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## Review

# Treatment of the benign inverted nipple: A systematic review and recommendations for future therapy



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BREAST

Q.M. Hernandez Yenty <sup>a</sup>, W.J.F.M. Jurgens <sup>a</sup>, P.P.M. van Zuijlen <sup>a, b, c</sup>, H.C.W. de Vet <sup>d, e</sup>, P.D.H.M. Verhaegen <sup>a, \*</sup>

<sup>a</sup> Department of Plastic, Reconstructive and Hand Surgery, VU Medical Center, Amsterdam, The Netherlands

<sup>b</sup> Department of Plastic, Reconstructive and Hand Surgery, Red Cross Hospital, Beverwijk, The Netherlands

<sup>c</sup> MOVE Research Institute, VU, Amsterdam, The Netherlands

<sup>d</sup> Department of Epidemiology and Biostatistics, VU Medical Center, Amsterdam, The Netherlands

<sup>e</sup> EMGO Institute for Health and Care Research, VU Medical Center, Amsterdam, The Netherlands

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#### ABSTRACT

The inverted nipple is a frequently encountered problem which can cause difficulties with breastfeeding, sexuality, and aesthetic dissatisfaction. Up to now, no consensus exists on a preferred treatment method. We performed a systematic review to identify the best treatment method for correction of benign inverted nipples. Treatment techniques were subdivided in the categories lactiferous duct preserving and lactiferous duct damaging.

A systematic review was performed using the PRISMA statement. Inclusion criteria were: female patients with congenital or acquired inverted nipples, a minimum sample size of 10 nipples, and studies reporting recurrence of inversion with a minimum follow-up of six months. Exclusion criteria were nipple inversion caused by malignancy.

Thirteen studies met the inclusion criteria which all had a level of evidence IV. No non-invasive treatment techniques were identified. In the duct preserving category eight studies were included with a recurrence rate of 0.6% (2/350) versus 9.9% (16/161) in the duct damaging category (n = 5). Other outcome parameters were not systematically reported in all studies.

Because of a small number of low quality studies with heterogeneous interventions and outcomes a meta-analysis could not be performed and no preferred treatment method was identified. Based on the available data there is no statistical evidence that duct damaging treatment is superior to duct preserving treatment. We recommend that the first method of choice should be a duct preserving treatment method. In the future, more studies of better methodological quality are required and recommendations were made on how these could be conducted.

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#### Introduction

The benign inverted nipple is a common phenomenon with a prevalence amongst women ranging from 1.7 to 3.5% [1–3]. The pathogenesis of the inverted nipple remains controversial: authors reported that inverted nipples may result from a failure of the underlying mesenchym to proliferate and to push the nipple out of its normal depressed position [3,4]. However, Han and Hong did not structurally find this soft tissue deficiency in the nipples that they

\* Corresponding author. VU Medical Centre, Department of Plastic, Reconstructive, and Hand Surgery, De Boelelaan 1118, 1081 HZ Amsterdam, The Netherlands. *E-mail address:* paulineverhaegen@hotmail.com (P.D.H.M. Verhaegen). histologically investigated [5]. Others currently agree that the major basis for inverted nipples is shortened, undeveloped breast ducts, combined with resistant collagen fibers [6,7]. The average projection of the nipple was measured to be 0.9 cm based on a study of 600 nipple measurements in adult women [2]. The manifestation of inverted nipples is classified based upon appearance and the ability of manually pulling out the nipple. The grading system according to Han and Hong is most often used in literature and classifies three degrees of nipple inversion (Table 1) [5]. Inverted nipples can cause functional problems such as difficulty or inability to breastfeed, problems with respect to sexuality, and aesthetic dissatisfaction [6,8–10]. For this reason patients often wish to undergo treatment for the correction of their nipple inversion.



Table	1
Table	

Grading system of nipple inversion developed by Han and Hong. Lactiferous ducts are indicated by vertical lines, fibrosis is indicated by small x [5].

Grade of nipple inversion	Preoperative	After traction	Clinical findings
I			The nipple can be easily pulled out manually and maintains its projection quite well. There is minimal or no fibrosis.
II			The nipple can be pulled out manually, but not as easily as in grade I. The nipple has difficulty maintaining its position and tends to retract. There is a moderate degree of fibrosis and mildly retracted lactiferous ducts.
Ш			The nipple is severely inverted and retracted. It is very difficult to pull out the nipple manually. Despite application of pressure on the nipple to force it to protrude, it promptly retracts. The fibrosis is remarkable and there are short and severely retracted lactiferous ducts with insufficient soft tissue.

According to Long et al. an ideal method of correcting inverted nipples should meet the following requirements: 1) regain normal shape; 2) keep visible scars at a minimum; 3) maintain normal sensation; 4) maintain an intact ductal system (enabling breastfeeding); 5) low recurrence rate; and 6) easy to perform [11]. Besides these six requirements, the patients' wishes are relevant in the decision-making process, e.g. 1) is there a preference for an invasive or non-invasive treatment method? 2) is there a wish to breastfeed (in the future)? 3) does the patient object to a treatment which may take several months? Since the first surgical correction of the inverted nipple by Kehrer in 1879 [12] a great variety of invasive and non-invasive treatment methods have been described. However, up to now no consensus exists on the preferred treatment method. There is no information available which treatment method has the lowest recurrence with the least complications. Therefore, the aim of this study was to assess the available literature on treatment methods of the benign inverted nipple and to identify a superior treatment method with respect to the recurrence and complication rates. Treatment methods were subdivided into two categories based on technical characteristics: lactiferous duct preserving and lactiferous duct damaging treatment techniques. Duct preserving treatment can be either a non-invasive treatment method (such as rubber bands or an external suction device) [8,9,13,14] or an invasive technique in which the lactiferous ducts are not damaged and fibrosis is not removed. Duct damaging treatment is characterized by an invasive technique in which the lactiferous ducts are damaged and fibrotic strands are transected. This categorization was performed: 1) to provide more evidence whether a duct preserving or a duct damaging treatment method is superior with respect to the recurrence rate; and 2) to provide a superior treatment method for women wishing to breastfeed, whereby a duct preserving therapy is preferred.

#### Materials and methods

To assure a systematic method of procedure the PRISMA statement (Preferred Reporting Items for Systematic Reviews and Meta-Analyses) was used [15,16].

#### Search strategy

Electronic searches were performed until August 29th 2015 in Pubmed, Embase, and Cochrane databases. Search terms included were "inverted nipple", "retracted nipple", and "depressed nipple". The included MeSH terms were "Nipple/Surgery" and "Nipple/Deformities". In addition, all references of the included articles were checked to identify additional relevant studies.

#### Inclusion and exclusion criteria

Studies investigating treatment methods of patients with congenital, acquired, bilateral, or unilateral nipple inversion were considered for inclusion. Both non-invasive and invasive treatment methods were considered. The minimum sample size was set at 10 nipples. Although the authors consider a sample size of 10 still relatively low, we anticipated that otherwise the eligible amount of studies would be too low for a systematic analysis. Inclusion and exclusion criteria are summarized in Table 2.

The one outcome parameter that was obligatory for inclusion of the study was registration of the recurrence of nipple inversion. In addition the following outcome parameters were systematically scored: nipple projection, nipple necrosis, nipple sensibility, ability to breastfeed after treatment, infection, hematoma, other complications, and patient satisfaction. Aforementioned clinical outcomes were selected based on requirements for an ideal method of correcting inverted nipples [11].

#### Data collection and extraction

Studies were collected and de-duplicated using Reference Manager 12 (Thomson Reuters, New York, USA). The selection process comprised three stages: selection by title, review by abstract, and review by full text. One author (QH) assessed all titles and abstracts identified by the literature search. The same titles and abstracts were assessed by one of the two other authors (PV and WJ) who each assessed half of the titles and abstracts. Potentially relevant studies were identified and full text articles were retrieved for final selection. Full text screening took place in the same fashion. If there was any disagreement between the reviewers, the final decision was reached by consensus, and when agreement could not be reached a fourth author (PvZ) decided upon inclusion.

Three authors (PV, QH, and WJ) independently extracted the following data from all included full text articles: study design, year of data collection, sample size, mean age, gradation of nipple inversion, nipple projection, and the type of treatment method. Complications such as nipple necrosis, loss of nipple sensibility,

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