Malposition of Teeth and Jaws in Patients with Congenital Superior Oblique Palsy

Zahn- und Kieferfehlstellungen bei Patienten mit kongenitaler Trochlearisparese

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Key words
- dental malposition
- head tilt
- superior oblique palsy

Schlüsselwörter
- Zahnfehlstellung
- Kopfzwangshaltung
- Trochlearisparese

Abstract

Background: Patients with congenital superior oblique palsy tend to adopt a head tilt to the contralateral side to maintain binocular single vision. It has long been recognised that facial asymmetries may be caused by a head tilt. The aim of this study was to describe the effect of habitual head tilt due to congenital superior oblique palsy on dental occlusion.

Patients and methods: The study was designed as a descriptive cohort study. Ten patients with congenital superior oblique palsy (3 female, 7 male; mean age 51.7 (y) ± 15.8 SD, ranging from 19 to 69 (y)) underwent orthodontic examination. Orthodontic findings and values for vertical, torsional and horizontal deviation measured with the Harms tangent screen and stereopsis using a random dot test were compared.

Results: Three orthodontic parameters were found to correlate significantly or at least as trend with orthoptic parameters. Midline deviation of the upper jaw to the face (rho = 0.623; p = 0.054) and anterior positioning of upper first molar in the sagittal plane (rho = 0.594; p = 0.07) correlate with the vertical deviation; overbite correlates with horizontal deviation measured in the primary position (rho = 0.768; p = 0.016).

Conclusions: In this small study, three orthodontic parameters correlated with orthoptic findings in patients with congenital superior oblique palsy. Further studies are needed to establish whether congenital superior oblique palsy is more frequent in patients exhibiting abnormal values of these orthodontic parameters.

Zusammenfassung

Hintergrund: Patienten mit kongenitaler Trochlearisparese nehmen eine Kopfneigung zur Gegenseite ein um das binokulare Einfachsehen zu bewahren. Gesichtsasymmetrien als Folge der Kopfzwangshaltung sind beschrieben. Wir untersuchten den Einfluss der Kopfzwangshaltung auf die dentale Okklusion.

Patienten und Methoden: Zehn Patienten mit kongenitaler Trochlearisparese (3 weiblich, 7 männlich; Alter 51,7 (J) ± 15,8, zwischen 19 und 69 (J)) wurden im Rahmen einer Kohortenstudie kieferorthopädisch untersucht. Die vertikalen, torsionellen und horizontalen Deviationen (Harmswand Tangenten Methode) und die Stereopsis (Lang-Test) wurden mit den kieferorthopädischen Daten verglichen.

Ergebnisse: Drei kieferorthopädische Parameter korrelierten signifikant oder als Trend mit orthoptischen Werten. Sowohl die Deviation der Mittellinie des Oberkiefers zur Mittelinie des Gesichtes (rho = 0,623; p = 0,054) als auch die anteriore Position der oberen ersten Molaren in der Sagittalebene (rho = 0,594; p = 0,07) korrelieren mit der vertikalen Deviation gemessen in Primärposition. Der Overbite korreliert mit der horizontalen Deviation gemessen in Primärposition (rho = 0,768; p = 0,016).

Schlussfolgerungen: In dieser Pilotstudie wurden drei kieferorthopädische Parameter gefunden, die mit orthoptischen Werten bei Patienten mit kongenitaler Trochlearisparese korrelieren. Weitere Studien müssen zeigen, ob in Personen mit diesen kieferorthopädischen Werten häufiger kongenitale Trochlearisparesen zu finden sind.

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Background

Patients with superior oblique palsy typically experience diplopia. To maintain binocular single vision patients tend to adopt a compensatory head tilt to the contralateral side. Especially patients with congenital superior oblique palsy (CSOP) develop a head tilt in early childhood, which may influence the development of the facial skull and thus lead to asymmetric proportions of the face. Even though CSOP is not a rare disease the diagnosis is not easily made. Therefore patients with CSOP often have a history of long-standing head tilt before being treated.

Facial asymmetry in patients with CSOP was first described by Wilson and Hoxie in 1993. They described a "facial atrophy" on the contralateral side of the paresis. They also reported one patient with dental malocclusion because of changes in the growth of the jaw [1]. Thereafter additional reports about facial asymmetry in patients with CSOP appeared in the medical literature [2–9]. Typically the orbit on the atrophic side is located deeper, the angle of the mouth higher than on the healthy side as seen in **Fig. 1**.

To the best of our knowledge this is the first study to examine a possible influence of longstanding head tilt due to CSOP on the jaw development with subsequent specific dental malocclusions. The purpose of this descriptive cohort study was to evaluate dental parameters in patients with CSOP.

Fig. 1 Photograph of a patient with a head tilt to the right. Note: Interpupillar line and dental occlusion line indicating Interpupillar dental occlusion line angle (IPDO-A).

Patients and Methods

Between 2000 and 2014 a cohort of 69 consecutive patients with CSOP were seen at the Orthoptic service of the Department of Ophthalmology of the University Hospital Zurich, Switzerland. Of these 69 patients 67 were contacted. Two were younger than 18 years and thus not allowed to participate in the study. The study protocol was reviewed and approved by the local and institutional ethics committee (KEK-ZH 2013–0342). The authors followed the Tenets of the Declaration of Helsinki. All patients signed a consent form. Patients with removable dental prosthesis were excluded from the study. Additional exclusion criteria were the absence of natural occlusion in the posterior teeth as well as lack of qualitatively sufficient documentation of preceding orthodontic treatment.

During the recruitment 12 patients had to be excluded because of an orthodontic treatment history without complete pre-orthodontic data. No patients had to be excluded due to removable dental prosthesis. One patient had insufficient pre-operative orthoptic documentation. 14 patients could not be reached due to invalid telephone number or postal address. 30 patients were not willing to participate in the study due to several reasons.

Ten patients with CSOP treated at the Department of Ophthalmology, University Hospital Zurich, Switzerland, were included in this descriptive cohort study and underwent subsequent dental examination. The examination included a clinical part, looking for facial asymmetries such as deviation of chin or nose to either side. In addition the deviation from the midline of the face (line through the middle of the nose and the philtrum) to the papilla incisiva between the two upper central incisors in the transversal plane was measured in mm (ML UJ/F). Furthermore evaluation of photographs and of plaster models formed out of two alginate impressions was performed. Photographs were analyzed for the presence of a head tilt and of a possible deviation of interpupillar (line through the pupils) and occlusal line (line along a wooden stick the patients were biting on). A head tilt was present if a visible deviation of the head compared to the upper trunk was present (**Fig. 1**). The interpupillar line and the occlusal line were drawn (**Fig. 1**). The angle between these two respective lines was measured (interpupillar dental occlusion = IPDO angle = IPDO-A). Evaluation of plaster models consisted of intra- and intermaxillary parameters. These parameters were measured in the sagittal, vertical and transversal dimensions.

In the sagittal plane the overjet (Oj) was measured at the level of the first right upper incisor. It points to the sagittal relation between the upper and lower front teeth. The sagittal position of the first upper molar was measured in mm (6 + 6 ant). 6 + 6 ant indicates which first molar is located more anteriorly. In the lower jaw the more anteriorly located buccal segment was determined (only right/left) (LJ BS ant).

In the vertical plane the overbite (Ob) was measured at the level of the mesial of the right central incisor (tooth 11). It points to the vertical relation between upper and lower front teeth. The curve of spee is the name of the curvature of the teeth of the lower jaw in the mesiodistal dimension. The dimension of the spee was measured on the right and on the left side. For that purpose a line from the buccal cusp of the first premolar of the lower jaw to the distobuccal cusp of the first molar was drawn and the deepest point of the spee curve in the vertical was measured in mm on each side.

In the transversal plane the deviation of the papilla of the two central incisors in the upper and the lower jaw (ML LJ/UJ) and of the palatal papilla of the upper central incisors to the median raphe was measured (ML UJ/Raphe). The transversal position of the first upper molars was measured in mm (6 + 6 pal). 6 + 6 pal indicates which molar is located more palatinal. 6 + 6 coordination in angle class I (6 + 6 class I) characterizes the transversal coordination of the upper and lower jaw. These dental parameters were compared to preoperative amounts of vertical, torsional and horizontal deviation in primary position using Harms tangent screen. "Harms tangent screen" is an orthoptic examination technique that serves to quantify the horizontal, vertical and torsional components of eye misalignment in nine different gaze positions [10]. In this study only measurements in primary position were analyzed, e.g. with the patient looking straight ahead in distance. Stereopsis was tested using a random dot test (Lang stereotest), in which pictures are used of stereo figures that are

Table 1 Correlation (spearman's rho) between the continuous variables of dental parameters and the orthoptic values measured in primary position of each eye using the Harms tangent screen. Negative values in horizontal deviation describe exo-deviation, positive values eso-deviation. Negative values in cyclodeviation describe incyclo-deviation, positive ones excyclo-deviation. Negative values in vertical deviation describe left over right position, positive values right over left. All values were adapted for right sided palsy.

		ML UJ/F	ML LJ/UJ	ML UJ/Raphe	Oj	Ob	6 + 6 class I	6 + 6 ant	6 + 6 pal	Spee right	Spee left	IPDO-A
Horizontal deviation	**rho**	− 0.498	0.127	− 0.367	0.166	0.768	0.400	− 0.068	− 0.034	0.412	0.474	0.105
	p-value	0.172	0.745	0.331	0.670	**0.016**	0.374	0.862	0.930	0.310	0.282	0.788
	N	9	9	9	9	9	7	9	9	8	7	9
Vertical deviation	**rho**	0.623	0.110	0.407	0.415	0.073	− 0.615	0.594	0.222	− 0.091	− 0.056	0.546
	p-value	**0.054**	0.762	0.243	0.233	0.840	0.105	**0.070**	0.537	0.815	0.895	0.102
	N	10	10	10	10	10	8	10	10	9	8	10
Cyclo-deviation	**rho**	0.069	0.175	− 0.259	− 0.295	0.138	0.205	− 0.188	0.381	0.481	0.169	− 0.111
	p-value	0.849	0.629	0.469	0.408	0.704	0.627	0.603	0.278	0.190	0.689	0.761
	N	10	10	10	10	10	8	10	10	9	8	10

Abbreviations: ML UJ/F = Distance of Midline (ML) of upper jaw (UJ) (between two upper incisors) and face (F) (middle of the nose and the philtrum) [mm]. ML LJ/UJ = Midline lower jaw (LJ) and upper jaw (UJ). ML UJ/Raphe = Midline upper jaw and median raphe palatina (Raphe). Oj = Overjet = distance between upper and lower incisor in sagital direction [mm]. Ob = Overbite = distance between upper and lower incisor in vertical direction [mm]. 6 + 6 = describing positions of the first molar teeth in angle class 1 (class 1) (meaning transversal distances between upper first molars and lower first molars), anterior (ant) or palatinal (pal) direction. Spee right and Spee left describes the curvature of the curved line of teeth alignment [mm] on each side. IPDO-A = Interpupillar dental occlusion line angle [°]. N = number of patients analysed (some missing data due to removed teeth. In one case no horizontal deviation was measured).

Table 2 Correlation between discrete variables of dental parameters and side of paresis using Pearson Chi-square Test (two-sided) (X2). Measured for deviations of the nose, the chin, the more anterior lower buccal segment (LJ BS ant) and the head tilt correlated to the side of the congenital superior oblique palsy (CSOP).

			Head tilt right/left X2 = 6.190 p = 0.045			Deviation nose X2 = 1.429 p = 0.490			LJ BS ant re/li X2 = 0.635 p = 0.728			Deviation chin X2 = 2.857 p = 0.240		
			left	neutral	right	left	neutral	right	left	neutral	right	left	neutral	right
Side of CSOP	left	N	0	3	4	4	1	2	1	4	2	0	5	2
		%	0%	43%	57%	57%	14%	29%	14%	57%	29%	0%	71%	29%
	right	N	2	0	1	1	0	2	1	1	1	1	1	1
		%	67%	0%	33%	33%	0%	67%	33%	33%	33%	33%	33%	33%

embedded in a background of random dots. Only subjects with normal stereopsis can recognize the presented pictures that seem to emerge in front of the plate.

For statistical analysis all values for horizontal, vertical and torsional deviation in primary position taken from Harms tangent screen measurements of patients having a left sided superior oblique palsy were adapted to the right side.

Data were coded in Excel (Microsoft Office Excel 2007, Microsoft Corporation Redmond, United States) and analyzed with SPSS Version 22.0 (PASW/SPSS IBM Corporation, New York, NY, USA). For statistical analysis Mann-Whitney-U test and spearman's rho correlation analysis for continuous variables and Pearson-Chi2-test for discrete variables were used.

Results with a p-value ≤ 0.05 were interpreted as statistically significant. A p-value ≤ 0.1 but > 0.05 was interpreted as statistical trend.

Results

Data of ten patients with CSOP were evaluated. The mean age was 51.7 years ± 15.8 SD, ranging form 19 to 69 years. Three female and seven male patients were included. In three cases the right side and in seven cases the left side was affected.

Comparing the dental data to preoperative amounts of deviation in primary position (using Harms tangent screen) and stereopsis (using a random dot test) three dental parameters showed significant correlation or at least a trend with orthoptic parameters (**Tables 1** and **2**).

The value of 6 + 6 ant, indicating which upper first molar is more anteriorly measured in the sagittal plane (**Fig. 2a**), showed a statistical trend to correlate with the vertical deviation measured in primary position (rho = 0.594; p = 0.07) (**Fig. 2b**). The molars of the paretic side were thus located more anteriorly.

Statistically significant correlation was found between overbite (measured from the mesial part of tooth 11) (**Fig. 3a**) and the horizontal deviation measured in primary position (rho = 0.768; p = 0.016) (**Fig. 3b**).

The value of ML UJ/F, indicating the deviation of the center of the upper jaw (midline between the teeth 11 and 21) and the face (center of the philtrum) (**Fig. 4a**) in the transversal plane showed a statistical trend to correlate with the vertical deviation measured in primary position (rho = 0.623; p = 0.054) (**Fig. 4b**).

Of the ten CSOP patients seven showed visible head tilt. In the group with the left eye affected (n = 7), four showed a head tilt to the contralateral side. Three patients showed no visible head tilt.

In the group with the right eye affected (n = 3), two patients showed a head tilt to the contralateral side. One patient showed a paradoxical head tilt to the ipsilateral side.

Drawing the interpupillar line (connecting the two pupils) and the occlusal line (a line drawn along the wooden stick the pa-

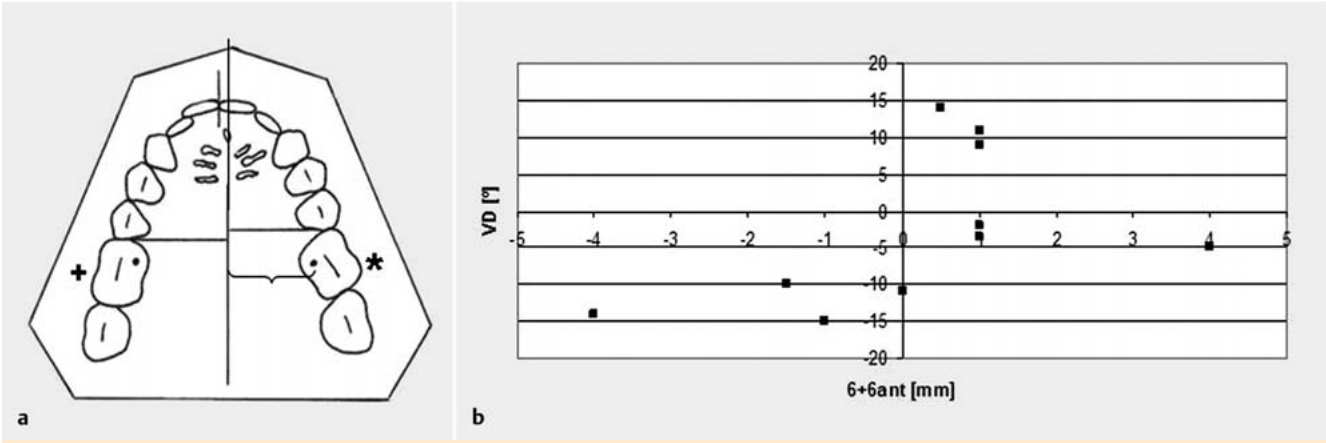

Fig. 2 **a** Schematic drawing showing the teeth position in the upper jaw and indicating the origin of "6 + 6 ant" as the sagittal position of the first upper molar. In this example the tooth 26 (*), which is the sixth tooth of the left half of the upper jaw, is located more anteriorly than tooth 16 (+), the sixth tooth of the right half of the upper jaw. **b** Correlation between 6 + 6 ant [mm] and vertical deviation (VD) [°]. All values were adapted for right sided superior oblique palsy. Negative values on the x-axis represent a more anterior location of the left molar compared to the right molar tooth.

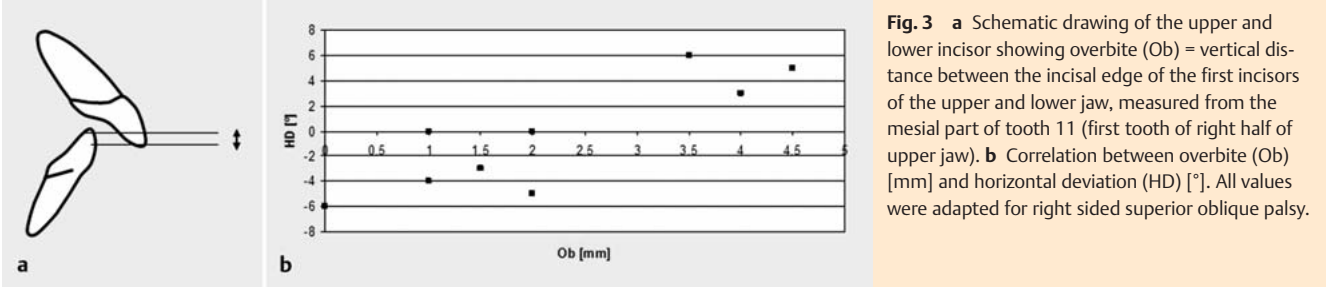

Fig. 3 **a** Schematic drawing of the upper and lower incisor showing overbite (Ob) = vertical distance between the incisal edge of the first incisors of the upper and lower jaw, measured from the mesial part of tooth 11 (first tooth of right half of upper jaw). **b** Correlation between overbite (Ob) [mm] and horizontal deviation (HD) [°]. All values were adapted for right sided superior oblique palsy.

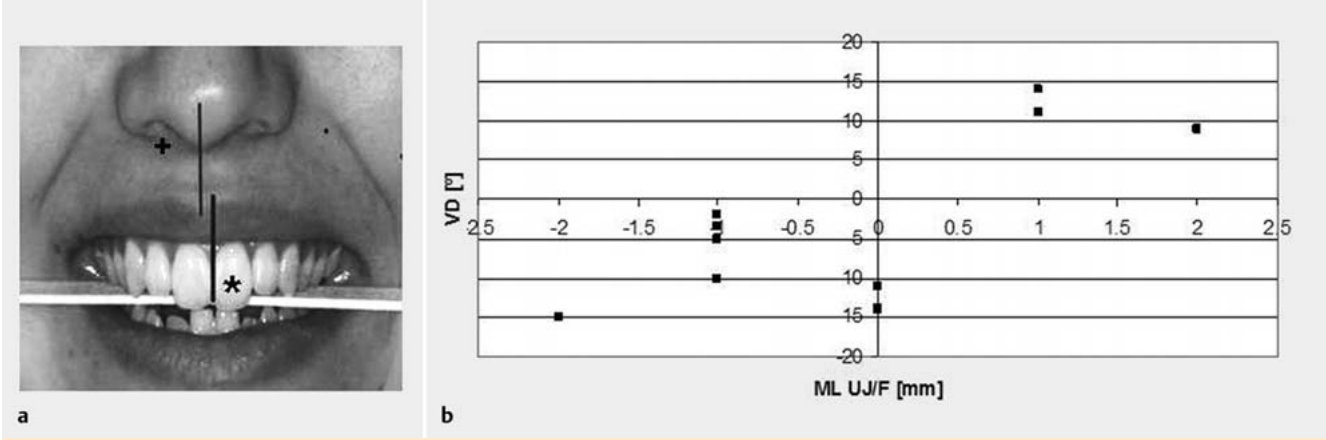

Fig. 4 **a** Photograph of upper and lower teeth and the nose marking the distance between the midline (ML) of the face (F) (line through the middle of the nose and the philtrum) and the midline between the two upper central incisors in the transversal plane (UJ), marked as ML UJ/F. The midline of the upper incisors (*) as well as the midline of the face (+) are marked. **b** Correlation between deviation of midline of upper jaw and midline of face (ML UJ/F) [mm] and vertical deviation (VD) [°]. All values were adapted for right sided superior oblique palsy. Negative values on the x-axis represent a leftward position of the upper jaw compared to the philtrum.

tients were biting on) on the photos revealed eight subjects with an angle of ≥ 1° between these two lines, thus indicating different degrees of facial asymmetry.

Discussion

The purpose of this study was to assess the presence of dental and jaw deformities due to longstanding head tilt in patients with CSOP. In our descriptive pilot study we determined three dental parameters (ML UJ/F, 6 + 6 ant and overbite) that correlated either significantly or showed a statistical trend to correlate

with orthoptic parameters such as vertical and horizontal deviation in primary position.

A small midline deviation in the direction of the head tilt might be caused due to reduced growth of the jaw of the non-paretic side. The smaller jaw on the side of the head tilt might cause an anterior displacement of the upper first molar (6 + 6ant) on the paretic side (opposite to the head tilt) with a respective facial midline deviation to the contralateral side. In this study six patients with an asymmetric position of the upper first molar had a more anterior positioning of the ipsilateral molar. In three cases the contralateral molar was more anterior.

Interestingly, eight patients showed a facial midline deviation, all to the paretic side. This may be explained by growth changes of the soft tissues. The philtrum is displaced contralaterally with respect to the incisal papilla, explaining a deviation of the midline to the paretic side. In these ten patients three had missing teeth. One patient had missing both first molars in the lower jaw. The other two patients had missing second molars (one also a first molar) on the ipsilateral side, which did not influence the facial midline position

Another explanation for our findings might be a slight mandibular rotation due to reduced growth of the non-paretic side. Further radiological investigation might be helpful to diagnose possible respective mandibular changes.

Facial asymmetry in patients with CSOP has previously been discussed in the literature [1–4, 7, 9]. In the present study seven patients showed an obvious head tilt. Six patients showed a head tilt to the contralateral side. In one case a paradoxical ipsilateral head tilt was found. Paysee et al found one patient out of 21 unilateral CSOP patients with a head tilt to the ipsilateral side [2].

Comparing the interpupillar and the occlusal line eight patients with converging lines were found (80%). In healthy subjects the lines are parallel, as opposed to patients with facial asymmetry in whom the lines converge and intersect at one point. 80% with facial asymmetry is much higher than in other studies [4, 8].

Further studies are needed to explore whether in subjects showing these orthodontic parameters CSOP occurs more frequently than in patients with good dental occlusion.

In Switzerland orthodontic treatment is very popular. 23% of the population undergo or underwent orthodontic therapy at some point in life. Particularly in the younger population group of 15–24 years of age orthodontic therapy is performed very frequently (56%) [11]. Thus orthodontists see more than every second child. If a specific combination of orthodontic symptoms could be verified at an early stage of development as being characteristic for CSOP, earlier attention to this disorder may be achieved by the help of the orthodontist and an adequate therapy might be initiated.

Conflict of Interest

None declared. The authors declare that they have no competing financial or proprietary interests and no funding sources or sponsors to declare. **Contributor ship statement:** All authors were substantially involved in conception and design of this study, as well as in data/sample acquisition or data/sample analysis. All authors contributed in drafting or revising the manuscript and approved the final version.

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