles of OPEC chemotherapy (vincristine 50 mg/kg, cyclophosphamide 200 mg/kg, cisplatin 20 mg/kg, and teniposide 50 mg/kg), a left thoracotomy (to remove the remaining tumor, which was adherent to the posterior middle thoracic wall and infiltrating the vertebral foramina) and six cycles of CYVADIC chemotherapy (cyclophosphamide 500 mg/m², decarbazine 250 mg/m², doxorubicin 50 mg/m², and vincristine 1.5 mg/m²).

At five-years follow-up, the patient was cancer-free, but showed no sensibility from the waist down (approximately from T9) and needed occasional bladder catheterization. MRI showed an extensive myelomalacia area (between T3 and T7) with cavitation.

CASE N. 3

A 1-year-old girl affected by rhabdomyosarcoma of the scapula was treated in Bambino Gesù Children's Hospital, in Rome. She underwent chemotherapy (protocol EpSSG 2005), radiotherapy and surgical excision. Nine years later, she presented a radiation-induced tumor of the VI rib; an open biopsy was performed and the histological diagnosis was osteosarcoma. The tumor was then surgically removed with no complications. However, the postero-lateral margin of the excised rib showed microscopic neoplastic involvement at histology; therefore, removal of the remaining portion of the rib was indicated. Preoperative anesthetic evaluation of the patient revealed no additional risk factors.

After induction of anesthesia, an intraspinal catheter was placed at T4 by a pediatric anesthesiologist and a continuous perineural infusion of ropivacaine 2% was started (0.2 mg/Kg/h), after 6 ml bolus of ropivacaine 0.375%. The surgical procedure consisted of removal of the postero-lateral portion of the sixth rib, with biopsy of the bone near the costo-vertebral joint, by thoracotomy (at the sixth intercostal space) and bleeding was controlled only by bone wax and oxidized and regenerated cellulose (tabotamp®).

Immediately after the procedure, the patient complained loss of sensibility and strength in her legs; neurological examination confirmed complete motor and sensory loss up to T4. Perineural infusion was stopped and urgent MRI was performed, highlighting an edema of the spinal cord at T5-T6. Electroneurography showed absence of sensory and motor evoked potentials.

A course of high-dose dexamethasone and rehabilitation program were started, with sensitive improvement at the T6-10 level and partial improvement of autonomy in the postural maneuvers, although intermittent bladder catheterization was still needed.

Two months after surgery, a selective angiography showed a normal flow in the arteria magna of Adamkiewicz and no improvement was observed on MRI.

Discussion

Paraplegia is a rare complication of thoracic surgery of adult and pediatric patients, occurring in 0.003% of cases^{1,2}. In a recent prospective audit of 10000 pediatric patients with continuous epidural analgesia infusions, only one case of neurological deficit was found⁶. Various mechanisms have been reported in order to explain this event, such as epidural hematoma, direct spinal cord trauma, spinal cord infarction, neurotoxicity by accidental subarachnoid injection³. Even though only MRI can clearly diagnose a medullar lesion⁵, many authors emphasize the importance of performing epidural anesthesia with conscious patients, because the early

detection of a neurologic dysfunction may activate appropriate salvage procedures^{7,8}.

Also patients not receiving thoracic epidural anesthesia can present paraplegia⁴, suggesting the presence of other underlying causes. Some of the contributing factors may be: bleeding at the costovertebral angle, migration of oxidized cellulose into the spinal canal, iatrogenic anterior spinal artery occlusion and hypotension. Bhuiyan refers about a case of paraplegia after thoracotomy in an adult patient who had a transient period of atrial fibrillation leading to embolism of the anterior spinal artery⁹. In another case, the mechanism was the ligation of intercostal vessels (supplying the arteria magna of Adamkiewicz) and the thrombosis of the anterior spinal artery².

In our study, we present three cases occurred in two different pediatric tertiary centers, in order to increase the awareness of this fearful complication also in children. Only five pediatric patients suffering from post-thoracotomy paraplegia, including ours, are reported in literature^{3,6} (Table II).

As described for adults, the mechanisms underlying this complication in children may be various and, in some cases, it's not possible to clearly identify the cause. Among the various pathogenesis causing the neurological deficit there are:

- vascular lesion during catheter insertion for epidural anesthesia, resulting in intraspinal hematoma¹⁰;
- intramedullary catheterization with narcotic injection inside the spinal cord⁷⁻¹¹;
- air injection in the vertebral canal;
- persistent bleeding originating from the vessels of the costovertebral angle;
- use of hemostatic material successively migrating into the vertebral canal¹²;
- lesion or closure of tributary intercostal arteries of the arteria radicularis magna (also known as artery of Adamkiewicz)^{2,13} (Fig. 1);
- anterior spinal artery thrombosis⁹;
- intraoperative hypotension;
- metastatic spinal canal invasion and intraspinal growth and extension of a primary neoplastic lesion (in particular a thoracic neuroblastoma)¹³.

Concerning epidural anesthesia, this procedure may be more challenging in pediatric patients, because its execution under general anesthesia deprives the anesthesiologist of patient's feedback; indeed, in presence of alarm signs, immediate interruption of the maneuver and therapeutic procedures can be carried out^{7,8}. Given the impossibility of practicing epidural anesthesia in awoken children, we suggest to always practice a complete neurological examination of patients undergoing thoracotomy with or without epidural anesthesia. If there are doubts of neurological deficits, an immediate neurosurgical evaluation with MRI should be mandatory¹⁴.

As regards thoracotomy, many authors revealed that a more posterior surgical accesses tend to increase the risk

TABLE II - Summary table of the cases reported in Literature

Authors	Year of publication	Age at diagnosis (years)	Sex	Diagnosis	Cause of paraplegia	Surgery	Neurological level loss	Outcome	Comment
Allison et al	2008	9	M	Ewings sarcoma	Thoracotomy/ Epidural anesthesia	Right third rib excision	T2-T3	No improvement	Surgical bleeding occurred in the vicinity of the costovertebral joint; bovine matrix hemostatic sealant was used (but no mass seen in the vertebral canal on MRI)
Llewellyn et al	2007	4 months	M	Not reported	Epidural anesthesia	Closure of stoma	Cauda equina syndrome	Improvement (1 year after surgery)	Born prematurely at 31 weeks gestation; partial neurological impairment preoperatively
Attar et al	1995	53	M	Bronchogenic carcinoma	Thoracotomy	Left upper lobectomy	T6-T7	No improvement	No massive bleeding, hypotension, nor any oxidized cellulose used for hemostasis
		73	M	Bronchogenic carcinoma	Thoracotomy	Left lower lobectomy	T6-T7	Improvement	
		40	M	Pulmonary tuberculosis	Thoracotomy	Left upper lobectomy	T6-T7 (detected only 72h after surgery)	No improvement	
		63	M	Tuberculous empyema	Thoracotomy	Decortication	T6-T7	No improvement	
		20	M	Stab wound	Thoracotomy	Control of bleeding	T6-T7	No improvement	
Kao et al	2004	81	M	Acute cholecystitis and common bile duct stones	Epidural anesthesia (intracord catheterization with subsequent local anesthetic injection)	Cholecystectomy and choledocholithotomy	T11	Partial improvement (after large-dose IV methylprednisolone and subsequent rehabilitation for 6 months)	MRI demonstrated intramedullary split-like lesion extending from T4 to T12 and intramedullary air bubble at T9
Bromage et al	1998	62	F	Not reported	Epidural anesthesia (Four unsuccessful attempts at epidural puncture made at or above the upper end of the laminectomy scar by the junior member of the anesthetic team)	Total arthroplasty of the right knee	T5	No improvement	Previous laminectomy (9 years before) for prolapsed intervertebral disc MRI revealed air bubble in the cord at T10 and region of increased T2-weighted signal in the anterior portion of the spinal cord between T4 and T5, consistent with infarction
Løvstad et al	1999	56	F	Hamartoma	Thoracotomy	Removal of the left lower lung (lobectomy or segmentectomy?)	T6-T7	No improvement	Patient suffering from ankylosing spondylitis Use of absorbable haemostatic sponge (Surgicel) between the 5th and 6th costa posteriorly against the muscular layer for bleeding from intercostal muscles. Laminectomy, performed 17 hours after primary operation, showed a large piece of Surgicel occupying 2/3 of the diameter of the spinal canal for 2 cm, compressing the spinal cord
Bhuiyan et al	1998	82	M	Squamous cell carcinoma	Thoracotomy	Right middle and lower bilobectomy	T3-T4	No improvement (Patient dead)	History of antero-inferior myocardial infarction (30 years before), deep vein thrombosis with an episode of pulmonary embolism (16 years before) and ankylosing spondylitis MRI failed to reveal any spinal or epidural haematoma or spinal cord ischaemia Post-mortem examination demonstrated spinal cord infarction and severely stenosed spinal arteries

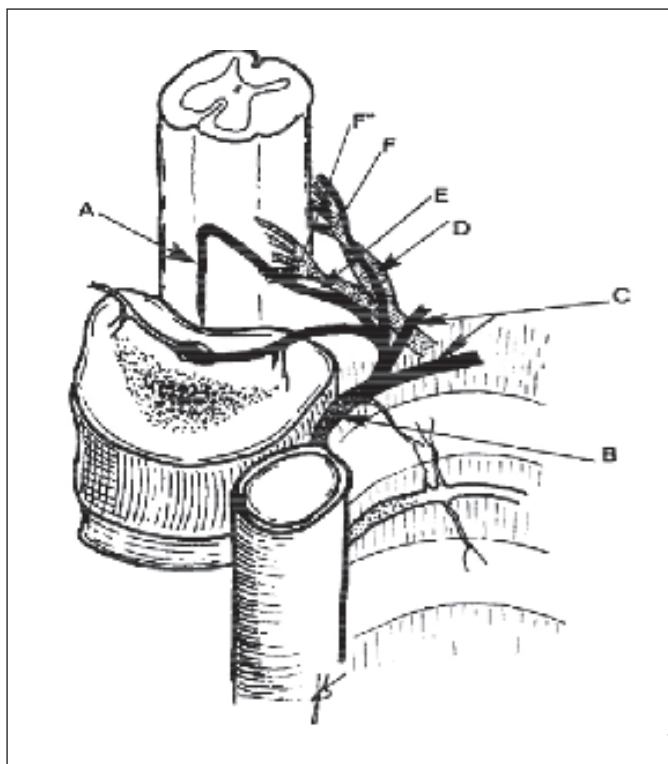


Fig. 1: Arteria radiculomedullaris magna (ARMM): the Adamkiewicz artery. Its contribution to the spinal cord supply at the level of T8–T12. A: ARMM. B: a. intercostalis communis. C: aa. intercostales propriae. D: a. radicularis posterior. E: a. radicularis anterior. F: and F’’: Radix dorsalis n. spinalis and radix ventralis n. spinalis. a, arteria; aa, arterial.

of a spinal cord damage with associated neurological deficit, given its greater proximity to the costo-vertebral angle and to the structures therein^{13,15}. In case of lesions involving the T8–T12 area, we suggest to practice a pre-operative arteriography, in order to highlight vascular anomalies and describe the path of Adamkiewicz artery¹⁶.

Furthermore, the use of hemostatic material or electrocautery in this area seems to expose the patient to the risk of spinal cord injury, due to the millimetric distance between pleural cavity and dura in the intraspinal compartment, through the intervertebral foramen. In particular, the hemostatic materials, once soaked in blood, tend to become gelatinous and to migrate into the vertebral canal (with unknown mechanism), increasing the intraspinal pressure and exposing the spinal cord to ischemic phenomena and damage¹². Therefore, in case of bleeding, hemostatic material should be used in minimal quantities and the excess must always be removed.

Conclusion

Paraplegia is a rare but fearsome complication of anesthetic and surgical procedures, especially thoracotomy

and epidural anesthesia. Many risk factors have been linked with this unfaithful event, such as the use of hemostatic material or electrocautery, a spinal cord trauma and the neurotoxicity of anesthetic drugs. The role of the clinician is crucial, since it must be aware of this rare possibility and its causes, suspecting the diagnosis, requesting the correct imaging study to confirm his hypothesis and directing the patient towards the most appropriate therapeutic pathway. This process should involve different professional figures (such as radiologists and neurosurgeons) as it must be as fast and precise as possible, in order to maximize the probability of functional improvement and recovery.

Riassunto

BACKGROUND: La paraplegia rappresenta una temibile complicanza delle procedure chirurgiche in anestesia generale ed è stata maggiormente associata alle toracotomie ed all'esecuzione di anestesia per via epidurale. Può avvenire in età pediatrica ed adulta e la diagnosi viene effettuata tramite Risonanza Magnetica Nucleare. L'obiettivo di questo studio è descrivere l'esperienza di due centri pediatrici in merito alla paraplegia post-operatoria e fornire una revisione della letteratura sull'argomento.

METODI: Abbiamo riportato tre casi di paraplegia post-operatoria in pazienti oncologici di età pediatrica, valutando le possibili cause e descrivendo le varie strategie di prevenzione.

RISULTATI: Dal nostro studio emergono due principali evidenze: 1) Il posizionamento di un catetere epidurale in età pediatrica può esporre ad un maggiore rischio di danno spinale permanente a causa della sua esecuzione in corso di anestesia generale, mancando il feedback da parte del paziente nel corso della procedura. 2) Nelle toracotomie, il rischio di paraplegia tende a crescere con l'avvicinarsi del campo operatorio all'angolo costo-vertebrale, soprattutto nel caso in cui vengano adottati l'elettrocoagulazione e materiali emostatici.

CONCLUSIONI: Una valutazione neurologica post-operatoria nei pazienti sottoposti ad anestesia epidurale o toracotomia dovrebbe essere sempre effettuata e, associato ad un approfondimento radiologico adeguato, può condurre ad una rapida presa in carico terapeutica ed una miglior prognosi.

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