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FOCUS SESSION

Cranioplasty in children: time for a paradigm shift

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Dear Editor:

In 2004, Grant et al. reported a risk of resorption of autologous bone flap of 50% in children and adolescents, namely under 19 years old [1], that is much higher than expected in adults. This evidence remained for a long time isolated. In the last years, the interest in cranial repair constantly increased, as a consequence of the introduction of customized solutions and implantable materials other than titanium and polymethilmethacrylate (PMMA) on one hand, and the awareness that cranioplasty is mandatory not only for anatomical and aesthetical functions but also to restore the physiological perfusion of the brain, on the other hand.

This increasing attention towards cranioplasty is confirmed by the trend in the number of papers published on this topic with almost 200 papers listed on Pubmed.gov in the last year and the organization of a large Consensus Conference on Cranioplasty (C3) in the context of the last International Conference on Recent Advances on Neurotraumatology (ICRAN), held in Naples (Italy) from 20 to 22 June 2018, with an expert panel dedicated to pediatric cranioplasty within C3. More recently, cranioplasty was one of the topics of the Consensus Conference of the European Society for Pediatric Neurosurgery (ESPN), held in Paris (France) from 28 to 1 March 2019.

Despites, doubts and controversial issues are still many in particular in the pediatric population, as confirmed by the difficulty to reach a consensus with statements still pending after months of cooperative work of the C3 group and by the stimulating discussions of ESPN members.

The higher risk of bone resorption in pediatric population is fairly established, but the investigation on the pathogenesis of this complication remains strongly focused on the method of storage. On these grounds, the cryopreservation has been for a long time blamed, though the problem of storage is shared with the adult counterpart with different outcome.

Then, the impact of age-related factors has been hypothesized, thus explaining the decreasing risk of bone resorption with age [2]. Although this figure has been recently confirmed by a large multicenter American study, the definition of these age-related factors remains faint [3].

Indeed, this evidence seems in contrast with the osteogenic capacity of children. Thus, we could hypothesize a severe impairment of this capacity, resulting from the etiology of the cranial defect and the technical aspects of our intervention. In fact, the osteogenic potential is strongly linked to the integrity and activity of dura mater and periosteal layer, due to the membranous ossification of the calvarium. In a cranial defect resulting from decompressive craniectomy for severe head injury, the scarring of the dura mater and periosteal layer leads to lose the role of the two main actors of osteogenesis. However, also, this condition is shared with adults and the higher rate of resorption of autologous bone remains an unsolved problem. Other forces should play a critical role and stratification of age is necessary to further investigate the variable impact of these factors in the different age groups.

When autologous bone fails, other cranioplasty solutions are proposed. In adults, cost-effectiveness analysis is pushing towards the use of synthetic materials, even discarding or at least questioning the use of autologous bone as first-line solution. In pediatric population, cranioplasty based on the use of autologous bone is still preferred. Harvesting autologous bone

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represents a valid solution for small- and medium-size defects but is burdened by complications at the donor site and is not feasible for large defects. Consequently, synthetic materials are used for cranial repair when autologous bone is not available. A large volume of literature deals with new materials, usually claiming good results in the short term. However, results in the long term are completely lacking. Our daily clinical practice teaches us that it is difficult to follow a patient after transition to adult age and our common sense teaches us that we are very reluctant in reporting complications, though we are aware that a cranial implant is a prosthesis and is burdened by a risk of complication lasting through the all life of the patient. A different approach relies on biomimetic materials, namely bioceramics, which try to overcome the limits of synthetic materials, by aiming to osteointegration. However, the rate of osteointegration is difficult to predict so far and effective strategies to accelerate and enhance this process are still lacking. Tissue engineering will eventually overcome these limitations, but the future is yet to come, and a first step ahead should be moved from where we are now.

The outline of this focus session stems from these considerations.

The first paper is a multicenter study that confirms that resorption of autologous bone flap is the main problem we have to face to in pediatric population. This, once again, indirectly highlights the role of age-related factors in the pathogenesis of this complication.

The second paper is a systematic review of the literature that shows that there is no ideal material in children to date. Another important conclusion is the low level of evidence of the studies published so far. We have to move forward from the institutional series with small group of patients, often heterogeneous by age and etiology. Multicenter studies are advocated and stratification of results by age is necessary.

The third paper introduces the discussion on age-related factors affecting the outcome of cranioplasty, focusing on the physiological cranial growth and its implications on cranial repair. In light of this, we should start discussing about cranial repair in different age groups. A proposal for stratification by age would be under 1 year of age, when the skull growth is extremely rapid, in 1–7 years of age, when the skull growth is slower but still significant, and over 7 years of age, when the skull could be assimilated to the "inert" adult skull.

The subsequent two papers focus the problem of cranial repair in the first years of life.

Indeed, the fourth paper further defines the peculiar problems resulting from opening the skull and dura mater at this age, firstly introducing the concept of acquired craniocerebral disproportion (ACCD) after decompressive craniectomy (DC). This condition, that is intimately related to the age of patient, to the physiological growth of the skull (that is a consequence of the age), and to the delay of cranial repair, may be very subtle. A variable degree of ACCD probably occurs in every child after DC and consequently may affect the outcome of cranioplasty.

The fifth paper suggests the research of different surgical solutions, namely decompressive craniotomy without removal of the bone, to avoid the complications of cranial repair in infants. Children are not little adults, infants are not little children, and newborns are not little adults, as Professor Raimondi taught us. Decompressive craniectomy with duraplasty could not be the best solution in all the age groups.

The sixth and last paper aims to discuss the aspects of cranioplasty concerning prevention and treatment of infections that are particularly important since we have to deal with patients staying long term in intensive care unit and in the hospital in general, often contaminated by antibiotic multiresistant germs.

I am very grateful to the editor for the support to this focus session and to the authors who have accepted to contribute, hoping that this focus session would mark a paradigm shift in this topic.

Compliance with ethical standards

Conflict of interest The author has no funding or conflict of interest to disclose.

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