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Multiple thoracic bilateral intersegmental arterial trunks

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SUMMARY

A bilateral intersegmental trunk is formed when a pair of left and right thoracic or lumbar intersegmental arteries share a common aortic origin. This rare variant – most frequently found at the low lumbar level – is exceptional in the thoracic region.

We describe five angiographic observations of multiple thoracic bilateral intersegmental trunks (average 4.6 per patient, range 2 to 8, with a predominance between T6 and T8).

The angiographic, comparative and developmental anatomy of thoracic BITs is discussed, as well as their clinical importance for angiographers and surgeons performing procedures that involve the thoracic aorta or the vertebral column.

Key words: Anatomic Variation [G07.049] – Angiography [E01.370.350.700.060] – Aorta, Thoracic [A07.231.114.056.372] – Embryology [H01.158.273.200.350] – Spinal Cord [A08.186.854]

INTRODUCTION

While thoracic and lumbar intersegmental arteries (ISA) generally arise from the dorsal wall of the aorta as individual branches, they can also participate in the formation of intersegmental trunks. Two types of trunks are recognized. Unilateral intersegmental trunks provide two or more adjacent ISAs. They can be observed at any vertebral level, but are most common in the upper thoracic (T2 to T5) and low lumbar (L3) regions (Adachi, 1928). Unilateral intersegmental trunks are further subdivided into complete and incomplete forms depending on the presence or absence of a full set of intersegmental branches (*i.e.*, medial, dorsal and lateral branches). An incomplete intersegmental trunk is typically associated with an isolated dorsospinal artery (Chiras et al., 1979).

A bilateral intersegmental trunk (BIT) is formed when a pair of left and right thoracic or lumbar intersegmental arteries share a common aortic origin. This rare variant – most frequently found at the low lumbar level (notably L4) – is rare in the thoracic region, at the possible exception of the Japanese population (Adachi, 1928). The possibility of multiple thoracic BITs appears even more exceptional, with a only one prior observation known to the author, made on an anatomical specimen by Ernst in 1899 (Ernst, 1899). Yet, a thor-

Table 1. Bilateral intersegmental trunk (BIT) distribution in five patients. X = BIT present, - = BIT absent, n = not investigated.

Patient	1	2	3	4	5
Gender	F	F	F	F	M
Age	45	10	23	47	58
T2	X	n	-	-	n
T3	X	X	-	-	-
T4	X	X	-	X	-
T5	X	-	-	X	-
T6	X	X	-	X	X
T7	X	X	-	X	X
T8	X	-	X	X	X
T9	X	-	-	X	-
T10	-	-	X	-	-
T11	-	-	-	-	-
T12	-	-	-	-	-

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ough understanding of the normal vascular anatomy and its variations is essential to the safe and efficient practice of angiography, notably during the investigation and treatment of spinal vascular anomalies. We therefore report our angiographic findings in five patients with multiple thoracic BITs.

CASE SERIES

Multiple BITs were observed during spinal digital

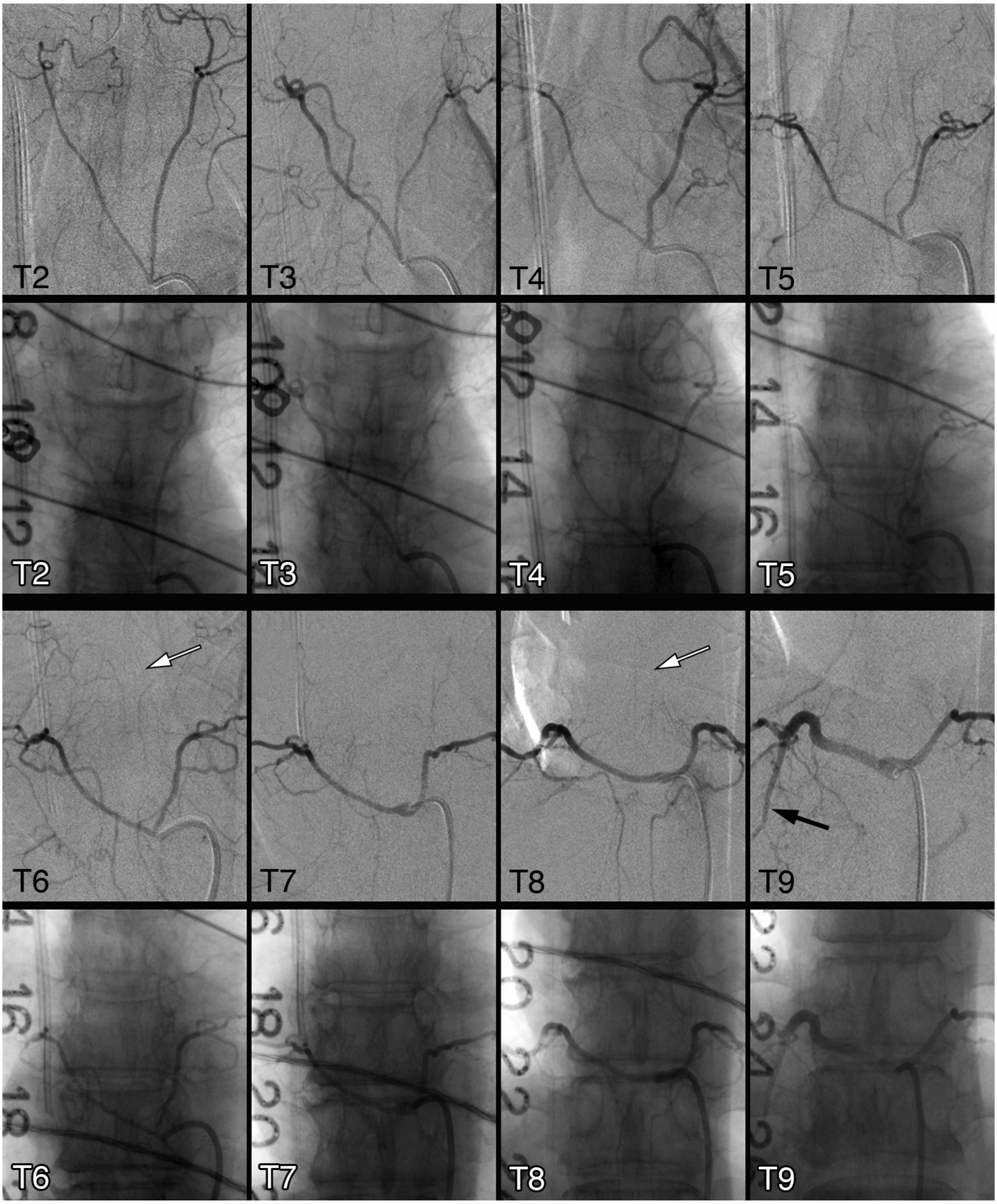

Fig 1. 45-year-old woman with 8 thoracic BITs (T12 to T9). This illustration shows subtracted (above) and native (below) posteroanterior projections of the selective catheterization of the thoracic ISAs between T2 and T9. Two anterior radiculomedullary arteries are noted at T6 and T8 (white arrows). The T9 BIT also provides the right T10 ISA (black arrow). Not that the arterial segment constituting the common trunk – short at the upper levels - increases in length in a craniocaudal progression.

subtraction angiography (SpDSA) in five patients (4 women and one man, average age=37, range 10 to 58) between October 2010 and July 2016.

BITs were found between T2 and T10 and were multiple in all instances (2 to 8 per patient, average 4.6); they were contiguous in 3 patients and skipped a level in the other two (Table 1). Other notable variations included four T12-L1 complete unilateral intersegmental trunks (in 3 patients) and one left L4 ISA of common iliac origin. Lumbar BITs were noted in only one patient.

Patient 1 – SpDSA was obtained in a 45-year-old woman with vertebral osteosarcoma. BITs were incidentally noted between T2 and T9 (Fig. 1). Note that the T9 BIT combines unilateral and bilateral configurations to also provide right T10.

Patient 2 – SpDSA was performed in a 10-year-old girl with a spinal epidural arteriovenous fistula. BITs were noted at T3, T4, T6 and T7. A right T12-L1 unilateral intersegmental trunk was observed as well. The right T3 BIT also provided a prominent bronchial artery (Fig. 2).

Patient 3 – A 23-year-old woman was investigated for a suspected thoracic epidural vascular malformation. SpDSA revealed several anatomical variants, including BITs at T8 and T10, the latter providing a prominent anterior radiculomedullary artery (Fig. 3). In addition, left and right L1-T12 unilateral intersegmental trunks, and a left L4 ISA arising from the left iliolumbar artery were also noted.

Patient 4 – SpDSA was obtained in a 47-year-old woman with a thoracic epidural hematoma. BITs were documented between T4 and T9 and between L2 and L4 (Fig. 4). The T5 BIT also provided a prominent bronchial artery.

Patient 5 – SpDSA was performed in a 58-year-old man with a spinal dural arteriovenous fistula. Angiography also documented BITs at T6, T7 and T8, as well as a right L1-T12 unilateral intersegmental trunk (not illustrated).

DISCUSSION

Bilateral intersegmental trunks

In spite of its importance, notably to spinal angiographers and spine surgeons, the mode of origin of thoracic ISAs as individual branches or trunks is rarely discussed. Early anatomists believed thoracic BITs to be common. Winslow (1776) noted for example that “quite often the intercostal [arteries] for both sides come from a single trunk” whereas Portal (1804) described them as “pairs united in very short trunks” (both authors being possibly influenced by observations made non-human mammals). Henle (1868) offered a more accurate description, noting that the thoracic and lumbar ISAs arose from the aorta as “two vertical rows” with rare instances of lumbar BITs. In the same work,

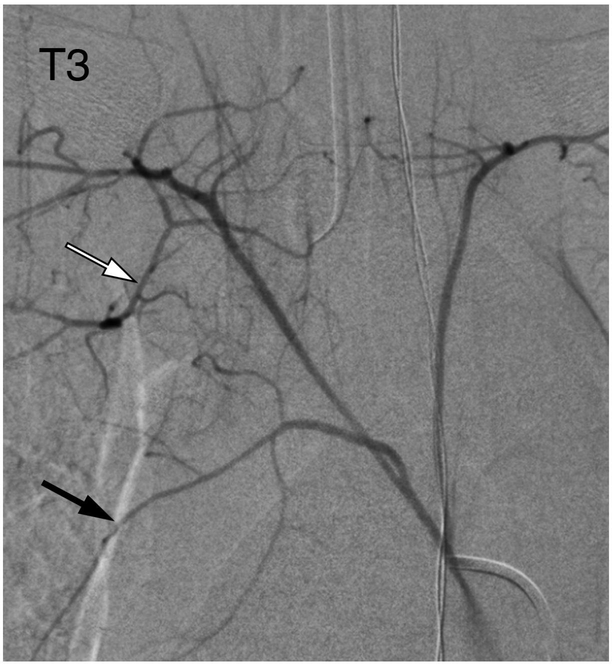

Fig 2. 10-year-old girl with 4 thoracic BITs (T3, T4, T6 and T7). This selective injection of the T3 BIT shows a prominent right bronchial branch (black arrow). The right T4 ISA (white arrow) is partially opacified via collateral channels but does not constitute a T3-T4 unilateral trunk.

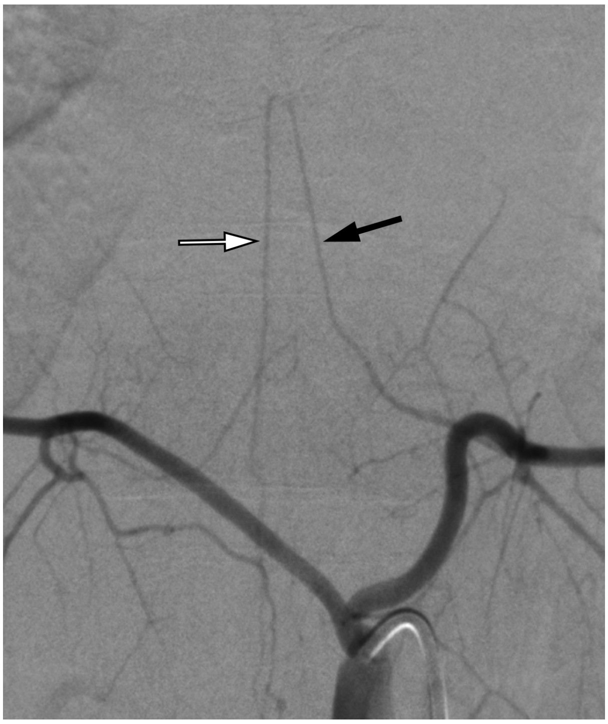

Fig. 3.- 23-year-old woman with two thoracic BITs (T8 and T10). This selective angiography of the left T10 BIT shows a prominent anterior radiculomedullary artery (artery of Adamkiewicz, black arrow) supplying the anterior spinal artery (white arrow).

Krause added that - as a variant - the left and right 12th ISAs can share a common origin and mentioned a case from Meckel in which the upper four lumbar arteries arose in that manner (Krause, 1868). While admitting that ISAs were "usually distinctly single vessels", Evans (1912) still believed BITs to be fairly common, although he mentioned – following Krause – that they were generally found in the lowest intercostal (T12) and lowest lumbar (L4) regions.

The mode of origin of ISAs from the aorta is generally not discussed in general anatomy textbooks, which sometimes mention that thoracic BITs are rare or exceptional (Poirier, 1912; Hovelacque et

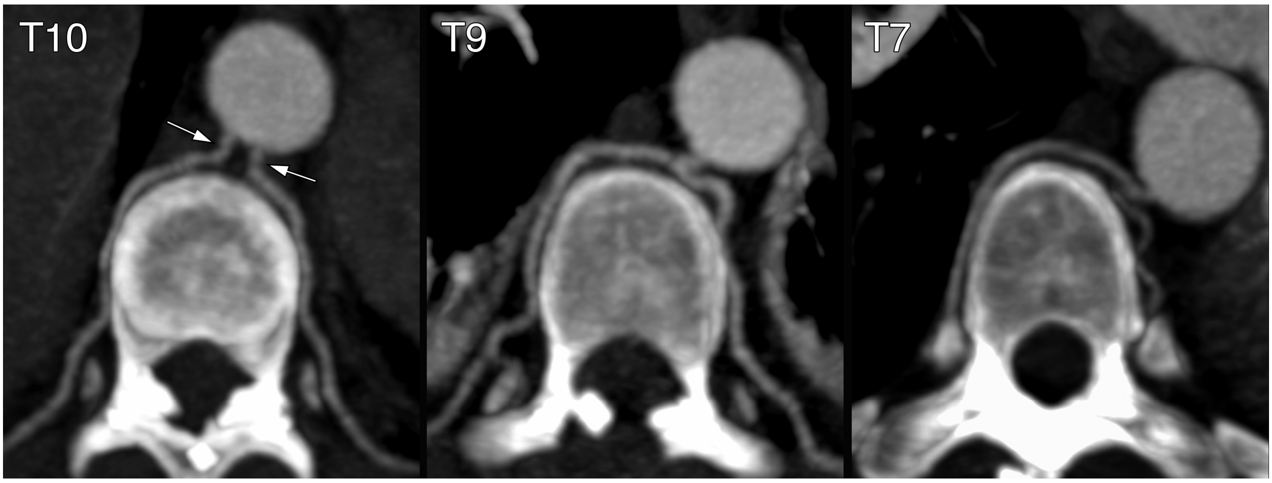

Fig 4. 47-year-old woman with 6 thoracic BITs (T4 to T9). These CTA images passing through T10 (left), T9 (middle) and T7 (right) document the appearance of the T9 and T7 BITs on axial imaging. The T10 ISAs originate as individual branches (white arrows), but the parallel course of their proximal segments illustrates well how a closer apposition during the merging of the primitive dorsal aortas into the adult aorta might have led to the formation of a single trunk. Note the progressive shift of the aorta from a paramedian to a leftward position, which leads to increasing stretching of the right-sided ISAs and tortuosity of the left-sided ones.

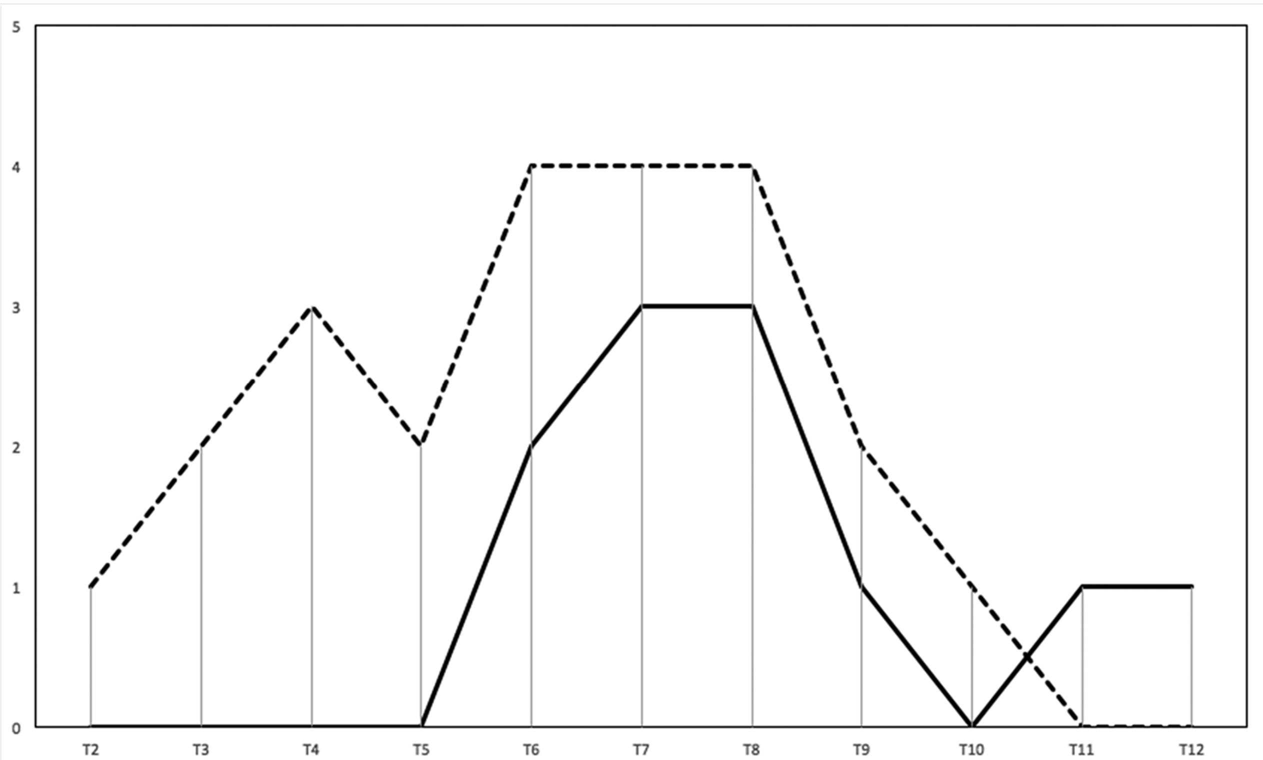

Fig 5. Graphic representation of the distribution of BITs in Adachi's specimens and the current cohort (vertical axis = number of observations, horizontal axis = vertebral level). In both series, a predominance was found between T6 and T8, while the purportedly more common low thoracic locations (T11 and T12) were seen only once each in Adachi's group and not at all in our patients. Continuous line = Adachi's data; Dotted line = current series.

al., 1937) but more often disregard that type of variation completely (Standring, 2016). More specialized works, on the other hand, typically consider the arterial supply to the vertebral column distally to its aortic take off (Crock, 1996; Parke et al., 2011). For example, Canavan (1943) studied the posterior intercostal arteries of 915 aortas without paying attention to their origin as individual branches or trunks, while reference works on spinal angiography published since its introduction in the nineteen sixties have added little or nothing to that specific topic (Djindjian et al., 1970; Lazorthes et al., 1973; Doppman et al., 1969; Chiras et al., 1979; Chiras et al., 1982).

Three recent studies - by Shimizu and co-authors (2005) investigating five human cadavers, by Kauppila (1994) examining 32 postmortem angiograms to the thoracolumbar junction (T10-T12), and by Kustan et al. (2016, unpublished data) analyzing of the distribution of intersegmental ostia in 40 full spinal angiograms – found no instance of thoracic BITs.

The most detailed information available today is still found in Adachi's masterpiece on arterial anatomy published in 1928 (Adachi, 1928). Adachi reported findings based on the investigation of 48 thoraxes and 63 abdomens. He found BITs to be generally rare compared to unilateral trunks, and noted an inverse relationship in regard to their respective occurrence. No BITs were seen in the upper thoracic region (T1-T5) and only a few in the low thoracic area, while they were somewhat more common at the mid-thoracic level. Overall, he found BITs at 11 of the 576 thoracic levels he studied (1.9%), the highest prevalence being noted at T7 and T8 (3 instances each, or 6.3%) (Fig. 5). Thoracic BITs were found in 2 of 10 women (20%) and 9 of 38 men (24%) (our series shows a clear female predominance (80%) but is too limited to draw firm conclusions). Unfortunately, Adachi did not mention the frequency with multiple thoracic BITs were observed in a single specimen.

BITs were more common in the lumbar region; Adachi saw 11 of them between L1 and L4 (out of a total of 252 vertebral levels, or 4.4%), with the following distribution: L1 – 1 case (1.6%), L2 and L3 – 3 cases (4.8%), and L4 – 4 cases (6.3%). His material does not include L4 ISAs originating from the median sacral artery, as he paid no attention to their mode of origin as individual branches or trunks.

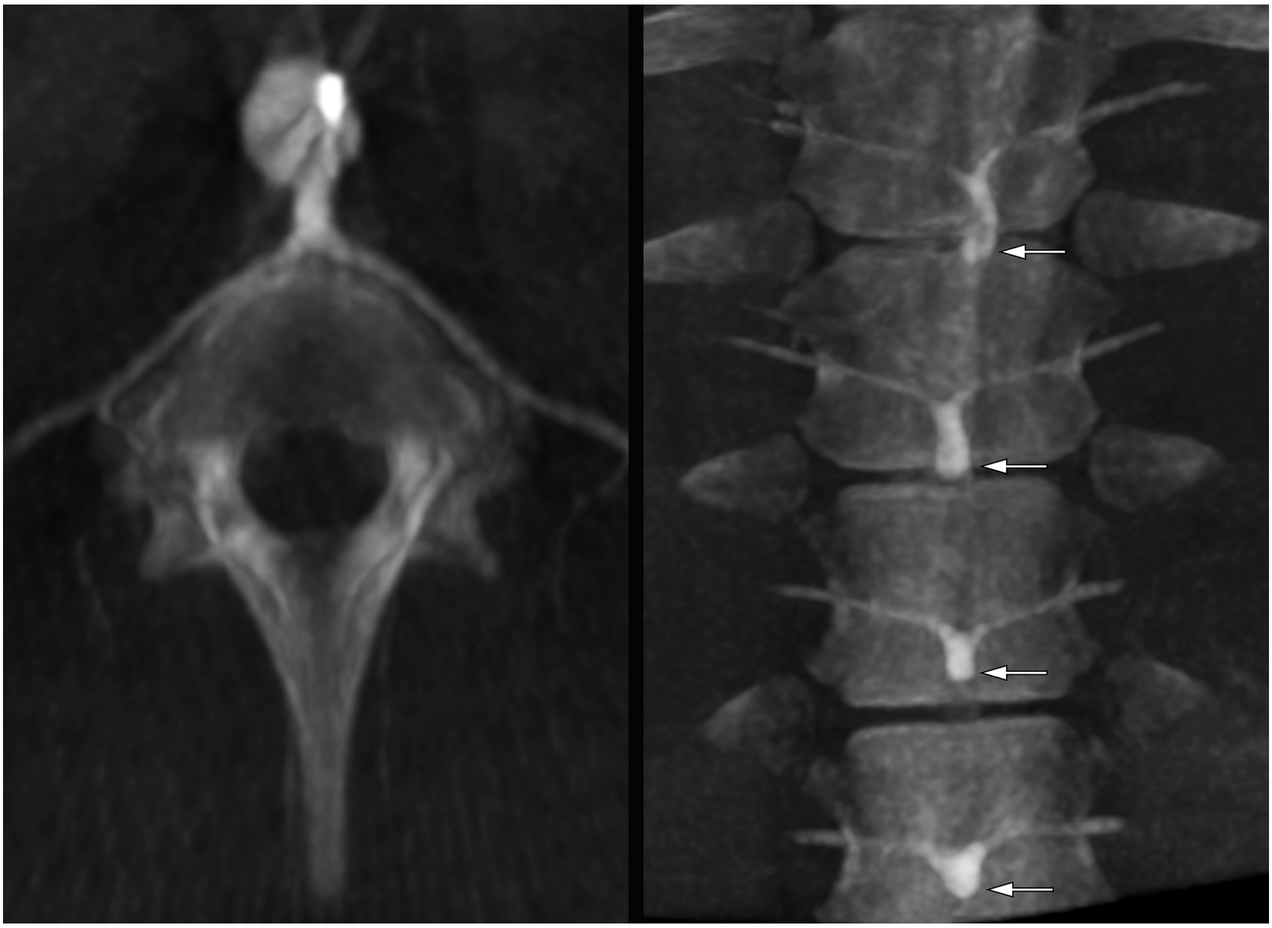

Fig 6. Thoracic BITs in the pig. These axial (left) and coronal (right) reconstructions of a three-dimensional aortic angiogram document the appearance of multiple BITs in the pig.

Bilateral thoracic intersegmental trunks

While BITs are infrequent, the existence of multiple thoracic BITs appear to be an even more exceptional configuration. Ernst (1899) reported what is to our knowledge the first and only published observation of multiple thoracic BITs extending from T4 to L4. As the aorta was cut along its length during a necropsy, Ernst remarked "that instead of two lines of ostia for the origin of the intercostal arteries only one existed, while the lumbar arteries arose in a single streak as well, so that the entire aorta was from top to bottom marked by a single row of holes."

One of the specimen studied by Khan and Haust (1979) while investigating variations in the origin of intercostal arteries seems to correspond to an instance of multiple thoracic BITs as well (T7 to T11), but the branches having been cut close to their origin, the authors were unable to ascertain the unilateral or bilateral nature of the variants (although the midline position of the ostia strongly suggest the latter).

Of note, multiple thoracic BITs should be distinguished from another rare variation in which a single dorsal aortic trunk with an ascending course provides a large number of bilateral ISAs (Chang et al., 2009; Brew et al., 2001; Jie et al., 2016).

Developmental and comparative anatomy

The adult aorta results from the fusion of two primitive dorsal aortas, each already carrying a set of ventral and dorsal branches:

"Both ventral and dorsal branches grow out before the primitive aortæ fuse, and consequently when this occurs an accurate apposition of the two aortae permits these branches to come off in pairs from the single aorta descendens." (Evans, 1912)

It has been proposed that BITs - similarly to the visceral arteries formed by the fusion of paired ventral branches – result from the fusion of two primitive dorsal ISAs rather than from a process of unilateral agenesis or disappearance. The observation of complete or incomplete septa within ISA trunks is consistent with that hypothesis (Ernst, 1899; Khan et al., 1979). Moreover, Ohale noted in the baboon a progressive craniocaudal transition of the dominant ISA configuration from paired ISAs (T3-T6) to common trunk with septations (T6-T7) to BITs (T9-T11), a pattern that matches an earlier report by Theile (1852), who saw BITs in the last three intercostal and in all lumbar levels of 4 specimen of Macaca (*Simia Inuus*). Ohale also found a corresponding craniocaudal trend when measuring the average distance separating the two branches of a single pair, from 0.3 mm at T3 to 0.01mm at T11, leading him to suggest "an initial gradual fusion in the cranial, to a more complete fusion in the caudal segments of the two dorsal aortas during early fœtal development"(Ohale, 1998). While this simple hypothesis is appealing, it fails to address the bimodal distribution (low thoracic and low lumbar) noted in humans and the reverse distribution observed in some mammals – ruminants and horses notably (Barone, 1996) – in which the most cranial aortic intercostal arteries, the fifth and sixth ones, "not uncommonly leave the aorta as a common stem" (Bradley, 1922). Holothoracic distributions have also been reported, for example in pigs (Fig. 6) (Barone, 1996). In order to explain these complex BIT patterns, Broman (1908) suggested the involvement of two embryological phenomena linked to the initial site of fusion of the paired primitive dorsal aortas (low thoracic) and the additional fusion process bringing the primitive roots of the common iliac arteries together (low lumbar).

In any case, the reasons behind both the rarity and – when present – the multiplicity of thoracic BITs in humans remains unclear, but our case series seems to suggest, at least for these patients, a locoregional disturbance rather than a focal variant, most likely in relation with the fusion of the paired primitive dorsal aortas into the single adult thoracolumbar aorta.

Clinical implications

A comprehensive knowledge of anatomical variations involving the vascular system is essential to the practice of angiography. Besides their purely morphological value, our findings are therefore of specific interest to the selective investigation of thoracic intercostal arteries, for example during diagnostic SpDSA or preoperative embolization of vertebral tumors. BITs can also play a role in the planning of surgical procedures involving the aorta and/or the vertebral column, since they can participate in the vascularization of the spinal cord (patients 1 and 3), while limiting the availability of collateral pathways after vessel sacrifice, particularly when they extend over several levels and/or are combined with unilateral intersegmental trunks. Finally, despite their median origin, BITs can provide significant bronchial branches (patients 2 and 4) and be involved in hemoptysis.

In summary, the occurrence of BITs at the thoracic level is extremely rare. When encountered, thoracic BITs are more common in the mid-thoracic region and tend to be multiple, on average 4.6 per individual in our series, with one reported instance of human holothoracic distribution in the literature.

Conflict of interest disclosure

There is no conflict of interest related to the submitted work. The author is a consultant for Codman Neurovascular and has received research grants from Siemens Medical.

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