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Clinical and radiological characteristics of Graves' orbitopathy patients showing spontaneous decompression

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ABSTRACT

Objective: To investigate clinical implications and radiological characteristics of spontaneous decompression in patients with Graves' orbitopathy (GO).
Methods: The medical records and images of GO patients showing spontaneous decompression in computed tomography (CT) scans without any other cause, such as orbital surgery or trauma were retrospectively reviewed. Clinical parameters, including clinical activity score (CAS), modified NOSPECS score, exophthalmometry results, extraocular muscle involvement, and the presence of optic nerve compression were evaluated. Paired orbit analyses of maximum recti muscle diameters, area of lamina papyracea, and number of ethmoid air cell septa were determined quantitatively in the unilaterally decompressed group.
Results: 77 orbits of 55 patients were found to present spontaneous decompression, which was observed only in the medial orbital wall in all cases. In the paired orbit comparison, maximal diameters of medial ($P = 0.009$) and lateral recti muscles ($P = 0.023$) were significantly larger in decompressed orbits than in non-decompressed orbits. However, the incidence of optic neuropathy was not significantly different ($P = 0.500$). There was no difference in anatomic features of lamina papyracea or ethmoid air cells between decompressed and non-decompressed orbits.
Conclusions: Spontaneous decompression occurred in the medial wall, associated with enlarged horizontal recti muscles, but not with structures of lamina papyracea or the ethmoidal sinus. Orbital bone remodeling by spontaneous decompression by lowering intraorbital pressure, might have provided a protective effect against the development of optic neuropathy.

1. Introduction

Graves' orbitopathy (GO) is an autoimmune inflammatory condition of orbital tissues associated with thyroid disease. The most common manifestations of GO include eyelid retraction, periorbital swelling and erythema, proptosis, and limited ocular movement (Epstein et al., 1993). GO can be complicated also by severe exophthalmos and compressive optic neuropathy, caused by the increase in the intraorbital pressure in a fixed bony orbit, due to the enlargement of extraocular muscles (EOM) and fat tissue (Koornneef et al., 1990; Bartalena et al., 2008). Decompression surgery is undertaken to reduce the intraorbital pressure in such complicated cases (Goh and McNab, 2005; Clauser et al., 2012). However, patients showing spontaneous decompression in CT scans, despite an absence of previous trauma or surgical history, are sometimes encountered.

Spontaneous decompression is an orbital wall fracture or flattened wall caused by any pathologically elevated intraorbital pressure without any other reason, such as trauma, orbital surgical history, or orbital bony pathology. Spontaneous decompression in GO is a rare event, and has been reported only in three case reports (Bhermi et al., 2006; Kashkouli and Pakdel, 2010; Detorakis, 2014). Bhermi et al. (2006) reported a case with spontaneous orbital floor fracture with developed pain, bruising, and infraorbital hypoesthesia, without any trauma history. Kashkouli and Pakdel (2010) reported a GO patient with an asymptomatic orbital floor fracture. Detorakis (2014) reported a GO patient with massive medial rectus muscle enlargement, displacing medially the medial wall, and thus creating an effective medial wall decompression. However, there are no large case series investigating clinical and radiological

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characteristics of GO patients showing spontaneous decompression.

In this study, we reviewed all the medical records and computed tomography (CT) images of patients who were diagnosed with GO at our institute, to identify spontaneous decompression. In those cases, we investigated the clinical and radiological features, and the association of spontaneous decompression with the development of compressive optic neuropathy.

2. Materials and methods

2.1. Patients

Consecutive Korean patients who visited Severance Hospital, Yonsei University College of Medicine, were diagnosed with GO, were followed up by one ophthalmologist (JSY), between January 2008 and June 2012, and were recruited for the retrospective study. Diagnosis of GO was made based on clinical ophthalmic examination, using disease history data, slit-lamp examination, intraocular pressure, exophthalmos measured with a Hertel exophthalmometer, Hess screen testing, binocular single vision tests, and CT scans. Diagnosis of Graves' disease was made by an endocrinologist, and was based on peripheral thyroid function, thyroid stimulating hormone (TSH) receptor antibody values, thyroid ultrasound pattern and/or thyroid scan, and clinical features, such as diffuse goiter or symptoms of hyperthyroidism.

The patients with spontaneous decompression were selected by review of CT scan images. Patients with a history of previous orbitofacial surgery, trauma, silent sinus syndrome, neoplasm, bony pathology, or chronic sinusitis were excluded. The study adhered to the tenets of the Declaration of Helsinki, and approval to conduct this study was obtained from the Institutional Review Board of the Severance Hospital of Yonsei University.

2.2. Ophthalmic manifestations of Graves' orbitopathy

GO activity was evaluated using the seven points of the clinical activity score (CAS) as described by Mourits et al. (1997). GO severity was assessed using the modified NOSPECS classification (Eckstein et al., 2006; Jang et al., 2012). The modified NOSPECS score included the following: eyelid retraction (class 1), soft tissue involvement (class 2), proptosis (class 3), EOM (class 4), and corneal (class 5) or optic nerve involvement (class 6). The scores ranged from 0 to 3, according to severity, and the sum of the scores constituted the modified NOSPECS score (range, 1–14) (Jang et al., 2013).

2.3. Quantitative CT measurements

CT scan (SOMATOM Sensation 16; Siemens Medical Solutions, Erlangen, Germany) results, with 2 mm axial and 2 mm coronal section thicknesses, were stored digitally. All measurements were performed using workstation software (GE Centricity, version 2.0; GE Healthcare, Milwaukee, WI, USA).

Spontaneous decompression was defined in CT scan images as a case with flattened orbital wall with displacement of the orbital wall toward the sinus (Fig. 1A), or fracture with herniation of soft tissue (Fig. 1B). To compare clinical and radiological features, patients with spontaneous decompression were divided into bilateral or unilateral groups, depending on whether decompression existed in both eyes, or in one eye. For the unilateral group, paired orbit analyses were performed between eyes with and without decompression, to determine why decompression only occurred in one of the two eyes. The CT features of EOM and angle of orbital apex were assessed quantitatively. Orbital angle was measured between 2 lines, on axial images, at the mid-globe level, and were quantified to evaluate the capacity of the bony apex (Fig. 2A). Maximum diameters of recti muscles were assessed using coronal scans for the inferior and superior recti muscles (Fig. 2B), and using axial scans for the medial and lateral recti muscles (Fig. 2C).

Lamina papyracea area, and number of ethmoid air cell septa were measured according to our previous report (Song et al., 2009). The anterior and posterior height of the lamina papyracea was estimated in coronal views, and the anteroposterior length was measured in the axial slice. The lamina papyracea was assumed to be a trapezoid, and the area was calculated as (anterior height + posterior height) × anteroposterior length/2. The number of ethmoid air cell septa was counted in the axial view transecting the optic foramen.

2.4. Statistical analysis

Data were analyzed with PASW statistical software version 20.0 (SPSS Inc., IBM Corp., USA). The categorical variables of NOSPECS and CAS were compared between groups, using Pearson's chi-square tests or Fisher's exact tests. The mean quantitative CT measurements were compared between groups by the Student's 2-tailed *t*-test. Paired *t*-tests and McNemar's tests were used to

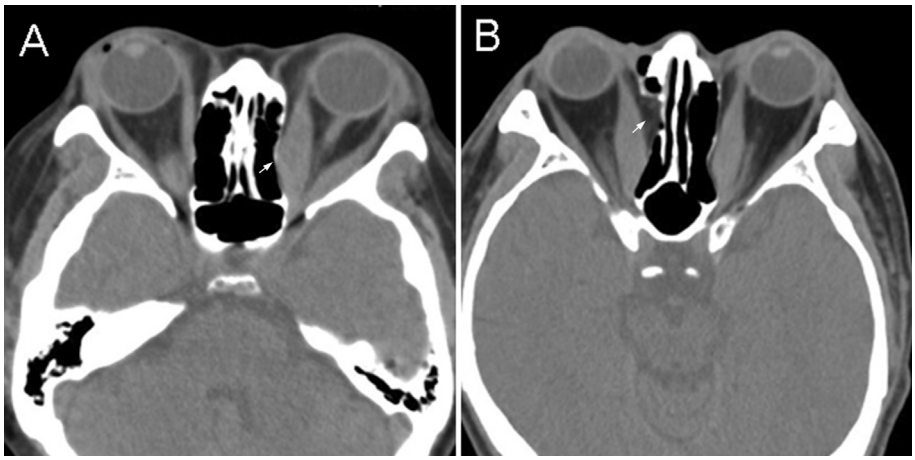

Fig. 1. Representative computed tomography (CT) images of spontaneously decompressed orbit. Axial CT scans showing the flattened orbital wall with nasal displacement of the left medial wall (A) and the fracture with herniation of soft tissue in the right orbit (B). White arrows indicate decompressed orbit.