Published in final edited form as:

J Pediatr Surg. 2014 January ; 49(1): 172–177. doi:10.1016/j.jpedsurg.2013.09.049.

“Trap-Door” and “Clamshell” Surgical Approaches for the Management of Pediatric Tumors of the Cervicothoracic Junction and Mediastinum

Emily R. Christison-Lagay, MD¹, David G. Darcy, MD¹, Eric J. Stanelle, MD¹, Stacy DaSilva, BA¹, Edward Avila, MD², and Michael P. La Quaglia, MD¹

¹Pediatric Service, Department of Surgery, Memorial Sloan-Kettering Cancer Center, New York, NY

²Department of Neurology, Memorial Sloan-Kettering Cancer Center, New York, NY

Abstract

Background/Purpose—For pediatric tumors of the cervicothoracic junction, an isolated cervical or thoracic surgical approach provides insufficient exposure for achieving complete resection. We retrospectively examined “trap-door” and “clamshell” pediatric thoracotomies as a surgical approach to these tumors.

Methods—We searched our database for pediatric patients with cervicothoracic tumors who underwent clamshell or trapdoor thoracotomy between 1991 and 2013, reviewing tumor characteristics, surgical technique, completeness of resection, morbidity, and outcome.

Results—Trap-door (n=13) and clamshell (n=4) thoracotomies were performed for neuroblastoma (n=9), non-rhabdomyosarcoma soft tissue sarcoma (n=4), germ cell tumor (n=2), rhabdomyosarcoma (n=1), and neuroendocrine small cell carcinoma (n=1). Fourteen of these cervicothoracic tumors were primary, and three were metastatic. Gross total resection was achieved in 15 patients (94%). Operative complications included vocal cord paralysis (n=2), mild upper-extremity neuropraxia (n=2), and hemidiaphragm paralysis (n=1). All but one involved encased nerves. Overall survival was 61% for the series and 80% for patients with primary tumors. Eleven (73%) of 15 patients who underwent gross total resection had no evidence of recurrence. Three patients with metastatic disease died of distant progression within 1.3 years.

Conclusions—Gross total resection of primary cervicothoracic tumors can be accomplished with specialized exposure in pediatric patients with minimal morbidity.

Keywords

trapdoor; clamshell; cervicothoracic tumor; neural monitoring

Corresponding Author: Michael P. LaQuaglia MD, Pediatric Service, Department of Surgery, Memorial Sloan-Kettering Cancer Center, 1275 York Avenue, New York NY 10065, Telephone: 212-639-7002, Fax: 212-717-3373, laquaglm@mskcc.org.

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Introduction

High thoracic tumors and tumors of the cervicothoracic junction are often not amenable to complete resection by either an isolated unilateral cervical or thoracic approach. Nonetheless, adequate surgical exposure of these tumors is essential to prevent injury to nearby nerves (brachial plexus, phrenic, vagus, recurrent laryngeal, and spinal accessory) and vascular structures (carotid, subclavian, and vertebral arteries, the thyrocervical trunk, and the subclavian and jugular veins), while assuring complete resection. Large mediastinal tumors that require access to both hemithoraces also present a challenge to complete resection with low post-operative morbidity. Series regarding pediatric bilateral anterior thoracotomy ("clamshell" approach) are found most often in the cardiothoracic literature. Isolated case reports have described excellent exposure for resection cervicothoracic junction tumors using a combined supraclavicular incision -sternotomy-anterior thoracotomy ("trap-door" approach); however, no series exist in the pediatric surgical literature describing indications, techniques, complications, morbidity, completeness of resection, or overall outcome.

Patients and Methods

With Institutional Review Board approval WA0402-11, we reviewed the pediatric surgical database at Memorial Sloan-Kettering Cancer Center for patients who had undergone a resection via a "clamshell" or "trap-door" thoracotomy during the period 1995 to 2013. Seventeen patients were identified, of whom 13 underwent a trap-door and four underwent a clamshell thoracotomy. A uniform operative approach was performed for each procedure

Trap-door

The patient is placed in the dorsal recumbent position with a roll beneath the shoulders, the arm ipsilateral to the lesion outstretched, and the contralateral arm tucked at the side. General anesthesia is induced, using either a double lumen endotracheal tube or a single lumen endotracheal tube with a bronchial blocker for intubation. The head is then rotated 30-45° away from the tumor. The patient is prepped from the ear to the umbilicus and a clear head and neck drape is used. A transverse incision is begun superior to the clavicle with a parallel course, or along the anterior border of the sternocleidomastoid with a descending course, to the mid-portion of the suprasternal notch, continued downward through the midline sternum to the fifth interspace, then laterally through the fifth interspace to the anterior-axillary line. The pectoralis is divided close to its point of insertion and the intercostal muscles divided at the fifth interspace. The pleural space is entered and the internal mammary vessels are isolated, ligated, and divided. The retrosternal space is bluntly dissected and a sternal saw is used to divide the sternum to the level of the fifth interspace, then laterally to the thoracotomy. Bleeding from the edge of the sternum is treated with bone wax. The sternal and a portion of the clavicular head of the sternocleidomastoid are divided close to their points of origin and marked with sutures for later approximation. The strap muscles are similarly divided. A Finochetto retractor is then placed between the cut edges of the sternum allowing excellent exposure. Neural monitors are placed as described below. Dissection then begins with mobilization of the thymus and identification of the phrenic,

vagus, and recurrent laryngeal nerves. The tumor is dissected piecemeal, in a cranial direction, and major vascular structures are controlled proximally and distally. For teratomas, an attempt to remove the whole tumor and prevent spillage of cells is optimal, although not always possible. When the thoracic duct can be identified, it is isolated, tied and ligated. Meticulous dissection continues with frequent use of neural monitoring, and marginal biopsies are taken after tumor extraction. At the conclusion of the dissection, a chest tube is placed and pectoralis flaps mobilized. The sternum is reapproximated with 3-4 sternal wires or non-absorbable sutures in small patients. Pericostal sutures are placed to reapproximate the ribs in the fifth interspace. The lungs are then inflated under direct vision and the pericostal sutures and sternal wires are secured. The mobilized edges of the pectoralis flaps are closed over the sternal wires. Attention is then directed to the neck where the sternal and clavicular heads of the sternocleidomastoid muscle is reapproximated with figure of eight sutures of heavy Vicryl. Finally, the subcutaneous and dermal layers of the incision are closed in layers.

Clamshell

The patient is placed in the dorsal recumbent position with a roll behind the midportion of the chest. The arms are abducted to 90° at the shoulder and elbow. General anesthesia is induced with a double lumen or single lumen endotracheal tube capable of lung isolation, and the patient is prepped from the chin to the umbilicus and transversely to the bilateral posterior axillary lines. An anterior curvilinear incision is made along the fifth interspace bilaterally from each anterior-axillary line, connecting at the midline. The pleural space is entered and the mammary vessels are isolated, ligated, and divided. The retrosternal space is then bluntly dissected, and the sternum divided transversely with a sternal saw. A Finochetto retractor is placed, allowing for exposure of both pleural cavities, from the pulmonary hilum to the posterior aspect of the diaphragm. When the tumor dissection is complete and hemostasis is assured, bilateral chest tubes are placed and the sternum is either approximated with wire or stabilized in the anterior-posterior dimension with a Steinmann pin. Pericostal sutures are used to approximate the ribs, and the subcutaneous and dermal layers of the incision are closed in layers.

Neural Monitoring

Continuous running electromyography (or free-run EMG) and direct neuronal stimulation techniques were used in 8 cases in this series. Free-run EMG evaluates interference patterns that are seen as motor unit potential firing at rates of 30-100 Hz and can be applied to any nerve in which its corresponding myotome can be accurately accessed [1]. Tactile stimulation, thermal activity, or mechanical traction of the nerve and axon cause neurotonic discharge. If the firing is self-limited and associated with a specific surgical maneuver, it is considered benign and simply serves as a warning sign, but if it persists after manipulation, it is considered pathologic and may be associated with a post-operative deficit.

Direct neuronal stimulation in the surgical field was also used to test axonal integrity and to distinguish tumor from nerve. In this case, stimulation of a nerve using a handheld monopolar or bipolar stimulator is then transmitted to a surface or intramuscular needle electrode which records a compound muscle action potential from the muscle innervated by

the stimulated nerve [2, 3]. For tumors of the cervicothoracic region, the C8/T1 nerve roots and vagus nerve were monitored. The C8 and T1 nerve roots, supplying the inferior aspect of the brachial plexus, were monitored by placing needles in the first dorsal interossei, and the thenar/hypothenar eminence, to monitor muscles innervated by the ulnar and median nerves. The vagus was monitored indirectly by the recurrent laryngeal nerve, using surface electrodes on a special endotracheal tube. It is possible, though not practical, to place diaphragmatic electrodes for monitoring of the phrenic nerve; rather, this nerve can be stimulated during the case and the response of the diaphragm verified visually.

Results

Pre-operative Patient and Tumor Characteristics

There were eight boys and nine girls. Median patient age at the time of resection was 5.9 years (range: 9 mo to 29.6 y). The oldest patient had been treated with chemoradiation therapy as a child and returned as a young adult for surgical resection of a recurrence; the second oldest patient was 16.8 years old. The tumor types were neuroblastoma (n=9), non-rhabdomyosarcoma soft tissue sarcoma (NRSTS; n=4), germ cell tumor (n=2), rhabdomyosarcoma (n=1), and neuroendocrine small cell carcinoma (n=1). In 12 patients, the chest was the primary site of disease, and in three patients, the primary site was the abdomen (neuroblastoma [n=2], malignant germ cell tumor [n=1]) with metastatic disease to the chest. One patient had a primary cervical neuroblastoma and one patient had a primary embryonal rhabdomyosarcoma of the neck and superior mediastinum. Shortness of breath (29%) and weakness (29%) were the most commonly reported presentations, followed by a palpable mass (24%), pain (24%), cough (24%), Horner syndrome (18%), and pneumonia (12%). Twelve patients (71%) received preoperative chemotherapy. Of those who did not, one patient had mature teratoma, one with ganglioneuroma, one with embryonal rhabdomyosarcoma (who received previous radiation) and two with neuroblastoma. Six of 17 patients (35%) were noted to have pre-operative Horner's syndrome, although this was not the presenting symptom in half of these children; in one child, no mention was made of ocular findings.

Intraoperative Details

The median operative time was 348 minutes (range: 163-495 min) and the median blood loss was 500 mL (range: 100-2200 mL). Dual-lumen endotracheal intubation was used in two of four clamshell procedures. Ten patients received an intraoperative transfusion of packed red blood cells. Two patients required patch angioplasty of the innominate vein. Neural monitoring was used in eight of 15 undergoing complete resection (53%), none of whom had an incidental injury. Two patients had encasement of neural structures resulting in operative sacrifice, including two phrenic and one right recurrent laryngeal nerve. The first patient had extensive internal mammary nodal disease involving resection of the second rib and phrenic sacrifice, resulting in left hemi-diaphragm paralysis with a moderate effusion, which resolved after thoracentesis. A sarcoma of the right thoracic apex encased the right phrenic nerve necessitating sacrifice, without post-operative complication. In the other patient, the right recurrent laryngeal was encased and could not be identified; post-operative vocal cord paralysis was verified, but the patient had full left vocal cord compensation. The

one inadvertent transection of a recurrent laryngeal nerve occurred in a patient with extensive cervical nodal disease and was immediately repaired by the microsurgical team; follow-up showed no vocal abnormalities. The procedure was aborted in a single patient deemed to have unresectable disease with tumor invading through the pericardium into the main pulmonary artery, which could not be identified on preoperative radiology. There were no intraoperative deaths.

Post-operative Details

Of the 16 patients undergoing operation for resection, a gross total or near gross total resection was achieved in 15 (94%). Median length of hospital stay was 7 post-operative days (range: 4-27 d). In each patient, a chest tube was left in place for a median of 3 days (range: 2-9 d). Seven patients required a post-operative blood transfusion, including three who did not receive an intraoperative transfusion. Blood transfusion information was missing from the records of four patients. There was no chylothorax as a complication in this series. Post-operative Horner's syndrome was noted in seven patients: in six with pre-operative pre-existing Horner's syndrome and in one patient undergoing resection of neuroblastoma involving the stellate ganglion. Three patients had post-operative arm weakness, the first being a patient with a pre-existing plexopathy from two prior posterior thoracotomy approaches, who demonstrated no change in function.

Two patients had new postoperative weakness, manifesting as fifth-digit weakness in one patient and generalized weakness which was markedly diminished at 2 months in the other; neither experienced any impact on their activities of daily living. Both patients had intra-operative neural monitoring for neuroblastoma that encased the inferior portion of the brachial plexus.

Recurrence

Of 16 patients undergoing a gross total resection of tumor, 12 (75%) had no evidence of disease recurrence. Of the four patients with a recurrence, three developed distant disease and died at a median of 18 months (range: 16 - 19 months) after resection. Recurrence was local and rapidly progressive in one patient with a primary undifferentiated sarcoma of the mediastinum; she died 2 months after her initial resection.

Survival

Five-year survival for the 17 patients in this series (intent to treat) was 61% (95% CI 33-89). In one patient, resection was abandoned due to invasion of the pulmonary artery. Five-year survival in the remaining 16 patients who underwent resection was 67% (95% CI 39-95). Patients who were treated for primary tumors and not metastatic disease had a 5-year survival of 80% (95% CI 54-106), which was significantly different than those treated for metastatic disease ($p = 0.003$). Of the three patients who presented with metastatic disease, all had prior posterior thoracotomy or median sternotomy approaches. The trapdoor approach made gross total resection possible in three. Survival in the patient with unresectable disease was 14 months.

Discussion

Large tumors of the superior mediastinum and cervicothoracic inlet represent a challenge to surgical resection. Neither a cervical nor thoracic approach alone is sufficient to achieve exposure of the tumor and its relationship to vital nerve and vascular structures of the head and neck. The unilateral anterior cervico-sterno-thoracotomy or “trap-door” incision was first described in 1979 by Masaoka et al. as a method of exposure of Pancoast tumors of the superior sulcus [4]. Since that time, the procedure (with minor alterations and variations) has remained an important method of approach for adult thoracic surgeons resecting pulmonary parenchymal tumors with apical involvement, tumors of the cervicothoracic junction, large tumors of the superior mediastinum, and tumors of the superior thoracic wall [5]. This incision allows access to the great vessels of the mediastinum and neck including, if necessary, intrapericardial control of the aorta and pulmonary vessels proximal to the level of the pulmonary hilum. Moreover, the approach preserves sternal clavicular articulation, which is sacrificed in anterior approaches, resulting in significant morbidity of the shoulder and chest girdle that may increase over years of pediatric growth [6, 7]. Finally, utilizing a fifth interspace incision for the anterior thoracotomy preserves innervation of sternal portion of pectoralis major by the medial pectoral nerve, leading to reduced atrophy of the pectoralis muscle. This branch of the medial cord of the brachial plexus descends inferiorly on the posterior side of pectoralis major to innervate inferior portion of the sternal head.

To date, reports of approaches to tumors of the cervicothoracic junction in the pediatric patient have been limited to small case series of fewer than four patients. The largest series, which describe approaches to complex spinal deformities, are published in the orthopedic literature [8]. In four patients with cervicothoracic neuroblastoma, Sauvat et al. used a transmanubrial osteomuscular-sparing technique popularized by Grunenwald to achieve >90% gross total resection [9, 10]. Complications included chylothorax in one patient and phrenic nerve palsy in two patients [9]. Similarly, Pimpalwar achieved a 99% gross total resection in a localized cervicothoracic neuroblastoma using a transmanubrial technique. This approach, however, requires resection of the first costal cartilage and mediastinal exposure is limited to the anatomy visible at the thoracic inlet, above the subclavian vein [11]. Parikh et al. reported the resection of cervicothoracic neuroblastoma in three patients using Darteville's approach, requiring resection of the medial clavicular head. In adult series, this has been associated with shoulder girdle instability, delayed post-operative recovery, and bony malunion in up to 42% of patients [11-13].

For large tumors arising in the mediastinum and extending into both hemithoraces, the clamshell incision provides excellent exposure extending to the level of the lung apices and thoracic outlet with minimal respiratory complications and a low rate of sternal nonunion or wound infection [14, 15]. A Steinmann pin can be used to further stabilize the sternum in the anterior-posterior dimension.

The majority of tumors resected by the trap-door approach in this series were neuroblastomas of the cervicothoracic inlet. An estimated 11% to 26% of neuroblastomas present in the thorax and fewer than 2% are cervical, typically arising from the stellate ganglion [16, 17]. There are conflicting data regarding a unique biology of thoracic

neuroblastoma [6, 17-19]. Regardless, achieving a gross total resection is associated with significantly improved progression-free and overall survival in these cases [20, 21]. In non-neuroblastic tumors, including germ cell tumors, rhabdomyosarcoma, and NRSTS, gross total resection is imperative for cure [14, 15].

In this series, we report the successful resection of 16 of 17 tumors of the cervicothoracic junction or bilateral mediastinum via either a “trap-door” or “clamshell” approach. This aggressive approach resulted in an overall survival of 80% in patients with primary mediastinal or cervical tumors. Control of the great vessels with this approach allowed dissection of encasing, difficult-to-access tumor growth. In one case, an aberrant vertebral artery was encountered, but an intact Circle of Willis with adequate backflow, as assessed by neurosurgery, allowed for prudent ligation. We attribute the absence of any complicating chylothorax in our series to the attempts made to specifically identify and ligate the thoracic duct and the generous use of MRI safe, non-ferromagnetic clips, while dissecting the left cervicothoracic region. In this series, patients whose cervicothoracic mass represented metastatic disease (two with neuroblastoma and one with a malignant germ cell tumor) did not fare as well. All succumbed to distant progression within 19 months of resection. While such a small sample size cannot be used to draw definitive conclusions regarding the efficacy of resection for metastatic disease, resection in this population should be approached with appropriate circumspection.

Operative and perioperative morbidity in this series was low and included a small risk of resection of a regional nerve or the need to reconstruct a great vessel. All patients had excellent functional recovery without any long-term evidence of weakness, neuropraxia, or vascular compromise. In summary, gross total resection of tumor mass of the cervicothoracic junction or bilateral mediastinum can be accomplished in most appropriately selected patients as either an upfront surgical approach or following chemotherapeutic cytoreduction. Complete resection offers an excellent chance of long-term survival with minimal morbidity for a variety of tumor subtypes, particularly when the tumor represents the primary site of disease.

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Figure 1.

A) Trap-door incision with ipsilateral arm abducted to 90° at the shoulder. The superior portion of the incision can follow the clavicle laterally or be extended along the sternocleidomastoid for tumors with a larger cervical component. B) Exposure of left cervicothoracic tumor investing the subclavian artery, phrenic and vagus nerves, after patch angioplasty of the innominate vein for tumor invasion. Divided ends of the sternocleidomastoid muscles have been marked with sutures for re-approximation.

Figure 2.

A) Clamshell incision with bilateral arms abducted to 90° at the shoulder. B) Large left-sided pleural tumor involving the pulmonary hilum with metastatic disease in the right hemithorax. C) Closure with Steinmann pin and wires.

Figure 3.

Distribution of tumor locations: Right and left cervicothoracic (3 and 4), right and left superior mediastinal extending into thoracic apex (2 and 6), superior mediastinal (1), inferior left pleural tumor with metastatic disease to the right chest (1).

Table 1
Patient and disease characteristics

	Survivors	Mortalities	Combined
Median Age at Resection (range)	6.4 years (1.3 – 29.6 y)	5.3 years (9 mo – 16.7 y)	5.9 years (9 mo – 29.6 y)
	N (% survivors)	N (% mortalities)	N (% total)
Total	12 (100%)	5 (100%)	17 (100%)
Sex			
Male	7 (58%)	1 (20%)	8 (47%)
Female	5 (42%)	4 (80%)	9 (53%)
Presenting Symptom			
Shortness of Breath	5 (42%)	0	5 (29%)
Weakness	3 (25%)	2 (40%)	5 (29%)
Palpable Mass	2 (17%)	2 (40%)	4 (24%)
Cough	4 (33%)	0	4 (24%)
Pain	2 (17%)	2 (40%)	4 (24%)
Horner Syndrome	2 (17%)	1 (20%)	3 (18%)
Pneumonia	1 (8%)	1 (20%)	2 (12%)
Neoadjuvant Therapy			
Chemotherapy	7 (58%)	5 (100%)	12 (71%)
Tumor Location			
Left Thoracic Inlet	6 (50%)	4 (80%)	10 (59%)
Right Thoracic Inlet	5 (42%)	0	5 (29%)
Bilateral Extent	1 (8%)	1 (20%)	2 (12%)
Primary Tumor Location			
Thoracic	10 (83%)	2 (40%)	12 (71%)
Cervical	2 (17%)	0	2 (12%)
Abdominal	0	3 (60%)	3 (18%)
Tumor Types			
Neuroblastoma	6 (50%)	3 (60%)	9 (53%)
NRSTS	3 (25%)	1 (20%)	4 (24%)
Germ cell tumor	1 (8%)	1 (20%)	2 (12%)
Rhabdomyosarcoma	1 (8%)	0	1 (6%)
Neuroendocrine Small Cell	1 (8%)	0	1 (6%)
Resection Status			
Gross Total Resection *	11 (92%)	4 (80%)	15 (88%)
Near-complete Resection †	1 (8%)	0	1 (6%)
No resection conducted	0	1 (20%)	1 (6%)

NRSTS: non-rhabdomyosarcoma soft tissue sarcoma.

* *Gross Total Resection* was defined as removal of all visible and palpable tumor.

[†] *Near-complete Resection* was defined as removal of >95% tumor.

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Table 2
Complications listed by patient, with CTCAE grade, and outcome

Patient	Complication(s)	CTCAE Grade	Outcome
2	Left Brachial Plexopathy *	1	Fifth-digit weakness, no effect on ADLs
5	Left Brachial Plexopathy **†	1	Resolved by 1 month post-op, no affect on ADLs
6	Phrenic Nerve Division *	1	Asymptomatic
10	Horner Syndrome *	1	Asymptomatic
13	Recurrent Laryngeal Nerve Palsy † Phrenic nerve division **†	2 2	No vocal abnormalities Asymptomatic
14	Recurrent Laryngeal Nerve Palsy **†	2	Full contralateral vocal cord compensation

ADL: Activities of Daily Living; CTCAE: Common Terminology Criteria for Adverse Events, version 4.0

* Nerve encasement resulting in operative sacrifice.

† Patient who presented for resection of recurrence following previous alternate approach (posterior thoracotomy or median sternotomy)