Diagnosis of thoracic outlet syndrome.
Value of angiography in the sitting position

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Abstract
Purpose: Thoracic outlet syndrome includes arterial, venous or neurological symptoms. Frequently difficult to diagnose clinically, confirmatory imaging studies are usually required. The purpose of this study is to review the diagnostic work-up during management of patients with thoracic outlet syndrome and demonstrate the value of angiography in the sitting position.

Materials and methods: Retrospective study of 81 surgical procedures for thoracic outlet syndrome, between 1997 and 2005, in 56 patients aged 17-57 years. Surgery was bilateral in 26 cases, with bilateral surgery in a single setting for 1 patient. All patients presented clinical symptoms confirmed on US, angiography, venogram or EMG. Angiography, from a transfemoral approach, was initially performed in the supine position, without and with dynamic maneuver, and in the sitting position with dynamic maneuver when needed.

Results: In 48 patients, supine rest angiography showed stenosis in 6% of cases and supine dynamic angiography showed stenosis in 81% of cases, with severe stenosis in only 35% of cases (stenosis >80% or arterial occlusion). Angiography in the sitting position was performed in 33 patients, showing worsening of stenosis in 91% of cases, with severe stenosis in 87%.

Conclusion: Angiography in the sitting position with dynamic maneuver improves the sensitivity for detection of thoracic outlet syndrome. This procedure may be considered in addition to other imaging modalities routinely used including Doppler US, CT and MRI.

Key words: Thoracic outlet syndrome. Angiography.


Thoracic outlet syndrome is relatively frequent and often overlooked (5-10% of patients with upper extremity pain). Diagnosis is difficult and requires direct correlation between a clinically confirmed compression syndrome and an anatomical cause for narrowing of the thoracic outlet, a naturally narrow and non-stretchable conduit that is widely variable in the general population. In other words, morphological and anatomical abnormalities observed at clinical evaluation and complementary work-up are only relevant when symptomatic. Multiple maneuvers have been described in the literature: Adson, Wright or Eden maneuvers, Roos test or Tinel sign. These are non-specific and not sufficient by themselves for diagnosis. Each clinical maneuver has an average sensitivity of 72% and specificity of 53%, but the combination of two maneuvers (Adson and Wright maneuvers for example) improves sensitivity (79%) and specificity (76%). As a result, indications for surgical management are difficult to confirm, and dedicated work-up with reliable complementary techniques is needed. The purpose of this paper...
is to review the imaging work-up of thoracic outlet syndrome and demonstrate the value of evaluation in the sitting position, corresponding to the typical symptomatic position, at the time of conventional angiography.

Materials and methods

Patient population

Retrospective study of 81 surgical interventions performed in 56 patients with thoracic outlet syndrome between 1997 and 2005. The patient population included 45 females and 11 males (sex-ratio of 4.5), aged 17-57 years (mean age of 37.7 years). Twenty-six patients underwent bilateral surgery, in two settings for 25 patients and in a single setting for 1 patient. Inclusion criteria were the presence of vascular or neurological symptoms or signs of thoracic outlet syndrome. Surgery was indicated for patients with persistent disabling symptoms in spite of optimal conservative management (NSAIDS, physical therapy, and local injections for a few patients). All patients underwent surgery using the transaxillary first rib resection procedure described by Ross, including transection of the subclavian, anterior and middle scalene and first and second intercostal muscles along with resection of an accessory rib when necessary. No surgical failure requiring re-intervention occurred in our patient population. Forty patients (72%) reported significant symptomatic improvement or complete symptomatic relief after a mean follow-up of 56.7 months.

Clinical features

Symptoms were most frequently mixed (with neurological component) in 41 patients (73.2%), arterial only in 8 patients (14.3%), and venous only in 6 patients (10.7%). Pain was generally the main symptom, considered severe when refractory to pain medication, daily or permanent, or nocturnal and causing insomnia. Pain was considered important when it evolved by recurring flare-ups and, while non-disabling, interfered with daily activities. Arterial symptoms were characterized by stress related ischemia (pain, cramps, upper extremity fatigue during daily activities). One patient presented with acute upper extremity ischemia requiring bypass surgery. Neurological symptoms included paresthesia, dysesthesia and hyposthesia, typically C7 and C8 in distribution, often worsened by positional maneuvers. Numbness was described by patients as a dead hand sensation with clumsiness. Weakness, mainly involving the intersosseous and hypothenar muscles, and rarely the biceps or triceps muscles, could cause patients to drop objects. Patients with venous symptoms complained of edema. Two patients presented with upper extremity venous thrombosis.

Symptoms were right sided in 19 patients (34%), left sided in 19 patients (34%) and bilateral in 18 patients (32%). Contralateral occurrence of symptoms following surgery was recorded in 9 patients (16%) over a 6 month to 5 year period. Symptom duration ranged from 1 month to 10 years (median of 18 months), with diagnosis often delayed.

In a number of patients, alternate diagnoses had been considered, including carpal tunnel syndrome (6 patients, 11%), ulnar nerve compression (4 patients, 7%), cervicobrachial neuralgia from cervical disk herniation (2 patients), tendinitis, and arterial dissection (1 patient). Symptoms occurred following trauma in 3 patients, including clavicular fracture (2 patients) and brachial plexus stretch injury (1 patient).

Imaging work-up

Thoracic spine radiographs were obtained in all patients to detect the presence of accessory ribs (6 patients, 11%), apophyseal spondylolisthesis (1 patient), or first rib synostosis (1 patient).

EMG was obtained in 22 patients with neurological deficits. This showed C7 or C8 radicular abnormality in 73% of cases. Several examinations were performed in patients with vascular symptoms. Venography was performed in patients with venous symptoms (15 patients, 27%). Bilateral injections were performed, with arms extended along the body (series 1), then with the arms elevated (series 2). Doppler US was performed in 7 patients (12%), positive in 5 patients (71%). Most patients underwent angiography (48 patients, 86%). Forty-six angiograms were obtained by the value of evaluation in the sitting position, corresponding to the typical symptomatic position, at the time of conventional angiography.

Results

Angiographic results are summarized in table I. In 9 patients (19%), the first angiographic series was negative and the second series was positive; in 39 patients (81%), the first series was negative and the second series was positive. Thirty three (69%) of 48 patients underwent a third angiographic series in the sitting position. This third series was not necessary for diagnosis in 13 cases (35% with stage 2+ or 3 on the first 2 series). Also, 2 patients underwent angiographic evaluation at outside facilities where evaluation in the sitting position was not performed. In the 33 patients who underwent all 3 angiographic series, none had 3 negative series. Only the third series was positive for 8 patients (24%). No patient showed stable stenoses on all three series. Worsening was demonstrated in 30 patients (91%) between series 2 and 3, including 87% of

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very significant stenoses (demonstration of stage 2+ or 3). No angiogram related complication was recorded.

Venography was performed in 13 patients. Five patients underwent a single positive series, and 8 patients underwent 2 series. In 6 patients (75%), the first series was negative and the second series was positive. Stenoses were stable on both series in two patients. Worsening was demonstrated between series 1 and 2 in 75% of patients, with very significant stenoses in 33% (demonstration of stage 2+ and 3).

**Discussion**

Angiography, similar to Doppler US, venography, CT and MRI does not reproduce the conditions under which symptoms occur (1) and dynamic evaluation is necessary in patients with thoracic outlet syndrome, where compression of the brachial plexus is much more frequent than compression of the subclavian artery (2% of cases) (2).

The thoracic outlet is a narrow conduit and traversing neurovascular structures may easily be compressed by physiologic or non-physiologic changes in its size, such as during shoulder, cervical spine and rib cage movements. The interscalene triangle is involved in 49% of cases. It is bound anteriorly by the anterior scalene muscle that may compress the artery during contraction, inferiorly by the upper margin of the first rib, especially in patients with anatomic variants, and posteriorly by the middle and posterior scalene muscles. The subclavian artery and brachial plexus traverse this triangle. The subclavian vein courses in the prescalene space. In 18% of patients, compression occurs at the costoclavicular space, bound superiorly by the clavicle, anteriorly by the subclavius muscle, and posteriorly by the first and second ribs, especially in hyperabduction (3).

The subclavian vein is ventral to the artery; the brachial plexus is more posterior. Finally, the subcoracoid tunnel or retropectoralis minor space is located dorsal to the pectoralis minor muscle, ventral to the subscapularis muscle, and under the coracoid process. Anatomical variants may be congenital (0.12-1% of the general population): complete or partial absence of regression of lower cervical ribs, hypertrophy of the C7 transverse processes, absence of the first rib, congenital bands and ligaments or acquired: impaired spine posture, first rib fracture or hypertrophic clavicle callus formation. Thin patients with drooping clavicles may be predisposed (4, 5).

We noted a female predominance for the arterial and neurological types, both in our

<table>
<thead>
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<th>Table I</th>
<th>Angiographic results.</th>
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<td>Stage 0</td>
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<td>Series 1 (n=48)</td>
<td>45</td>
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<tr>
<td>Supine</td>
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<td>Series 2 (n=48)</td>
<td>9</td>
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<td>Supine, arms up</td>
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<td>Series 3 (n=33)</td>
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<td>Sitting, arms up</td>
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patients (45 females and 11 males) and in
the literature (6). This may be explained
by the more vertical orientation of the
first rib in females and a more thoracic ty-
p of respiration compounded by the estro-
gen and progesterone impregnation caus-
ing muscular and ligamentous laxity (7).
Age also plays a role, as shown in our se-
ries (mean age of 37.4 years for females and
42.5 years for male), and reports from
the literature (7). This may be explained
by several morphological changes of the
rib cage (8, 9), narrowing of the costoclav-
icular space (10) and progressive drop-
ning of the clavicles.

Most reports (7) indicate the more fre-
quent right sided involvement as well as
the presence of bilateral symptoms in
10-60% of cases. In our series, involve-
ment was bilateral in 18 patients (32%),
left sided in 19 patients (34%) and right si-
ded in 19 patients (34%).

In the management of thoracic outlet
symptoms, complementary examinations
may be performed, but none provides dia-
gnostic certainty. These examinations are
mainly helpful for differential diagnosis.
A frontal radiograph of the thoracic spine
should always be obtained (11) to detect
bone abnormalities (present in eight pa-
tients or 14% of our patients), especially
cervical ribs. EMG may confirm the dis-
tribution of nerve involvement (12), fre-
cently C7 or C8, the severity of involve-
ment, and allows exclusion of a differential
diagnosis (13, 14). In our study, EMG was
not always performed, but it was helpful
in 73% of cases when performed.

Doppler US is a non-invasive technique
that allows differentiation between ve-
nous thrombosis and spasm in patients
with venous symptoms, demonstration of
a collateral circulation, and detection of
arterial abnormalities, plaque or emboli-
genic aneurysm. US is easy to perform
and its sensitivity is higher for patients
with more pronounced clinical findings
(15, 16), especially with the help of the
Adson maneuver (arm abduction and re-
trpulsion, hand behind neck, contralat-
eral head rotation and deep inspiration)
that mobilizes the thoracic outlet. Dyna-
mic maneuvers may also be performed,
preferably those reproducing the symp-
toms, at the time of scanning to demons-
trate variations in vascular flow. A gradual
decrease in arterial flow to complete ab-
sence of flow indicates compression of the
subclavian artery. Dynamic arterial occlu-
sion is abnormal when occurring at low to
moderate amplitude movements. Dyna-
mic arterial occlusion in hyperabduction
may be physiologic and occur in 30-50% of
normal subjects (17). This technique is
operator dependent and requires a cer-
tain degree of expertise along with ex-
cellent knowledge of the underlying pa-
thology, with reported sensitivity and
specificity values of 84% and 89% in the
literature (18). Even though US does not
allow optimal evaluation of all regional
anatomical structures, it can easily comple-
ment physical examination as a first line
imaging study. In our study, Doppler US
was performed in only 7 patients. Early
on, the surgical team requested Doppler US
in all patients. However, because of the
discordance between results from US and
combined clinical findings and dynamic
maneuvers, angiography with dynamic
maneuvers and sitting acquisition soon
replaced Doppler US as a complementary
evaluation in the work-up of patients
with thoracic outlet syndrome. Other cen-
ters propose CT or MRI evaluation in all
patients. This work-up is justified since
surgery is invasive and surgeons require
the most sensitive imaging studies availa-
ble to confirm the diagnosis. The situa-
tion is similar to patients with carotid
artery stenosis where the surgeon fre-
cently requires CTA or MRA confirma-
tion after US prior to surgical management.

MRI and CT evaluation allow excellent de-
lineation of the site and cause of symp-
toms and functional anatomy of the thoracic
outlet while enabling acquisition of excel-
ent angiographic images. These are usually
second line imaging techniques, even thou-
gh their indications are increasing. Images
with the arm extended along the side of the
body and elevated must be acquired.
CT allows multiplanar reconstructions
(MPR) and angiography with the help of
iodinated contrast material injected in-
travenously in the arm opposite the symp-
tomatic side (19). Sagittal reformatted
images are useful to evaluate the arteries
and detect the presence of post-stenotic
aneurysmal dilatation. In addition, CT is
optimal for bone evaluation and may pro-
vide imaging guidance for procedures in
patients with radicular compression (20).
Radiation exposure is reduced by the use
of several techniques (tube current modu-
lation for example) while maintaining ex-
cellent image quality (21). MRI provides
improved evaluation of soft tissues, espe-
cially the brachial plexus, fibrous bands
and muscle anomalies with the help of sa-
gittal and coronal T1W acquisitions (22,
23). Time of flight or contrast material en-
hanced angiographic acquisitions complete
the MRI evaluation (24). A limitation to MR
remains the longer duration of the ex-
amination. A pitfall for both CT and
MRI is the supine acquisition and impai-
red ability to perform dynamic maneu-
vers due to bore diameter (23). Supine ac-
quisions may lead to an increase in the
rate of false negative results (23) with re-
duced sensitivity compared to US (22, 24).

Nonetheless, the availability of open MR
scanners could address some of these pitfalls.
Venography is excellent in the evaluation
of patients with venous symptoms becau-
es of its high sensitivity (93% in our series)
and ease of performing dynamic maneu-
vers. On the other hand, it cannot charac-
terize the nature of the compression. In
addition, venous compression has been des-
bribed in asymptomatic patients, re-
quiring that results be interpreted with
caution (22).

In our series, 91% of patients who unde-
rwent angiography in the sitting position
showed worsening of stenosis (87% with
very significant stenosis). In addition,
8 patients (24%) with stenosis had negati-
ve angiograms in the supine position, il-
lustrating the improved sensitivity of the
sitting acquisition. Angiography is not
routinely performed in patients with tho-
racic outlet syndrome without specific
vascular symptoms, and it should prefera-
ibly be performed in patients with obvious
arterial involvement such as aneurysm or
occlusion (19). However, angiography
may be helpful in patients with negative
results at non-invasive imaging and posi-
tive clinical findings or when the surgical
team requires confirmation of findings by
this sensitive examination prior to invasi-
ve surgery. The acquisition of angiogra-
phy in the sitting position was proposed
based on the results of angiography in the
supine position. Results from supine an-
giography were well below those obtai-
ned at clinical examination in the sitting
position with dynamic maneuvers. Be-
cause the only difference was the sitting
position, angiography in the sitting posi-
tion with use of similar maneuvers was
attempted. The variation in the degree of
stenosis between supine and sitting posi-
tions on angiography and other imaging
studies (US for example) suggests that
gravity during arm abduction in the sit-
ting position plays a substantial role in the
pathophysiology of arterial stenosis. As
such, the weight of the shoulder presumably contributes to compression of the neurovascular bundle at the thoracic outlet. Nonetheless, angiography is not without limitations. Angiography does not allow diagnosis of the cause of compression, a fact that probably is of little clinical significance since it does not affect surgical management of patients with positive clinical findings. Angiography is invasive, exposes patients to ionizing radiation, and requires the administration of iodinated contrast material. In addition, the potential morbidity of angiography in the sitting position using a transfemoral approach and the Seldinger technique is probably superior to that of angiography in the supine position. Finally, it should be noted that 10 patients (18%) referred for unilateral symptoms had bilateral positive findings on angiogram. Nine (16%) of these patients presented with contralateral recurrence of symptoms between 6 months and 5 years after surgery. This result does not raise questions about the role of angiography because, in our study, the goal was not to demonstrate the presence of positional arterial variations at the thoracic outlet (the prevalence of positive results at invasive positional examination in the general asymptomatic population is unknown) but to identify symptomatic patients that would benefit from surgery. These are biases in our study, typical of retrospective studies, but also patient selection biases, since all underwent surgery.

Conclusion
Angiography, in patients with thoracic outlet syndrome, allows demonstration of intrinsic lesions responsible for symptoms, which is important in the presurgical period, with the use of dynamic maneuvers and acquisitions in the sitting position that increase the sensitivity of the examination, as shown by our results. Angiography is more sensitive than Doppler US and other cross-sectional imaging modalities (CT or MRI) to demonstrate some vascular abnormalities though the latter allow improved evaluation of the regional anatomy of the thoracic outlet. However, the purpose of our study was not to promote angiography in the routine work-up of patients with thoracic outlet syndrome. Angiography is indicated in patients with diagnostic difficulties unres

res by non-invasive imaging modalities. It is reasonable to propose that all patients with possible thoracic outlet syndrome first undergo clinical evaluation with dynamic maneuvers performed in the sitting position, complemented by Doppler US and thoracic spine radiographs. If US and radiographs are sufficient and the surgeon does not require additional imaging because of the correlation with clinical findings, surgical management could then be entertained. If US is negative (discordant with clinical findings) and/or the surgeon wishes to obtain additional imaging to determine the site and cause of symptoms, visualize the functional anatomy of the thoracic outlet syndrome to finally ascertain the indication for surgical management, CT or MRI including angiography would be performed. If CTA or MRA examinations are negative and the clinical suspicion remains, angiography with dynamic maneuvers in the sitting position should then be performed.

References