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NEURO-OPHTHALMOLOGY

Kestenbaum procedure on the vertical rectus muscles with simultaneous compensation of the induced cyclodeviation for nystagmus patients with chin-up or chin-down head posture

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Abstract
Background Kestenbaum surgery is performed for nystagmus-related abnormal head posture, and symmetrically changes the position of both eyes to shift the null point to the primary position. Most patients with infantile nystagmus have their null point zone in a lateral gaze position. Less frequently, surgery can be performed to reduce chin-up or chin-down head posture. We report indications for, and the results of eight consecutive interventions performed according to the Kestenbaum principle for the reduction of a chin-up or chin-down head posture.
Methods In a retrospective study, the clinical findings for eight patients who consecutively underwent treatment in the University Eye Hospital of Cologne between 2001 and 2007 were investigated. The patients were aged 6 to 16 years; median age was 6.5 years. For all patients, surgery was to correct a chin-up or chin-down head posture due to infantile nystagmus. Preoperatively, five patients showed a chin-down, three a chin-up head posture. All vertical rectus muscles were recessed or tucked between 6 and 7 mm; the resulting cyclodeviation was reduced by an intervention on the superior oblique muscles (6 to 8 mm tucking, in the case of chin-down, or recession in the case of chin-up head posture).
Results Surgery was successful in seven of the eight patients, with a reduction of the vertical head posture to less than 10°. In the cases of chin-down posture, head posture was reduced to between 0° and a maximum of 20° in one case postoperatively (before the operation 20° to 35°); in the cases of chin-up posture, to less than 8° (before the operation 25° to 35°). One case showed no postoperative improvement in chin-down posture but a head turn to the left of up to 20°; another case had a remaining chin-up posture of 8° with a right turn of 15°. Binocular vision was better or the same in all cases after surgery.
Conclusion For nystagmus patients with chin-up or chin-down head posture, surgery for bilateral parallel shifting of the eyes can considerably improve the head posture. It is possible to compensate the induced cyclodeviation at the same time by bilateral surgery on the superior oblique muscles.

Keywords Kestenbaum procedure · Chin-up head posture · Chin-down head posture · Nystagmus · Vertical musculi recti

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Introduction

In 1953, Kestenbaum [7] and Anderson [1] independently developed a surgical procedure for the treatment of nystagmus patients with abnormal head posture. Surgery for nystagmus dampening according to the Kestenbaum principle changes the position of both eyes in the orbits to shift the null point zone to the primary position.

Therefore, after successful surgery the innervation required for lowest frequency and/or amplitude of nystagmus is found in the primary position. Most patients with infantile nystagmus have their null point in the horizontal gaze position. Kestenbaum procedures are therefore mainly performed on the horizontal recti to reduce head turns to the right or left. Less frequently, surgery following the Kestenbaum principle is done to reduce chin-up or chin-down head posture. In these cases, surgeons must be aware of the cyclodeviation induced by the parallel shifting of the gaze position by the vertical rectus muscles, and be aware that this may require compensation by interventions on the oblique muscles [16].

There are few reports on interventions for chin-up or chin-down head postures following the Kestenbaum approach. Furthermore, only a few indications are given in the literature on the management of the cyclotropia which may be induced [2, 3, 11, 13, 16, 17].

We therefore report our experience with indications and the results of eight consecutive interventions according to the Kestenbaum principle for the reduction of a chin-up or chin-down head posture.

Methods

In a retrospective study, the clinical findings, before and after surgery, of eight patients were investigated who consecutively underwent surgery according to the Kestenbaum principle at the University Eye Hospital of Cologne between 2001 and 2007. The median postoperative follow up was 2 months (0.5–8 months).

In all patients, seven males and one female, the head posture was due to infantile nystagmus. The diagnosis was idiopathic nystagmus in four cases and sensory nystagmus in four cases; the etiology of the sensory nystagmus was one case each of ocular albinism and macula hypoplasia and two cases of rod–cone dystrophy. The median age was 6.5 years (6–16 years). Before the operation, five patients showed a chin-down, three a chin-up head posture. For further details see Table 1, where the patients' preoperative characteristics are summarized, including age at surgery, diagnosis, nystagmus direction and type, binocular visual acuity, binocular function and head posture. None of the patients showed an alphabet incomitance in the eye motility test or heterotropia in the cover test before the operation. The eye motility and cover tests were performed with the head adjusted to a straight position pre- and postoperatively. Additionally, the Hirschberg Test and tests for binocular function (Bagolini striated glasses, Lang I or II Stereo Test, Titmus Stereo Test) were performed in the head posture adopted. The best-corrected monocular and binocular visual acuity was examined pre- and postoperatively at a distance of 5 metres and reading distance (0.3 metres) in head posture. Head posture was measured with a goniometer pre- and postoperatively, with the patient fixing the smallest possible age-appropriate target at a distance, under binocular and monocular conditions (numbers/text for children who were already able to read, tumbling “E” Test for the smaller ones). As the values for monocularly tested head posture were very similar to those for binocularly tested posture, only binocularly tested posture was considered in this study. Preoperatively, homonymous based prisms were used to assess the possible benefit from surgery. The prism base was therefore put in the direction of the head posture, opposite to the direction of gaze in which nystagmus dampening occurred. For example, a patient with chin-down posture would be given prisms with base down to correct the head posture.

Surgery was performed under general anaesthesia. Based on the Kestenbaum principle, all four vertical rectus muscles were recessed or tucked; the expected cyclodeviation was reduced by a simultaneous intervention on the superior oblique muscle tendons.

Surgery for chin-down posture

In case of surgery for chin-down posture, the anterior margin of the superior oblique muscle was tucked 6 to 7 mm first, before surgery on the superior rectus muscle was performed. Then, the superior rectus muscle was recessed 6 to 6.5 mm, and fixated again to the sclera anterior to the tendon of the superior oblique muscle. Thus, the superior oblique tendon remained between rectus and

Table 1 Patients´preoperative characteristics

Case	Age (years)	Diagnosis	Nystagmus direction, type	Binocular visual acuity	Binocular function	Head posture
1	14	ocular albinism	horizontal, pendular jerk	0.5	Lang +, Titmus fly +, rings 1–2 +, animals A–B +	30°° chin-down
2	6	rod-cone dystrophy	horizontal, pendular	0.1	Lang -, Titmus animals A–C +, rings 1–2 +	25° chin-down
3	16	idiopathic	horizontal, pendular	0.4	Lang +, Titmus fly +, rings 1–4 +, animals A–C +	20° chin-down
4	6	idiopathic	horizontal, pendular jerk	0.32	Lang -, Titmus fly +, rings 1–2 +	35° chin-down
5	6	idiopathic	horizontal, pendular jerk	0.4	Lang -, Titmus fly-	35° chin-up
6	7	macula hypoplasia	horizontal, jerk	0.32	Lang +, Titmus fly+, animal A +, rings 1–4 +	25° chin-down, 20° L turn
7	8	rod-cone-dystrophy	horizontal, jerk	0.5	Lang +	30° chin-up, 10° R tilt
8	6	idiopathic	horizontal, jerk	0.63	Lang +	30° chin-up,30° R turn

sclera and not above the rectus muscle after surgery. In a final step, the inferior rectus muscle was tucked 6 to 6.5 mm. Then, the same procedure was performed on the second eye.

In the first case (case 1) the inferior rectus muscles were tucked by 6 mm; the superior rectus muscles were recessed by 12 mm. The surgeons expected to induce an excyclorotation by weakening the superior rectus, which also is an incyclorotator, and attempted to compensate the apprehended cyclodeviation by recessing and transposing the insertion of the superior rectus muscles by 6 mm temporally. However, an additional intervention on the superior oblique muscles was necessary because of the subjective perception of tilted images, and because of objective excycloduction in both eyes, when measured with the Maddox rod and at the tangent screen of Harms under Maddox dark red glass. Another patient (case 7) had had previous Kestenbaum surgery to correct a horizontal head turn 1 year earlier. Because of this time interval, and of the patient´s young age, and because in both interventions a recess-tuck technique was performed, an anterior segment ischemia as a follow-up to surgery on more than two recti felt to be unlikely.

Surgery for chin-up posture

In the case of surgery for chin-up posture, the inferior rectus muscle was recessed 6 to 7 mm first. Second, the superior oblique muscle tendon was fully recessed 6 to 8 mm. The superior rectus muscle was then tucked 6 to 7 mm. Both eyes were operated in the same manner.

For further details on surgery see Table 2; further details on the surgical technique are given by Rüssmann et al. [14].

Results

Surgery was successfull in seven of eight patients enabling a reduction of the vertical head posture to less than 10°.

Surgery for chin-down posture

Preoperatively, chin-down posture ranged from 20° to 35°, and was reduced after the operation to between 0° and a maximum of 20° in one case. The median individual effect on chin-down head posture was 20° (5°–30°). Details are given in Fig. 1.

One patient (case 5) showed no postoperative improvement in chin-down posture but a head turn to the left of up to 20°.

In case 1, an additional intervention on the superior oblique muscles was necessary after 3 days, because the cyclodeviation that was measured as an excyclodeviation of 19° did not decrease spontaneously (Table 2).

Surgery for chin-up posture

In the three cases of chin-up posture, the amount of abnormal vertical head posture ranged from 25° to 35°, and was reduced to less than 8°. The median individual effect on chin-up head posture was 30° (22°–35°). Details are given in Fig. 1.

Table 2 Details of surgery and results

Preop head posture	Bilateral surgery	Previous surgery	Further surgery	Postop head posture
30° chin-down	recess SR 12 mm, transposition of insertion to temporal 6 mm; tuck IR 6 mm	-	bilateral tuck SO anterior 8 mm	5° L turn
25° chin-down	tuck SO anterior 7 mm, recess SR 6 mm, tuck IR 6 mm	-	-	20° chin-down
20° chin-down	tuck SO anterior 6 mm, recess SR 6 mm, tuck IR 6 mm	-	-	none
35° chin-down	tuck SO anterior 7 mm, recess SR 6 mm, tuck IR 6 mm	-	-	10° chin-down
35° chin-up	recess SO 6.5 mm, recess IR 6 mm, tuck SR 7 mm	-	-	20° L turn
25° chin-down, 20° L turn	tuck SO anterior 7 mm, recess SR 6.5 mm, tuck IR 6.5 mm	-	-	10° chin-down, trace L turn
30° chin-up, 10° R tilt	recess SO 8 mm, recess IR 7 mm, tuck SR 7 mm	OD: recess LR, tuck MR; OS: recess MR, tuck LR, 6 mm each	-	trace R tilt
30° chin-up, 30° R turn	recess SO 6 mm, recess IR 6 mm, tuck SR 6 mm	-	-	8° chin-up, 15° R turn

SR = superior rectus muscle, IR = inferior rectus muscle, MR = medial rectus muscle, LR = lateral rectus muscle, SO = superior oblique muscle, IO = inferior oblique muscle

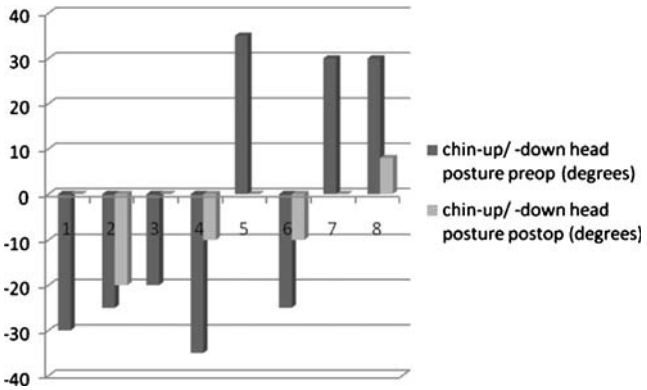

Fig. 1 Preoperative (*dark*) and postoperative (*light*) chin-up and -down head posture of the eight patients in degrees: chin-up posture is shown in positive values, chin-down posture in negative values

One patient showed a remaining chin-up posture of 8° with a 15° right turn.

None of our patients showed significant up or down gaze deficits after the operation, nor did they have eyelid malpositions such as lower eye lid retraction which may occur after inferior rectus recession, or upper eye lid retraction which may appear after superior rectus recession. All patients were orthotropic postoperatively in the primary position, except in the case described (case 1). After the second intervention, the patient showed a hyperphoria of 3 prism dpt.

Discussion

Nystagmus patients often adopt an abnormal head posture to benefit from the null point position in which the amplitude and/or frequency of the nystagmus are most compatible with high visual acuity. Usually, patients with infantile nystagmus have their null point in a horizontal gaze position, and therefore adopt a head turn to the right or to the left. In 1953, Kestenbaum [7] and Anderson [1] were the first to describe a new procedure for surgery on nystagmus patients, that is, the principle of bilateral parallel shifting of the eyes with the aim of reducing an abnormal head posture. Since 1953, dosage on the horizontal rectus muscles has frequently been modified, and literature has dealt extensively with nystagmus surgery for reduction of a horizontal face turn. Kestenbaum [7] was the first to advocate a combined recess–resect procedure on all four horizontal rectus muscles. Recession or resection of the two horizontal muscles by a total of 4 to 10 mm on each eye was recommended, to the same extent in millimetres as the corneal limbus moves when the head is turned from its abnormal posture to primary position—a dosimetric model which neglects the elastic forces of the orbital tissues and therefore did not yield sufficient effect. Parks [8, 10] subsequently suggested a modified procedure for all four horizontal muscles. For a horizontal abnormal head posture of about 20° he performed a total of 26 mm of surgery, with a distribution of 5-6-7-8 mm for the rectus muscles. Pratt-Johnson [12] already in 1991 reported results of surgery to modify the null-zone position in congenital nystagmus by relatively high dosage. Initially, patients with a horizontal face turn of about 35° were treated with 5 mm recess-resect procedures on both eyes, but as this had only limited success, the amount of surgery was increased to 10 mm on each horizontal rectus muscle. Nelson et al. [9] reported in 1984 on surgical management for abnormal head position in nystagmus by an augmented Kestenbaum dosage. Because of the high rates of recurrence and undercorrection, the dosages suggested by Parks were augmented by either 40% for patients with head turn of 30°, resulting in surgery of 7 mm, 8.4 mm, 9.8 mm, and 11.2 mm on the rectus muscles, or 60% for patients with head turn of 45°, which results in surgery of 8 mm, 9.6 mm, 11.2 mm and 12.8 mm. Recently, larger studies, for example by Gräf et al. [4, 5] confirmed the need for higher dosages for the correction of an abnormal horizontal head posture. Gräf et al. concluded that surgery in mm on each eye should be two-thirds of the horizontal head turn in degrees, i.e. 10 mm of surgery on each horizontal muscle for a head-turn of 30°.

Less frequently, nystagmus patients have their null point zone in an up or down gaze position, and adopt a chin-down or chin-up head posture. Fewer reports exist regarding a surgical approach in these cases. Similar to recommendations for horizontal head posture, recent studies dealing with vertical head posture advise a need for high dosages of surgery of up to 10 mm on each vertical rectus muscle for head postures up to 30°.

In 1973, Parks [10] recommended bilateral recessions of the superior or inferior rectus muscles for a chin-up or chin-down posture of less than 25°, and surgery on all four vertical rectus muscles for postures of more than 25°. He himself started with 4 mm recessions and 4 mm resections, but subsequently recommended combined 8 mm recessions and 8 mm resections due to residual or recurrent abnormal head postures in his patients.

A survey of management strategies for nystagmus patients with vertical or torsional head posture, conducted by Sigal and Diamond among AAPOS members in 1990 [15], confirmed that the vertical head posture associated with congenital nystagmus is relatively infrequently dealt with. The participating surgeons used two main surgical procedures: either bilateral average vertical rectus recession of 4.8 mm, 5.9 mm, and 7.3 mm for mild (10°), moderate (20°) and severe (30°) head posture respectively, or, as a more common procedure, a bilateral vertical rectus recess-resect procedure with average measurements for recession

and resection of 4.5 and 4.3 mm for mild, 5.3 and 5.3 mm for moderate, and 7.7 and 6.4 mm for severe head postures. Success rates were not given in this survey.

Parks' findings for the need of higher measurements for total surgery were confirmed by reports from Roberts et al. [13] in 1996, who suggested the need for a very large surgical correction for patients with a chin-up head posture of more than 30°. Bilateral inferior rectus muscle recessions of at least 8 mm, combined with superior rectus muscle resections of at least 8 mm, were suggested as a primary procedure, due to the fact that their patients (four cases) with chin-up head posture manifested some degree of residual posture after the operation. In the case of a chin-down posture, Roberts et al. preferred weakening the two elevators to a large recess–resect procedure. None of the three patients treated showed postoperative complications such as upper eyelid retraction, superior oblique muscle overaction or an A-Pattern, but all of them had up-gaze deficits.

In a newer review from 2004, of 20 patients, Yang et al. [17] concluded that in vertical head postures, combined recess–resect and recession followed by resection procedures were more predictable and successful than recession alone. Furthermore, no late postoperative changes in vertical abnormal head posture were observed when all four vertical rectus muscles had been operated. Accordingly, combined recess-resect procedures of all four vertical rectus muscles with a total recession and resection of at least 12 mm per eye were preferred, as this procedure gave good results. In their surgical guidelines, Yang et al. recommend vertical recess-resect surgery of 12, 16, and 20 mm on each eye for vertical abnormal head posture of 10°–15°, 20°–25°, and 30° or more respectively.

Gaze deficits occurred in three patients. Furthermore, two patients developed a transient retraction of the upper eye lids after maximal inferior rectus muscle resection and superior rectus muscle recession. Although large corrections on the vertical rectus muscles are expected to result in torsion problems, only two patients reported symptomatic torsion that needed to be corrected by additional surgery on the oblique muscles. Neither Yang et al. nor Parks described lower eyelid retraction after inferior rectus muscle recession.

Due to the secondary functions of the vertical rectus muscles (the superior rectus muscle is also an incyclorotator, and the inferior rectus muscle an excyclorotator [6]), an additional cyclotorsional effect can be expected after recess-resect surgery on the vertical eye muscles. In 1991, Stapper and Rüssmann [16] reported significant excyclodeviation in two of three patients after pure vertical rectus muscle surgery for chin-down head posture. Excyclodeviation had to be reduced by bilateral inferior oblique muscle recession. Personal communications from other strabismus surgeons were reported which suggest that with Kestenbaum surgery of more than 10 mm combined recess-resect procedure on the vertical muscles, the expected cyclotorsional effect must be compensated for by a simultaneous intervention on the superior or inferior oblique muscles.

The superior oblique and superior rectus muscles act in a synergistic way for cyclodeviation, but antagonistically for vertical movement. Thus, after compensation of the cyclodeviation, the surgery on the oblique muscles has an additional positive effect on the head posture and increases the Kestenbaum effect. This might explain why good reduction of the head posture was reached with less surgery than reported in previous studies given on vertical and horizontal procedures [4, 5, 17]. So simultaneous procedures on the oblique muscles not only compensate for cyclodeviation, but the intended effect on head posture is also increased so that dosage of surgery on the vertical recti might be kept relatively low to prevent adverse side-effects.

However, in the chin-down cases, where only the anterior margin of the superior oblique tendon was tucked, it is questionable whether a strong additional vertical effect can be achieved by this procedure. Possibly, part of the potential excyclodeviation caused by the described method of vertical rectus surgery for chin-down posture may also be induced by displacement of the superior oblique tendon from its original to a more sagittal path, thus changing the ratio between the incyclo and the depressive torque towards the latter. The nasal edge of the new rectus insertion might act like a hypomochlion between the trochlea and the insertion of the superior oblique tendon. Tucking of the superior oblique tendon thus would compensate for this loss of cyclo torque and increase the depressive effect of surgery.

Although surgeons are often wary of complications such as upper or lower eyelid retraction, anterior segment ischemia and gaze deficiencies, none of these occured in our patients. Our patients did not show any lid abnormalities, and there was no significant limitation in vertical motility. Yang et al. [17], who recommended doses of 16–20 mm on each eye, reported upper lid retraction in two cases. Upper lid retractions are thought to be caused by increased innervation due to the increased effort required to elevate the eye after surgery. To avoid induction of cyclotorsion, Pierse [11] combined bilateral inferior rectus recession and superior oblique tenectomy or recession respectively, to reduce chin-up head posture in two patients. Roberts et al. [13] successfully used superior rectus recession combined with inferior oblique muscle anterior transposition in cases with chin-down posture. Yang et al. [17] described two patients who reported symptomatic torsion after surgery for abnormal vertical head posture. The cyclodeviation had to be corrected with additional surgery of the oblique muscles in both cases.

In summary, for patients with chin-up or chin-down head posture due to idiopathic motor as well as sensory nystagmus, surgery according to the principle of bilateral par-

allel shifting of the eyes in the orbit can considerably improve the head posture. It is possible to compensate the induced cyclodeviation at the same time by bilateral surgery on the superior oblique muscles. For head postures of 20° to 30° we recommend a maximum surgery of 14 mm on the vertical rectus muscles per eye, and 6 to 8 mm of surgery on each of the two superior oblique muscles.

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