LETTER TO THE EDITOR

Neurological Sciences

Reply to “The effect of filum terminale sectioning for Chiari 1 malformation treatment: systematic review”

Abbreviations: ACM, Arnold-Chiari malformation; ACMI, Arnold-Chiari malformation type I; ISM, Idiopathic syringomyelia; SOC, suboccipital craniectomy.

Dear Editor-in-Chief,

In the review by Jerônimo Buzetti Milano et al. “The effect of filum terminale sectioning for Chiari 1 malformation treatment: systematic review” published in the journal “Neurological Sciences” [1], the following points must be considered:

Firstly, the authors Milano et al. refer to this condition as "Chiari Malformation 1", or simply "Chiari" or "Chiari syndrome". Suppressing “Arnold”, in English and change the Roman numeral to Arabic, this means disregarding recent international rules and a step toward misunderstandings, which the WHO nomenclature aims to avoid, since there are other entities which can cause confusion, such as: Chiari-Fromel syndrome, Budd-Chiari syndrome, Chiari osteotomy. In the WHO International Statistical Classification of Diseases and Related Health Problems 11th revision, WHO, Geneva, 2018), the English term "Arnold-Chiari malformation type I" is stated for this disease (LA07.4, ICD-11.).
Secondly, the authors Milano J.B. et al. refer as motivation for the article: “The Brazilian Ministry of Health requested an appraisal of the technique to the Brazilian Neurosurgery Society spine department in 2018 in regard to increasing judicial demands. To address the Brazilian Health National Institutes ‘request to clarify treatment effects in FT releasing for patients harboring CM, a review of published data related to efficacy and safety of the procedure was performed to elaborate recommendations related to this surgical technique’ [1].

They later add; “In Brazil, the Federal Constitution has established the duty to ensure health care as a citizen’s right in all levels of complexity. Because of this, the state is often asked to pay for surgical interventions that may not be available in the national setting.”[1].

Reports for the Ministry of Health may have multiple legitimate purposes, but do the motivations that induce the review of Milano J.B. et al. not imply an economic motivation, which can condition a preconceived scientific result, contravening the ethical norms of a medical publication?

Thirdly, of the numerous proposals on the levels of evidence and grades of recommendation currently used, the Oxford Center for Evidence-Based Medicine (CEBM) was chosen in the Milano et al. article for the analysis of the level of evidence in only two publications by the same author, Royo-Salvador, the titles of which are “A new surgical treatment for syringomyelia, scoliosis, Arnold-Chiari malformation, kinking of the brainstem, odontoid recess, idiopathic basilar impression and platybasia” published in 1997 and “Results of the section of the filum terminale in 20 patients with syringomyelia, scoliosis and Chiari malformation” published in 2005.

It is textually suggested In the explanation of the CEBM that: “the levels of evidence be interpreted with a dose of common sense and good judgment”, which is possible to achieve by conducting a systematic and exhaustive search of the scientific literature which allows the obtaining of the relevant articles and a thorough critical analysis of the literature, before assessing the evidence and applying it in practice if it so merits it. [2].

In the case of Milano J.B. et al., the CEBM program is only applied to two publications by Royo-Salvador M.B., without considering that they are part of a 45-year long investigation divided into Primary Research starting with
the doctoral thesis confirmed in its arguments by six professors of the pertinent speciality with a qualification of “cum laude” in 1992 [3] followed by a numerous series of publications up to 2019, the list of which is too long to mention in the current letter.

Milano J.B. et al., by not considering the previous publications from the author Royo-Salvador M.B., which are fundamental for the understanding and justification of the surgical treatment of SFT for ACMI, implies the incorrect use of the CEBM program, which invalidates the conclusions of their review.

Fourthly, by considering the “h index” which is a system proposed by Jorge Hirsch of the University of California for the measurement of the professional quality of physicists and other scientists, depending on the number of citations that have received their scientific articles and applying it to the publications related to the ACMI of the first author of the review, Milano J.B., results in a new demonstrative “thematic h-index” of “0”, since there are no publications and citations in relation to the ACMI of this author.

Milano J.B., with no previous publication in relation to ACMI, evaluates motivated by the Brazilian Ministry of Health two publications by Royo-Salvador M.B., disregarding his doctoral thesis and other seventeen publications related to ACMI.

And finally, how can Milano J.B. et al. justify the application of suboccipital craniotomy (SOC) in ACMI [1], when the mortality rate and serious complications from this surgical intervention exceed the natural course of the disease?

The most extensive bibliographical review regarding SOC applied for ACMI and ISM is by Arnautovic et al. 2015 with a numerous series of 8605 in which a 3% mortality and 4.5% complication rate were found [4]. This represents a total of 258 dead patients with regards to the surgical technique, being mainly SOC.

In the last three decades, eight deaths related to the spontaneous progression of the ACMI/ISM complex have been published [5]. Considering that every year 75 million people are born throughout the world, this represents 2.25 billion individuals born in the span of three
decades and considering that the incidence of the ACHI/ISM complex is of 1:1000 births (numbers from the 19th and 20th century), the number of cases with the ACHI/ISM complex in three decades is of 2.25 million. Taking the 8 recorded deaths into account; this indicates a 0.0004% spontaneous mortality rate, which in Arnautovic’s caseload would amount to 0.034 dead patients if no patient would have undergone surgery. On the other hand, with the mortality index of 3% found by Arnautovic, this would represent 7500 iatrogenic deaths due to SOC, as opposed to the very low amount of spontaneous mortalities due to the ACMI/ISM complex.

While SOC with a 3% mortality rate can be justified in fatal pathologies such as tumors, hematomas, cysts and others with a high mortality rate, it cannot be justified in ACMI and ISM, as it significantly exceeds and adds to the mortality rate of the spontaneous progression of the disease. The presumed improvement with the application of SOC applied to ACMI and ISM does not justify the death of even a few patients. It can therefore be deduced that the SOC with an average iatrogenic mortality of 3%, and an average complication rate of 4.5% is not indicated but rather contraindicated as the standard treatment for ACMI and ISM. Nevertheless, the application of the SOC can be considered for ACMI when inaction represents a greater danger than the SOC.

Conclusion

Royo-Salvador M.B’s doctoral thesis, qualified cum laude by a board of professors and doctors from a prestigious European university, demonstrates and justifies the theory on which the application of the SFT in the ACMI is based. Hence, this procedure ceases to be experimental and the following related publications are a logical continuation of what has been demonstrated, so no further demonstration is required for each subsequent publication. This fundamental basis should not be ignored in related publications as Milano J.B. et al. have done. The reason for the review from Milano J.B. et al. is evident: to give a tool to the Brazilian Ministry of Health to deny the reimbursement of the expenses that Brazilian patients have made, make and will make, to be treated outside their borders. They try this with a letter whose first author
has no publications in relation to the ACMI and by discrediting a small part of the extensive work with numerous publications on ACMI by Royo-Salvador M.B.; incorrectly using a protocol on the levels of evidence and grades of recommendation. While this review from Milano J. B. et al. is justifiable as an internal report for the Ministry of Health, it is not as a medical publication, as it contravenes the ethical principles of medical publications, given the existence of economic reasons involved.

The SOC applied by Milano J.B. et al. for ACMI is contraindicated given its higher mortality and morbidity index than the spontaneous evolution of the disease.

References


