

Transverse Dimension of Maxillary Arch in 5-Year-Old Children with Unilateral Cleft Lip and Palate: A Retrospective Study

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Abstract

Hypoplastic and distorted maxilla in children with unilateral cleft lip and palate (UCLCP) has always been a challenging feature for orthodontic rehabilitation. Series of orthodontic and surgical collection took place over span of 20 years. Morphology of malformed arch in the early stage influenced the later treatment options greatly. This study aimed to present characteristics of maxillary arch in transverse dimension of 5-year-old, UCLCP children, and to compare those in the cleft side to the non-cleft counterpart. Measurements were performed on 55 dental models of maxillary arch which were mature primary dentition, belonged to non-syndromic UCLCP children selected from the Khon Kaen University (KKU) Cleft Center's archive since 2002-2020. The measurement included intercanine width, intermolar width at primary first molars (D) and second molars (E) levels, arch height and arch perimeter by only one researcher (Intraclass correlation coefficient =0.96). Analysis of the whole arch showed the mean values (SD) in millimeters, including the intercanine width =28.86 (3.37); intermolar width-D =33.42 (2.75) and -E =39.73 (2.31); arch height =14.11 (2.23), and arch perimeter =68.35 (6.09). The comparison of arch dimensions between the cleft side and non-cleft side, means of intermolar width at both D and E levels were significantly wider (Wilcoxon signed rank test; $p < 0.01$). Similarly, significantly larger of the mean arch perimeter was observed on the cleft side (Paired t-test; $p = 0.01$, 95% CI: 0.23-1.64). There was no significant difference of means of the intercanine width and arch height between both sides. Results demonstrated that at this stage of full primary-dentition, arch dimensions of the cleft side appeared to be, at least, comparable to the non-cleft side. The significantly wider transverse dimension observed in the posterior section might contribute to the larger arch perimeter of the cleft side. Our findings suggested that maxillary arch collapse in the transverse plane was not obvious at this stage.

Keywords: Unilateral cleft lip and palate (UCLCP)/ 5 years old/ Transverse arch dimension/ Cleft side/ Non-cleft side

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Introduction

Prevalence of clefts in Thailand during the past three decades, has been varied, between 0.84-2.49 in 1,000 live births.¹ The prevalence reported in the Northeastern region ranks in the higher end of this range.²⁻³ Malformation of lip and palate disturbed the development of surrounding structures and hence, compromised their appearances and functions.⁴ Children born with orofacial cleft need comprehensive health care from at birth, in some cases from prenatal stage, through adulthood.⁵ To maximize the treatment outcomes, interprofessional guideline required multidisciplinary management to rehabilitate all

aspects of these congenital anomalies. The evidence-based treatment guidelines aim to maximize growth of the child to one's potential and restore the functions and esthetic of the face, which help promoting patients' quality of life and self-esteem so that our patients would be able to blend in with the community.

Complete cleft lip and palate has been considered as the most severe form on all types. However, unilateral cleft lip and cleft palate (UCLCP) may be the most difficult to manage due to asymmetrical of maxillary arch and distortion of the

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arch form.^{4,6} In Thailand, the prevalence of UCLCP is accounted for 50-70% of the orofacial clefts.^{1-3, 7,8} UCLCP children were born with developmental defects both in the skeletal and dentoalveolar structures, which affected their maxillary growth in the antero-posterior, vertical and transverse planes. Depending on the severity, the cleft arch will be, firstly, orthopedically-orthodontically modified and the remaining gap closed by orthodontist and surgeon using nasoalveolar molding device, follow by closing of oronasal gap and palate. These treatments happened before their second birthday. It is recommended that outcomes of the earlier comprehensive treatment could be accurately monitored at 5 years old, that would serve as the predictor for future prognosis.⁹ Later in the mixed dentition prior to the subsequent phase of orthodontic treatment commenced, anterior and/or posterior crossbite usually became more noticeable.¹⁰⁻¹¹ Malposition of either greater or lesser segment makes the maxillary arch appeared disproportionate in the transverse direction. It is assumed that the transverse arch dimension of cleft sides would be more severely collapsed.¹²⁻¹³ Whereas those of the non-cleft side might be, more or less, similar to those of unaffected children. Deficiency of the transverse dimension of UCLCP arch at 5-year-old could be a consequence of palatoplasty. Severity of the UCLCP arch during the 5-year-old stage greatly influences the later treatment options and the treatment outcomes in the permanent dentition.¹⁴ After 5 years old, the orthodontic treatment aims to modify the growth and align the teeth so that upper and lower arches of cleft patient are coordinated, but not to be comparable to those of the non-cleft individuals. Previous studies showed that either side of the UCLCP arch reacted to the treatment e.g. surgery and orthodontic, in different manners.¹⁵⁻¹⁷ Transverse arch-expansion, therefore, is one of the great concerns for orthodontic plan during mixed

dentition stage for children with UCLCP. The degree of maxilla deficiency of the asymmetrical UCLCP arch has not been reported in Thailand. Therefore, the aims of this study are to examine the transverse arch dimension of 5-year-old children with UCLCP, and to compare these parameters between cleft and non-cleft sides using models available in our Center. Data derived from this study will provide the genuine extent of the maxillary arch transversal defect and differentiate the discrepancy between the cleft and non-cleft sides, that may contribute to decision making of the transverse arch expansion.

Materials and methods

This study was approved by Center for Ethics in Human Research, Khon Kaen University (HE642122).

Sample

Samples were dental model of children born with non-syndromic UCLCP, who registered with the Center of Cleft Lip-Cleft Palate and Craniofacial Deformities Incorporation with Tawanchai Foundation and attending Faculty of Dentistry, Khon Kaen University for treatment according to KKU Cleft protocol (https://kkucleft.kku.ac.th/?page_id=1654) since 2002-2020. All samples in the archive were derived from patients who had surgical repair of lip and palate according to KKU Cleft protocol. As part of the 5-year index records, the studied models were derived from children aged approximately at 5 years old.⁵ All available maxillary models that had mature primary dentition, where 2nd molars fully erupted, were selected. These included models with carious cavity on crowns. Excluded models were 1) presented with permanent tooth, 2) broken or had defects, which unable to identified all the reference points.

Data collection

General data of the sample were recorded as follows; gender, cleft laterality, and cleft size. Cleft size was measured at the widest interalveolar gap.

Arch dimension measurement

Transverse dimension was measured on cleft and non-cleft sides separately by midline. The model midline was defined as follows; In the anterior, reference points were the median labial frenum and the point of incisive papilla. Posterior boundary was set by a line between right and left tuberosity.¹⁸⁻²⁰ A straight midline (M in Figure 1A) was then drawn from anterior reference points, perpendicularly, to the posterior boundary. Additional reference points were determined (Figure 1A) as follows; intercanine width $\{(C-M \text{ Line})+(M \text{ line}-C')\}$: distance from midline to the

tip of right and left primary canines; intermolar width $\{(D-M \text{ line})+(M \text{ line}-D')\}$ and $\{(E-M \text{ line})+(M \text{ line}-E')\}$: distance from midline to central fossa of right and left primary first and second molars and arch height: distance between a line intermolar width of the right and left primary first molar to M line and contact point of the primary central incisor. In case that any index tooth was absent, mid-point of the width of alveolar ridge at the referred position was used. Arch perimeter (a+b and c+d): sum of linear distances from a to b and c to d as shown in Figure 1B.

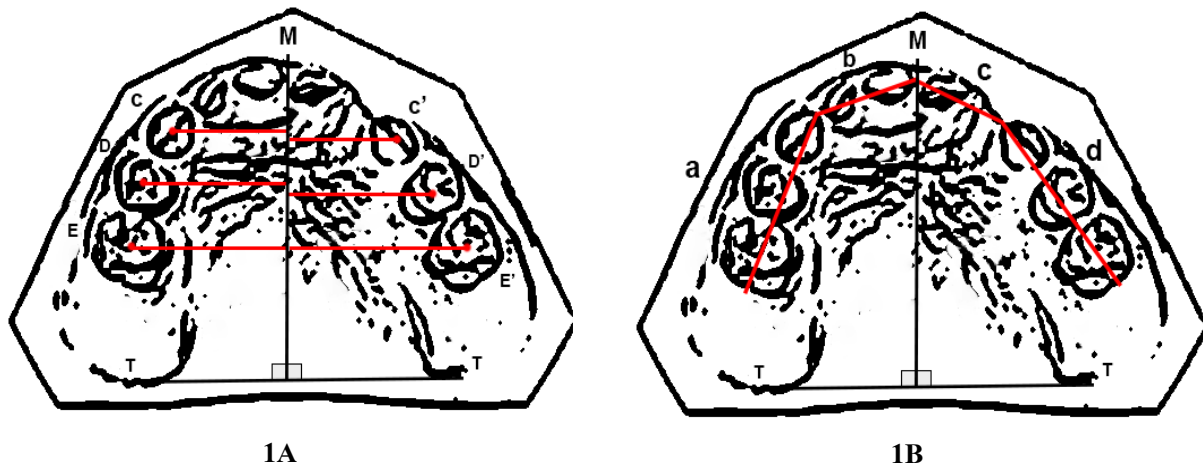


Figure 1 Diagram depicts measurements of transverse arch dimension 1A) and arch perimeter 1B) on the maxillary arch of a UCLCP dental model. Transverse arch dimension of each side was measurement of the lines from 3 reference points from non-cleft side (*i.e.* C, D, E) and those from cleft side (*i.e.* C', D', E') perpendicularly to the midline (M). The upper case letters with (') in 1 A and "c" and "d" in 1B, designate for measurements on the cleft side.

Calibration of the examiners

One researcher was trained and calibrated to collect the data with an experienced orthodontist. On dental models, sharp black pencil ($\varnothing=0.5 \text{ mm}$) was used to locate each reference point under the light setting. Variables were measured using orthodontic transparent template and a manual divider. Twenty dental models were randomly selected to conduct for intra-examiner reliability test. Intraclass correlation coefficient for intra-examiner reliability was 0.96, considering as good. Each measurement was performed in triplicate and mean values were used.

Statistical analysis

Statistical analysis was performed using IBM® SPSS® Statistics version 27.0 (IBM Corp., New York). General characteristics, laterality and cleft size were reported as frequencies, percentages and mean and standard deviation (SD). Wilcoxon signed test was used to compare the intercanine width and intermolar width of the cleft side to those of the non-cleft side. While, the mean of arch height and arch perimeter were compared using paired t-test. Statistical significance difference was set at $p<0.05$.

Results

Fifty-five models of UCLCP patient met the inclusion criteria and undergone the analysis (Table 1). There was a slightly higher proportion of girl (52.7%). Cleft occurred mainly on the left side (67.3%). The mean cleft width was 1.89 (1.62) mm. and the width range of cleft size was 0.5 - 5.5 mm. In 28 studied models (50.9%), at least one tooth was absent from the ridge.

Table 1 General characteristics of the studied samples (n = 55)

Variables	Frequency	Percentage	
Gender	Male	26	47.3
	Female	29	52.7
Cleft site	Right	18	32.7
	Left	37	67.3

Analysis on whole transverse arch dimension showed that the mean (SD) of intercanine width was 28.86 (3.37) mm., and means of intermolar width for D-D' was 33.42 (2.75) mm. and for E-E' was 39.73 (2.31) mm. The mean of arch height was 14.11 (2.23) mm. and was at 68.35 (6.09) mm. for arch perimeter. When compared the transverse dimension between affected and unaffected sides, the cleft side was wider at the posterior part (Table 2). The means of intermolar width of the cleft side, at both levels of the first and second molars were significantly wider than those in the non-cleft side. However, the mean of intercanine width was comparable on both sides.

The mean of arch perimeter of the cleft side was more than that on the non-cleft side, statistically significant. The mean of arch height of the cleft side was slightly longer, but not statistically significant.

Table 2 Comparison of arch dimensions between cleft side and non-cleft side

Arch dimension (mm.)		Cleft side	Non-cleft side	p-value	
		n = 55	n = 55		
Intercanine width (mm.)	mean (SD)	14.54 (2.74)	14.32 (1.69)	0.44 ^a	
	Median (Q1 - Q3)	14.50 (13.00 - 16.50)	14.50 (13.00 - 15.00)		
Intermolar width (mm.)	(D)	mean (SD)	17.34 (2.17)	16.07 (1.67)	< 0.01* ^a
		Median (Q1 - Q3)	17.00 (16.00 - 19.00)	16.00 (15.00 - 17.50)	
	(E)	mean (SD)	20.76 (2.37)	18.96 (1.48)	< 0.01* ^a
		Median (Q1 - Q3)	21.00 (19.00 - 22.50)	19.00 (18.00 - 20.00)	
Arch height (mm.)	mean (SD)	14.48 (2.67)	13.74 (2.65)	0.06 ^b	
	Median (Q1 - Q3)	14.50 (12.00 - 16.50)	14.00 (12.00 - 15.00)	(95% CI; -0.05-1.52)	
Arch perimeter (mm.)	mean (SD)	34.64 (3.32)	33.71 (3.31)	0.01* ^b	
	Median (Q1 - Q3)	34.50 (32.00 - 37.00)	33.50 (31.00 - 36.00)	(95% CI; 0.22-1.64)	

Different superscript letters indicate significant differences in the comparison method using Wilcoxon signed rank test (a) and the Paired t-test (b). (*) indicates mean difference is significant at the 0.05 level (2-tailed)

Discussions

Inherently, the maxilla of UCLCP was hypoplastic. The alveolar ridge was discontinued with deviation of lesser segment from the greater segment.²¹ From the edentulous arch, at birth, to fully developed primary dentition, growth of the cleft arch was, in part, the outcome of the prior treatments, which influenced by the timing and protocol.²²⁻²³

Initially, those segments required alignment by presurgical orthopedic/orthodontic appliance. Cleft defects at lip and palate were later surgically repaired, which inevitably affected the transverse arch dimension. In most cases, arch expansion would be provided by orthodontic appliance at late-mixed or permanent dentitions. Well-aligned and expanded

arches gained huge benefits on both esthetic and function as well as facilitated surgeons on the bone grafting.^{5,24} In each well-established center, the same principle protocol e.g. orthodontic and surgical plan, has often been calibrated among the team to maximize the treatment outcomes, including the technique and the timing for treatment. The variation in the treatment methods usually is due to complexity of the cleft, rather than the operator/surgeon's preference. The systematic treatment outcomes are commonly monitored when patient is approximately at the age 5, 10, 15 and 20 years.⁵

The analysis of dental models at 5-year-old has been recognized as a reliable index to monitor growth and treatment outcomes at the early stage.⁹ This study addressed the intensity of transversal problem of the UCLCP maxilla by examining the cleft side and compared to the non-cleft side of the same child using the model according to the 5-year index. Dental model had captured the arch dimension for later re-analysis. Measurement of arch parameters using dental model was an accurate and reliable method, which has been used across the disciplines. The data can be compared to other previous studies. One of the drawbacks of model analysis is limited compliance of such young children, which make the whole arch impression taking became impossible in some cases. Others include the damage of the models over time and even loss of some models. In this study, individual's information including radiographs and medical conditions, other than cleft types, have not been analyzed.

In this study, cleft presented more frequent on the left side of the arch similar to the epidemiological report.²⁵ Our studied samples, deriving from UCLCP children, at approximately 5 years of age, who were homogeneously treated patients under the KKU Cleft Protocol within the Center. They were in mature primary dentition of which all the primary second molars, the last teeth to

emerge, had fully erupted. Evidence has showed that at the age range of 4-5 years old the dimension of maxillary arch was very much unchanged and a significant growth would resume as the first permanent teeth come through.²⁶⁻²⁷

The maxillary arch with UCLCP, in this study, were smaller in all dimensions when compared to the average measurement reporting in a cohort study of non-cleft Thai children at 5 years old.²⁸ Arch perimeter was reported to be 73.49 (3.30) mm. as compared to ours at 68.35 (6.09) mm. The reported width of intercanine at 30.24 (1.78) mm. and intermolar at 44.24 (2.16) mm in non-cleft Thai children, also wider than those of our study at 28.86 (0.45) mm. and at 39.73 (2.31) mm., respectively. Arch height was not reported in the cohort study so the antero-posterior plane was unable to compare with our data. Studies from other ethnicities also showed that maxillary arch dimension of UCLCP children in their primary dentition was relatively underdeveloped when compared with the non-cleft children. However, they were closely resembled to the dimension of the unilateral cleft lip children^{10, 29} and the cleft palate only.^{11, 28} In view of interarch relationships, discrepancy occurred both the antero-posterior and the transversal dimensions. The reduction of maxillary transverse dimension led to posterior arch discrepancy, featured as posterior crossbite. It has been shown that cleft and non-cleft sides affected differently by the surgery/orthodontic treatments,^{13, 15-16} which resulted in further distortion of the UCLCP maxilla. Planning for arch expansion was, therefore, required analysis of all these factors. A recent study by the Cleft Care UK found that the transverse dimensions of maxillary arch of 5-year-olds had improved in small but positive direction due to the continuing development of cleft protocols and better quality of training. The transverse dimensions of lesser segment (cleft side) was one of the

outcomes that was reported to be increased at both primary canine and second molar levels.³⁰

Based on the study's aim, data was analyzed to compare between affected and unaffected sides. The transverse dimension of cleft side was significantly larger, particularly at the posterior portion, whereas the anterior width was comparable. The findings supported that the growth of anterior portion, either on affected or unaffected side, which undergone restriction immediately after surgical lip repaired, had caught up at around 5 years old and became relatively least affected in UCLCP children.^{21, 29} The wider of the posterior transversal width at the cleft side could result from position of the lesser segment. The lesser segment, which contained the molars, tended to be positioned backward and outward, diverted from the midline. The position of primary (D') and secondary (E') molars that served as reference points had, therefore, drifted distally and buccally. Larger arch perimeter and intermolar width could, therefore, be expected on the cleft side. The posterior transverse dimension at 5 years old in this study was shorter by approximately 5 mm. than the non-cleft Thai children,²⁸ similar to other reports.^{21, 29} At an earlier stage, the comparison of UCLCP to, non-cleft controls and to other cleft groups, was controversial.^{10, 31} This might be due to the different treatment protocols e.g. palatoplasty, in various centers. As the development progresses into mixed dentition, the posterior dimension might be able to approach the normality, still with certain degree of constriction, without orthodontic treatment.¹⁴ By analyzing cleft and non-cleft sides separately, we found that transverse arch dimension of the affected side was at least comparable, and not smaller than that measured on the unaffected side at 5 years old. A simple comparison of the average transverse dimension of the non-cleft, Thai children from a cohort study demonstrated that either cleft or non-cleft side was smaller than maxilla of the non-cleft. This finding emphasizes that growth of the non-cleft side is also deficient in these UCLCP children.

Some features could also impact the maxillary arch dimension. A larger cleft size located mesially to the canine, has been associated with less development of crossbite at the posterior and anterior segments in the primary dentition.³² The larger cleft gap seemed to keep the transverse constriction less severe. The average cleft width presented in the studied samples was around 2 mm. The sub-analysis could not be performed at this stage of study, as there were only 20% of the samples had cleft gap greater than 3 mm.

Based on findings from this and other studies, there are several combinations of internal and external factors that could variously influence the growth and development of the maxillary arch in children with UCLCP. Some of which, could be controlled. Attempts by all disciplines in the team should be made to reduce collapsing of the maxilla and to facilitate patient's arch growth to one's potential.

Conclusions

Transverse arch dimension of children with repaired UCLCP, treated under a homogenous protocol, was reported. The transversal width and arch perimeter of the cleft side are greater at the posterior level, without signs of the segmental collapse on the cleft side at 5 years old as shown by increased intermolar width. Altogether, our findings suggested that close monitoring and early intervention may be implemented in UCLCP cases that predisposing factors for arch dimension loss are detected.

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นันทรัตน์ วิจิตร* ญัฐวีร์ เผ่าเสรี** *** เข็มพร กิจสรวงศ์** พรทิพย์ ผงจวีรียาทร** ปฎิมาพร พิงชาญชัยกุล*** **

บทคัดย่อ

ขากรรไกรบนของเด็กที่มีภาวะปากแหว่งเพดานโหว่ด้านเดียวมีลักษณะการเจริญพร่องและผิดปกติซึ่งเป็นปัญหาหนึ่งของการรักษาฟันที่ ต้องได้รับการรักษาทางทันตกรรมจัดฟันและศัลยกรรมช่องปากเป็นช่วงเวลาต่อเนื่องยาวนานถึง 20 ปี โดยรูปร่างของส่วนโค้งขากรรไกรที่ผิดปกติในระยะแรกส่งผลต่อทางเลือกการรักษาในภายหลังอย่างมาก การศึกษานี้มีวัตถุประสงค์เพื่อรายงานลักษณะของส่วนโค้งกระดูกขากรรไกรบนในมิติแนวขวางในเด็กอายุ 5 ปีที่มีภาวะปากแหว่งเพดานโหว่ด้านเดียว และเปรียบเทียบลักษณะต่างๆระหว่างด้านที่มีและไม่มียรอยโหว่ นำแบบจำลองฟันขากรรไกรบนจำนวน 55 ชิ้นของเด็กที่มีภาวะปากแหว่งเพดานโหว่ด้านเดียวที่ไม่มีความพิการอื่นร่วมด้วยในศูนย์ปากแหว่งเพดานโหว่ฯ มหาวิทยาลัยขอนแก่น ตั้งแต่ปี ค.ศ. 2002-2020 มาวัดขนาดส่วนโค้งของขากรรไกร คือ ความกว้างส่วนโค้งแนวฟัน ความสูงส่วนโค้งแนวฟัน และความยาวโดยรอบกำหนดให้ผู้วิจัยเป็นผู้วัดแบบจำลองฟัน (ค่าสัมประสิทธิ์สหสัมพันธ์ภายในชั้น = 0.96) การวิเคราะห์ขนาดส่วนโค้งแสดงเป็นค่าเฉลี่ย และส่วนเบี่ยงเบนมาตรฐาน หน่วยเป็นมิลลิเมตร ดังนี้ ความกว้างส่วนโค้งแนวฟันเขี้ยวข้างบน, ฟันกรามน้ำนมซี่ที่หนึ่ง และฟันกรามน้ำนมซี่ที่สอง เท่ากับ 28.86 (3.37), 33.42 (2.75) และ 39.73 (2.31) มิลลิเมตร ตามลำดับ; ส่วนความสูงของส่วนโค้งแนวฟัน เท่ากับ 14.11 (2.23) มิลลิเมตร และความยาวโดยรอบ เท่ากับ 68.35 (6.09) มิลลิเมตร เมื่อนำค่าขนาดส่วนโค้งขากรรไกรด้านที่มีรอยโหว่เทียบกับด้านที่ไม่มีรอยโหว่ พบค่าเฉลี่ยของความกว้างส่วนโค้งแนวฟันกรามน้ำนมซี่ที่หนึ่ง และฟันกรามน้ำนมซี่ที่สองของด้านที่มีรอยโหว่มากกว่าด้านที่ไม่มีรอยโหว่อย่างมีนัยสำคัญทางสถิติ ($p < 0.01$) เช่นเดียวกับค่าเฉลี่ยของความยาวโดยรอบส่วนโค้งแนวฟัน ($p = 0.01$, ช่วงความเชื่อมั่นที่ร้อยละ 95: 0.23-1.64) ในขณะที่ไม่พบความแตกต่างอย่างมีนัยสำคัญของความกว้างส่วนโค้งแนวฟันเขี้ยวข้างบน และความสูงของส่วนโค้งระหว่างทั้งสองด้าน ผลการศึกษาแสดงให้เห็นว่าในระยะชุดฟันน้ำนม ขนาดส่วนโค้งแนวฟันด้านที่มีรอยโหว่มีขนาดใกล้เคียงกับด้านที่ไม่มีรอยโหว่ และความกว้างที่มากขึ้นในขากรรไกรด้านหลังของด้านที่มีรอยโหว่ส่งผลให้ความยาวโดยรอบของส่วนโค้งแนวฟันมีปริมาณมากขึ้น จากการศึกษาชี้ให้เห็นว่าการหดตัวของสันกระดูกขากรรไกรบนไม่ชัดเจนในระยะนี้

คำไชรหัส: ภาวะปากแหว่งเพดานโหว่ด้านเดียว/ อายุ 5 ปี/ ส่วนโค้งขากรรไกรในแนวขวาง/ ด้านที่มีรอยโหว่ของปากแหว่งเพดานโหว่/ ด้านที่ไม่มีรอยโหว่ของปากแหว่งเพดานโหว่

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