Strabismus Surgery for Abnormal Head Position: 7-Year Experience at a Tertiary Care Center

Cirurgia de Estrabismo para Corrigir Posição Anómala Cabeça: 7 Anos de Experiência num Hospital Terciário

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Recebido/Received: 2021-12-05 | **Aceite/Accepted:** 2022-06-05 | **Publicado/Published:** 2022-06-30

DOI: https://doi.org/10.48560/rspo.25975

ABSTRACT

INTRODUCTION: Traditionally, the goal of strabismus treatment has been to realign the visual axes in order to eliminate diplopia, to produce, maintain, or restore binocular vision or for aesthetic reasons. Nowadays, surgery to improve an abnormal head position (AHP) is also a well-accepted indication for surgery. AHP or torticollis is a frequent encountered sign in routine ophthalmology practice, especially in pediatric patients. The present study aims to characterize the patients of a tertiary hospital who were submitted to surgery for an AHP caused by an ocular condition, as well as the results obtained by the treatment provided to them.

MATERIAL AND METHODS: The medical records of patients who were admitted to surgery for an AHP in the Ophthalmology department of Centro Hospitalar Universitário de São João between July 2014 and July 2021 were retrospectively studied. The patients underwent ophthalmologic examinations pre and post-operatively. Characterization of the AHP was subjectively evaluated.

RESULTS: In total, 24 patients were studied, 8 (33.3%) of whom were female and 16 (66.6%) were male. Mean age of patients was 13.7 years (range 3-61 years). There were 7 patients with superior oblique palsy, 7 patients with Duane syndrome (type 1), 6 patients with nystagmus, 3 patients with third nerve paralysis and 1 patient with Brown syndrome. Face turn was the most frequent AHP (14 patients), followed by head tilt AHP (5 patients). Four patients had mixed AHP (head tilt and face turn) and 1 patient presented with chin up. A total of 95.8% of patients had their AHP improved with surgical treatment. Nineteen patients were submitted to one surgery and a second surgery was necessary in 5 patients.

CONCLUSION: Overall, almost all patients improved after surgery. Nevertheless, it is important to highlight that occasionally it is necessary more than one surgery to achieve the best result.

KEYWORDS: Duane Retraction Syndrome; Head; Nystagmus, Congenital; Ocular Motility Disorders; Posture; Strabismus/surgery; Torticollis.

RESUMO

INTRODUÇÃO: Tradicionalmente, a cirurgia de estrabismo é realizada com o objetivo de realinhar os eixos visuais de forma a eliminar a diplopia, melhorar a visão binocular ou melhorar a apresentação estética. Atualmente, a cirurgia pode ser também realizada para melhorar a posição anómala da cabeça (PAC). A PAC é um sinal frequentemente encontrado na prática clínica em Oftalmologia, especialmente em crianças. O presente estudo tem como objetivo caracterizar o grupo de doentes de um hospital terciário que foram submetidos a cirurgia por PAC causada por uma condição oftalmológica, bem como apresentar os resultados cirúrgicos encontrados.

MATERIAL E MÉTODOS: Foram retrospetivamente estudados os dados clínicos dos doentes que foram submetidos a cirurgia para corrigir a PAC no departamento de Oftalmologia do Centro Hospitalar Universitário de São João entre Julho 2014 a Julho 2021. Foi realizado exame oftalmológico antes e após a cirurgia. A caracterização da PAC foi feita de forma subjetiva.

RESULTADOS: No total foram estudados 24 doentes – 8 (33,3%) do sexo feminino e 16 (66,6%) do sexo masculino. A idade média foi de 13,7 anos (entre 3-61 anos). Foram identificados 7 doentes com paralisia do obliquo superior, 7 doentes com síndrome de Duane (tipo 1), 6 doentes com nistagmo, 3 doentes com paralisia do terceiro par craniano e 1 doente com síndrome de Brown. Cabeça rodada foi a PAC mais frequente (14 doentes), seguido de cabeça inclinada (5 doentes). Quatro doentes apresentaram PAC mista (cabeça rodada e inclinada) e 1 doente apresentou PAC com mento elevado. Do total dos doentes, 95,8% melhoraram a sua PAC com o tratamento cirúrgico. Dezanove doentes foram submetidos a uma cirurgia e uma segunda cirurgia foi necessária em 5 doentes.

CONCLUSÃO: No geral, quase todos os doentes melhoraram a sua PAC com o tratamento cirúrgico. No entanto, é necessário salientar que por vezes é necessário mais do que uma cirurgia para obter o melhor resultado cirúrgico.

PALAVRAS-CHAVE: Cabeça; Estrabismo/cirurgia; Nistagmo Congénito; Perturbações da Motilidade Ocular; Postura; Síndrome da Retração Ocular; Torcicolo.

INTRODUCTION

Traditionally, the goal of strabismus treatment has been to realign the visual axes to eliminate diplopia or to produce, maintain, or restore binocular vision.[1] Additionally, surgeries to improve an abnormal head position (AHP), eliminate abnormal eye movements or restore the standard anatomical position of the eyes are well-accepted indications for surgery.[1]

AHP or torticollis is a frequently encountered sign in routine ophthalmology practice, especially in pediatric patients. The cause of the AHP can be orthopedic, ocular, neurologic, or postural.[2] The majority of children who present with torticollis during the first year of life have congenital muscular torticollis secondary to unilateral fibrosis of the sternocleidomastoid muscle.[2] Numerous ocular conditions can cause AHP or "ocular torticollis". Among them: superior oblique muscle palsy, lateral rectus muscle palsy, nystagmus, vertically incomitant horizontal strabismus (A or V patterns), Brown's syndrome, Duane's syndrome, refractive errors and dissociated vertical deviation.[3] Superior oblique palsy (SOP) is the single most common cause of ocular torticollis.[4]

The presence of AHP during sleep or despite closing one eye is the most important finding used to detect non-ocular origins of torticollis. If the AHP has an ocular basis, there is no preference to sleep on one side, and on the other hand, the head returns to its natural position upon closing one eye.[5]

The AHP is adopted to improve visual acuity, maintain binocular single vision or center residual visual field with the body. Face turn is the most frequent AHP.[6]

The consequences of the AHP are diverse. For example, AHP can cause neck pain, self-esteem issues or facial asymmetry. Facial asymmetry is more frequent in AHP due to cyclovertical muscle palsy, including midface hypoplasia, nasal tip or septum deviation.[7]

The present study aims to characterize the patients in a tertiary hospital who were submitted to surgery because of an AHP caused by an ocular condition.

MATERIAL AND METHODS

The records of twenty-four patients submitted to surgery because of an AHP caused by an ocular condition from July 2014 through July 2021 were reviewed in a computerized patient database. The main goal of surgeries was to improve the AHP.

Patients submitted to strabismus surgery before July 2014 and patients with AHP submitted to surgery primarily because of diplopia were excluded from the study.

Age, sex, ocular diagnosis, type of AHP, age at surgery and duration of follow-up were recorded.

AHP was assessed when the patient was fixating an object in the distance.

The patients underwent complete ophthalmologic examination pre-operatively, including clinical assessments of visual acuity (with cycloplegic refraction if appropriate), nystagmus assessment, and evaluation of the oculomotor equilibrium (Hirschberg test, Cover test and extraocular movements). Best-corrected visual acuity was measured with patients' heads in the preferred postures.

The characterization of the AHP after surgery was subjectively evaluated (0 - worse, 1 - equal, 2 - better, 3 - much better, 4 - resolved). Surgical success was defined as total resolution of AHP (result 4) and partial surgical success as partial resolution of AHP (result 2 or 3).

The authors have followed the protocols of their work center on the publication of data. The study was conducted in accordance with the Helsinki Declaration.

RESULTS

Twenty-four patients were studied, eight (33.3%) female and sixteen (66.6%) male. The mean age of patients was 13.7 years (range 3-61 years).

Seven patients with superior oblique palsy, seven patients with Duane syndrome (type 1), six patients with nystagmus, three patients with third nerve paralysis and one patient with Brown syndrome were evaluated.

Face turn was the most frequent AHP (fourteen patients), followed by head tilt (five patients). Four patients had mixed AHP (head tilt and face turn) and one patient presented with chin up.

A total of 95.8% of patients had their AHP improved ("better", "much better" or "resolved").

SUPERIOR OBLIQUE PALSY

The clinical characteristics of patients with AHP who were diagnosed with superior oblique palsy are shown in Table 1. The mean age was 10.6 years (range 3-32 years).

Anterior transposition of the ipsilateral inferior oblique was made in 71.4% of patients. A sequential surgery was necessary for patient no. 1 (Fells' modified Harada-Ito procedure), no. 2 (Right medial rectus Faden operation for ipsilateral esotropia) and no. 4 (Fells' modified Harada-Ito procedure). After the first surgery, the AHP evaluation was "equal" for patient no. 1, "better" for patient no. 2 and "equal" for patient no. 4.

They all continue being followed in our department, except for patients no. 4 and no. 6.

DUANE SYNDROME

The clinical characteristics of patients with AHP who were diagnosed with Duane syndrome are shown in Table 2.

All patients were diagnosed with type 1 Duane syndrome, with left eye affected and they all presented with the same AHP - head turn to the left. The mean age was 8.71 years (range 4-13 years).

Asymmetric bilateral medial rectus recession was made in all cases. There was no additional surgery in all patients and five of them continue being followed in our department (Patients no. 8, 10, 11, 12 and 14).

NYSTAGMUS

The clinical characteristics of patients with AHP who were diagnosed with nystagmus are shown in Table 3.

The mean age was 10.8 years (range 3-34 years). Five cases were congenital and one was acquired at six months of age due to meningitis. In all cases, there was no additional surgery and they all continue being followed in our department.

In patient no. 15, right lateral rectus resection of 8 mm with infraplacement of 3 mm, right medial rectus recession of 5 mm with supraplacement of 3 mm, left medial rectus resection of 6 mm with infraplacement of 3 mm and left lateral rectus recession of 7 mm with supraplacement of 3 mm was executed to improve torsional deviation. In patient no. 17, right inferior oblique transposition was associated with the Kestenbaum procedure because inferior oblique muscle overaction was objectified.

Table 1. Patients with AHP and superior oblique palsy.

Patient	Sex	Age, Y	Eye	Etiology	Type of AHP	Follow-up pre-op, Y	Surgery	Post-op AHP	BVCA
1	M	4	OS	Congenital	Head tilt to the right	1	1) IOAT + 2) Harada-Ito	4	OD 1.0 OS 1.0
2	M	6	OD	Congenital	Head turn and tilt to the left	4	1) IOAT + 2) MR Faden operation	4	OD 1.0 OS 1.0
3	M	32	OS	(Decompensated) Congenital	Head tilt to the right	1	OD IR & OD SR recession with half tendon nasal transposition and slanting	4	OD 1.0 OS 1.0
4	F	3	OS	Congenital	Head tilt to the right	2	1) IOAT + 2) Harada-Ito	4	OD 1.0 OS 1.0
5	M	15	OS	Congenital	Head tilt to the right	3	IOAT	4	OD 1.0 OS 1.0
6	M	3	OS	Congenital	Head tilt to the right	1	IOAT	4	OD 0.8 OS 0.8
7	M	11	OD	Congenital	Head turn and tilt to the left	5	IO Recession	3	OD 1.0 OS 1.0

AHP - abnormal head position, BCVA - best-corrected visual acuity, F- female, IR - inferior rectus, IO – inferior oblique, IOAT- inferior oblique anterior transposition, M - male, MR- medial rectus, OD - right eye, OS - left eye, Post-op - post-operative, SR- superior rectus, Y- years

Table 2. Patients with AHP and Duane syndrome.

Patient	Sex	Age, Y	Eye	Type of AHP	Follow up pre-op, Y	Surgery	Post-op AHP	BCVA
8	F	7	OS	Head turn to the left	4	Asymmetric bilateral MR recession	4	OD 1.0 OS 1.0
9	M	4	OS	Head turn to the left	3	Asymmetric bilateral MR recession	3	OD 1.0 OS 1.0
10	F	11	OS	Head turn to the left	3	Asymmetric bilateral MR recession	4	OD 1.0 OS 0.6
11	F	6	OS	Head turn to the left	3	Asymmetric bilateral MR recession	4	OD 1.0 OS 1.0
12	M	8	OS	Head turn to the left	3	Asymmetric bilateral MR recession	3	OD 1.0 OS 1.0
13	M	13	OS	Head turn to the left	4	Asymmetric bilateral MR recession	3	NR
14	M	12	OS	Head turn to the left	1	Asymmetric bilateral MR recession	3	OD 1.0 OS 1.0

AHP - abnormal head position, BCVA - best-corrected visual acuity, F - female, M - male, MR - medial rectus, NR – no record, OD - right eye, OS - left eye, Post-op - post-operative, Y- years

Table 3. Patients with AHP and nystagmus.

Patient	Sex	Age, Y	Etiology	Type of AHP	Follow up pre-op, Y	Surgery	Post-op AHP	BCVA
15	M	8	Congenital	Head turn and tilt to the right	1	Kestenbaum (8+5+6+7) + Vertical transposition	3	0.63 OU
16	M	3	Congenital	Head turn to the left	1	Kestenbaum (8.5+7.5+6+9.5)	3	0.32 OU
17	M	7	Congenital	Head turn to the left	7	Kestenbaum (7+6+5+8) + RIO transposition	4	0.32 OU
18	F	8	Congenital	Head turn to the left	7	Kestenbaum (8.4+7.2+6+9.6)	4	OD 0.125 OS 0.2
19	F	34	Meningitis 6 months	Head turn to the left	1	Kestenbaum (8.0+6.5+5.5+9)	4	0.63 OU
20	M	5	Congenital	Head turn to the right	5	Kestenbaum (9+5.5+8.5+10)	4	0.32 OU

AHP – abnormal head position, BCVA - best-corrected visual acuity, F- female, M- male, OD - right eye, OS - left eye, OU - both eyes, Post-op - post-operative, RIO – right inferior oblique muscle, Y - years, surgical amounts (right lateral rectus + right medial rectus + left medial rectus + left lateral rectus)

THIRD NERVE PALSY

Three patients with AHP were diagnosed with third nerve palsy (Table 4). A sequential surgery was necessary for patient no. 21 (ptosis surgery) and no. 23 (lateral rectus 6 mm recession).

BROWN SYNDROME

One patient with AHP was diagnosed with Brown syndrome (Table 5).

Table 4. Patients with AHP and third nerve palsy.

Patient	Sex	Age, Y	Eye	Etiology	Type of AHP	Follow-up pre-op, Y	Surgery	Post-op AHP	BCVA
21	F	61	OS	Adquired	Chin up	3	1) OD SR Faden operation + 2) Ptosis surgery	3	OD 0.8 OS 0.8
22	F	60	OD	Adquired	Head turn to the left	1	OD LR Recession + OD MR 4mm Resection and anteriorization	4	OD 1.0 OS 1.0
23	M	5	OD	Congenital	Head tilt to the right and turn to the left	2	1) OD SR Faden operation + 2) LR recession	1	OD 1.0 OS 1.0

AHP – abnormal head position, BCVA - best-corrected visual acuity, F - female, LR – lateral rectus, M - male, MR – medial rectus, OD - right eye, OS - left eye, Post-op - post-operative, SR – superior rectus muscle

Table 5. Patient with AHP and Brown syndrome.

Patient	Sex	Age, Y	Eye	Etiology	Type of AHP	Follow-up pre-op, Y	Surgery	Post-op AHP	BCVA
24	M	3	OD	Congenital	Heal tilt to the left	1	RSO Recession + Tenotomy	3	OD 0.6 OS 0.6

AHP – abnormal head position, BCVA - best-corrected visual acuity, RSO – right superior oblique, M - male, OD - right eye, Post-op - post-operative, Y – years

DISCUSSION AND CONCLUSION

Khawam studied 158 patients with ocular torticollis.[8] 70% was caused by incomitance and 17% by nystagmus. Of the incomitance group - 39% had superior oblique palsy, 24% had Duane syndrome, 14% had lateral rectus muscle palsy, 8% had double elevator palsy, 5% had Brown syndrome and only 4 patients were considered to have AHP due to refractive errors. Comparing to our study, 75% of the AHP was caused by incomitance and 25% by nystagmus. Of the incomitance group – 39% had with superior oblique palsy, 39% had Duane syndrome (type 1), 17% had third nerve paralysis and 5% had Brown syndrome.

In the present study, several types of AHP were identified: head tilt to the right or left, head turn to the right or left, chin up and the combination head turn/tilt. Head turn was the most frequent. They were associated with superior oblique palsy, Duane syndrome, nystagmus, third nerve paralysis or Brown syndrome.

The majority of patients had <18 years, which can be justified by congenital being the most frequent etiology of the diagnosis.

Overall, the AHP improved with surgical treatment in 95.8% of patients – Only one patient (Patient no. 23) had her final AHP evolution worse than "better".

The authors think it is important to highlight that occasionally more than one surgery is necessary to achieve the best result. Indeed, more than one surgery was required in five cases (Patients no. 1, 2, 4, 21 and 23). This detail should be clearly informed and discussed before surgery.

AHP in SOP is adopted to compensate for the excyclotorsion and/or hypertropia effect. The typical AHP in this situation is a contralateral head tilt used to regain binocular vision.

In the superior oblique palsy group, the most frequent surgery was an anterior transposition of the ipsilateral inferior oblique. This technique was popularized by Elliott and Nankin[9] but based on theoretical work by Scott.[10] Scott suggested that transposing the inferior oblique anteriorly and placing it at the temporal corner of the inferior rectus muscle would increase the effect of a recession. In addition, it is theorized that the anterior position creates a vector for depression as the muscle is placed anterior to the equator.[10]

It has been shown that if the corrective surgery for SOP is delayed, the AHP can be maintained due to changes in facial and neck muscles and bones.[11]

Type 1 is the most common subgroup of Duane syndrome. It is characterized by limited abduction and relatively normal adduction. The abduction often improves with elevation or depression of the affected eye. There is globe retraction and narrowing of the palpebral fissure on adduction and widening of the fissure on abduction.[12]

In our study, every patient with Duane syndrome was classified as type 1 and had left eye affected.

The best surgical approach to esotropic Duane syndrome is not consensual. Jampolsky was the first to propose asymmetric bilateral medial rectus recession with more recession in the uninvolved eye to induce "fixation duress" via introduced adduction limitation in the normal eye.[13] Our research showed positive results with this technique – two patients had their AHP improved and four resolved their AHP.

Nystagmus is defined as an involuntary rhythmic oscillation of the eyes. Nystagmus can be grouped into infantile nystagmus (IN), which usually appears in the first three-six months of life, and acquired nystagmus, which appears later.[14] For those who have IN, this can be idiopathic or associated with another eye disease, such as retinal disease, albinism, low vision or visual deprivation in early life, due, for example, to congenital cataracts or optic nerve hypoplasia.[14] In our study, the IN cases were idiopathic.

The null zone is the position of gaze where the intensity of the nystagmus is minimal. Some patients discover that their vision is at its best when their eyes are placed in the position of least ocular instability and so adapt an AHP to bring the zone of best vision to the straight-ahead position.[15]

Kestenbaum and Anderson were the first-to-perform procedures aiming to shift both eyes' null zone into the primary position to eliminate the AHP.[16,17] Anderson proposed to weaken the agonists horizontal rectus muscles activated during the slow phase of the nystagmus because they were thought to have a greater tone than their antagonists.[17] Kestenbaum procedure combines 5 mm recessions of the agonist muscles with resections of the antagonist muscles (5-5-5-5 technique).[16] However, it led to a high rate of hypocorrections, so variants of this technique have been experimented. Parks modified the Kestenbaum technique to "5-6-7-8" or the "Classic Maximum" procedure in which is made in the abducted eye a 7 mm recession to the lateral rectus and a 6 mm resection to the medial rectus and in the adducted eye a recession of 5 mm to the medial rectus and an 8 mm resection to the lateral rectus.[18] Therefore, each eye gets a total of 13 mm of surgery. This surgical option is adequate for head turns up to 30°. When the head turn is between 30-45°, the surgical amounts are augmented 40% (7, 8.4, 9.8, 11.2 mm). When the head turn is more than 45°, the surgical amounts are increased 60% (8, 9.6, 11.2, 12.8 mm).[15]

In patients no. 15 and 17, the Parks modified technique was performed. In patients no.16 and no.18 it was a Parks+20%. In patient no. 19 the chosen approach was approximately a Parks+10%. In patient no. 20 there was a 30-45° head tilt and a right exotropia, so the standard amounts were adjusted accordingly.

Patients with nystagmus and an AHP that use glasses do not wear them through the optical center. This aspect can cause optical aberrations and interfere with refraction correction, ultimately interfering with vision development. Hence, surgical improvement of the AHP can also improve visual acuity by making it possible to look through the optical center of the glasses.

Isolated third nerve palsy may be unilateral or bilateral, complete or partial, pupil involving or pupil sparing, and congenital or acquired. Therefore, clinical management of third nerve palsy is challenging.[19] This diversity means that symptoms, including AHP, may be unique on each case, as can been seen in Table 4. Therefore, surgical management must be considered individually.

Brown syndrome is an ocular motility disorder characterized by a limitation of elevation in adduction, which Brown first described in 1950. The severe mechanical restriction is present during attempts to elevate the adducted eye passively in the forced duction test, but the elevation in abduction is normal.[20] Surgical correction is based on the weakening or

lengthening of the superior oblique muscle. In our case (case no. 24), a SO tendon recession to the anteromedial quadrant and posterior fibers tenotomy was performed.

One limitation of the present study is the subjective assessment of the AHP pre and post-operative. In past studies, evaluation of the AHP pre and post-operative was done using the goniometer.

As it is a retrospective study, the investigation depends on written information, which can consequently justify that some cases, such as lateral rectus muscle palsy, were not identified.

We have demonstrated different types of AHP and their respective ocular diagnosis and surgical approach. Overall, almost all patients improved after surgery.

CONTRIBUTORSHIP STATEMENT / DECLARAÇÃO DE CONTRIBUIÇÃO:

JSO, RSS and AM: Research design, data acquisition, data analysis, manuscript preparation and final approval.

PFC, FFR, JB: Research design, manuscript preparation and final approval.

RESPONSABILIDADES ÉTICAS

Conflitos de Interesse: Os autores declaram a inexistência de conflitos de interesse na realização do presente trabalho.

Fontes de Financiamento: Não existiram fontes externas de financiamento para a realização deste artigo.

Confidencialidade dos Dados: Os autores declaram ter seguido os protocolos da sua instituição acerca da publicação dos dados de doentes.

Proteção de Pessoas e Animais: Os autores declaram que os procedimentos seguidos estavam de acordo com os regulamentos estabelecidos pelos responsáveis da Comissão de Investigação Clínica e Ética e de acordo com a Declaração de Helsínquia revista em 2013 e da Associação Médica Mundial.

Proveniência e Revisão por Pares: Não comissionado; revisão externa por pares.

ETHICAL DISCLOSURES

Conflicts of Interest: The authors have no conflicts of interest to declare.

Financing Support: This work has not received any contribution, grant or scholarship

Confidentiality of Data: The authors declare that they have followed the protocols of their work center on the publication of data from patients.

Protection of Human and Animal Subjects: The authors declare that the procedures followed were in accordance with the regulations of the relevant clinical research ethics committee and with those of the Code of Ethics of the World Medical Association (Declaration of Helsinki as revised in 2013).

Provenance and Peer Review: Not commissioned; externally peer reviewed.

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