
TORSIONAL DIPLOPIA IN GRAVES' ORBITOPATHY

THREE CASE REPORTS

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SAMENVATTING

Torsionele diplopie wordt vaak miskend bij patiënten met Graves' orbitopathie omwille van de frequent geassocieerde verticale en/of horizontale deviatie. Torsionele diplopie bij Graves' patiënten kan het gevolg zijn van strabismechirurgie en/of orbitadecompressie, maar kan ook primair zijn. Aan de hand van drie gevallen en een kleine literatuurstudie proberen we inzicht te krijgen in het ontstaansmechanisme van de cyclodeviatie.

SUMMARY

Torsional diplopia may frequently accompany Graves' orbitopathy and is often not recognized. Some patients with manifest cyclotorsion do not complain of torsional diplopia because it is concealed by the large vertical and/or horizontal deviation. Torsional diplopia can occur spontaneously or appear after decompression or strabismus surgery. We discuss some interesting points arising from three cases. This will be followed by a discussion of the literature on the pathogenesis of cyclodeviation in Graves' orbitopathy.

RÉSUMÉ

Chez les patients avec une orbitopathie de Graves, la diplopie torsionnelle est fréquemment méconnue parce qu'il y a souvent simultanément une grande déviation verticale et/ou horizontale. Cette diplopie torsionnelle peut se manifester spontanément, mais

également après la chirurgie de strabisme ou après une décompression de l'orbite. A l'aide de trois cas cliniques, et suivant la littérature nous avons étudié la pathogénèse de la cyclodéviation.

KEY-WORDS

Graves' orbitopathy - torsional diplopia - pathogenesis

MOTS-CLÉS

Orbitopathie de Graves- diplopie torsionnelle - pathogénèse

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INTRODUCTION

In patients with hyperthyroidism who develop thyroid associated orbitopathy, fibrosis of the extraocular muscles causes restrictive strabismus. These patients frequently complain of diplopia. We are familiar with horizontal or vertical diplopia caused by restriction of the rectus muscles, but torsional diplopia constitutes a more difficult problem. Both oblique and rectus muscles may contribute to the genesis of torsional diplopia. We present and discuss three cases with torsional symptoms.

CASE REPORTS AND COMMENT

CASE 1

A 47 year old man with hyperthyroidism since december 1997 was referred to our department because of increasing orbitopathy and diplopia. He was treated with antithyroid drugs (Strumazol®). In 1998 he received orbital radiotherapy (20 Gy). He complained mostly of horizontal and vertical diplopia. Orthoptic examination showed a 14 prism diopter left hypertropia. There was no horizontal deviation. There was a symmetric limitation of the abduction and adduction in both eyes. Depression was limited in abduction in both eyes. There was a symmetrical proptosis.

After euthyroid status was achieved, a bilateral coronal orbital decompression was performed. The left hypertropia was increased (30 prism diopters) and there was now a 6 prism diopter esotropia. Ocular rotations were restricted in abduction (right more than left), with a 'secondary overaction' of the inferior oblique muscle (left more than right). Elevation in abduction was limited in both eyes, depression in abduction appeared normal in both eyes. The left hypertropia increased in dextroversion. Double Maddox rod test showed a 5° right excyclotorsion and a 20° left excyclotorsion.

In order to correct the vertical deviation, we recessed the right inferior rectus muscle (4 mm + hang-back suture of 1 mm diameter) and tucked the left superior oblique muscle five months after the orbital decompression. To cor-

rect the esotropia we recessed the left medial rectus muscle (5 mm).

Postoperatively the diplopia persisted. The left hypertropia was now 7 prism diopter in primary position and 15 prism diopter in dextroversion. There was a 10° excyclotropia of the left eye. With prismatic correction comfortable fusion was obtained in primary position.

Six months after the first strabismus surgery a recession of the left superior rectus muscle (4 mm) was performed to correct the vertical deviation. Postoperatively the left hypertropia remained 6 prism diopters in primary position. In dextroversion the vertical deviation was still 15 prism diopters and a 10° excyclotropia persisted. There was no significant limitation of ocular ductions.

Six months later, a disinsertion of the left inferior oblique muscle was performed and the left superior rectus muscle was recessed (insertion on 11 mm from the limbus with a hang-back suture of 3 mm).

Postoperatively there was no diplopia in primary position, but the patient complained of persisting diplopia in dextroversion. Because of the increase of the left hypertropia in dextroversion, a final operation (five months after the third strabismus operation) consisting of a resection of the right superior rectus muscle was performed. Double Maddox rod test showed a 6° excyclophoria of the right eye without complaints.

CASE 2

This 65 year old male patient suffered from hyperthyroidism and orbitopathy since 1996. After treatment with radioactive iodine, he became euthyroid. A recession of the right inferior rectus muscle (amount unknown) had been performed in 1998, prior to the first examination in our department.

Orthoptic examination preoperatively showed a 12 prism diopter left hypertropia and a 5° incyclotropia of the left eye. There was a limitation of abduction and elevation in the right eye and a limitation of depression in the left eye. The proptosis was more marked in the right eye compared to the left eye.

In 2000 an asymmetrical anterior two-wall orbital decompression was performed. Postoperatively the elevation of the left eye became re-

stricted as well, the other orthoptic findings remained unchanged.

In primary position the right hypotropia of 12 prism diopters persisted.

Because of this, and because of the fact that the forced elevation of the right eye under anaesthesia was restricted, it was decided to enlarge the recession of the right inferior rectus muscle by means of a hang-back suture. This operation was performed 8 months after the orbital decompression.

At the end of this procedure the forced elevation under general anaesthesia remained restricted, and indeed, after stabilisation the orthoptic findings were essentially unchanged.

The limitation of elevation of the right eye was more pronounced, especially in abduction. One month later, a recession of the inferior oblique muscle of the left eye was performed because the left hypertropia was more pronounced in dextroversion.

This diminished the left hypertropia in dextroversion but the over-all result was insufficient: there remained an important left hypertropia and incyclotropia in dextroversion.

Three months later, we decided to re-explore the region of the inferior rectus muscle of the right eye and to act according to the intraoperative findings: the inferior rectus muscle was found at 9 mm from the limbus. The muscle was rerecessed by means of a large hang back suture, but this time this procedure was combined with a 4 mm resection of the superior rectus muscle of the right eye.

Postoperatively the eyes were well aligned. There remained a discrete limitation of elevation of the right eye without functional complaints.

CASE 3

A 49 year old woman with Graves' orbitopathy was referred to our department for orbital decompression surgery. She was already euthyroid on referral. She had no diplopia and pre-operative orthoptic examination showed symmetric limitations of ab- and adduction of both eyes. There was a small limitation of depression of the right eye. Double Maddox rod test showed 2.5° incyclotropia of the right eye. The proptosis was the same on both eyes. CT scan showed thickening of the medial and inferior recti.

A symmetric coronal orbital decompression was performed.

At follow-up examination postoperatively she reported diplopia. Cover testing disclosed a 25 prism diopter esotropia with an important A pattern and a 3 prism diopter left hypertropia. There was limitation of elevation of the right eye and limitation of abduction of both eyes. Five months after the orbital decompression the medial rectus muscles were recessed 5 mm each and simultaneously an upwards transposition of 1/2 tendon width was performed because of the A pattern. The right inferior rectus muscle was also recessed (3 mm).

Postoperatively no horizontal deviation remained. Abductions were improved as well and the A pattern was corrected. Cover testing showed an 8 prism diopter right hypertropia and there was limitation of depression of the right eye.

Postoperative torsional symptoms led to double Maddox rod testing and disclosed a 15° right incyclotropia.

Two months after the first strabismus operation, the medial recti muscle insertions were put back on the 3 and 9 o' clock positions respectively. The left inferior rectus muscle was recessed 4 mm.

Postoperatively Double Maddox rod testing showed an 8° right incyclotropia. The right hypertropia was unchanged, but the downgaze was more symmetrical. A small A pattern reappeared.

Advancement and full tendon width temporal transposition of the right inferior rectus muscle was performed eleven months after the second strabismus operation.

After the last operation the torsional symptoms persisted. A 10 prism diopter right hypertropia and esotropia and an 8° right incyclotropia persisted. A 12 diopter base-down and out prism was prescribed for the right eye.

With this prism, there was comfortable sensory fusion and there was no need for additional treatment.

COMMENT

Patient 1 presented an excyclotropia of 25° after orbital decompression.

In this patient, relaxing the left inferior rectus muscle together with a tucking of the left su-

perior oblique muscle was not sufficient to correct this excyclotropia. The left hypertropia was also undercorrected, and because sensory fusion could be obtained with prisms, our second procedure was primarily aimed at correction of the vertical deviation (recession of the left superior rectus muscle).

The hypertropia persisted, in the subsequent intervention both elevators of the left eye were weakened. This eliminated the vertical deviation in primary position and fusion of the residual excyclodeviation was obtained.

A final resection of the right superior rectus muscle improved the field of binocular single vision in dextroversion without increasing the excycloduction

In patient 2, incyclotropia was noted on the first examination in our department. It should be mentioned that he had already undergone a recession of the right inferior rectus muscle. The cyclodeviation was not altered by the orbital decompression. We performed a recession of the right inferior rectus muscle which turned out to be insufficient: the vertical deviation and limitation of elevation didn't improve. We decided to perform an anteropositioning of the left inferior oblique muscle because the hyperdeviation increased in dextroversion. An anteropositioning procedure decreased the vertical component insufficiently. This procedure didn't diminish the cyclodeviation.

Exploration of the right inferior rectus muscle showed that the muscle had migrated forwards, which explained the poor result. A recession combined with a resection of the right superior rectus muscle finally led to a satisfactory result. Theoretically, this procedure should increase the incyclotropia, but because fusion was restored, our patient was comfortable.

In patient 3 the limitation of elevation in the right eye was treated with a recession of the right inferior rectus muscle. The limitations of abduction were treated with recessions of the medial recti muscles and the A pattern was improved with upwards transposition of the medial recti. Unfortunately the transposition procedure increased the incyclodeviation and fusion was insufficient to overcome this incyclotropia.

We had to place the medial recti downwards to the original level. This resulted in a decrease of the incyclotorsion from 15° to 8°. The left

inferior rectus muscle was recessed to compensate for the limitation of depression of the right eye. Even with prismatic correction fusion was not possible because of the large residual incyclotropia.

The final operation consisted of a temporal transposition of the right inferior rectus muscle. After this surgery fusion could be obtained with prisms. Under binocular conditions the patient did not complain of incyclotropia.

An interesting point that arises from these 3 cases is the role of the fusional system.

It is conceivable that the slow progression of the thyroid orbitopathy allows the fusional system to adapt itself to the new situation and indeed enlarged fusional amplitudes may be found in these patients. Nevertheless horizontal, vertical and torsional deviations caused by the mechanical restrictions of the ocular motility may form obstacles for sensory fusion. It is important to note that in all our patients cyclofusion could be obtained with sufficient correction of the horizontal and vertical deviations. In this respect, prisms are a useful tool to evaluate the fusional capacity to compensate torsion after correction of the horizontal and vertical deviation.

If the cyclodeviation appears minor (patient 1 and 2), it may be sufficient to correct the horizontal and vertical deviations and to improve the ocular motility. Care should be taken to avoid iatrogenic increase of the cyclodeviation. However, if the cyclodeviation is more important, the surgical plan should include specific measures to diminish the cyclotropia (see patient 3).

Another point worth mentioning concerns the effect of surgery on the oblique muscles in these patients. In patients 1 and 2, oblique muscle surgery was performed to correct certain vertical incomitancies. It was remarkable that these procedures had less effect than expected on both the vertical- and the cyclodeviation.

The beneficial effect of resections of the superior rectus muscle in two of these patients is surprising. In general, resections are not recommended for the surgical treatment of Graves' orbitopathy. In our patient 2, who underwent

an ipsilateral recession of the inferior rectus muscle prior to the orbital decompression surgery and had a resection of the superior rectus muscle afterwards, the resection didn't jeopardize the downgaze. We think that in this case the orbital decompression caused a certain degree of hypoglobus, and that the resulting hypotropia really needed a resection.

Hangback sutures may be used to achieve a maximal weakening of the rectus muscles, while keeping the functional insertion before the equator (3). However, as illustrated by patient 1, the muscle postoperatively may migrate towards its original insertion and then the effect will be reduced. A simultaneous resection of the ipsilateral antagonist decreases this risk. However, we do not recommend such a combination as a standard procedure because as a primary procedure, the effect tends to be unpredictable and overcorrections occur generally.

The presence of an A or V pattern may complicate the planning of surgery. For instance, in patient 3, the A pattern was very important, so we decided to treat it with an upward transposition of the medial recti. This did correct the A pattern, but increased the incyclodeviation. Vertical repositioning of the medial recti decreased the incyclotropia, but the A pattern reappeared. Temporal transpositioning of the right inferior rectus muscle was performed. This diminished the incyclotropia without changing the residual A pattern.

DISCUSSION

A lot of information can be found on diplopia in Graves' orbitopathy and on the incidence of horizontal and/or vertical motility disorders (4,7-10). However only few articles concerning torsional diplopia have been published (2,6). In our experience torsional diplopia occurs more frequently in patients who underwent an orbital decompression procedure. Spontaneously, most patients complain only of horizontal and/or vertical diplopia, possibly the large horizontal and/or vertical deviations conceal the torsional component.

EXCYCLOTROPIA

There is a discussion about the origin of excyclotropia in Graves' orbitopathy. Excyclotropia is frequently seen in patients who didn't undergo previous surgery (2).

Garrity et al. state that it is caused by restriction of the inferior rectus muscle, which simultaneously depresses, adducts and excycloducts (2).

Kushner believes that this excyclotropia is caused by an overaction of the ipsilateral inferior oblique muscle secondary to its working against the contracted inferior rectus muscle (6). The differential diagnosis should be made by looking in which field of gaze the excyclotropia increases. If there is a 'secondary' overaction of the inferior oblique muscles, there will be an increased excyclotropia with upgaze. However, the instances of excyclotropia in patients with contracted inferior recti muscles which persist in downgaze, out of the field of the inferior oblique muscle, mitigate for a more direct causal role of the inferior rectus muscle.

According to almost all authors, relaxing the restriction of the inferior rectus muscle eliminates the excyclotropia and the apparent overaction of the inferior oblique muscle. In our patient 1 this procedure was combined with a tucking of the contralateral superior oblique muscle and the result was nevertheless insufficient. Simultaneous inferior rectus muscle recession and inferior oblique muscle weakening should not be performed because of the risk of overcorrection of the torsion resulting in an incyclotropia postoperatively (6).

INCYCLOTROPIA

Contrary to excyclotropia, incyclotropia is observed more frequently after strabismus surgery or after orbital decompression (2).

Three pathophysiological factors are proposed as possible reason for incyclotropia after orbital decompression: hypoglobus, globe recession and inferior-medial prolapse of orbital tissue into the opened antral and ethmoidal sinuses (2,5).

Hypoglobus after decompression may induce tension on the superior oblique muscle and tendon, which are suspended at the trochlea. In-

cycloduction may result from this and from tension on a restricted superior rectus muscle. Excessive proptosis reduction as a result of the orbital decompression could also lead to increased tension on the superior oblique tendon. However, the correlation between the amount of proptosis reduction and the onset of torsional diplopia is not proved (2).

Finally, a study by Abramoff showed that the osteotomies in decompression surgery cause a centrifugal displacement of the path of the medial and inferior rectus muscle, with secondary hypotropia or esotropia (1). Earlier Koornneef demonstrated the existence of these paths (5).

This esotropia can allow for enhanced action of the superior oblique muscle. In clinical practice it is not always possible to determine the factors contributory to the appearance of torsion.

As has been claimed by Kushner, strabismus surgery can increase incyclotropia if downgaze is impaired postoperatively.

Any situation that impairs downgaze (an excessively weakened inferior rectus muscle or a severely restricted superior rectus muscle) may recruit the superior oblique muscle to assist with downgaze and induce incycloduction.

In his view large recessions of the inferior rectus muscles may cause a muscle weakness and thus lead to a secondary overaction of the ipsilateral and contralateral superior oblique muscles. He recommends to perform simultaneously a weakening of the superior oblique muscle if recessions of more than 6 mm are necessary (6). However, in patient 2 recession of the inferior rectus muscle and even resection of the superior rectus muscle did not influence the cyclodeviation. In patient 3 the incyclotropia increased after the vertical transposition of the medial rectus muscle. Although this patient also underwent a 3 mm recession of the inferior rectus muscle, we don't think that this played a significant role in the increase of incyclotropia.

Garrity et al. (2) state that in patients with impaired downgaze, treatment for incycloduction should be based upon weakening of the incycloductors (superior oblique muscle or superior rectus muscle) or advancing a previously

recessed inferior rectus muscle. According to them the choice depends on the vertical deviation in primary position:

- If there is an ipsilateral hyperdeviation, a recession of the superior rectus muscle should be performed.
- If there is an associated hypodeviation and an A pattern, a tenectomy of the superior oblique muscle is recommended.
- In cases where there is no associated vertical deviation, recession of the superior rectus muscle in combination with an ipsilateral superior oblique tenectomy should be recommended in order to prevent the postoperative vertical deviation.

Alternatively a previously recessed inferior rectus muscle may be advanced.

CONCLUSION

Torsional diplopia may frequently accompany thyroid disease and is often not recognized. Most patients with manifest cyclotorsion do not complain of torsional diplopia because it is concealed by the large vertical separation of the images. Therefore, orthoptic examinations in Graves' patients should always include a double Maddox rod test or an examination with the synoptophore. Prisms may be a useful tool to evaluate the fusional capacity to compensate torsion after correction of the vertical and/or horizontal deviation.

Torsional diplopia in Graves' orbitopathy can occur spontaneously or appear after decompression or strabismus surgery. Excyclotropia appears often without previous surgery and is probably caused by overaction of the ipsilateral inferior oblique muscle against the restricted inferior rectus muscle. On the contrary, incyclotropia occurs more often after orbital decompression or strabismus surgery.

Certain guidelines in planning strabismus surgery may minimize the incidence of postoperative symptomatic torsion in most cases. If the cyclodeviation is minor, the surgical plan is aimed at correction of the horizontal and vertical deviation so that the fusional system is able to correct the torsional component. If the cyclodeviation is important, surgery is aimed at decreasing the cyclotropia.

While resections are generally not recommended with restrictive strabismus, it may be be-

neficial in the patients with hypoglobus after decompression surgery. In selected cases, resections of the ipsilateral antagonist may also be advisable in combination with hang-back sutures to reduce the risk for forward migration of these sutures.

Surgery of the oblique muscles seems less efficient for correction of both the vertical and torsional component in these patients.

Treatment of torsional diplopia associated with Graves' orbitopathy remains a great challenge for the physician.

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