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Parry Romberg syndrome: A long-term retrospective cohort study of 10 patients

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ABSTRACT

We performed a retrospective cohort study of patients included in the database of the department of Oral & Maxillo-Facial Surgery between 2006 and 2016 at the University Hospitals Leuven with Parry Romberg syndrome to assess and compare initial presentation, diagnostic methods, and different surgical and non-surgical approaches. Primary outcomes were functional and esthetic results using intra- and extra-oral images, clinical orthognathic measurements, radiological cephalograms, and cone beam computed tomography, including three-dimensional cephalometry analyzing the facial symmetry of hard tissues. The secondary outcome was patient quality of life using the Ferrans and Powers Generic Quality of Life Index. Ten patients were included; two had medical treatment, three had reconstructive surgery, four had orthognathic surgery, and three had lipofilling. Two patients had post-surgical infectious complications. Two patients developed neuropathic pain. Three-dimensional cephalometry showed no significant difference with regards to anatomical bony landmarks between the affected and non-affected sides. A volumetric analysis showed a significant difference ($P=0.04$) in maxillary volumes. Seven patients were satisfied with their aesthetic and functional outcomes. Mean follow up was eleven years. In conclusion, this study should be interpreted carefully due to small sample size. We feel most patients can be treated conservatively or with minor aesthetic corrections using fat grafting methods. Use of allografts and osteosynthesis materials was associated with an increased risk of postoperative superinfection. Fat grafts produced predictable, noncomplicated results and can be used during disease progression. Three-dimensional analysis showed acceptable symmetry of the bony framework on follow-up.

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1. Introduction

Parry Romberg syndrome is a rare progressive disease causing hemifacial atrophy. It affects more women than men, and its etiology is still largely unknown [1]. The diagnosis is made on a clinical basis. The expert opinion is that it should be considered a spectrum disease of linear scleroderma. Consequently, it is possible to categorize this disease according to severity, aiding in the choice of treatment. Treatment options vary from conservative anti-inflammatory drug therapy to

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lipofilling and reconstructive orthognathic surgery. In this article, we present all our cases with an extensive discussion supported by current literature.

2. Materials and methods

2.1. Data collection

We conducted a search of all patient records at the Oral and Maxillo-Facial Department of the University Hospitals, Leuven, between 2006 and 2016, using following search terms: hemifacial atrophy, Parry, Romberg, and morphea en coup de sabre. Information from the files was extracted and patients were contacted for a follow-up consultation (Fig. 1). During the follow-up consultation, the following data was obtained: history, current complaint, general clinical exam, clinical orthognathic measurements, VECTRA[®] 3D imaging, intra- and extra-oral images, TRIOS[®] intraoral scans, T-scan[™], panoramic x-rays, cephalometric x-rays, and cone beam computed tomography (CBCT).

2.2. Grading of Parry Romberg

All patients were graded according to following criteria. Morphea is a unilateral linear scleroderma of the frontoparietal region. Mild Parry Romberg syndrome: presence of soft tissue atrophy in one trigeminal area of the lower face without rotation of the occlusal plane. Moderate Parry Romberg syndrome: more than one trigeminal dermatome is involved as well as the nasal ala, with a deviation of the oral commissure; the occlusal plane remains more or less horizontal and bony atrophy is not apparent. Severe Parry Romberg syndrome: rotation of the occlusal plane with atrophy of the skeletal framework involving the maxilla, mandible and zygoma; chin and nose deviate to the affected side.

2.3. Three-dimensional cephalometry

After the follow-up consultation, DICOM files from the CBCT images were extracted and analyzed using Materialise Mimics[®] 20. Three-dimensional anatomical landmarks were marked according to Swennen et al. [2]. Symmetry analysis was performed using the methods from Gateno et al. (2011) defining the intrinsic symmetry of the maxilla and mandible [3]. The

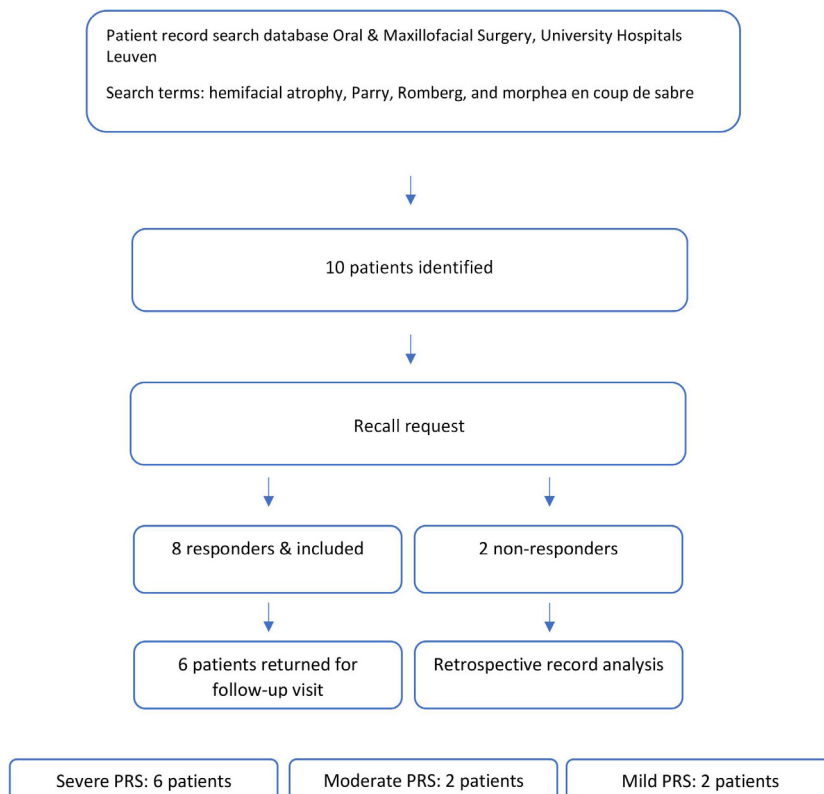


Fig. 1. Patient selection and study recruitment flowchart. (NOT IN COLOR).

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