Frontal lifting using a tissue expander in pachydermoperiostosis: A case report

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Abstract
Pachydermoperiostosis, a rare condition, is characterized by pachydermia, finger clubbing, and periostosis. We present an unusual treatment for frontal rhytids, for which we used a tissue expander that contributed to thinning of the skin and the depth of the rhytids prior to frontal lifting. The results were maintained after one year.

1 | INTRODUCTION

Pachydermoperiostosis is the primary form of hypertrophic osteoarthropathy, which is characterized by pachydermia, finger clubbing, and periostosis. It should be differentiated from the secondary form of hypertrophic osteoarthropathy, which is more common and often associated with pulmonary bronchiectasis, bronchogenic carcinoma, congenital heart disease, and malignant neoplasms of the thyroid and gastrointestinal tracts.² It was first described in 1868 by Friedrich, who reported two brothers with what he termed hyperostosis of the entire skeleton. In 1935, three dermatologists—Touraine, Solente, and Golé (for whom the syndrome was named afterward)—distinguished its three forms: complete form (with periostosis, pachydermia, and finger clubbing), incomplete form (without pachydermia), and forme fruste (pachydermia with minimal skeletal changes).²³ Its pathogenesis is not completely elucidated but is related to an increase in the secretion of prostaglandin E2. Regardless of the subtype, all patients present with acropathy; 20%–40% of patients present with arthralgia (associated or not with arthritis), mainly in the knees, hips, and hands, and joint effusion may also be present.³⁵ Cutaneous manifestations include thickening of the skin, mainly of the face and scalp, with prominent wrinkles in the form of cutis verticis gyrata. Edema and palpebral ptosis may be present due to the proliferation of dermal fibroblasts.³

Symptoms usually begin at puberty and tend to stabilize with time. Pachydermoperiostosis predominantly occurs in male individuals at a ratio of 9:1, with a familial inheritance pattern, and both autosomal dominant inheritance and autosomal recessive inheritance have been described.⁴⁶ Various treatments, such as rhytidoplasty, rhytidectomy, and botulinum toxin injections, have been described in the literature.⁷⁻⁹ Two cases involving frontal region treatment associated with the use of tissue expanders prior to frontal rhytidoplasty have also been reported.⁹¹⁰

A search was performed of the PubMed and Embase databases using the following terminology: pachydermoperiostosis, “Touraine Solente and Golé Syndrome,” treatment, surgical treatment, frontal lifting, subperiosteal lifting, tissue expander, and combinations of these. We aimed to report an unusual surgical treatment, the third case in the literature to the best of our knowledge, involving the frontal region in a patient with pachydermoperiostosis using a tissue expander before rhytidoplasty to thin the skin and decrease the prominent wrinkles and folds that are characteristic of the disease.

2 | CASE REPORT

A 25-year-old hypertensive man presented with a history of skin thickening mainly on the face and scalp from the age of...
14 years associated with prominent skin wrinkles and folds (Figure 1). He also presented with joint edema in the hands and knees and finger clubbing. His main complaint was premature aging of his face, specifically the wrinkles in the frontal region. He had previously received botulinum toxin injections with unsatisfactory results. Preoperative examinations did not reveal any changes. Placement of a rectangular tissue expander was proposed to thin the skin and reduce the depths of the skin wrinkles and folds that are characteristic of the syndrome (Figure 2).

Subperiosteal lifting was chosen, as the disease pathophysiologically manifests with thickened periosteum. In an attempt to thin the skin and decrease the prominent wrinkles and folds that are characteristic of the disease, as well as achieve greater and improved lengthening of the forehead skin over time compared with the punctual lengthening achieved via surgery, a rectangular expander measuring $8.5 \times 5$ cm with a 200 mL volume was initially placed on the forehead in a submuscular position. Slow and gradual expansion was then performed until the 200 mL volume was filled (Figure 3). Six months after the placement of the expander, the patient underwent a subperiosteal frontal lift, wherein a precapillary incision was made, the expander and its capsule were removed, and multiple incisions were made in the forehead flap to improve stretching. Resection of the thickened periosteum, superior flap traction, and removal of the excess skin were also performed, maintaining the previous capillary line.

3 | OUTCOME

The patient presented significant improvements in the skin folds and wrinkles of the forehead, with this result being maintained 1 year after the surgical treatment (Figure 4). Slight scars were noted. The patient complained of hypoaesthesia in the frontal region until the third month after the final surgery.

4 | DISCUSSION

The classic methods of rejuvenation of the upper third of the face, such as botulinum toxin injections, are not effective in cases of pachydermoperiostosis as the primary cause of deep wrinkles is thick skin (mainly the dermis), rather than the repetitive use of the frontal and corrugator muscles. Rhytidoplasty with subperiosteal detachment of the upper third of the face is effective for the treatment of deep wrinkles, as it treats its cause, that is, thickened periosteum. The use of a tissue expander contributes to thinning of the skin and reductions in the depths of rhytids.

The syndrome described herein is rare and presents with phenotypic variability. Reports of new cases and treatments of pachydermoperiostosis (Touraine-Solente-Golé syndrome)
are important to provide new individualized treatments according to the deformities presented.

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This research was funded by the authors. In this case, we used a rectangular tissue expander measuring 8.5 × 5 cm with a 200 mL volume, costed by the public health system. The authors do not have any association with the tissue expander manufacturer and will not make any profit from the sale of tissue expanders. Published with written consent of the patient.

**CONFLICT OF INTEREST**
The authors declare no commercial associations or financial relationships that might create a conflict of interest.

**AUTHOR CONTRIBUTION**
DJDC: participated in surgery, patient management, and drafting the manuscript. RMRP: participated in surgery, patient management, and drafting the manuscript. JLDVN: participated in surgery, patient management, and drafting the manuscript. EXOJ: participated in surgery, patient management, and drafting the manuscript. JLMR: participated in surgery, patient management, and drafting the manuscript.

**ETHICAL APPROVAL**
This study was approved and complied with the World Medical Association Declaration of Helsinki on Ethical Principles for Research Involving Human Participants. Written free and informed consent was taken.

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