VIA MEDICA

LETTER TO THE EDITOR

Folia Morphol.
Vol. 82, No. 2, pp. 445–446
DOI: 10.5603/FM.a2022.0087

ISSN 0015–5659
eISSN 1644–3284
journals.viamedica.pl

Comment on "A left circumflex aorta with a displaced thoracic duct in a 94-year-old male cadaver: a case report with discussion on embryology"

D. Borthakur*, F. George*, R. Dhingra*

All India Institute of Medical Sciences, New Delhi, India

[Received: 21 August 2022; Accepted: 28 September 2022; Early publication date: 14 October 2022]

We read the article "A left circumflex aorta with a displaced thoracic duct in a 94-year-old male cadaver: a case report with discussion on embryology" by Ostrowski et al. [7] with great interest. In fact this article was selected for presentation in the weekly journal club organized by Department of Anatomy at All India Institute of Medical Sciences (AIIMS), New Delhi, India. The meticulous dissection by authors showing the anomalous left circumflex aorta (LCA) and other associated structures was appreciated by the faculty. The article is well written about an interesting case report. The authors discussed their findings in details; however, we wanted to add few additional points. The authors hypothesized that the concomitant presence of scoliosis although apparently seemed to displace the LCA to the right side, but it is more prudent to consider it an anomaly during embryogenesis, specifically regression of the left dorsal aorta and its retro-oesophageal course. Agreeing with the authors and erstwhile observations by Sanchez Torres and Roldan Conesa [9], we also believe that the LCA anomaly is inborn and related to embryological deviation. It is not less than a miracle to witness such an extremely rare anomaly in an individual who lived 94 years without obvious clinical symptoms and surgical procedure, which raises a question here. It also emphasizes the need of modified approach towards the art of eliciting past medical history from the kin at the time of body donation. The aorta is seen significantly larger than average throughout its course, which can be due to increase in external diameter or aortic wall thickness with simultaneous reduction in elastin density in the tunica media with advancing age. Studies show that the number of elastic lamellae and the physiological circumferential stress per unit area of the circumferential lamellae remains always constant [2]. Therefore some degree of age-related ectasia is expected in the vessel, but significant dilatation without any compressive symptoms is difficult to interpret. Probably histology of the same could have revealed the state of degeneration of elastin fibres, their fibrosis along with elastin fragmentation in the tunica media as well as internal and external elastic laminae [5]. Considering the triad of LCA, scoliosis and aberrant right sided thoracic duct, we want to draw attention to an important gene *TBX1* (T-box DNA binding transcription factor) involved in 22q11 micro deletion syndrome (also known as the Di-George syndrome with multiple congenital defects) and Goldenhar syndrome with pharyngeal arch artery defects [4]. *TBX1* has been shown to be crucial for mesenteric lymphangiogenesis by regulating expression of vascular endothelial growth factor receptor 3 (VEGFR3) in endothelial cells [3]. An important observation was made that in *TBX*–null conditions, lymph angiogenesis did occur but was highly disorganized, which suggests that *TBX1* is not merely crucial for lymph angiogenesis but also important for its maintenance [3]. Thus, *TBX1* is not essential for lymphangiogenesis per se; rather, it is required for the devel-

Address for correspondence: Prof. Dr. R. Dhingra, PhD, Department of Anatomy, All India Institute of Medical Sciences (AIIMS), New Delhi, Ansari Nagar-110029, India, e-mail: renudhingraaiims@gmail.com

*All authors contributed equally to this work.

opment of the lymphatic network. This could explain the right sided thoracic duct. *TBX1* is also reported to be extensively expressed in the endothelium of blood vessels [8, 10]. Evidences suggest that genetic variant of *TBX1* is linked with idiopathic scoliosis and optimal expression of *TBX1* is required during pharyngeal arch artery development [1, 6]. Therefore a genetic study investigating the expression *TBX1* gene can be undertaken in similar cases to elucidate the underlying molecular embryonic regulatory mechanism. Also an attempt to explore the lymphatic drainage of left side could also have been done as the authors did not find any lymphatic duct draining into left venous angle (according to Ostrowski et al. [7] "No vessel draining into the left venous angle was visualised by macroscopic dissection."). A proofreading error which drew our attention is seen in the abstract section where thoracic duct is mentioned to be draining into the right internal carotid vein (according to Ostrowski et al. [7] "However, the thoracic duct was placed on the right, and drained into the right internal carotid vein.") which might perplex readers. Such an inadvertent error, although minor might confuse the readers. We hope our concerns will be considered and we will appreciate additional clarification in this regard. We are eagerly waiting for author's further investigative report to solve the molecular mystery of LCA embryogenesis.

Conflict of interest: None declared

REFERENCES

1. Anderson RH, Bamforth SD. Morphogenesis of the mammalian aortic arch arteries. Front Cell Dev Biol. 2022; 10: 892900, doi: 10.3389/fcell.2022.892900, indexed in Pubmed: 35620058.
2. Changes S, Human IN, Aortas T, et al. Mechanical and structural changes in human. Acta Biomater. 2020; 103: 172–188, doi: 10.1016/j.actbio.2019.12.024.
3. Chen Li, Mupo A, Huynh T, et al. Tbx1 regulates Vegfr3 and is required for lymphatic vessel development. J Cell Biol. 2010; 189(3): 417–424, doi: 10.1083/jcb.200912037, indexed in Pubmed: 20439995.
4. Glaeser AB, Santos AS, Diniz BL, et al. Candidate genes of oculo-auriculo-vertebral spectrum in 22q region: A systematic review. Am J Med Genet A. 2020; 182(11): 2624–2631, doi: 10.1002/ajmg.a.61841, indexed in Pubmed: 32893956.
5. Komutrattananont P, Mahakkanukrauh P, Das S. Morphology of the human aorta and age-related changes: anatomical facts. Anat Cell Biol. 2019; 52(2): 109–114, doi: 10.5115/acb.2019.52.2.109, indexed in Pubmed: 31338225.
6. Li Y, Wu Z, Xu L, et al. Genetic Variant of TBX1 Gene Is Functionally Associated With Adolescent Idiopathic Scoliosis in the Chinese Population. Spine (Phila Pa 1976). 2021; 46(1): 17–21, doi: 10.1097/BRS.0000000000003700, indexed in Pubmed: 32947497.
7. Ostrowski P, Popovchenko S, Bonczar M, et al. A left circumflex aorta with a displaced thoracic duct in a 94-year-old male cadaver: a case report with discussion on embryology. Folia Morphol. 2023; 82(2): 400–406, doi: 10.5603/FM.a2022.0043, indexed in Pubmed: 35481702.
8. Paylor R, Glaser B, Mupo A, et al. Tbx1 haploinsufficiency is linked to behavioral disorders in mice and humans: implications for 22q11 deletion syndrome. Proc Natl Acad Sci U S A. 2006; 103(20): 7729–7734, doi: 10.1073/pnas.0600206103, indexed in Pubmed: 16684884.
9. Sánchez Torres G, Roldán Conesa D. [Left aortic arch without a circumflex segment and a right descending aorta: a hypothetical case and a real example]. Arch Inst Cardiol Mex. 1989; 59(2): 125–131, indexed in Pubmed: 2764632.
10. Vitelli F, Morishima M, Taddei I, et al. Tbx1 mutation causes multiple cardiovascular defects and disrupts neural crest and cranial nerve migratory pathways. Hum Mol Genet. 2002; 11(8): 915–922, doi: 10.1093/hmg/11.8.915, indexed in Pubmed: 11971873.