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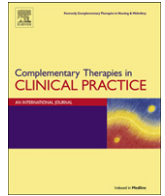


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Exploring the impact of osteopathic treatment on cranial asymmetries associated with nonsynostotic plagiocephaly in infants

Sylvie Lessard^a, Isabelle Gagnon^{b,c,*}, Nathalie Trottier^a

^a Collège d'Études Ostéopathiques de Montréal, 7400, boul. Saint-Laurent bureau 211, Montréal, Qc, H2R 2Y1 Canada

^b School of Physical and Occupational Therapy, McGill University, 3630 promenade Sir William Osler, Montreal, Qc, H3G 1Y5 Canada

^c Trauma and Child Development Programs, Montreal Children's Hospital, 2300 Tupper, Montreal, Qc, H3H 1P3 Canada

A B S T R A C T

Keywords:

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Objectives: To document the evolution of cranial asymmetries in infants with signs of nonsynostotic occipital plagiocephaly (NSOP) who were to undergo a course of four osteopathic treatments (in addition to the standard positioning recommendations) as well as to determine the feasibility of using this methodology to conduct a randomized clinical trial investigating the impact of osteopathic intervention for infants with NSOP.

Design: Pilot clinical standardization project using pre-post design in which 12 infants participated. Ten infants presented an initial Oblique Diameter Difference Index (ODDI) over 104% and five of them had an initial moderate to severe Cranial Vault Asymmetry (CVA) (over 12 mm).

Interventions: Infants received four osteopathic treatments at 2-week intervals.

Main outcome measures: Anthropometric, plagiocephalometric as well as qualitative measures were administered pre-intervention (T1), during the third treatment (T2) and two weeks after the fourth treatment (T3).

Results: Participants showed a significant decrease in CVA ($p = 0.02$), Skull Base Asymmetry (SBA) ($p = 0.01$), Trans-Cranial Vault Asymmetry (TCVA) ($p < 0.003$) between the first and third evaluations.

Conclusions: These clinical findings support the hypothesis that osteopathic treatments contribute to the improvement of cranial asymmetries in infants younger than 6.5 months old presenting with NSOP characteristics.

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1. Introduction

Occipital plagiocephaly is usually described as a cranial deformity involving primarily a unilateral flattening of the occiput. Literature indicates two types of plagiocephaly: (1) synostotic plagiocephaly (malformation due to premature fusion of the cranial sutures)¹ and (2) nonsynostotic occipital plagiocephaly (NSOP).^{2–4} NSOP is generally defined as including the presence of one or more of the following: a Cranial Vault Asymmetry (CVA) of 3 mm or more,⁵ a Trans-Cranial Vault Asymmetry (TCVA) of 4 mm or more² and an Oblique Diameter Difference Index (ODDI) of 104% or more.^{6,7} NSOP is reported to be caused by mechanical strains such as traction or compression applied before, during or after birth, therefore causing a unilateral flattening of the occiput (Fig. 1).^{8–10} Since the “Back to sleep” campaign initiated by the American

Academy of Pediatrics (AAP) in 1992 to reduce the incidence of Sudden Infant Death Syndrome,¹¹ this type of skull deformity has become more prevalent and could be as high as 19.7% of healthy newborns.¹⁰

A number of observational studies suggest that plagiocephaly may be related to delays and impairments in various aspects of an infant's functioning.^{12,13} Available findings show that children with this condition are more prone to developing problems related to: postural compensations,^{14,15} musculo-skeletal dysfunction,¹⁶ visual perception and ocular dysfunction^{17,18}; temporo-mandibular articulation¹⁹ and developmental achievements.^{20,21} Despite popular belief, research findings suggest that without intervention, NSOP, in the majority of cases, does not resolve in time.^{22–24} Interventions for NSOP usually include: counter-positioning indicated for mild cases of plagiocephaly,²⁵ physical therapy added to counter-positioning when torticollis is associated with the presence of skull deformity,²⁴ and cranial orthosis, particularly for moderate to severe cases of NSOP or when counter-positioning has failed to correct the asymmetries.^{14,26,29} Earlier intervention has been shown to increase the potential for NSOP correction.^{16,24,26,27,30,31}

* Corresponding author. McGill University Health Center-Montreal Children's Hospital, Trauma Program, 2300 Tupper, Montreal, Canada H3H 1P3. Tel.: +1 514 412 4400x22001; fax: +1 514 412 4398.

E-mail address: isabelle.gagnon8@mcgill.ca (I. Gagnon).



Fig. 1. Characteristic shape of plagiocephaly in a 2 month-old infant.

In addition to traditional therapies, parents have been found to seek alternative modes of intervention such as osteopathy. Osteopathic manual therapy recognizes the importance of treating the body as a whole functional unit. It aims to optimize symmetry in the growing child. Some key elements of the osteopathic treatment for NSOP thought to be particularly useful include; the normalization of the skull base and particularly the strains, the optimization of vertebral alignment and normal mobility of the head/neck (without using thrust technique), the normalization of cranial membranes, cranial sutures and intraosseous lesions.

Osteopathic manual studies include in its curriculum the treatment of the cranium as part of its total body approach. The bones of the cranium start out as tiny centers of ossification scattered throughout a connective tissue matrix. At birth, many of these bones are in parts and most of them are still cartilaginous. There are six major fontanelles, located between adjoining bones of the cranial vault. The sutures between the vault bones are quite plastic and flexible, so the bones may overlap each other during the delivery process. The goal of osteopathic treatment is to remove any impediments to these mechanisms and facilitate the homeostatic processes of the body.^{32,33}

The objectives of this pilot project were to document the evolution of cranial asymmetries in infants with signs of NSOP who were to undergo a course of four osteopathic treatments (in addition to the standard positioning recommendations) as well as determining the feasibility of using this methodology to conduct a randomized clinical trial investigating the impact of osteopathic intervention for infants with NSOP.

2. Methods

2.1. Participants

Twelve infants (mean age 4.1 months \pm 34 days) with signs of NSOP participated in this project. The age for initial consultation was consistent with that reported in the literature for initial presentation.²⁴ Infants evaluated and treated in the project were either referred by pediatricians or family doctors or seen through direct access. Inclusion criteria for the infants were: to be younger than 6.5 months at the time of the first evaluation, to have a diagnosis or present signs of NSOP (as per recognized definition) and to be at term corrected age if born prematurely. Infants were excluded if there was a documented craniosynostosis, an ongoing cranial orthosis treatment and if presenting with any medical condition

Table 1
Clinical characteristics of participants at baseline.

Characteristics		%
Sex	Male	75
	Female	25
Side of occipital flattening	Right	92
	Left	8
Ear displaced anteriorly	Right	92
	Left	8
Age at first osteopathic assessment (month)	Less than 4 months	58
	Between 4 and 6.5 months	42
Mean age (month)	4.1 months	
Sleep position	Supine	100
Supine position (hours/24 hours)	20–24	58
	15 to <20	33
	0 to <15	8
Restriction in cervical mobility (rotation)	Right > Left	8
	Left > Right	92
Delivery	Vaginal delivery	83
	Cesarean section	17
Birth presentation	Vertex	83
Mother's first pregnancy		7

judged inappropriate by a physician (e.g. an infant with lower extremity casts, vertebral or skull birth defects). The characteristics of the infants are presented in Table 1.

2.2. Measures

All measures used for this clinical standardization project were those used as part of standard care for infants presenting with NSOP in our institution. At the initial assessment, as done for all infants seen in this clinic, a routine detailed history was taken including obstetrical, perinatal and postnatal information. In the context of standard assessment for this clientele, tests administered to the infants included anthropometric and plagiocephalometric measurements. Anthropometric measurements, to document the CVA, the SBA and the TCVA, were done using a digital spreading caliper.^{d,3,34} Landmarks are presented in Fig. 2. This method was shown to have good intra-rater reliability and is recognized for its ease of administration in clinic, its safety and low cost.⁵ Clinical adaptation of plagiocephalometric measurements, to document the evolution of the oblique diameter left (ODL) and oblique diameter right (ODR), were taken via cranial circumference molds using a low-temperature thermoplastic material (sansplint), as described by Van Vlimmeren et al.^{6,7} This is a non-invasive, reliable, easy to apply method to assess skull asymmetry with low cost application.⁶ From the ODL and ODR measurements, an Oblique Diameter Difference Index was derived and was used for analyses. Osteopathic evaluations consisting of mobility, vitality and positioning of different anatomical structures of the child's body in a holistic manner were also included. Finally, a parent questionnaire, collecting information on general health of the child, pregnancy condition of the mother, and perinatal data was administered and parents were also asked to fill a daily log of infant activity.

Measurements were performed at three different times: Prior to the intervention (pre-test at T1), at the time of the third treatment (T2) and two weeks after the fourth treatment (T3). A blind evaluator performed all the measurements in the clinic where children

^d In our practice, an engineer guided the team to permanently fix the arms (with rounded tips) of a spreading caliper (ERP Group, #1062) to a digital caliper with a resolution of 0.01 mm.

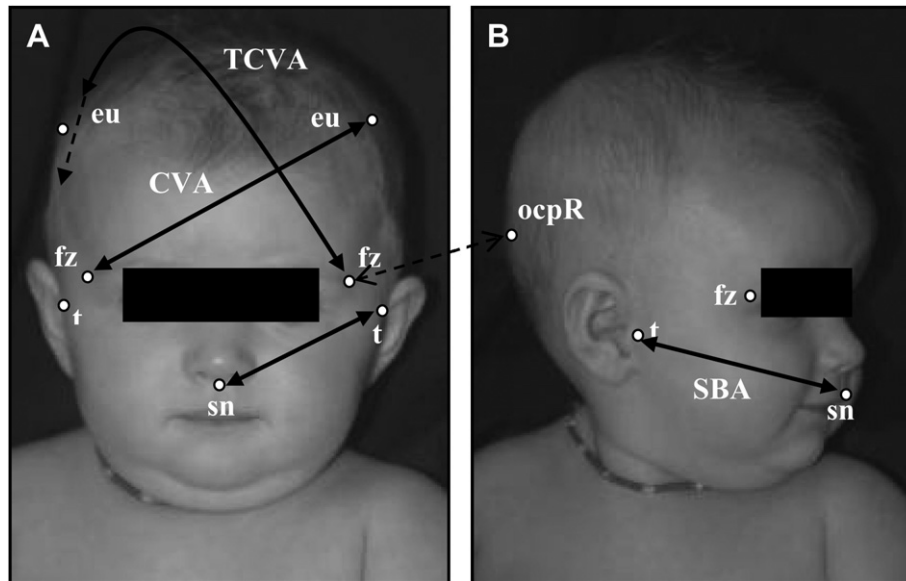


Fig. 2. Frontal (A) and lateral (B) views of the main measurements and anthropometric landmarks used to quantify asymmetries in NSOP, adapted from Kolar and Salter (1997).³⁴ Skull Base Asymmetry (SBA), sn-t R/L: Difference of measurements between the subnasale (sn) landmark (junction of the septum nasal and skin of superior lip in the midline) and the tragus (t) (located at the notch above the tragus of the ear, the cartilaginous projection in front of the external auditory canal, where the upper edge of the cartilage disappears into the skin of the face). Cranial Vault Asymmetry (CVA), fzR-euL, fzL-euR: Difference of measurements between frontozygomaticus (fz) (most lateral point on the frontozygomatic suture) and eurion (eu) (most lateral point on the head in the parietal region). Trans-cranial vault asymmetry (TCVA), fzR-ocpL, fzL-ocpR.^{1,14,24,39} Difference of measurements of the longer and the shorter oblique diameter (from the frontozygomaticus-occipital prominence/flatness).

were receiving the intervention. This project had received approval from the Collège d'Études Ostéopathiques de Montréal and all parents reviewed and signed informed consent forms for their child's data to be included in our analysis.

2.3. Intervention

Each infant received four osteopathic treatments (of 60 mins) scheduled 15 days apart (± 4 days). Osteopathic manipulative therapy (OMTh) is defined as a therapeutic application of manually guided forces by an osteopath (non-physician) to improve physiological function and homeostasis that has been altered by somatic dysfunction. By addressing the whole body, the primary dysfunction and the main tensions will be treated using direct correction techniques (see Appendix A for glossary of techniques). The frequency and content of OMTh provided were similar to those provided routinely to that clientele. Standard recognized protocols for identifying osteopathic problems and determining osteopathic treatment content were followed as per recommendations from the Collège d'Études Ostéopathiques, Montréal, Canada. OMTh employs a wide variety of techniques and the most frequently used were interosseous de-compression techniques and suture releases, intraosseous moulding work, correction of sphenobasilar symphysis strains (mostly lateral), vault normalization, basilar expansion, membranous reciprocal tension techniques, occipital condyles and falx cerebri normalization, myofascial release techniques, treatment of the pelvic and shoulder girdles. Advice on counter-positioning and stimulation such as those suggested by the CPS⁴² were demonstrated by the clinician, practiced by parents and given in writing to optimize time spent in prone during awake hours while supervised as well as to stimulate rotation of the head on the side opposite to the flattening.

2.4. Data analysis

Data from all testing sessions were compiled visually and analyzed for trends. To address the study objectives, measures of

the infants at each testing session were compared using one-way ANOVAs with repeated measures on the time factor. Specific differences between each assessment times were analyzed using paired *t*-tests (T1–T3; T3–T5; T1–T5). SAS software was used for analyses.^e

3. Results

All 12 infants enrolled in this project were able to complete the assessments and interventions included in this project, thus confirming the feasibility of using such a methodology to undertake a clinical trial. The evaluations revealed that 11 of the participants presented a right occipital flattening, and this is consistent with what is reported in the literature.^{2,29,35}

The descriptive statistics for anthropometric measures are presented in Table 2. The means of all three anthropometric indices measured (CVA, SBA, TCVA) were decreased over the follow-up period. From an anthropometric aspect, the ANOVA performed on the CVA ($F = 5.20$; $p = 0.02$), SBA ($F = 5.72$; $p = 0.01$) and TCVA ($F = 7.97$; $p < 0.003$) all revealed a main effect for the time factor, present both between T1 and T2 as well as between T2 and T3.

When assessed with plagiocephalometric measures, 10 of the participants presented an Oblique Diameter Difference Index (ODDI) of $>104\%$, a value recognized as clinically significant illustrating obvious clinical asymmetry.^{6,7} Over the course of the four treatments, the ODDI of 8 of those 10 participants decreased below the 104% threshold. The average ODDI of the 12 participants evolved from 107.9% (T1) to 103.9% (T3). The ANOVA performed on the ODDI revealed no time effect ($F = 2.79$; $p = 0.08$) but there was a tendency towards significance between T1 and T3 values as illustrated by the paired *t*-test values performed on those times alone ($t = 2.35$; $p = 0.03$). Furthermore, visual examination of the moulds suggests that increased skull growth over the course of the follow-up period was primarily at the site of the occipital flattening.

^e SAS version 9.1, 2006, North Carolina.

Table 2
Anthropometric and plagioccephalometric outcomes measures at T1, T2 and T3.

Variable	Pre-treatment (T1) (mean ± SD)	After 3rd treatment (T2) (mean ± SD)	p (Paired t-test T1–T2)	Post-treatment (T3) (mean ± SD)	p (Paired t-test T2–T3)
SBA (mm)	5.1 (±2.5)	2.5 (±2.1)	0.012	2.2 (±2.0)	0.006
TCVA (mm)	12.7 (±5.3)	9.0 (±3.3)	0.048	5.6 (±3.6)	<0.001
CVA (mm)	7.4 (±4.2)	3.3 (±3.1)	0.030	2.1 (±1.6)	0.006

3.1. Secondary outcomes

Although those were not performed by a blind evaluator and could not be used for statistical analyses due to their lack of psychometric properties, osteopathic assessments revealed that the right sided occipital flattening present in 11 of the participants was associated with a right lateral strain for seven of them just as suggested by Sergueef (2006)³⁰ who found a correlation between flattening of the occiput (right/left) and the lateral strain pattern of the sphenobasilar symphysis. Also linked with the occipital flattening, was a limitation in assisted active cervical rotation to the side opposite of the flattening in all the infants. Finally, as reported by Solano (2002),³⁷ we noticed that in the presence of NSOP, the dura-mater membranes were not in balanced tension (this situation has been noted for the 12 infants).

4. Discussion

The main objective of this pilot project was to document the evolution of cranial asymmetries in infants with signs of NSOP who were to undergo a course of four osteopathic treatments (in addition to the standard counter-positioning recommendations). The overall findings suggest that asymmetries found at the time of initial exam were significantly decreased over the short follow-up period in which 12 infants received the osteopathic intervention.

Results of the anthropometric measurements targeting the CVA, SBA and TCVA were all found to show significant decrease after the fourth treatment. In addition, all three measured asymmetries were also found to have significantly decreased between T1 and T2 suggesting a noticeable improvement after only two treatments. We notice, as did Terpenning et al. (2001),³ that the relative correction (% correction from T1 to T3) of the SBA occurs relatively more slowly than the other asymmetries. We believe that the fact that the base is more resistant to the correction may be partially explained by its cartilaginous embryological origin in contrast to the membranous origin of the skull vault.³⁸

The anthropometric measurements of the TCVA appears to be the most clinically relevant since it allows the visualization of the parallelogram pattern and the degree of asymmetry (occipital prominence/flatness) for which parents usually seek interventions. Many others^{1,24,39} refer to the TCVA measurement with which practitioners can notice the evolution of the parallelogram head shape characterizing NSOP. According to Biggs (2004)⁴⁰ the “parallelogram” shape is pathognomonic for plagiocephaly and it was indeed present in every child in our sample ($n = 12/12$). Moreover, TCVA is used by some insurance companies to determine the severity threshold for treatment²⁴ (e.g. Fallon Community Health Plan).

Results obtained from the clinical adaptation of plagioccephalometric measurements revealed significant improvement in the ODDI throughout the follow-up period. The molds are usually found to be clinically relevant as they reflect the parallelogram shaped head. They enable parents and service providers to objectively and easily note the evolution of asymmetries. Vlimmeren et al. defined an ODDI score of 104% or higher as clinically relevant asymmetry of the skull.⁴¹ Although plagioccephalometry allows

only for bi-dimensional measurements, the decrease of the ODDI under the 104% threshold among eight participants after only four treatments, was in fact clinically associated to parents satisfaction regarding the esthetic aspect of the problem.

From an osteopathic point of view, the observable aspects characterizing NSOP are only part of the problem. Osteopathic philosophy suggests that the compression forces which deforms the skull are absorbed and transmitted to the body as a functional unit and because of this, the global aspect of osteopathic treatment deserves particular attention. The impact of the force which contributed to deforming the skull is not thought to be limited to the level of the head or cervical region. The NSOP is a problem of asymmetry and the flattened occiput transforms the axes and planes of movements of the condyles with regard to the atlas.³⁰ These asymmetries of the occiput may create postural compensations in order to ensure horizontal gaze which the body strives to maintain. Asymmetries and immaturity of certain movement components were also noted in our sample and by correcting the main dysfunctions (in order of primary) found in each infant, all of them showed improvement of cervical mobility and tolerance in prone.

Recommendations of American Academy of Pediatrics (1996)¹¹ and Canadian Pediatric Society and Canadian Paediatric Society (2004)⁴² to place infants in supine to sleep in order to prevent sudden infant death syndrome are largely implemented. It is the responsibility of all service providers to inform parents of the importance of prone play during awake hours. Once the diagnosis of NSOP is made however, our results suggest that early referral to osteopathy professionals could improve asymmetries and facilitate recovery.

Because of the small sample size and the absence of a control group, no definitive conclusions can be made that the decrease in asymmetry observed in our sample were due to the osteopathic treatment alone. However, it is reasonable to say that any improvement superior to that reported with positioning alone, the widely accepted intervention for NSOP, could be attributed, at least partially, to the intervention. Indeed, infants in our sample improved their CVA by 62%, their SBA by 48%, and their TCVA by 60%, as well as improving their ODDI by 4%. Other studies measuring the efficacy of helmet therapy reported that the asymmetry of the CVA diminished of 41.56%²⁹ and 62.5%²⁶ using anthropometric measurements. Mulliken et al. (1999)¹ recorded an improvement of the TCVA (42%) in 53 infants either positioned or helmeted (mean follow-up 4.6 months). Vlimmeren et al. (2008)⁴¹ using plagioccephalometry, measured a decreased of the ODDI of 1.5% in the experimental group after a four month standardized intervention program of pediatric physical therapy (children with positional preference aged between 7 weeks and 6 months, children with congenital muscular torticollis were excluded) in comparison to an increase of 0.2% in the control group who received care as usual. While comparing the results from the present pilot study to the previous research on the subject it suggests that osteopathic treatments could contribute to modifying the cranial asymmetries of infants younger than 6.5 months old presenting with signs of NSOP. A larger sample, using a randomized controlled design is recommended to confirm the results of the

present exploratory study. Furthermore, our clinical experience and our understanding of the problem leads us to believe that there is a need for more research of the impact of skull asymmetries on the child's development.

5. Conclusion and clinical impact

This project demonstrated statistically and clinically significant improvements in cranial asymmetry of infants with NSOP following four osteopathic treatments (in addition to the standard positioning recommendations), provided over a two month period. These results suggest that osteopathic treatments could contribute to modifying the cranial asymmetries of infants younger than 6.5 months old presenting with signs of NSOP. Though it must be considered a preliminary project, to our knowledge, this is the first report to objectively measure the changes in plagiocephaly with osteopathic treatment. This project successfully utilized clinical procedures that could contribute to the creation of standardized protocol both clinically and with a larger-scale randomized controlled trial.

Conflict of interest

The authors have no conflict of interest to declare.

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Appendix A. Glossary of key osteopathic terms^f

De-compression technique: is utilized to normalize tissue around a segment, especially cranial sutures. The osteopath guides the segments away from one another in a gentle distraction force. Once again the osteopath will hold this position until the tension releases indicating a normalization of tissue stress.³³

Direct method (D/DIR): any osteopathic treatment strategy by which the restrictive barrier is engaged and a final activating force is applied to correct somatic dysfunction. (glossary, p. 29).

Lateral strain of the speno-basilar symphysis: sphenoid and occiput have rotated in the same direction around parallel vertical axes; lateral strains of the SBS are named for the position of the basisphenoid, right or left. (glossary p. 55).

Membranous balance: the ideal physiologic state of harmonious equilibrium in the tension of the dura mater of the brain and spinal cord. (glossary p. 25).

Methodology for osteopathic manipulative therapy (OMTH) according to the Collège d'Études Ostéopathiques: standard recognized protocols for identifying osteopathic problems and determining osteopathic treatment content were followed as per recommendations from the Collège d'Études Ostéopathiques, Montreal, Canada: Restore vitality if needed, treat compressions and intraosseous dysfunctions, correct non physiological dysfunctions without respect to axes, then non physiological dysfunctions with respect to axes, physiological dysfunctions, restrictions and myofascial tensions.

Molding, intraosseous: The technique for ameliorating the abnormality induced in any cranial bone by restrictive mechanical conditions which have prevented the development of the usual contours, or inhibited its proper mechanical function. Such methods involve a slow, gradual restoration of normal shape and function by cooperating with the natural forces of growth and development. See deformity, parallelogram; horn, frontal; "twig, bent."⁴³

Myofascial release (MFR): system of diagnosis and treatment first described by Andrew Taylor Still and his early students, which engages continual palpatory feedback to achieve release of myofascial tissues.

Direct MFR, a restrictive barrier is engaged for the myofascial tissues; the tissue is loaded with a constant force until tissue release occurs. (glossary p. 31).

Normalization: the therapeutic use of anatomic and physiologic mechanisms to facilitate the body's response toward homeostasis and improved health. (glossary p. 27).

Primary somatic dysfunction: primary s. d., (1) The somatic dysfunction that maintains a total pattern of dysfunction. See also key lesion. (2) The initial or first somatic dysfunction to appear temporally. (glossary, p. 54).

Somatic dysfunction: impaired or altered function of related components of the somatic (body framework) system: skeletal, arthrodial, and myofascial structures, and related vascular, lymphatic, and neural elements. Somatic dysfunction is treatable using osteopathic manipulative treatment. (glossary, p. 53).

Sphenobasilar synchondrosis (symphysis), somatic dysfunctions of: Any of a group of somatic dysfunctions involving primarily the inter-relationship between the basilar portion of the sphenoid (basisphenoid) and the basilar portion of the occiput (basiocciput). (glossary, p. 54).

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^f Glossary of Osteopathic Terminology. 2009 AACOM, or by citing the Foundations of Osteopathic Medicine 3rd edition which has the most recent edition of the glossary. <http://publish.aacom.org/resources/Pages/glossary.aspx>.

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