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Research Article

Cervico-Thoraco-Brachial Crossing Syndrome: Results of Surgery in 5 Cases Operated on in the Thoracic and Cardiovascular Surgery Department in Senegal

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Abstract

Introduction: Cervico-thoraco-brachial outlet syndrome is a rare condition, affecting 1% of the population. It results from the compression of the vasculo-nervous structures, most often by the presence of a cervical rib or fibrous tissue. The place and importance of surgery remains debated by these authors who advocate conservative treatment. *Patients and methods*: This was a retrospective descriptive study of five patients with cervico-thoraco-brachial crossing syndrome treated in the Thoracic and Cardiovascular Surgery Department of National University Hospital Center of Fann in Senegal from January 1, 2006 to December 31, 2020, over a period of 15 years. *Results*: Five patients, aged on average 33 years (extreme 18 and 50 years), were operated on. The predominance was female (1 man and 4 women). The symptoms were neurological in all patients, with upper limb claudication (3 cases), cervico-brachialgia and paresthesias (5 cases). The right side was the most affected with 3 patients. Right supraclavicular swelling was noted in two patients. The Adson test was positive in 4 patients. No vascular manifestations were observed. The average duration of symptoms before the intervention was 4 years (Extremes: 1 and 5 years). Imaging revealed: a cervical rib in 3 patients. This was bilateral in 2 patients; and right unilateral in 1 patient. In the two other cases, it was an apophysomegaly of the 7th cervical vertebra and a fibrous dysplasia of the first left rib. The electromyogram performed in the 3 patients confirmed a C6 C7 C8 (1 case) and C6 C7 (2 cases) plexopathy. The preferred surgical approach was supraclavicular. For fibrous dysplasia of the first rib, a double sus and subclavicular approach was made. Compression was due to a cervical rib (3 cases), dysplasia of the first rib (1 case) and apophysomegaly (1 case). There was fibrous tissue (2 cases) and the anterior scalene muscle (1 case). Medical treatment and physiotherapy were instituted in all patients. This allowed us to have generally satisfactory results after an average follow-up of 8 years (2 – 15 years). *Conclusion*: cervico-thoraco-brachial crossing syndrome is rare. The most common anatomical modifications are the presence of a cervical rib or fibrous tissue. Surgery combined with physiotherapy improves the quality of life of patients. A more exhaustive multicenter study would allow a better evaluation of the syndrome.

Keywords

Cervico-Thoraco-Brachial Outlet Syndrome, Surgical Results, Supraclavicular Approach

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1. Introduction

The cervico-thoraco-brachial crossing syndrome (CTBCS) is the set of clinical manifestations resulting from the compression of the nervous and / or vascular structures during their passage at the level of the thoraco-brachial passage [1-3].

Compression is linked to changes in the cervical outlet and / or the presence of a cervical rib. Compression generally comes from the presence of a cervical rib, abnormal scalene fascicles or fibromuscular abnormalities [4, 5]. Presence of a cervical rib occurs in 0.1 – 1% of the population. It is bilateral in 8-50% according to authors [3-6].

We distinguish: the complete cervical ribs, attached to the sternal manubrium by a cartilage individualized or confused with that of the first rib; incomplete cervical ribs, which may have a free end which floats in the supraclavicular hollow or be extended forward by a fibrous bunch which attaches to the first rib; Cervical pseudo-ribs are C7 processes extended by a solid fibrous cord and often associated with a cervical rib [5-7].

The diagnosis is clinical based on neurological manifestations (pain, paresthesias and paresis of the neck, upper limb, chest wall and interscapulo-thoracic region) and/or vascular (more or less marked signs of limb ischemia thoracic: (pain and paresthesia, edema, reduction or abolition of pulse, heaviness, coldness and cyanosis) The cervicothoracic x-ray and CT scan highlight the anatomical abnormalities [1, 6-9].

The aim of surgical treatment is to decompress the vascular-nervous bundle at the level of the thoraco-brachial process. The results are always punctuated by pain and sequelae paresthesias [10-15]. Thus, the recent use of physiotherapy has made it possible to improve the symptoms relegating surgery to the background [16-18].

The aim of our work is to evaluate the results of surgical treatment of cervico-thoraco-brachial crossing syndrome. We will analyze these results in light of the literature.

2. Patients and Method

This was a retrospective study of five patients operated on for cervico-thoraco-brachial crossing syndrome between January 2004 and December 2021 in the Thoracic and Cardiovascular Surgery department in the National University Hospital Center of Fann in Senegal. only patients operated on for cervico-thoraco-brachial crossing syndrome and followed in the department were included.

Pre- and post-operative pain was assessed using the visual analog scale (VAS), as well as whether symptoms recurred or not. The parameters studied were: sociodemographic characteristics, history, clinical signs and radiological lesions (radiography and chest CT), electromyography results, surgical treatment, evolution. The degree of patient satisfaction rated on a scale of 1 to 10 as for the VAS. The patients were classified into three groups. The result was fair if the score was less than 4. It was good if the score was between 5 and 8. A score above 8 was considered an excellent result. The analysis was descriptive. Quantitative variables were expressed by means and limits.

3. Results

Five patients, aged on average 33 years (extreme 18 and 50 years), were operated on. The majority was female (1 man and 4 women). The symptomatology was neurological in all patients marked by cervico-thoraco-brachial pain; paresthesias. Upper limb claudication was noted in 3 patients. The average duration of symptoms before the intervention was 4 years (Extremes: 1 and 5 years). The right side was the most affected with 3 patients. Right supraclavicular swelling was noted in two patients. The Adson test was positive in 4 patients. No vascular manifestations were observed.

Imaging revealed a cervical rib in 4 patients. This was bilateral in 3 patients; and right unilateral in 1 patient. In the last case, it was fibrous dysplasia of the first left rib. The Electromyogram (EMG) performed in the 4 patients with cervical ribs confirmed C6 C7 C8 (1 case) and C6 C7 (1 case) plexopathy. The EMG was normal in the other two patients. EMG was not indicated in the last patient with fibrous dysplasia of the left first rib. Table 1 summarizes the diagnostic aspects of the patients.

Table 1. *Diagnostic aspects according to patients.*

Patients	Sex	Age years	Clinical Data			Para-clinical data			Diagnosis retained
			Functional	General	Physical	X-ray	Scanner	EMG	
1	W	30	-Right cervvico-brachialgia -right upper limb lameness - Paresthesias -Since 1 year	General condition preserved	-ADSON (+)	Bilateral cervical rib	-Bilateral cervical rib. -Synostosis with the first right rib. -Risk block narrowing the costoclavicular process to 6 mm on	Plexopathy C6, C7, C8 on the right	Right root compression by a cervical rib Gruber type 3

Patients	Sex	Age years	Clinical Data			Para-clinical data			Diagnosis retained
			Functional	General	Physical	X-ray	Scanner	EMG	
							the right compared to 16 mm on the left.		
2	W	45	-Left cervvico-brachialgia -right upper limb lameness - Paresthesias -Since 5 years	General condition preserved	-Painful left su-pracla-vicular mass -ADSON (+)	Bilateral cervical rib	-Bilateral cervical rib	Plexopathy C6 and C7 on the left with radicu-lar pain	Left root com-pression by a cervical rib Gruber type 3
3	W	18	-Right cervvico-brachialgia -right upper limb lameness -Since 5 years	General condition preserved	-Right supracla-vicular mass -ADSON (+)	Bilateral cervical rib	Bilateral cervical rib without vascular lesion	Not done	Right radicular syndrome by a cervical rib Gruber type 3
4	W	29	Right cervvico-brachialgia Paresthesias of the 4th and 5th fingers Since 4 years	General condition preserved	ADSON (+)	C7 apophy-so-megaly	C7 apophyso-megaly	Normal	C7 apophyso-megaly With right root compression. Gruber type 3
5	M	50	-Left cervvi-cobrachialgia. -Since 4 years Paresthesias	General condition preserved	no mass. ADSON (-)	Fibrous dysplasia of the 1st left rib.	Fibrous dysplasia of the 1st left rib.	Not indicated	Fibrous dys-plasia of the 1st left rib.

C = Cervical rib; EMG = Electrmyogram; W = Women; M = Male.

Patients with a cervical rib were operated on via a supra-clavicular approach. For fibrous dysplasia of the first rib, a double sus and subclavicular approach was made.

Exploration revealed compression of the brachial plexus by the cervical rib (once). In two cases, the compression was due to fibrous tissue stretched from the end of the cervical rib to the first rib. In one case, it was a compression by an extension of a fibrous tissue of the anterior scalene. In one patient (5th case), there was agenesis of the anterior scalene.

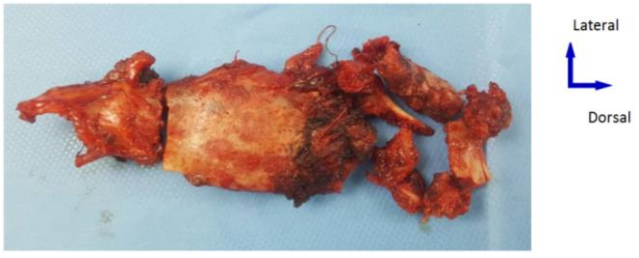

Figure 1. *Surgical specimen for fibrous dysplasia of the first rib.*

In one patient, the procedure was a resection of the cervical rib and the extending fibrous tissue (figure 1); or the 1st rib (4 cases). A partial anterior scalenectomy was associated in one patient.

There was a right pleural rupture which progressed well after 72 hours with chest drainage. The average length of hospitalization was 3 days (range: 2 and 5 days). Postoperative pain management was based on injectable paracetamol associated or not with tramadol. The postoperative course was simple.

After an average follow-up time of 8 years (range: 1 year – 17 years), one patient presented with residual pain relieved by analgesics. It was classified as a good result. In 2 patients, pain with residual paresthesia was noted at 2 months and 3 months postoperatively respectively. The EMG confirmed C7 C6 and C8 C7 C6 plexopathy. Analgesic treatment and vitamin B1 associated with motor physiotherapy were instituted with a disappearance of the pain after 6 months and 8 months respectively. They were classified as good results. Another patient who had undergone a previous scalenectomy had intermittent, persistent residual pain that was bearable under paracetamol combined with codeine or with oral tramadol. The latter was classified as fair. The last patient had

no postoperative pain. His result was excellent. Table 2 illustrates the therapeutic and evolutionary aspects of the patients.

Table 2. *Therapeutic and evolving aspects according to the patients.*

Patients	Surgical Data			Evolution and results				
	Way	Exploration	Gestures	Duration of hospitalization	Evolution	EMG	Post-operative treatment	Results
1	Right supraclavicular	Right cervical rib compressing the right brachiocervical plexus	Resection of the right cervical rib.	2 Days	Fatigability at Y3 Brachial paresthesia at Y10 Residual pain	Axonopathy C6, C7, C8 on the right	Vitamin B Motor physiotherapy	Good
2	Left supraclavicular	Left cervical rib + fibrous tissue attaching to C1 and compressing the right brachiocervical plexus + right pleural rupture	Resection of fibrous tissue and left cervical rib + pleural drainage	Days	Left antebrachial monoparesis at Y17 Residual pain	C8 and D1 axonopathy on the left. Sensory axonal neuropathy of the left ulnar nerve with C8 and D1 radicular pain on the left	Vitamin B Motor physiotherapy Analgesics	Good Paresthesias at A3 EMG shows a clear improvement in previous sensory and motor amplitudes
3	Right supraclavicular	Right cervical rib + fibrous termination of the anterior scalene on C1 compressing the right brachiocervical plexus	Right cervical rib resection + partial anterior scalenectomy.	Days	M2: regression of symptomsM5: pain after long work Y1: normal examination Y2: intermittent neurological symptoms Y11: Cervicobrachialgia and anesthesia of the right shoulder stump	Normal	Vitamin B Motor physiotherapy Analgesics	Fair
4	Right supraclavicular	Right cervical rib with fibrosis sheathing nerve threads of the brachial plexus	Cervical rib resection.	4 Days	None at Y2	Not done	Vitamin B Motor physiotherapy Corticotherapy Analgesics and Anti-inflammatories	Good
5	Left supra and subclavicular	Fibrous dysplasia of the left 1st rib compressing the right brachiocervical plexus	Resection of the 1st left rib.	Days	None at Y1	Not done	Analgesics	Excellent

C = Cervical rib; D = Dorsal rib; EMG = Electromyogram; Y = Year.

4. Discussion

The cervico-thoraco-brachial crossing syndrome has been the subject of publications and research for a long time but remains a rare condition [4-8]. This rarity, proven by the short series found in the literature, is confirmed by the small size of our series. In his 2013 publication, San é AD et al [19] found only 2 cases in Senegal. The anatomical anomalies and variations of the cervico-thoraco-brachial region are the causes of the symptoms of the cervico-thoraco-brachial crossing syndrome, the management of which gives rise to numerous controversies [20, 21].

The female predominance has been noted by several authors [9, 13, 20, 22], which corroborates our results. Neurological forms are more the prerogative of women while vascular forms affect both men and women [9, 13]. In our series, all our patients presented neurological forms consistent with the female predominance described in the literature. [1-3, 23].

The cervical rib is bilateral and symptomatic in 8-50% according to the authors [6, 24]. We observed the presence of bilateral cervical rib in 3 patients. It was unilateral right in one patient. Other anomalies outside the cervical rib can be the cause of the cervico-thoraco-brachial crossing syndrome, as is the case with pseudarthrosis of the first rib reported by Borrelly J et al [25]. These rare etiologies responsible for the neurological forms of cervico-thoraco-brachial crossing syndrome described in the literature were also found in our short series. This is the case in our 5th patient. Fibrous dysplasia of the first rib has not been found in the literature as a cause of cervico-thoraco-brachial crossing syndrome.

The neurological symptoms were dominated in all our patients by paresthesia. There was intermittent claudication in the 2 patients. Compression of the lower trunk of the brachial plexus is responsible for sensory and motor disorders in the ulnar territory [8, 10]. In crude forms, the Adson test can highlight the existence of nervous irritability [1-4]. This was the case in all our patients with a cervical rib. It was negative in the 5th patient.

Table 3. *Gruber's classification [22].*

Type	Description
Gruber 1	Slight increase in the cervical vertebra not extending beyond the transverse processes.
Gruber 2	Cervical rib protrusion beyond transverse processes with free or floating end
Gruber 3	Projection of the cervical rib beyond the transverse processes reaching the cartilage of the 1st rib by a ligament
Gruber 4	Formation of a neo-articulation between the cervical rib, costal cartilage and sternum

Standard radiography made it possible to make the diagnosis in all cases and place them in the Gruber classification: Table 3 [22]. Our 4 patients with a cervical rib were classified as Gruber type 3 (Table 3). Other complementary examinations such as the cervicothoracic scan carried out in all our patients made it possible to look for associated vascular and bony abnormalities. The correlation between radiological signs and clinical manifestations is difficult to establish as reported by all authors [1, 3, 23-25] and this was confirmed in our series.

All our patients had undergone surgery associated with functional motor rehabilitation and medical treatment based on vitamins, non-steroidal anti-inflammatory drugs and analgesics. For several years, this surgical treatment has sparked controversy among certain authors due to new conservative approaches in the management of cervico-thoraco-brachial crossing syndrome [16-18, 24]. These new approaches consist of different functional rehabilitation protocols finding some indications in certain cases of cervico-thoraco-brachial crossing syndrome. These protocols were only effective in the absence of anatomical lesions responsible for the conflict with the vascular-nervous structures of the cervicothoracic region. This was not the case in any of our patients. The indications for surgery have been well elucidated in the literature [21, 26].

Surgical resection of the cervical rib remains the standard treatment. The first approaches were Ross's axillary approach [15, 27]. It has the advantage of being esthetic and less painful. Recognized as a major cause of vascular and nervous complications with a high recurrence rate, it was abandoned especially since it did not allow optimal control over the vascular and nervous elements at the time of the surgical procedure [22, 27, 28]. Thus, it is currently reserved for vascular forms of cervico-thoraco-brachial crossing syndrome [2, 12, 27]. The new recommendations suggest the supraclavicular route which respects the insertion of the anterior scalene and the 1st side. It is currently the most used by surgeons. It allows total disinsertion of the cervical rib, resection of the middle scalene and other ligamentous, aponeurotic and muscular structures as needed [8, 13, 22, 24].

All our patients were operated on via the supraclavicular route. It allowed for a complete resection of the cervical rib (4 patients). In fibrous dysplasia of the first rib, it was associated with a complementary subclavicular approach for resection of the sternal end of the first rib. Rib resection was associated with partial anterior scalenectomy in one patient because of the close relationship between the anterior scalene and the brachial plexus.

Scalenectomy allowed complete release of brachial plexus compression [28]. A deltopectoral or subclavicular approach could be carried out for the exploration of the clavipectoral process in order to section the aponeurosis of the subclavicular muscle and / or the coracoclavicular and coraco-costal ligaments if necessary [1, 8, 13]. Most often, scalenectomy concerns the middle scalene in order to prevent lateral migra-

tion of the vascular-nervous bundle which can be compressed between the clavicle and the first rib [28].

The most common operative incident in the literature is pleural rupture, which progresses well with chest drainage [29, 30]. This is the case in one of our patients. Furthermore, it allowed better drainage of the supraclavicular cavity reducing the risk of postoperative pain [30]. The immediate postoperative course was marked by complete remission of pain and paresthesias. Healing was achieved 15 days postoperatively. Before 2 months postoperatively, we noted a complete remission of the symptoms in all our patients. This was found among several authors [15, 29-30]. On the other hand, in 3 patients, we noted the recurrence of symptoms such as paresthesia of the upper limb from 2 months after the intervention. This could be explained by irritation of the brachial plexus or incomplete resection of the anterior scalene during surgery [15, 19, 30] which was not the case in our series. This recurrence has been reported in the literature, some authors of which proposed medical treatment associated with physiotherapy.

This rehabilitation, if systematically indicated after surgery, could risk aggravating the lesions if it was too violent [10, 13, 22]. Indeed, exercises should never trigger or increase pain during or after sessions. The indication for functional motor rehabilitation was aimed at patients who presented painful manifestations that were more or less bothersome on a functional level [13, 22-24]. All our patients benefited from rehabilitation for three months with two to three sessions per week. Satisfaction was noted in all patients. The symptoms persisted after 2 years in one patient but were controlled by analgesics. The regularity of rehabilitation sessions favored success and regression of symptoms [10, 29, 30].

The evaluation of the degree of satisfaction with surgery in our patients was made by residents using a visual scale as recommended by the authors [10, 15]. It was generally satisfactory for 4 patients, 3 of whom were classified “good” and one classified “excellent”. The fair result was mentioned in the patient with residual pain after a follow-up of 2 years. Our results confirm, after an average follow-up of 8 years, the improvement in the quality of life after surgery of patients suffering from cervico-thoraco-brachial crossing syndrome [10, 15, 30, 31].

5. Conclusion

The cervical rib is no longer an isolated clinical entity. It is rather part of the cervico-thoraco-brachial crossing syndrome “thoracic outlet syndrome”. The management of symptomatic cases requires multidisciplinary involvement. However, cervical rib resection is most often associated with an anterior scalenectomy. It made it possible to avoid resection of the first rib, which is a cumbersome procedure with serious complications. Resection of the first rib is only justified in cases of proven anatomical anomaly of the latter. Primary surgery in the management of cervico-thoraco-brachial crossing syndrome offers the best results and remains standard treatment. Surgery associated with rehabilitation is increasingly gaining a place of choice in the therapeutic arsenal of this pathology but recurrences are always possible, motivating long-term follow-up of patients.

Abbreviations

C	Cervical Rib
CTBCS	Cervico-Thoraco-Brachial Crossing Syndrome
D	Dorsal Rib
EMG	Electromyogram
M	Male
VAS	Visual Analog Scale
W	Women
Y	Year

Author Contributions

Kondo Bignandi: Conceptualization, Data curation, Formal Analysis, Funding acquisition, Investigation, Methodology, Writing – original draft, Writing – review & editing

Souleymane Diatta: Funding acquisition, Project administration, Resources, Supervision, Validation

Moussa Seck Diop: Methodology, Software, Validation, Visualization

Pape Amath Diagne: Formal Analysis, Validation, Visualization

Pape Ousmane Ba: Formal Analysis, Methodology, Resources, Visualization

Gabriel Amadou Ciss: Formal Analysis, Methodology, Supervision, Visualization

Conflicts of Interest

The authors declare no conflicts of interest.

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Research Fields

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