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ATYPICAL FORMS OF STRABISMUS AND THEIR TREATMENT

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Abstract

Atypical forms of strabismus, including Brown syndrome and Duane syndrome, are complex and varied oculomotor disorders that require a careful diagnostic approach and individualized treatment. This article presents current data on the etiology, clinical presentation, diagnostic methods, and modern treatment options for these conditions. **Brown syndrome** is characterized by restricted elevation of the eye during adduction, due to mechanical difficulty of the superior oblique tendon passing through the trochlea. This is confirmed by limitations of elevation in adduction, a V-pattern deviation, and positive traction test results, supported by MRI/CT findings. Treatment may be conservative (observation, steroid injections in inflammatory forms) or surgical (tenotomy, tenectomy, silicone expander implantation). In contrast, **Duane syndrome** is a congenital dysinnervation disorder featuring limited horizontal eye movements, globe retraction, and often caused by aplasia or hypoplasia of the abducens nerve (cranial nerve VI), with compensatory innervation of the lateral rectus by the oculomotor nerve (cranial nerve III). Diagnosis is clinical, with MRI or genetic testing used as needed. Therapy includes correction of refractive errors, amblyopia treatment, use of prisms, and, in cases of significant cosmetic or functional impairment, surgical intervention such as muscle relaxation, transposition surgery, Y-splitting, and more. Both conditions are characterized by a wide range of manifestations and the absence of a universal surgical algorithm. Therefore, treatment strategy must be based on thorough clinical analysis and patient-specific considerations.

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Introduction

Strabismus is one of the most common ophthalmic disorders encountered by pediatricians and ophthalmologists. While typical forms of strabismus (comitant strabismus) are characterized by the same degree of deviation in all directions of gaze, atypical forms represent a more complex group that includes incomitant strabismus and dysinnervational disorders. Brown syndrome and Duane syndrome are particularly noteworthy among them—pathological conditions requiring a deep understanding of mechanisms, clinical presentation, and treatment approaches.

1. Clinical and Pathophysiological Features of Brown Syndrome

Brown syndrome, first described by Harold Brown in the mid-20th century, is characterized by restricted elevation on adduction due to mechanical obstruction of the superior oblique tendon's passage through the trochlea. Clinically, it manifests as a V-pattern gaze, hypotropia in the primary position, a characteristic head posture (chin-up posture with head turn), and a positive forced duction test. Pathophysiologically, fibrous thickening of the superior oblique tendon restricts its smooth gliding through the trochlea, and congenital dysinnervation anomalies may also contribute. The syndrome occurs in approximately 1 in 450 strabismus cases, with a female predominance and 10% bilateral forms. Some cases show spontaneous improvement with conservative management, especially in congenital non-functional forms. However, in the presence of hypotropia or significant compensatory posturing, surgery is often necessary. Tenotomy or tenectomy of the superior oblique tendon, and less commonly silicone expander implantation, are the most frequent procedures. Surgical correction shows significant improvements in ocular motility and gaze elevation—particularly with temporal tenotomy in cases of limited elevation. In inflammatory or acquired cases (e.g., trochlea inflammation, sinusitis, rheumatoid arthritis), treating the

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underlying disease with steroid injections (local or systemic) and immunosuppressants such as adalimumab is indicated.

2. Clinical and Etiopathogenetic Characteristics of Duane Syndrome

Duane syndrome is a congenital form of restricted horizontal strabismus, including globe retraction, narrowing of the palpebral fissure on adduction, and possible upshoot/downshoot on attempted gaze shifts; it is classified as a congenital dysinnervation disorder. It is etiologically caused by aplasia or hypoplasia of the abducens nerve (VI), with compensatory innervation of the lateral rectus from a division of the oculomotor nerve (III), which causes cocontraction of the medial and lateral rectus muscles and retraction of the globe. It accounts for 1–5% of all strabismus cases, is usually unilateral, and is slightly more common in females (about 60:40). Haber's classification identifies three types: Type I (most common, ~75–85%) involves restricted abduction; Type II involves restricted adduction; and Type III involves restriction of both movements.

Diagnosis relies on ophthalmic examination—evaluation of motility, forced duction testing, refraction assessment, amblyopia detection, compensatory head posture, and binocular function. Genetic testing is used when there is a family history or suspicion of syndromic forms. Management is individualized: conservative observation, refractive correction, prism use, and occlusion therapy for amblyopia are viable in mild or asymptomatic cases.

Surgery is considered in cases of marked deviation, glob retraction, or abnormal head posture. Techniques include muscle relocation (relaxation of medial or lateral rectus), vertical transposition, lateral rectus fixation to the periosteum, often using adjustable sutures. These procedures primarily aim to improve eye position in the primary gaze and cosmetic appearance rather than to fully restore motility.

Analysis of Current Data

Evidence confirms that atypical forms of strabismus, such as Brown and Duane syndromes, are distinct nosological entities with specific clinical phenotypes,

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pathophysiologies, and management requirements. Unlike typical (comitant) strabismus, these atypical variants involve motility restriction (incomitance), mechanical or neurological disruption, diagnostic complexity, and demand tailored therapy.

1. Relevance and Clinical Significance

Brown syndrome presents with limitations in elevation during adduction, V-pattern deviation, hypotropia, and compensatory head posture. Although rare (~1:450 strabismus cases), it significantly impacts function and aesthetics, requiring long-term monitoring and timely intervention. Duane syndrome, a common atypical form (1–5% of strabismus cases), results from lateral rectus dysinnervation due to VI nerve hypoplasia and III nerve compensation. Restricted horizontal movements, globe retraction, and abnormal head posture severely affect binocularity, visual comfort, and patient psychology.

2. Diagnosis: Key Differences

Brown syndrome diagnosis is based on elevation restriction in adduction, with imaging (MRI, CT) used when needed. Observation is important in congenital forms without significant functional or aesthetic impairment due to potential spontaneous improvement. In inflammatory forms, addressing the primary disease through steroid therapy and immunomodulation is essential. Duane syndrome requires detailed history, ocular motility assessment, recognition of dysinnervation signs, use of genetic testing when indicated, and evaluation for amblyopia, binocular function, and head posture. Differential diagnosis includes VI nerve neuropathy, syndromic strabismus, and mechanical restrictions.

3. Treatment: Conservative vs. Surgical Approach

Brown Syndrome: Conservative management is appropriate for mild forms without hypotropia or pronounced head posture, recognizing the potential for spontaneous improvement. In inflammatory cases, systemic or local therapy (steroids, NSAIDs, immunosuppressants) may be effective. Surgery is indicated

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when there is significant hypotropia or compensatory head posture. Common procedures include tenotomy or tenectomy, sometimes with silicone expanders. Conservative care involves refractive correction, occlusion for amblyopia, prisms for mild head tilt, and follow-up in cases with binocular function in primary gaze. Surgical interventions—muscle relaxation, transposition, periosteal fixation, adjustable sutures—aim to minimize deviation and improve head posture and aesthetics, rather than restore full motility.

Duane Syndrome: Management is individualized; mild cases may suffice with refractive correction and prisms. Significant deviations, retraction, or abnormal head posture merit surgery—relaxing the medial or lateral rectus, vertical transposition, or periosteal fixation (often with adjustable sutures). The aim is to optimize primary gaze alignment and appearance rather than full motility restoration.

4. Individualized Approach and Future Perspectives

There is no universal treatment algorithm. Diagnosis, therapeutic strategy, and surgical decisions must be guided by detailed clinical evaluation, disease severity, treatment goals, patient age, and prognosis. Dynamic monitoring and a multidisciplinary approach (involving ophthalmology, neurology, rehabilitation) enhance outcomes. Prognosis should account for amblyopia risk, compensatory abilities, patient tolerance for surgery, and potential requirement for repeat interventions (especially in Brown syndrome, which may involve persistent supraduction issues post-tenotomy).

Conclusion

Atypical strabismus forms—Brown and Duane syndromes—are intricate ophthalmic disorders with distinct mechanisms, clinical manifestations, and treatment needs. Contemporary evidence reinforces that optimal outcomes are achieved through precise diagnosis, tailored treatment planning, prudent surgical decision-making, and realistic expectations. Timely intervention enhances visual function, cosmetic outcomes, and overall patient quality of life.

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