

Quality of life and nasal splints after primary cleft lip and nose repair: Prospective assessment of information and tolerance



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ABSTRACT

Splints are commonly used after primary cleft surgery in order to secure the position of the nasal cartilages. Although the importance of splints is more and more stressed in the literature, many questions remain unanswered relative to the psychological impact of this device on children and families.

Two questionnaires, *Information* and *Tolerance*, were used in order to measure the quality of life (QoL) associated with the use of nasal splints after primary cleft surgery. *Information* assessed the understanding of the parents the day before the procedure. *Tolerance* assessed their experience 3 months after splint placement. We prospectively included 41 consecutive patients from a Paris cleft center, 21 consecutive patients from a Russian center (Moscow) and 10 consecutive patients from another French center (Nantes). In Paris and Nantes, an initial fixed splint was placed during the procedure until day 10, and then replaced by a removable splint for a period of 4 months. In the Moscow group, removable splints were used primarily for a total period of 4 months. Three types of removable splints were considered: commercial anatomical self-retentive splints (Nose-Fit™, Moscow, Russian Federation), in-house anatomical self-retentive splints and commercial Talmant-type splints requiring taping (Sebbin, Boissy-l'Aillierie, France). The data was analyzed as Likert scales and internal consistency was assessed using the Cronbach coefficient. Age at surgery, uni- or bilateral cleft, type of splint, number of splint changes and complications were tested against the scores of the questionnaires using multivariate models.

We did not find correlations between the factors assessed by the multivariate analysis and the splint type. *Information* and *Tolerance* scores were high and showed satisfactory QoL associated with the use of splints. The internal consistency of the combination of the two forms was good. While the effects of splints on nasal morphology still need to be confirmed based on a controlled prospective study, we show here that this device is well tolerated by families and is not associated with specific complications.

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1. Introduction

The cleft nose is considered by many as the most tricky issue in cleft repair. In order to stabilise the morphological results of

the rhinoplasty associated with primary cleft surgery, various types of nasal splints have been developed (Ivanov and Khonsari, 2011). These devices consist of flexible rubber tubes generally connected by a columellar bridge (Fig. 1). Splints are inserted into the nasal cavities during the primary procedure and are believed to provide two benefits: (1) an active morphological role by molding the alar cartilages and (2) a functional role by ensuring upper airway patency and favouring nasal breathing.

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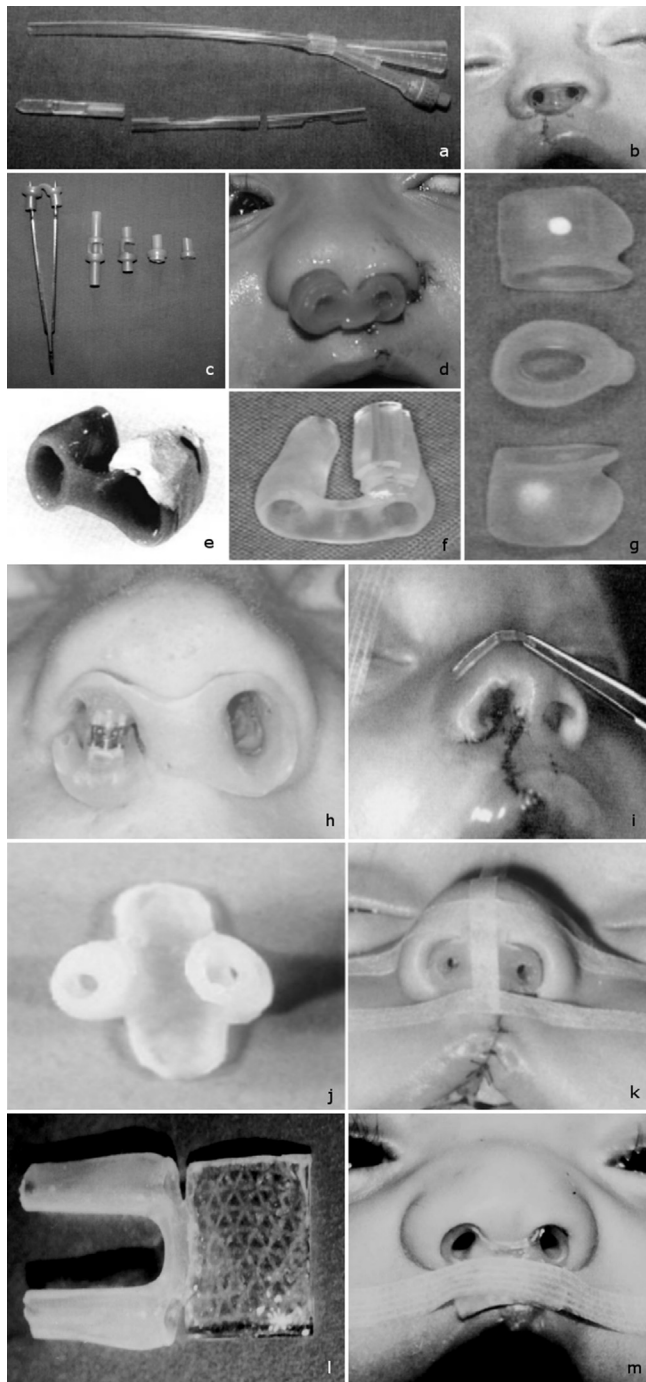


Fig. 1. Overview of different splint types available in the literature. (a, b) in-house splint made with a silicon urine catheter (from Yildirim and Ak_oz, 2001); (c, d) in-house splint made with the rubber of an infusion set (from Tan et al., 2006); (e) modification of a Koken (Koken Co., Tokyo, Japan) splint using quick silicon set (from Nakajima et al., 1990); (f) modification of a Koken splint using silicone sheets cut from silicone tubings of 1-mm thickness (from Chang et al., 2010); (g) nostril splints without extension into the nasal cavities (from Yuzuriha et al., 2001); (h) dynamic nostril splint with expansion screws (from Cenzi and Guarda, 1996); (i) adsorbable internal splint (from Wong et al., 2002); (j, k) modification of a Koken splint with two butterfly wing for taping (from Cobley et al., 2000); (l, m) modification of a Koken splint with a philtral plate for taping and scar sheeting (from O'Zyazgan and Eskitaş çiog_lu, 2000).

Splints are an important component of the functional approach to cleft surgery, where the central procedure is primary cheilorrhinoseptoplasty at 4–6 months of age followed by a period of

3–4 months of nasal conformation (Markus and Delaire, 1993; Talmant et al., 2016).

Actual effective molding of cartilages using external mechanical forces has been mostly reported during the first three months of life, in situations such as pre-operative naso-alveolar molding for cleft patients (Greives et al., 2014) and in external ear moulding (Tan et al., 1997; Doft et al., 2015). Circulating maternal estrogen levels are higher in children before 6 weeks of age; maternal estrogens favor the responsiveness of the cartilage to deformation in the newborn due to an increased production of hyaluronic acid, responsible for the malleable nature of the neonatal cartilages (Byrd et al., 2010). For instance, an 'inner splint' formed by a congenital intranasal fibrochondroma (Fig. 2) supports the fact that the neonatal nostril shape may be plastic: during the first 6 months of life, until surgical removal, the fibrochondroma induced a significant deformation of the nostril outline.

Nasal splints are used later in the life of cleft patients, generally between 4–6 months of age and 8–10 months of age, that is long after the period during which the neonate is exposed to maternal estrogens. There is no current prospective assessment of their effect on long-term nasal shape, even though retrospective investigations seem to indicate that they have a positive esthetic effect on cleft nose morphology (Yeow et al., 1999; Greives et al., 2014). As their direct effect on cartilage shape is unlikely – based on the current knowledge of the plasticity of the neonatal cartilage – splints, if efficient, could play a role at two levels: (1) by positioning and maintaining the alar cartilages in the position decided by the surgeon during the primary rhinoplasty, splints may direct the post-surgical healing and help to minimize the resulting deformation and (2) by providing better conditions for nasal ventilation versus oral ventilation, splints could stimulate the naso-maxillary growth.

Many splint types have been described in the literature: fixed splints for the immediate post-operative period and various commercial and in-house removable devices for long term conformation (Fig. 1). Four types of splints were used in this study. The two French teams from Paris and Nantes used an initial fixed Talmant-type splint (FT) for the first 10 days after surgery (Fig. 3). After this initial period of 10 days, three types of splints were used by the two French teams: (1) commercial anatomical splints (CAN), which were removable self-retentive silicon medical devices (Nose-Fit™, Fig. 4a), (2) in-house anatomical splints (LAN) which were locally manufactured removable self-retentive silicon medical devices (Fig. 4b) and (3) commercial removable Talmant-type splints (RT) from Sebbin (Boissy-l'Aillierie, France), which were commercial anatomical removable silicon medical devices requiring taping over the upper lip for retention and scar molding (Fig. 4c)

In order to assess quality of life related to the use of nasal splints, we designed two forms (*Information* and *Tolerance*) and prospectively included patients from three cleft centers – Paris, Nantes and Moscow – who applied the same surgical protocol: primary surgery at 4–6 months of age (cheilorrhinoseptoplasty) followed by a period of 4 months of nasal splinting. The Paris team used FT after the procedure for 10 days, followed by 4 months of removable splinting with either CAN, LAN or RT. The Nantes team always used FT initially followed by RT for a period of 4 months. The Moscow team used CAN for the whole splinting period.

Information was assessed before the primary procedure and *Tolerance* was assessed at the end of the splinting period. We evaluated *Quality of Life* based on these two questionnaires and screened for specific issues related splint types and for complications using a multivariate model.



Fig. 2. Intranasal fibrochondroma acting as a natural splint and inducing a deformation of the nostril, at 1 month of age (left) and 6 months of age immediately after surgery (right).

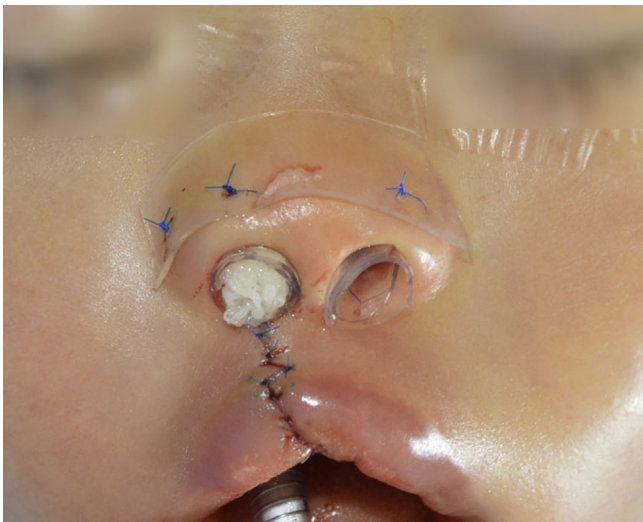


Fig. 3. Fixed splint (Talmant-type fixed splint, FT) used for the first 10 post-operative days by the two French teams (Paris and Nantes). A silicon sheath was stitched to the nasal dorsum and two silicon rolls were inserted into the nasal cavities and stitched through the septum and to the dorsum. The splints were removed under local anesthesia or during a short general anesthesia during stitch removal, at day 10. A removable splint was then used for a total period of 3–4 months.

2. Materials and methods

All patients operated on for the primary closure of total unilateral or bilateral cleft lip or cleft lip and palate over a period of 12 months in three centers (Paris, Nantes and Moscow) were prospectively included into the study. Two forms were completed by the parents: (Tables 1, 2)

- (1) the *Information* form the day before primary surgery, with 8 items rating 1–5;
- (2) the *Tolerance* form, three months after the surgical procedure, with 13 items rating 1–5.

Exclusion criteria were incomplete clefts, syndromic clefts and cognitive and/or motor impairment. The forms were analyzed as Likert scales (Tourangeau et al., 2013). The internal consistency of the scale was measured using the Cronbach α coefficient (Bland and Altman, 1997). The consistency of the form depended on the value of α . For $\alpha < 0.5$, the form was considered unacceptable, while forms with $0.5 \leq \alpha < 0.6$ were considered poor and forms with $0.6 \leq \alpha < 0.7$ were considered questionable. For $0.7 \leq \alpha < 0.8$, the form was considered acceptable; for $0.8 \leq \alpha < 0.9$, the form was considered good and for $\alpha \geq 0.9$, the form was considered excellent. Cronbach α values were computed for each form – *Information* and *Tolerance* – and also for the two forms combined together (*Quality of Life* form). Cronbach α values were also computed for each form by removing items sequentially to test the dependence of α on specific questions for each form. A score for each form /40 for the *Information* form and /65 for the *Tolerance* form – and a total score /105 were computed by adding the results obtained for each item.

Linear and logistic multivariate models were built with the total score for each form as the explanatory variable and the six following parameters as response variables: (1) age at surgery, (2) gender, (3) unilateral or bilateral cleft, (4) country (France or Russia), (5) number of splints used and (6) splint type (CAN, LAN, FT, RT). Complications – wound dehiscence in the contact zones with the splint, allergy or splint ingestion by the child – were recorded at the end of the conformation period. The logistic regression model used the average of the total scores as the 'satisfaction threshold'. The parameters were compared to 0 using



Fig. 4. Three different types of removable splints: (a) commercial anatomical splint (CAN), Nose-Fit™ (Moscow, Russian Federation), (b) in-house anatomical splint (LAN) and (c) commercial removable Talmant-type splint (RT), Sebbin (Boissy-l'Aillerie, France). CAN and LAN were self-retentive. RT required taping with the midline silicone tongue exerting scar sheeting on the columella.

Table 1
Information form used to assess the quality of medical information provided by the medical and nursing team before primary surgery. Eight items rated from 1 to 5: Strongly disagree (1/5), Disagree (2/5), Neutral (3/5), Agree (4/5) and Strongly agree (5/5) – Total score:/65.

Information form
1. I am well informed about the importance of nasal conformation after primary lip and nose surgery.
2. I am convinced of the importance of nasal conformation after primary lip and nose surgery.
3. I am informed about potential difficulties that may arise when using the retainers.
4. The surgeon of my child showed me how to handle the retainer.
5. The nurse of my child showed me how to handle the retainer.
6. I am worried that I will eventually have to manipulate the conformer myself.
7. I am afraid that the retainer might hurt my child.
8. I know whom to call in case of difficulties when using the retainer.

Table 2
Tolerance form assessing general repercussions of the use of retainers during the first 3 months following primary surgery. Thirteen items rated from 1 to 5: Strongly disagree (1/5), Disagree (2/5), Neutral (3/5), Agree (4/5) and Strongly agree (5/5) – Total score:/105.

Tolerance form
1. I was afraid that my child would swallow his retainer during his sleep.
2. I am satisfied with using the retainer.
3. I am satisfied about the medical follow-up regarding the retainer.
4. I do not regret using the retainer for my child.
5. Things went as expected regarding the use of the retainer.
6. I have little confidence in the results of the use of the retainer.
7. I felt that my child was not safe when using the retainer.
8. I had additional problems because of the retainer when my child got sick.
9. I had problems feeding my child because of the retainer.
10. I had problems putting my child to sleep because of the retainer.
11. I felt that my child had to carry the retainer for a very long period of time.
12. Using the retainer had a negative impact on our everyday life (for instance: going to nursery school, resting at home, shopping).
13. I confess not using the retainer everyday as prescribed by the surgeon of my child.

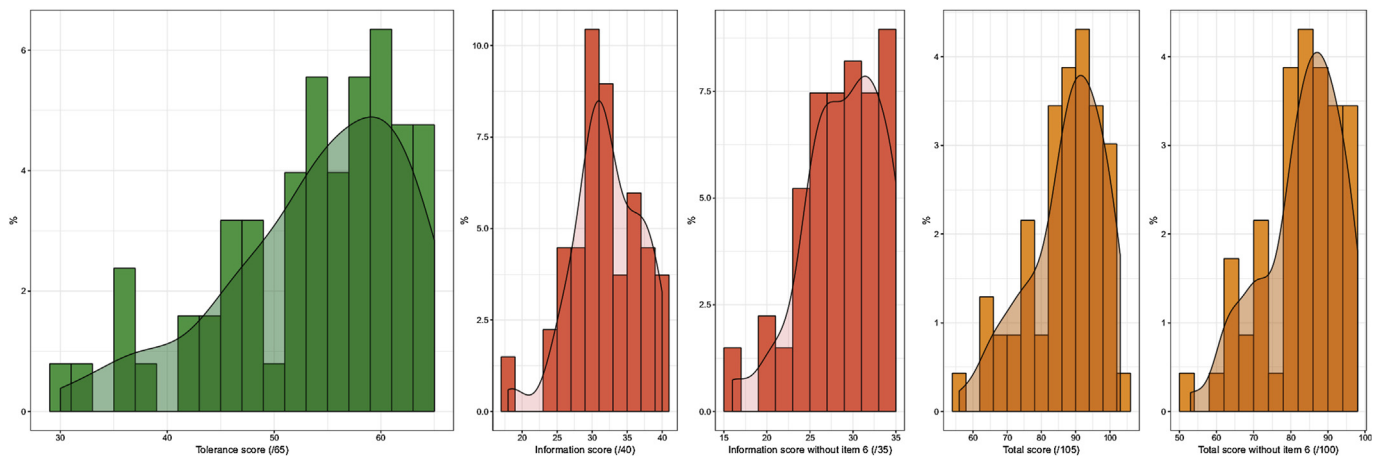


Fig. 5. Distributions for Tolerance score, Information score, Information score without item $N^{\circ}6$, Total score and Total score without item $N^{\circ}6$.

Student tests with a threshold value $p < 0.05$. Statistical analyses were carried out on R (R Core Team, R Foundation for Statistical Computing, Vienna, Austria, 2017, <https://www.R-project.org>) with the following packages: (1) *nlme* for mixed models (Pinheiro et al., 2018), (2) *ggplot2* for plots (Wickham, 2009), (3) *likert* for Likert scale analyses (Bryer and Speerschneider, 2016) and (4) *psy* for Cronbach α computations (Falissard, 2012).

3. Results

A total of 72 patients were included into the study: 41 in the Paris center, 21 in the Moscow center and 10 in the Nantes center. The sex ratio was 43 boys for 29 girls; cleft types were 39 unilateral

and 16 bilateral (17 cleft types were not recorded). The age at surgery was 6.722 months (± 0.409); this information was not available for 15 patients. An average of 2.68 (± 0.464) splints were used per patient; this information was not available for 22 patients. The type of splints used were: CAN for 57 % of cases, FT for 36 % of cases, RT for 45 % of cases and LAN for 31 % of cases. The FT group (24 patients) consisted in 15 boys and 9 girls, from the Paris and the Nantes centers, with 11 unilateral clefts and 2 bilateral cleft (data on laterality was missing for 11 patient). The average number of splints used per patient was 3.00 (data were missing for 16 patients). After FT, 33 % of patients had CAN, 61 % of patients had RT and 42 % had LAN (data on splint type after FT was missing for 6 patients). No specific local or general complication related to the

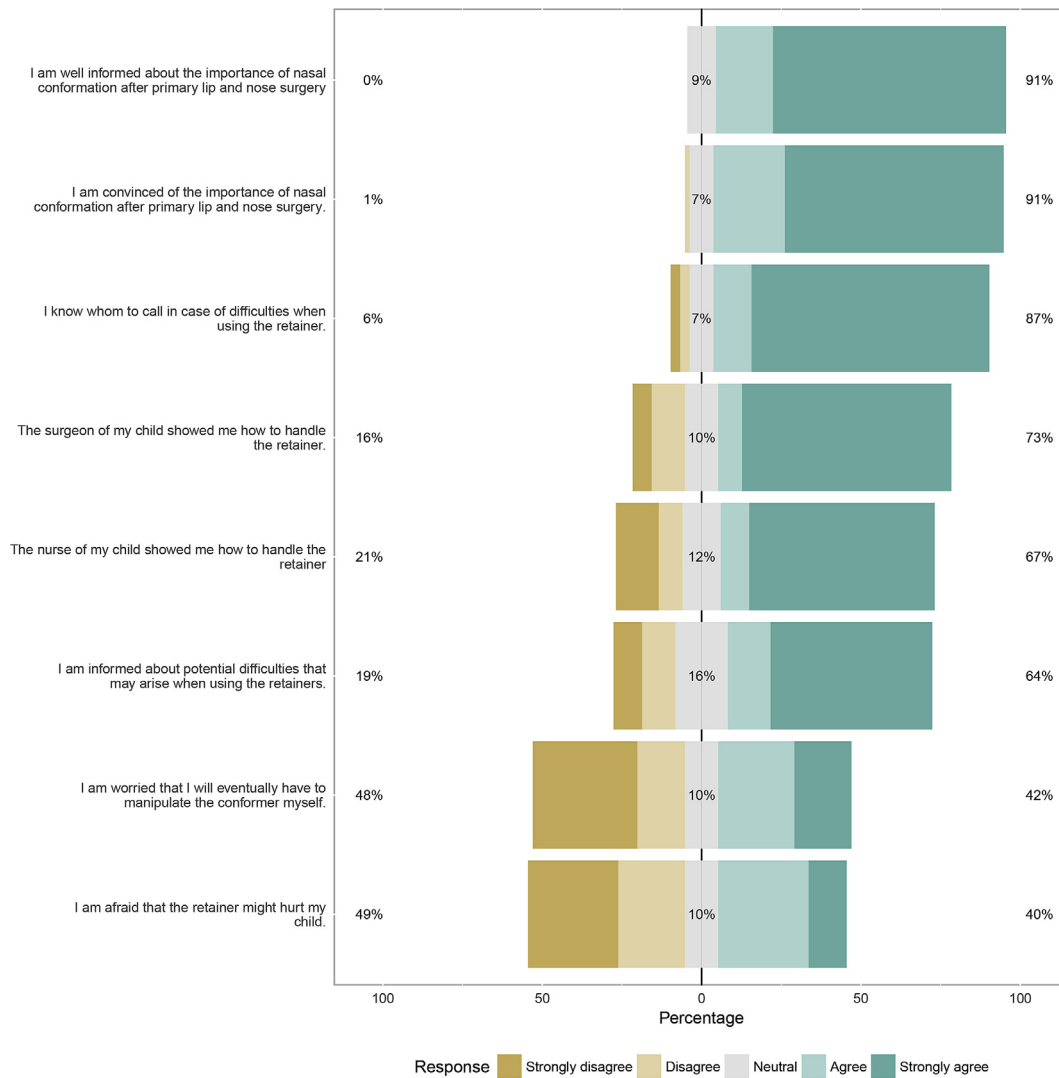


Fig. 6. Likert scale for the Information score.

use of splints was recorded. No major specific local or general complication related to the use of splints was recorded, except from 16 splint losses, 3 minor irritations of the nostril rim and one most probably intercurrent rhinopharyngeal infection.

The Information score (/40) was 32.15 (±4.834) and the Tolerance score (/65) was 53.97 (±8.525). The total Quality of Life (Information + Tolerance) score (/105) was 87.16 (±11.157).

The Cronbach α was 0.547 (poor) for the Information form and was 0.773 (acceptable) for the Tolerance form. By removing item N \geq 6 from Information form, α increased to 0.628 (questionable). The Cronbach α for the Quality of Life form (combination of the Information and Tolerance forms) was 0.786 (acceptable) and raised to 0.804 (good) when the item N \geq 6 was excluded from the Information form. (See Figs. 5–7).

4. Discussion

Residual nostril deformation after lip repair is a major issue in the management of cleft patients. Nasal splinting after surgery has been proposed by numerous teams in order to mold the alar cartilages and maintain the nostril shape obtained during the repair procedure but prospective controlled studies assessing splint

efficiency are still missing. Here we show that this device is not associated with issues in quality of life and does not seem to cause specific complications. Of note, item N \geq 6 from Information form was not informative as the answers to this item were contradictory. Item N \geq 7 from Information form had a nearly similar answer pattern with less contradictory results and had no negative influence on Cronbach α values.

Splints can be subdivided into two groups: (1) **group A**: non-removable splints, such as FT (Fig. 3) and (2) **group B**: removable splints, such as RT, CAN and LAN (Fig. 4). Group B splints can be subdivided into three categories: (1) **type B1** = in house splints developed locally by cleft teams, often using various silicon medical devices such as catheters or infusion sets (Fig. 1a–d); (2) **type B2** = commercial splints, the most commonly used worldwide being the Koken splint (Koken Co., Tokyo, Japan) and (3) **type B3** = modified commercial splints, generally in order to obtain nostril shape hypercorrection (Fig. 1e and f) and support for taping and/or scar sheeting (Fig. 1j–m). A final characteristic of splints is their need for fixation; group B splints can thus be separated into: (1) **B(r)** = retentive splints, such as CAN and LAN and (2) **B(nr)** = non-retentive splints, such as RT (Fig. 4c).

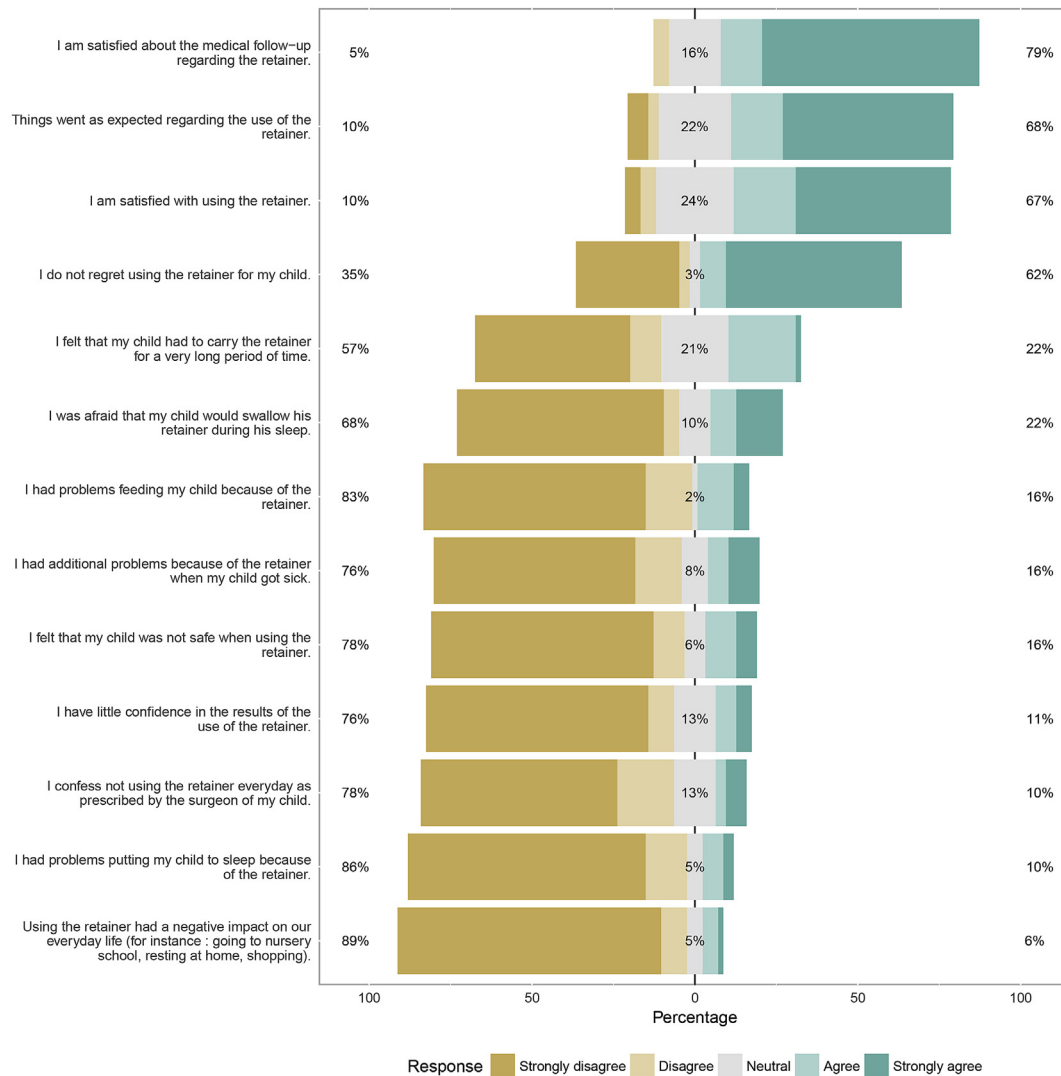


Fig. 7. Likert scale for the Tolerance score.

5. Conclusion

Our study does not show specific issues related to the primary use of **A** splints and/or secondary **B1(nr)** splints, even though these two types of device seem more invasive and difficult to handle than **B1(r)** or **B2(r)**. We thus do not have arguments related to the quality of life and complications that would tend to recommend the primary use of removable stents and the primary and/or secondary use of self-retentive devices. Nevertheless, until prospective studies are available on the morphological benefits of stenting, we recommend using simple commercial splints such as **B2(r)**, in order to comply with local medical device regulations. Encountering complications secondary to the use of **B1** or **B3** splints would not be defensible on legal grounds based on the current scientific literature, as the benefits of splints are not formally proven and as various certified commercial devices are available.

Circulating maternal estrogen levels may be crucial in maintaining the plastic properties of the neonatal cartilage during the first 6 weeks of life (Byrd et al., 2010; Cottler et al., 2017). The level of maternal estrogens rapidly decreases to levels similar to those in older children after 6 weeks of age, but the use of general or topical

estrogens in animal models seems to prolongate the bio-mechanical effects of this hormone (Kyriazis and Tsaltas, 1971; Matsuo et al., 1984; Oh et al., 1999). Breast-feeding could furthermore maintain the levels of circulating maternal estrogen in newborns beyond 6 weeks of age, most probably secondary to the presence of maternal estrogen in breast milk, and could have an influence on the bio-mechanical properties of the cartilage (Tan et al., 1997). By analogy with data on the external ear cartilage, these two parameters – breast feeding and the use of topical estrogens – could be interesting perspectives to investigate for improving the effects of nasal stenting after primary cleft repair.

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Conflicts of interest

AI and RHK have designed and manufacture a commercial anatomical retainer used in this study (Nose Fit™, Moscow, Russian Federation).

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Appendix A. Supplementary data

Supplementary data related to this article can be found at <https://doi.org/10.1016/j.jcms.2018.07.022>.

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