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Positional Preference: Prevalence in Infants and Follow-Up After Two Years

Magda M. Boere-Boonekamp, MD, PhD*, and Lida T. van der Linden-Kuiper, MD†

ABSTRACT. *Objectives.* 1) To determine the prevalence of positional preference in the general population of infants up to the age of 6 months; 2) to gather information on possible risk factors; 3) to determine the percentage of children with positional preference undergoing diagnostic evaluation and/or treatment; and 4) to assess the overall outcome of positional preference in infants and toddlers, with currently used diagnostic and treatment practices.

Setting. Infant health care centers in The Netherlands.

Methods. Seven thousand six hundred nine infants below the age of 6 months were screened for positional preference (cases: $n = 623$). Anamnestic data and physical signs of asymmetry of the range of motion and the shape of the head were recorded. These data were also registered of an immediate next child visiting the infant health care center with the same sex and about the same age but without positional preference (controls: $n = 554$). In a first follow-up study, 6 to 8 months after the original study, 468 of the 623 children with positional preference were reexamined for asymmetry of the range of motion and the shape of the head. In a second follow-up study, 24 to 32 months after the original study, 129 of 259 children who still had shown signs of asymmetry in the first follow-up study were again reexamined.

Results. The prevalence of positional preference was 8.2% and was highest in children below 16 weeks of age. The boy:girl ratio was 3:2. Firstborns, premature children, and children with breech position at the time of delivery proved to have a higher risk for positional preference. The supine sleeping position of the child and a strong preference in offering the feeding always from the right or the left side were positively correlated with positional preference. In the first follow-up study, 12% still showed restricted active range of motion, 8% restricted passive range of motion, 47% asymmetric flattening of the occiput, and 23% of the forehead. Thirty-two percent of the children with positional preference had been referred for diagnostic/therapeutic intervention. In the second follow-up study, active range of motion was restricted in 6%, passive rotation in 2%, 45% had an asymmetric flattening of the occiput, and 21% of the forehead.

Conclusion. Positional preference is frequently observed (8.2%) in The Netherlands. It leads to referral, additional diagnostics and, if necessary, treatment of al-

most 1 of every 3 affected children. Extrapolated to the original population in 1995, 2.4% of all children would still have a restricted range of motion and/or flattening of the skull at the age of 2 to 3 years. The high prevalence of positional preference in infancy, the persistency of accompanying signs, the large number of children referred for further diagnostic and/or treatment, and the resulting high medical expenses strongly call for a primary preventive approach. *Pediatrics* 2001;107:339–343; *positional preference, deformational plagiocephaly, asymmetry, infants, population-based study.*

ABBREVIATION. IHC, infant health care.

Positional preference in infants has become more prevalent in The Netherlands since the early 1990s. In children with this condition, the head is turned toward one side most of the time. Active movements of the head to the opposite side are scarce and passive mobility is often restricted.¹ In addition to this, deformational plagiocephaly with asymmetrical flattening of the occipital skull, protruding of the forehead, and a so-called bat ear on the ipsilateral side is often observed.² Less frequent accompanying signs in infants with positional preference are scoliosis, limited abduction of the contralateral hip, and foot abnormalities.

Depending on the perspective of the author, the combination of signs is described under several and different headings: torticollis,³ scoliosis of infancy,⁴ molded baby syndrome,⁵ habitual unilateral supine position,⁶ squint baby syndrome,⁷ turned head, abducted hip, truncal curvature syndrome,⁸ and Kopf-gelenk Induzierte Symmetrie Störungen syndrome.⁹

Prevalence figures of positional preference in infants have never been published. However, several authors did report an increased frequency of referrals of children with deformational plagiocephaly to tertiary craniofacial centers in the past decade. As a consequence of this, the therapeutic management of these children was extensively discussed.^{10–16} In the United States, the rising number of referrals has been attributed to the increased frequency of supine sleeping in response to the “Back to Sleep” campaign, triggered by the American Academy of Pediatrics in 1992.^{11,15,17} In The Netherlands, a comparable campaign in which prone and side sleeping positions for infants are strongly discouraged to prevent the occurrence of sudden infant death syndrome was started in 1989.¹⁸

The infant health care (IHC) program in The Neth-

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erlands is set apart from curative health care. The physicians on the program are mainly involved in monitoring and surveillance of normal development and have a counseling role. They are not permitted to initiate diagnostic procedures or to treat children under their care. If specialist involvement of any sort is deemed necessary, the child must be referred for this. It has become common practice in the IHC program to communicate handling suggestions to parents of children with positional preference. Children with serious motion deficits and those who do not improve on handling suggestions are referred for additional diagnostic evaluation to exclude underlying pathology (cervical anomalies, neurological defects, ocular torticollis, developmental dysplasia of the hip). If no pathology can be identified, usually an expectative attitude is adopted or physical therapy to stimulate symmetric development is started.

In 1995, the setting of the nonclinical surveillance environment of the regular Dutch IHC program was chosen to perform an epidemiologic survey with the following objectives: 1) to determine the prevalence of positional preference in the general population of infants up to the age of 6 months; 2) to gather information on possible risk factors; 3) to determine the percentage of children with positional preference undergoing diagnostic evaluation and/or treatment; and 4) to assess the overall outcome of positional preference in infants and toddlers, with currently used diagnostic and treatment practices.^{1,17}

METHODS

Original Survey

On their regular visit to the IHC center, a total of 7609 infants below the age of 6 months were screened for the presence of positional preference in the month of September 1995 (Fig 1). A large number of 167 IHC physicians across The Netherlands took part in this effort. Of every infant with positional preference (cases: $n = 623$), anamnestic data and physical signs were recorded. The same data were also to be registered of an immediate next child visiting the IHC center with the same sex and about the

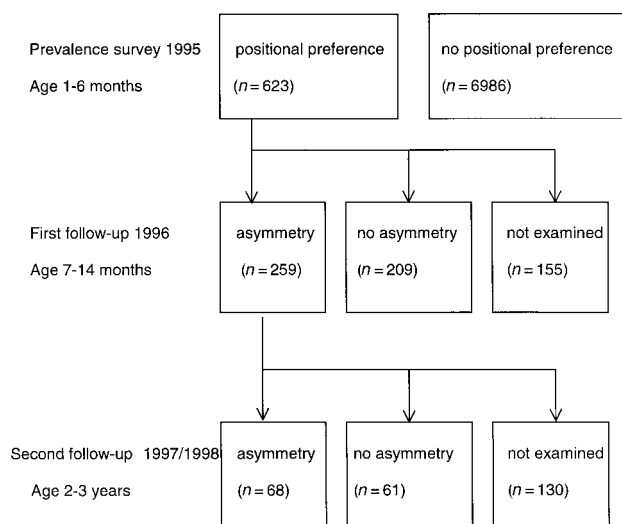


Fig 1. Schematic presentation of the numbers of children in the original survey for the prevalence of positional preference in 1995 and the 2 follow-up studies in 1996 and 1997–1998 for asymmetry in range of motion and shape of the head in children who had shown positional preference in infancy.^{1,19}

same age but without positional preference (controls: $n = 554$). Within the time fixed, it was not feasible on these conditions to find a matching control child for every case child.

First Follow-Up Study

In March and April 1996, ie, 6 to 8 months after the original survey, it proved to be possible to reexamine approximately three quarters of the 623 children ($n = 468$) with positional preference (Fig 1). The IHC physicians were requested to look for persistent signs of a restricted range of motion of the head as well as for signs of deformational plagiocephaly.

Second Follow-Up Study

Between September 1997 and April 1998, ie, 24 to 32 months after the original survey, a second follow-up examination was undertaken (Fig 1). Through this, follow-up data became available for approximately one half of the group of the 259 children ($n = 129$) who still had shown a restricted range of motion and/or signs of deformational plagiocephaly in the first follow-up study.

Definition of Positional Preference

Positional preference was defined as the condition in which the infant, in supine position, shows head rotation to either the right or the left side for approximately three quarters of the time of observation. Active rotation of the head over a range of 180 degrees cannot be accomplished; this is tested by provoking the child to follow a person or an object with its eyes and head. Passive rotation of the head to the nonpreferent side is usually possible but the range of motion is restricted.

Data Registration

In the original survey the following data were collected: 1) baseline characteristics: gender, birth rank (including stillbirths); 2) general characteristics on pregnancy and delivery: single/multiple birth, duration of pregnancy, amount of amniotic fluid (oligohydramnion, normal, hydramnion, unknown), delivery mechanism (vaginal, cesarean section), presentation at delivery (breech, other), use of ventouse or forceps on delivery, complications (fracture of the clavicle, shoulder dystocia); and 3) specific characteristics on nursing habits: sleeping position in the first week of life, after the first week, and in the days preceding the examination (supine, prone, or side); the position of the principal fosterer when taking care of the child (always from same side, alternate positions, infant's feet pointing toward the fosterer), the method of feeding (breast or bottle), right-/left-handedness of the principal fosterer when offering the bottlefeeding.

By means of physical examination the following data were collected by the IHC physician: 1) spontaneous movements, the active range of motion when following a person or object with the eyes and the head, passive range of motion of the head; 2) deformational plagiocephaly: visual assessment of the symmetry of the occipital skull and forehead and of the position of the ears; and 3) presence of a muscular torticollis (a shortened sternomastoid muscle, palpable and visible as a tight rope), scoliosis, limited abduction of the hips or abnormalities of the feet (club feet, pes adductus).

It was also recorded whether any handling suggestions were communicated to the parents with regard to the side and/or position in which the infant should be nursed. Any additional diagnostic evaluation and/or treatment that had taken place was registered as well. In the first and second follow-up study, data were collected on the spontaneous movements of the infant, the active and passive range of motion, and the symmetry of the occipital skull and forehead.

Analysis

The observed frequency distributions were compared using the Pearson χ^2 analysis. Odds ratios were calculated for the possible risk factors.

RESULTS

Prevalence

The prevalence of positional preference in infants below the age of 6 months was 8.2% ($n = 623$). The

prevalence in children younger than 8 weeks was 10%, between 8 and 16 weeks was 11%, and between 16 and 26 weeks was 3%. The boy:girl ratio was 3:2. In 68% of the 623 children, the preference was to the right side, in 27% to the left side, in 5% the direction was not reported. In 30% of the 623 children, the positional preference was accompanied by a restriction of the passive range of motion of the head.

The prevalence of specific physical signs of the condition in children with defined positional preference compared with control children is presented in Table 1. A muscular torticollis could be held responsible for the positional preference in 7.6% of the 623 children. Based on the percentages in cases and controls presented in Table 1, the prevalence of flattening of the occiput in the population of 7609 infants can be calculated at 9.9%.

$$((76\% * 623) + \{4\% * (7609 - 623)\} / 7609).$$

Baseline, General, and Specific Characteristics

The supine sleeping position of the child (after the first week of life and in the days before the examination) and a strong preference in offering the feeding always from the right or the left side were positively correlated with positional preference (Table 2). Firstborns, premature children, and children with a breech presentation at delivery proved to have a higher risk for positional preference.

Multiple/single pregnancy, amount of amniotic fluid, delivery mechanism, use of ventouse or forceps, complications in vaginal delivery, sleeping position of the child in the first week of life, and position of the fosterer when taking care of the child were not related to the prevalence of positional preference.

Follow-Up

The first follow-up study of 468 children at the age 7 to 14 months who had shown positional preference in the first half year of life demonstrated substantial improvement of the condition. However, 55% of the children ($n = 259$) still had signs of asymmetry of the shape and/or the movements of the head: 12% still showed a restricted active range of motion, 8% a restricted passive range of motion, and 47% asym-

metric flattening of the occiput and 23% of the forehead.

In the second follow-up study at the age of 2 to 3 years of 129 children of the group of 259 children who had shown signs of asymmetry of the movements and/or shape of the head in the first follow-up study, 53% ($n = 68$) still had signs of asymmetry: active range of motion was restricted in 6%, passive range of motion in 2%, and 45% had an asymmetric flattening of the occiput and 21% of the forehead.

Despite the fact that a considerable number of children were not followed up, there was no proof of any obvious positive or negative selection of cases in the first and second follow-up study. Therefore, the prevalence data of the second follow-up study were used to extrapolate the expected prevalence figures of the 623 cases at the age of 2 to 3 years. In the last column of Table 3 the thus extrapolated prevalences are presented to allow comparison with the prevalence figures in the original survey and in the first follow-up study. Of all the 623 cases, 3% would still have a restricted active range of motion, 1% a restricted passive range of motion, 25% an asymmetric flattening of the occiput, and 12% of the forehead at the age of 2 to 3 years. In the original aselect population, 2.4% of the 7609 children would still have one or more symptoms of a restricted range of motion of the head and/or deformational plagiocephaly at the age of 2 to 3 years.

$$(\{0.53 * 259 / 468\} * \{623 / 7609\} = 2.4\%).$$

Additional Diagnostics and Treatment

As was expected, handling suggestions with regard to the position in bed and the side and/or position in which the infant should be taken care of were communicated to 96% of the parents of children with positional preference. In the original survey, it was recorded that 107 of the 623 children (17%) with positional preference underwent some form of additional diagnostic evaluation and/or treatment. Between the original survey and the first follow-up study, another 15% of the children had been referred. For a number of 149 children, who underwent additional diagnostics, data on the nature of a therapeutic intervention became available: 66 children received physical therapy, 3 children were treated with a helmet, and at least 13 children proved to have developmental dysplasia of the hip and were treated accordingly. As could be expected, the signs of asymmetry were more serious and persistent among those children who received treatment for the condition.

DISCUSSION

The 8.2% prevalence of positional preference in this population-based study of children younger than 6 months of age in The Netherlands proved to be very high. In a previously published report the prevalence of deformational plagiocephaly was estimated at .3%.²⁰ The prevalence of flattening of the occiput (nearly 10%), the most common sign of deformational plagiocephaly, was far higher than expected. Of all children with signs of plagiocephaly, none proved to have the nondeformational type (cal-

TABLE 1. Prevalence Percentages of Specific Signs on Physical Examination of 623 Children Younger Than Six Months of Age With Positional Preference and of 446 Control Children

Physical Sign	Children With Positional Preference ($n = 623$)	Control Children ($n = 446^*$)
Asymmetric flattening of the occiput	76.0	4.0
Asymmetric flattening of the forehead	34.8	2.0
Asymmetric position of the ears (bat ear)	11.4	1.2
Muscular torticollis	7.6	.2
Scoliosis	18.4	1.1
Limited abduction of the hip(s)	8.0	1.4
Foot abnormality	2.4	.2

* Sufficiently detailed data of the physical examination were only available in 446 of the 554 control children.

TABLE 2. Data on Pregnancy, Delivery, Sleeping Position, and Method of Feeding of 623 Children Younger Than Six Months of Age With Positional Preference and of 554 Control Children, With Odds Ratios

Characteristic	Percentage of Children		Odds Ratio (95% CI)
	With Positional Preference (<i>n</i> = 623)	Control Children (<i>n</i> = 554)	
First child	48.1	39.6	1.4 (1.1–1.8)
Second and next child	51.9	60.4	
Prematurity	10.7	6.3	1.8 (1.2–2.8)
At term	89.3	93.7	
Breech position	8.6	5.2	1.7 (1.1–2.7)
Other position	91.4	94.8	
Supine sleeping position after first week	99.8	96.6	20.9 (2.8–157)
Other position	.2	3.4	
Supine sleeping position in the days before the examination	97.1	93.7	2.3 (1.2–4.1)
Other position	2.9	6.3	
Preferential side of feeding	64.2	54.0	1.5 (1.2–2.9)
No preference (including breastfeeding)	35.8	46.0	

CI indicates confidence interval.

TABLE 3. Prevalence of Asymmetry of the Head in 623 Infants With Positional Preference: at the Age of 1 to 6 Months,¹ 7 to 14 Months,¹ and 2 to 3 Years¹⁹; Only the Children With Asymmetry of the Range of Motion of the Head and/or Deformational Plagiocephaly Were Included in the First and Second Follow-Up Study

Sign	Prevalence			
	Original Survey Age 1 to 6 Months (<i>n</i> = 623) <i>n</i> (%)	First Follow-Up Study Age 7 to 14 Months (<i>n</i> = 468) <i>n</i> (%)	Second Follow-Up Study Age 2 to 3 Years	
			(<i>n</i> = 129) <i>n</i> (%)	(<i>n</i> = 623) (%)*
Restricted active range of motion of the head	430 (69)	54 (12)	8 (6)	(3)†
Restricted passive range of motion of the head	186 (30)	37 (8)	3 (2)	(1)‡
Asymmetric flattening of the occiput	473 (76)	220 (47)	58 (45)	(25)§
Asymmetric flattening of the forehead	217 (35)	108 (23)	27 (21)	(12)

* The prevalences for the whole group of 623 children at the age of 2 to 3 years were calculated through extrapolation to compensate for the children that were normal in the first follow-up study or not examined in both follow-up studies: the percentages of the second follow-up study were first converted into absolute numbers for the total of 259 children; these numbers were then converted into percentages for the total of 468 children in the first follow-up study; the thus calculated percentage also represents the expected prevalence in the group of 623 children of the original survey; this extrapolation is permissible because there were no indications of selection of the population in the first and second follow-up study.

† $(6 \times 259 / 100) / (468 \times 100) = .06 \times 259 / 468 = 3\%$.

‡ $.02 \times 259 / 468 = 1\%$.

§ $.45 \times 259 / 468 = 25\%$.

|| $.21 \times 259 / 468 = 12\%$.

culated prevalence in literature: .003%¹⁶). These findings strongly support the opinion that the prevalence of positional preference and deformational plagiocephaly has risen dramatically in the past decade. The observation that boys are more frequently affected than girls and the more common preferential direction to the right side are in line with other published data.^{4,6,10,21}

The higher prevalence of positional preference in firstborn children and after breech delivery is attributed to a higher compression of the fetus toward the end of pregnancy.² Positional preference was observed more frequently in children who are always bottle-fed with either the right or the left hand; offering the feeding (bottle or breast) alternately from the right or left side proved to be a protective factor. In premature children, the lower tonus, a limited possibility to move, and an asymmetrical nursing care are plausible explanations for a more frequent occurrence of positional preference.^{22,23} The prone

and side sleeping position seemed to have a protective effect for positional preference. In an unselected population in Sweden, 2.4% of the children with a prone sleeping position and 19% of children with a supine sleeping position showed positional preference.⁶ Supine sleeping was described as a predisposing factor for positional preference by several other authors.^{4,10,12,15,16,23,24}

Although not proven by this observational study, a causal relationship between supine sleeping position and positional preference seems likely. Whether the relationship between preferential side of feeding and positional preference is also of a causal nature is not clear. One may argue that a child with positional preference will typically nurse better on one side and the mother will continue to nurse the infant on this side. A preferential feeding pattern may thus be either the result of a positional preference or a cause for it.

In this study, the evaluation of the long-term ef-

fects of positional preference in infancy was restricted to the range of motion of the head and the symmetry of the skull. At the age of 2 to 3 years, asymmetry of the range of motion and deformational plagiocephaly had not disappeared in nearly one third of the infants with earlier signs of positional preference. Asymmetric flattening of the occiput proved to be the most persistent sign.

A US study showed that children sleeping in the prone position can roll over at an earlier age than those sleeping on the back or the side.²⁵ In a Dutch study sleeping and playing in the prone position were accompanied by a more advanced motor development in healthy mature-born infants at the age of 5 months.²⁶ It seems, however, that the described differences in motor development are only temporary and will disappear before the age of 18 months.²⁷ It is not clear to what extent positional preference plays a role in the transient slower motor development of supine sleeping children.

The high prevalence of the condition in infants, the persistency of accompanying signs, the large number of children referred for additional diagnostic evaluation and/or treatment, and the resulting high medical expenses strongly call for a primary preventive approach. Influencing the modifiable associated factors through primary preventive campaigns seems to be an appropriate response to the risen prevalence. Some kind of nursing advice with regard to the handling and positioning of the infant was already communicated to almost all parents of children with positional preference. Despite this, a high number of children underwent additional diagnostic and/or therapeutic interventions (32%). This is a rather alarming observation, because these interventions cause much anxiety in parents and lead to high medical expenses. Unfortunately, the framework of our observations does not allow any conclusions to be drawn with regard to the preventive effects of handling suggestions or the effects of therapeutic interventions. It is clear that only a prospective randomized trial will be able to provide an answer to this.

In the IHC program in The Netherlands, a preventive guideline with accompanying educational material has been designed and is used now in the counseling of parents on positioning, handling, and nursing of the infant from the neonatal period onwards. Of course, the content of this guideline does not contradict in any way with the rightly justified recommendations on the sleeping position of infants in the Dutch equivalent of the "Back to Sleep" campaign. To determine whether such a primary preventive approach is beneficial, it will be necessary to evaluate the effects of the use of the guideline on the prevalence of positional preference in infants in the years to come.

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