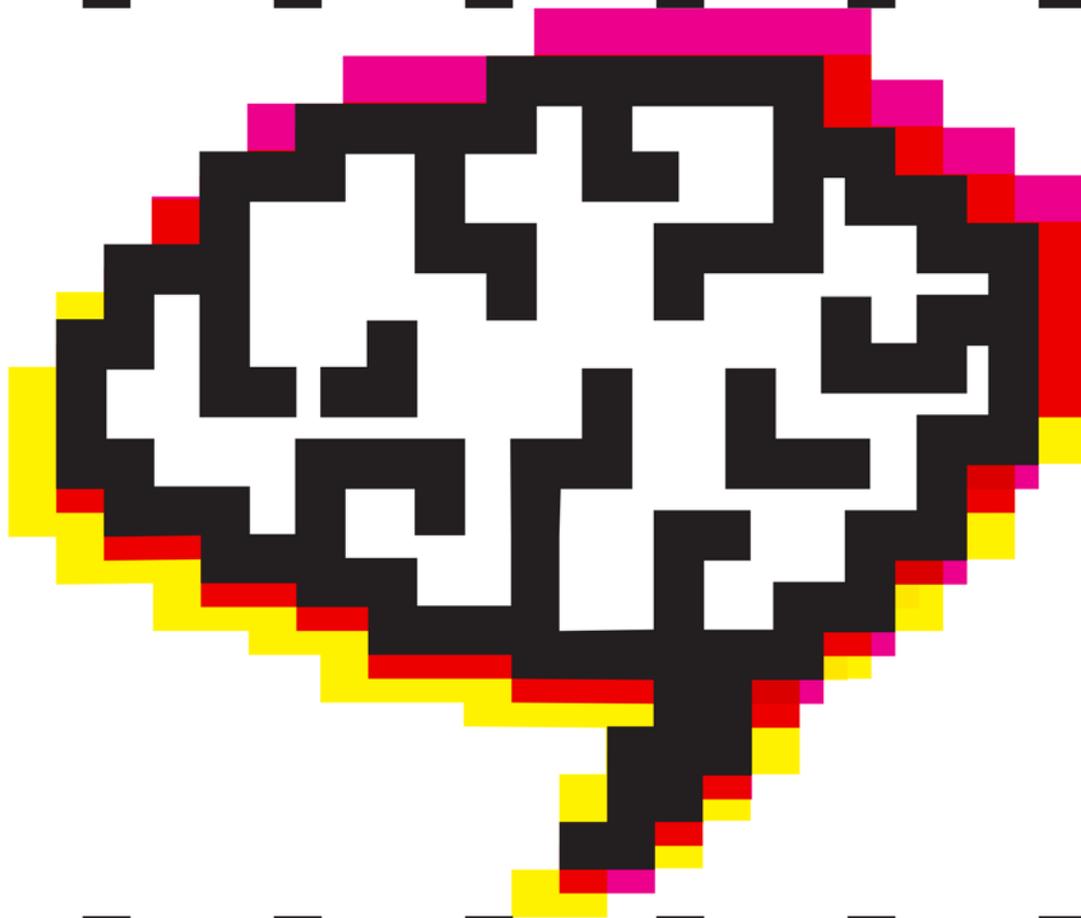


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Case report

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Surgical management of craniosynostosis in a second-level hospital. Five years' experience

Manejo quirúrgico de craneosinostosis en hospital de segundo nivel.
Experiencia de 5 años

Abstract

Introduction: craniosynostosis is caused by the early closure of skull sutures in the child, this can be classified in relation to affected sutures or if it is associated with a genetic syndrome. The definitive diagnosis is made with tomography and the management must be surgical, together with other disciplines. There is evidence of the management of this pathology in hospitals of third level of care but scarce in a second level.

Objective: to characterize patients with craniosynostosis surgically treated in a second level hospital during the 2011-2016 period.

Method: retrospective study where 24 records were reviewed, the variables of sex, age at surgery, type of craniosynostosis, surgical technique used, use of absorbable miniplates and postoperative complications were analyzed, after authorization by the Local Research Committee and the director of the hospital.

Results: we analyzed 24 cases of patients with a diagnosis of craniosynostosis, surgically operated in the HGP / MF 31 of IMSS in Mexicali, B.C., during the period 2011-2016, of which 9 were girls and 15 children. The mean age at the time of surgery was 10.5 months. The type of craniosynostosis by gender, in girls the previous plagiocephaly was the most common in 16.6%. In children, scaphocephaly predominated in 29.2%. The age at the time of surgery varied in relation to the type of craniosynostosis; the most used surgical technique was modified calvarectomy in 37.5%. In all surgeries, absorbable miniplates were used, except for modified calvarectomy. In only one case surgical wound infection occurred.

Conclusion: this study shows that this type of surgeries can be performed in a hospital of second level of care, as long as they have trained staff and the necessary equipment.

Keywords

Glymphatic system, blood-brain barrier, neurovascular unit, immune privilege.

Resumen

Introducción: la craneosinostosis se genera por el cierre temprano de las suturas del cráneo en el niño, esta puede clasificarse en relación a las suturas afectadas o si está asociada a un síndrome genético. El diagnóstico definitivo se hace con tomografía y el manejo debe ser quirúrgico aunado a otras disciplinas. Existe evidencia del manejo de esta patología en hospitales de tercer nivel de atención, pero escasa en un segundo nivel.

Objetivo: Caracterizar a los pacientes con craneosinostosis intervenidos quirúrgicamente en un hospital de segundo nivel durante el periodo 2011-2016.

Métodos: estudio retrospectivo donde se revisaron 24 expedientes, se analizaron las variables de sexo, edad al momento de cirugía, tipo de craneosinostosis, técnica quirúrgica utilizada, uso de mini placas absorbibles y complicaciones postoperatorias, previa autorización por el Comité Local de Investigación y de la directora del hospital.

Resultados: se analizaron 24 casos de pacientes con diagnóstico de craneosinostosis, intervenidos quirúrgicamente en el HGP/MF 31 del IMSS en Mexicali, B.C., durante el periodo 2011-2016, de los cuales 9 fueron niñas y 15 niños. La edad promedio al momento de la intervención quirúrgica fue de 10.5 meses. El tipo de craneosinostosis por género, en las niñas la plagiocefalia anterior fue la más común en un 16.6%, en los niños predominó la escafocefalia en el 29.2%. La edad al momento de la cirugía, varió en relación al tipo de craneosinostosis, la técnica quirúrgica más utilizada fue la calvarectomía modificada en el 37.5%. En todas las cirugías se usaron mini placas absorbibles, excepto en la calvarectomía modificada. Solo en un caso se presentó infección de herida quirúrgica.

Conclusión: con este estudio se demuestra que este tipo de cirugías pueden realizarse en un hospital de segundo nivel de atención, siempre y cuando se cuente con el personal capacitado y el material necesario.

Palabras clave

Craneosinostosis, plagiocefalia, calvarectomía.

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Introduction

Craniosynostosis is a pathology characterized by the premature fusion of the sutures of the skull. It is estimated that the overall incidence is 1 in 2,000-2,500 live births.¹⁻² The etiology is associated with genetic factors and there are several theories, the most accepted of which is the hypothesis by Moss.³ It can be classified as syndromic and non-syndromic, and, in relation to the number of sutures affected, as simple, where only one suture is affected, or complex, where more than two sutures are affected. The simple can be anterior or posterior plagiocephaly or scaphocephaly, and the complex can be trigonocephaly or brachycephaly, among others.³⁻⁴ The diagnosis is clinical; however, to characterize the condition it is necessary to perform a CT of the skull. This pathology causes aesthetic alterations and central nervous system complications, which is why it requires multidisciplinary treatment, and surgical intervention is a priority depending on the type of craniosynostosis.⁵ The age range at which surgical intervention is recommended is from three to six months old, although surgery is still permissible prior to the first birthday.⁶ Among the main postoperative complications are bleeding, infection of the surgical site, early reossification, injury to neurovascular structures, possible need for another surgical intervention, and death.⁷ Currently, the mortality rate after surgery for craniosynostosis is low and the prognosis is usually good—aesthetically and functionally—if the operation is performed at the right time and with the appropriate technique.⁸

Imaging studies that assess the skull three-dimensionally are recommended for follow-up. It has been noted that the linear measurements are not adequate because there is a growth restriction perpendicular to the affected suture in these pathologies. The skull compensates by growing parallel to that suture, so measurements such as the cephalic perimeter are not affected. These patients are usually within the normal percentiles (5 and 95) for their age, pre- and postoperatively. The only exception to this is

oxycephaly, where there is a restriction to the growth of the skull.⁹⁻¹⁰

To date, there is a scarcity of published evidence regarding craniosynostosis in a hospital of second level of care, so we intend to show the experience in the surgical management of this pathology with this case series.

Methods

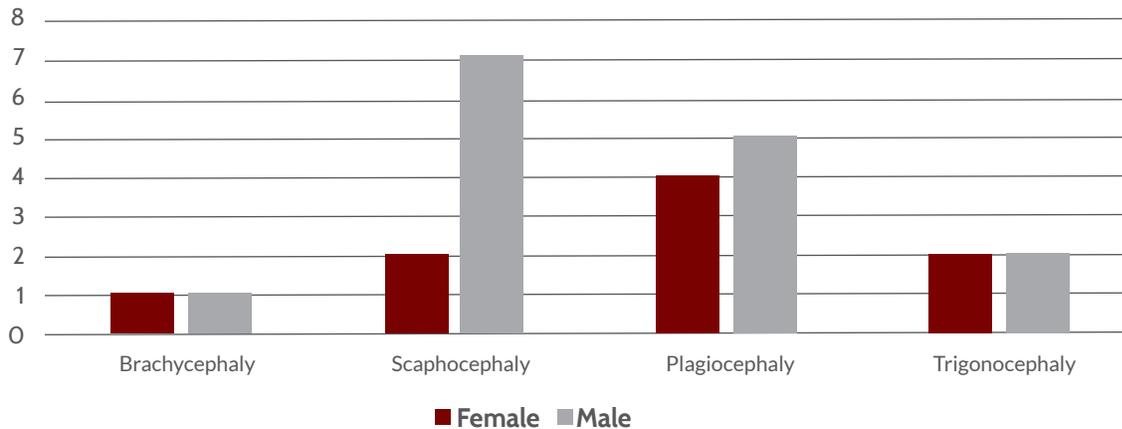
The study was developed adhering to the 2013 Declaration of Helsinki and was approved by the Local Research Committee (R-2017-201-13) and the hospital director. Because it was a retrospective study, it did not require an informed consent letter though the patient's confidentiality was still safeguarded. The information was collected from the physical files of the pediatric patients diagnosed with craniosynostosis who underwent surgery at the Gyneco-Pediatrics and Family Medicine Hospital No. 31 in the 2011-2016 study period. The information was collected in an Excel database sheet and included the following items: age, gender, age of surgical intervention, diagnosis, surgical technique used, implant utilization, and postoperative complications. The data was then analyzed with the statistical program SPSS Version 21.

Results

We reviewed 25 cases of patients diagnosed with craniosynostosis who had surgery at the Gyneco-Pediatrics and Family Medicine Hospital No. 31 of the Mexicali, BC, IMSS in the period from 2011-2016. One file was eliminated due to incomplete information. Of these 24 cases, nine were girls and 15 were boys.

The average age at the time of surgery was 10.5

Figure 1. Type of craniosynostosis by gender.



months (minimum 4 months, maximum 84 months). Sorting the type of craniosynostosis by gender, 16.6% of the girls presented anterior plagiocephaly (three cases of left anterior plagiocephaly and one case of right anterior plagiocephaly), 8.3% scaphocephaly, 8.3% trigonocephaly, and 4.1% brachycephaly. In boys, scaphocephaly predominated at 29.2%, followed by anterior plagiocephaly with 12.5% (the three cases were left anterior plagiocephaly), posterior plagiocephaly and trigonocephaly were 9.4%, and one case of brachycephaly with closure of both lambdoid sutures. (Figure 1)

The age at the time of surgery varied depending on the type of craniosynostosis: the brachycephaly at four and 14 months, scaphocephaly between five and 30 months, plagiocephaly from four to 15 months (and one at 84 months), and trigonocephaly from 11 to 60 months. (Figure 2) Only one case of plagiocephaly which was operated at six months required surgical reintervention at 30 months due to the development of scaphocephaly.

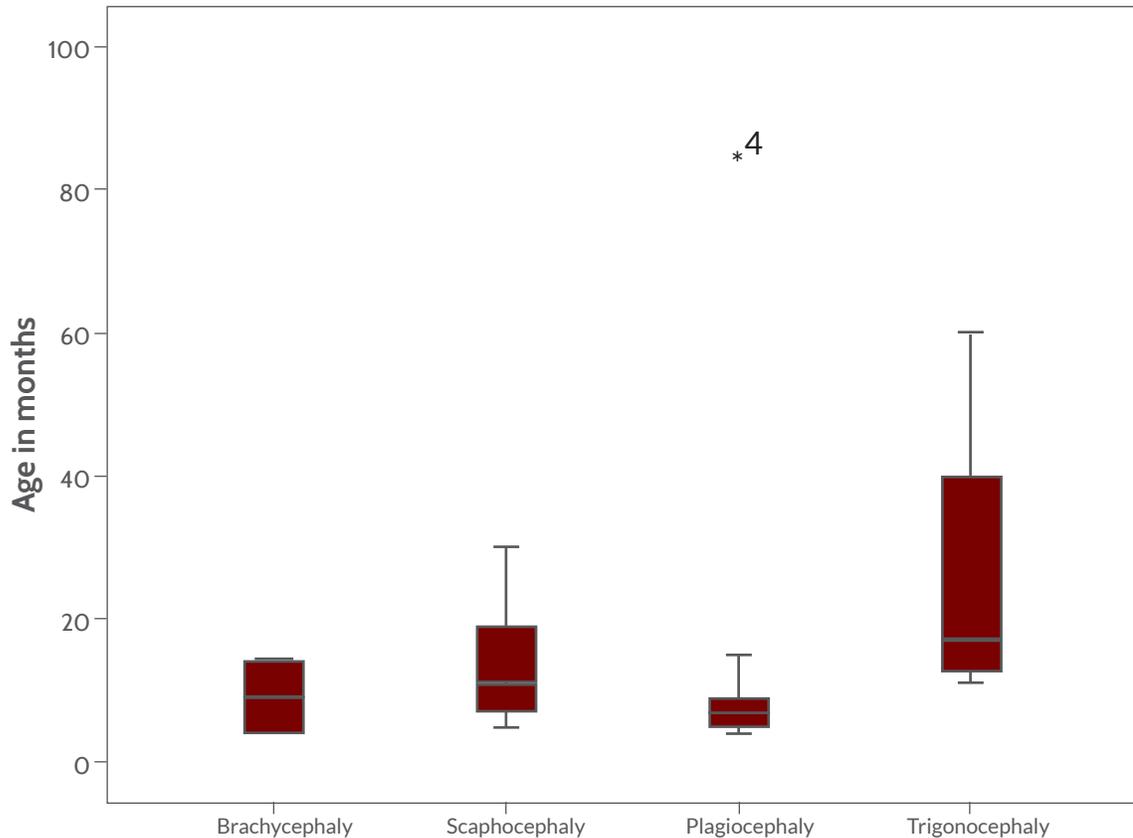
The surgical technique performed was chosen according to the type of craniosynostosis. The most frequent was a modified calvarectomy in 37.5%. (Table 1)

The modified calvarectomy performed for scaphocephaly does not use absorbable mini-plates, which were used in the rest of the surgical techniques. (Figure 3)

Table 1.

Type of craniosynostosis	Surgical technique	Frecuency
Brachycephaly	Bilateral orbital cranial advance	1 (4.16%)
Right anterior plagiocephaly	Bifrontal craniotomy + right fronto-orbital advance + cranial remodeling	3 (12.5%)
Left anterior plagiocephaly	Bifrontal craniotomy + left fronto-orbital advance + cranial remodeling	4 (16.6%)
Trigenocephaly	Bifrontal craniotomy + bifrontal orbital advance + cranial remodeling	4 (16.6%)
Scaphocephaly	Modified calvarectomy	9 (37.5%)
Posterior brachycephaly with closure of both lambdoid sutures	Parieto-occipital cranial remodeling	1 (4.16%)
Posterior plagiocephaly	Sun rise	2 (8.33%)
Total		24 (100%)

Figure 2. Type of craniosynostosis by age.



Only one case presented a postoperative complication of infection of the surgical wound, which was treated satisfactorily.

Discussion

This series of cases is one of the first of its kind published in the national literature in relation to a second-level care hospital. The study population consisted of 24 postoperative craniosynostosis patients, where males predominated at 62.5% vs 37.5% of females, similar to reports from 2014 at the Hospital de Juárez in Mexico. The craniosynostosis presented was non-syndromatic.¹¹

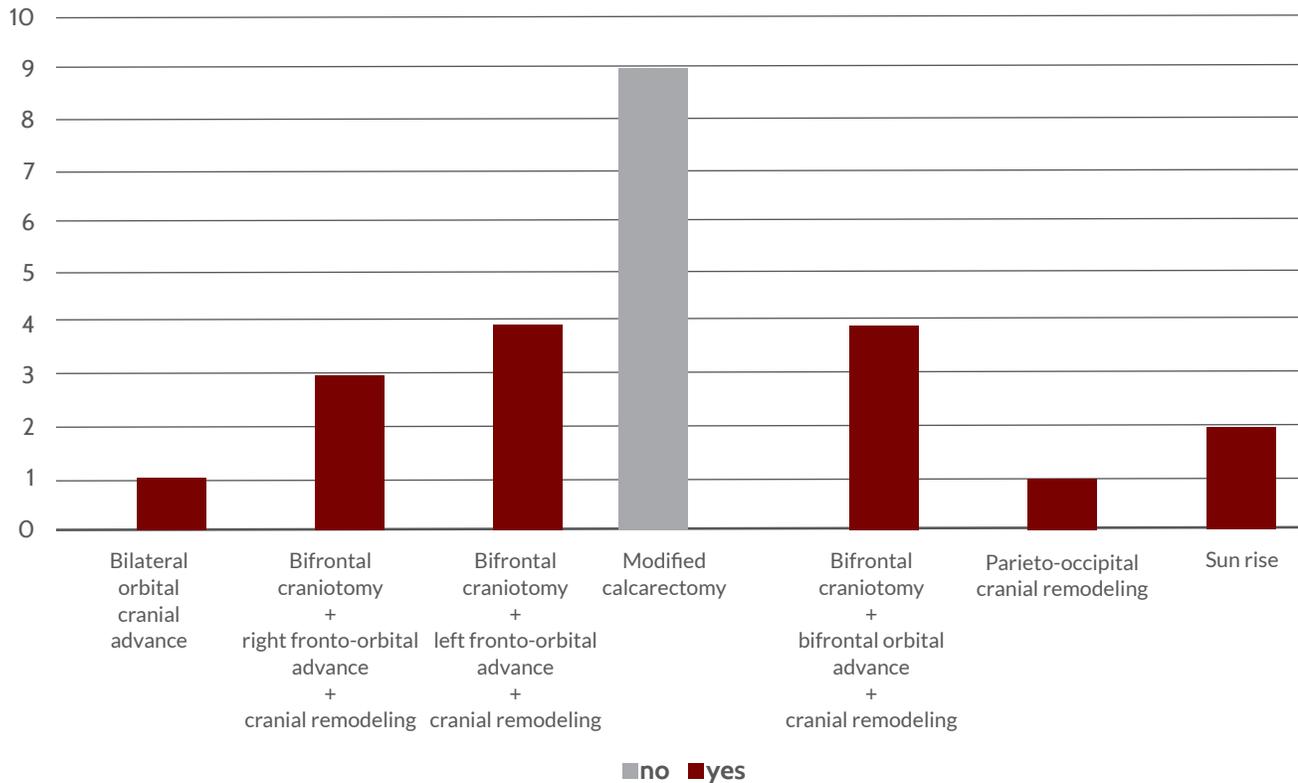
In this study, simple craniosynostosis predominated. Scaphocephaly occurred in 37.5% of cases, which coincides with the literature. Anterior

plagiocephaly presented in 29.1%, trigonocephaly in 17.7%, and brachycephaly, as well as posterior plagiocephaly, in 8.2%.^{11,12}

The diagnostic approach and decision-making (Figure 4) is based on the Clinical Practice Guidelines for the diagnosis, treatment, and rehabilitation of nonsyndromic craniosynostosis in the three levels of care.¹³ In this series, diagnosis was clinical and by CT of the skull, some with three-dimensional reconstruction for better assessment of the affected sutures and the type of surgical intervention to be performed.¹⁴

With respect to scaphocephaly, the majority of the patients had the surgical intervention at an optimal time; however, there were some who had the operation at ages older than 12 months, contrary to what is recommended in different publications.¹⁵⁻¹⁶ Regarding plagiocephaly, the recommended

Figure 3. Use of absorbable mini-plates.



surgery time is between eight and 12 months of age,¹⁶ and in this series, there were surgical interventions on patients older than 12 months—the delay of surgical age possibly due to the lack of timely diagnosis in the initial consultation.

Regarding the surgical techniques used, calvarectomy was performed on scaphocephaly cases since it has shown better results in relation to morphological correction. Panchal *et al.* observed that calvarectomy achieved a normal cephalic index in the majority of children who had surgery in the first 13 months of age.¹⁷ The use of absorbable mini-plates was deferred in these patients, considering there is evidence of satisfactory procedures without their use, which coincides with this series.^{18,19}

For trigonocephaly and anterior plagiocephaly, the techniques used are consistent with those reported in other studies, performing fronto-orbital advancement on the majority of these

pathologies.²⁰ The surgery performed in brachycephaly was a bilateral orbital cranial advance. This type of patients usually presents an associated syndrome; however, it should be noted that this case was isolated.²¹

The most frequent complications reported with a greater number of cases were surgical wound infections, subgaleal abscesses, dural tears, residual deformity, and transurgical hemorrhage.²²⁻²⁴ In this study, a case of surgical site infection was reported and there were no deaths. The mortality data is similar to that reported by a third-level hospital, differing only with regard to postoperative complications where they reported postoperative bleeding.²⁵ Although it is true that transurgical bleeding is one of the most frequent complications in this type of surgery, this risk and its impact on the patient's health is controlled once the appropriate pre-, trans-, and post-operative protocols are followed. These protocols include keeping blood derivatives available in

the surgical room to transfuse at the indicated time, having a neuro-anesthesiologist trained in the precise replacement of this type of blood products, keeping strict hemodynamic control during and after surgery, and having a specialist in pediatric intensive care capable of recognizing and correcting residual hemodynamic alterations after surgery. Proceeding this way has led us to have no complications related to bleeding.

Within the national literature, postoperative complications (of which postoperative bleeding is the most common) are reported in approximately 19.5% of cases—we place below the national statistics with 4%.²⁵⁻²⁶

The case that presented infection in the surgical site started with fever on the fourth postoperative day, hyperemia of the surgical wound, and subgaleal blood collection. A percutaneous puncture was performed to cultivate the blood content, which was positive for *S. aureus*. On the seventh postoperative day he underwent a thorough surgical lavage and the use of an antiseptic solution, evolving satisfactorily in the subsequent days with no recurrence or bone resorption. A patient that had surgery at six months old for anterior

plagiocephaly was reoperated at two and a half years old due to developing scaphocephaly.

All the patients reported in this series were referred to the pediatric neurology service for cognitive assessment. Until now, no condition has been reported to affect any of these patients. This coincides with reports in the literature stating that children with non-syndromic craniosynostosis have an intellectual coefficient within normal ranges.^{27,28}

It should be noted that, despite being a second-level hospital, the results concerning complications and mortality in this series of cases are favorable.

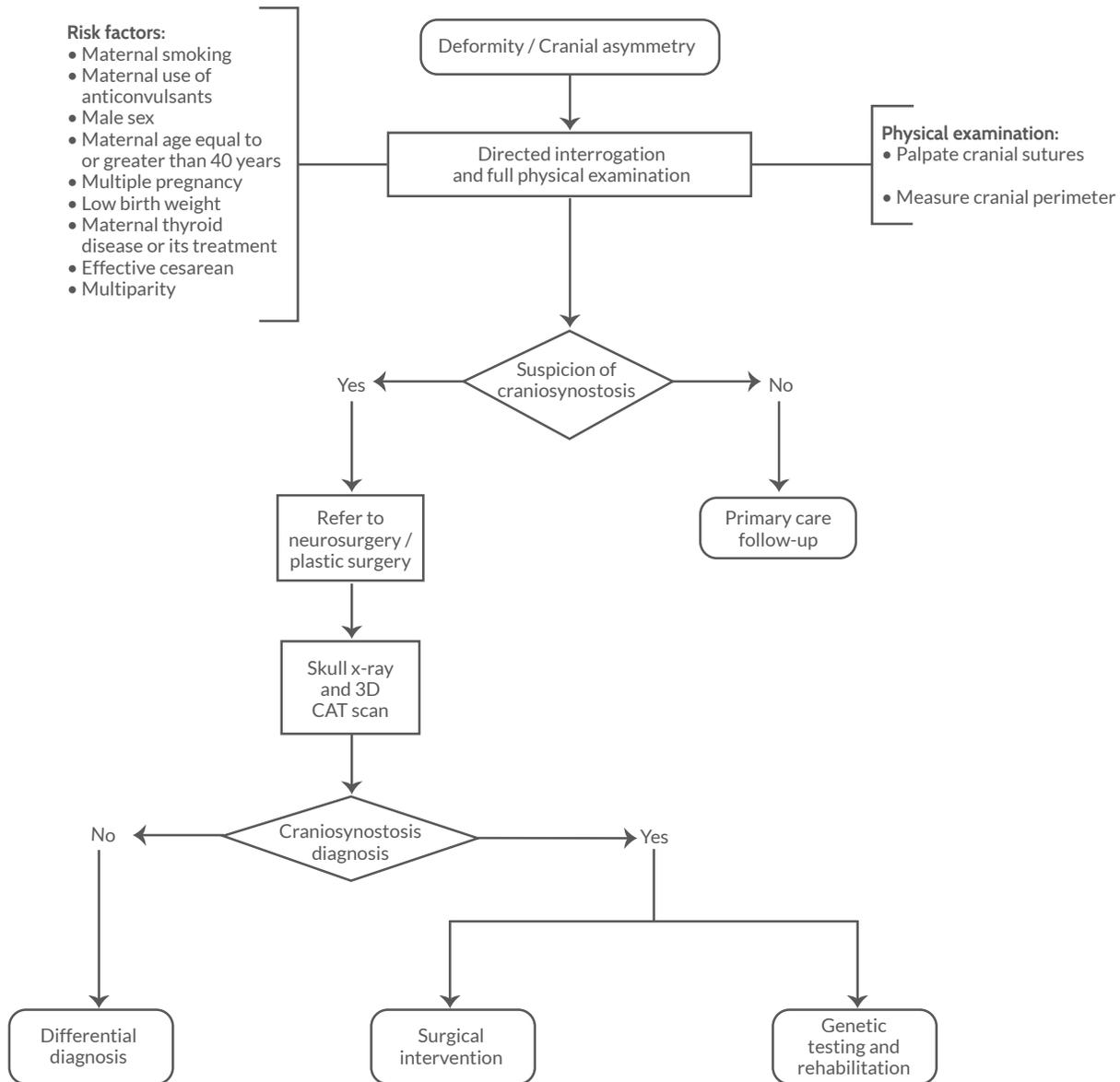
Faced with the inability of our national health system to deal with complex pathologies due to the large number of cases, the need has arisen for hospitals, in theory, of a lesser level of specialization to undertake the task of developing the means (human and infrastructural) to be able to treat pathologies of a certain level of complexity at appropriate times before the critical periods of treatment expire. Previously, patients passed the critical treatment periods waiting for space at third-level centers to be operated.

Conclusion

It is necessary to make an opportune diagnosis at a primary care level to avoid a delay in the diagnosis of these patients and, subsequently, the age of the surgery.

Craniosynostosis surgeries can be performed in a second-level hospital as long as it has trained staff and the necessary materials. This would improve the early care of these patients since they would not have to wait for attention in third-level hospitals.

Figura 4. Algorithm for the diagnosis and treatment of craniosynostosis.



Taken from the Guide of Clinical Practice for the Diagnosis, Treatment and Rehabilitation of Craniosynostosis in Three Levels of Care. CENETEC. Mexico.

Conflicto de intereses

The authors declare no conflicts of interest.

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