CASE STUDY

Sandifer Syndrome: Improved Health Outcomes in an Infant Undergoing Care for Vertebral Subluxation

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Abstract

Objective: To present a case study of conservative subluxation-based chiropractic care of an 11-week-old female that presented with a group of symptoms associated with Sandifer syndrome.

Clinical Features: An 11-week-old female presented with a history of reflux, digestive problems, and possible seizures. Prescription medications included Zantac. Chiropractic analysis and thermography revealed specific patterns of subluxations.

Intervention & Outcomes: Subluxation-based chiropractic care was performed utilizing the Torque Release Technique® analysis and protocol via the Integrator. Adjustments were delivered specifically at C1, C2, T6, Coccyx and Sacrum accordingly. The patient showed improved developmental changes, reduction in reflux, and better bowel function and is no longer congested, fidgety or having issues with lying supine.

Conclusion: This case describes the chiropractic care for a rare syndrome that is associated with a mirage of symptoms. There are few studies in the chiropractic literature on Sandifer syndrome. Additional research is needed on the chiropractic treatment of Sandifer syndrome to raise awareness and treatment of this condition.

Key Words: *Chiropractic, Sandifer syndrome, Torque Release Technique, gastro esophageal reflux, vertebral subluxation, adjustment, spinal manipulation*

Introduction

Sandifer syndrome (SS) consists of complex contortions of the head and neck, sometimes the chest and abnormal postures[1] seen among children suffering from gastro-esophageal reflux with or without hiatal hernia in an effort to relieve their symptoms.[1] They are thought to be involuntary and due to torsion dystonia.[2] They consist of a sudden extension of the head and neck and the head may be twisted from side to side and the upper part of the trunk bent acutely from side to side.[2]

The initial patients were under the care of a neurologist, Dr. Paul Sandifer[3] and although Sandifer initially observed the association, Kinsbourne and Oxon first reported it based on the observations of Sandifer.[4] The syndrome is most certainly under recognized and delays in diagnosis are due to atypical presentations or cases in which the diagnosis is not part of the differential.[3,4]

The original five children ranged from age 4 years to adolescence. All had hiatal hernia with gastro esophageal reflux (GER) associated with vomiting and abdominal pain. Onset of symptoms ranged from the neonatal period to age 3 years. Hyperextension and lateral position of the neck began as early as age 20-months.[3] Early reports of Sandifer syndrome noted the association of hiatal hernia with abnormal head and neck posturing.[3,4] Since then, other reports have indicated hiatal hernia need not be present even though gastro esophageal reflux is a consistent finding.[4]

The exact pathophysiology is not known.[4] The head and neck posturing is thought to be a response to the pain associated with gastro-esophageal reflux.[5] It is also possible that these children have an acute esophageal sensitivity to refluxed gastric acid.[5]

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Gastro Esophageal Reflux

Gastro esophageal reflux (GER) is defined as the physiologic passage of gastric contents into the esophagus.[6] The phenomenon is very frequent in neonates since it occurs in more than two-thirds of otherwise healthy infants during the first months of life.[6] These reflux events can have an impact on health because between 10% and 60% of infants show GER-related symptoms.[6] Given that vagal activity dysfunction is observed in both the presence and absence of inflammatory changes in the esophagus, some researchers have suggested that parasympathetic dysfunction is the prime factor in the etiology of GER and not just the consequence of esophageal inflammation.[6] It has further been hypothesized that disturbances in ANS activity could impair contraction of the lower esophageal sphincter, which normally acts as a reflux barrier, and may be involved in transient lower esophageal sphincter relaxation and therefore also GER.[6,7] Lastly, it has been suggested that ANS changes have a role in GER in neonates.[8]

Differential Diagnoses

The differential diagnoses include congenital anomalies of cervical vertebrae, congenital muscular torticollis, traumatic causes, non-traumatic inflammatory conditions of head and neck, neurologic disorders and psychological causes.[9] The differential diagnosis also includes dystonia due to cerebral lesions and is obviously important, but it may be equally disastrous if the abnormal movements are thought to be of an epileptic nature.[10] The syndrome has been reported among adult patients, and has been misdiagnosed as a case of refractory partial seizures in an adult.[10]

Case Report

History

A 12-pound 11-week-old baby girl presented to the clinic with reflux, digestive problems, possible seizures, and possible colic. She was diagnosed by her pediatric neurologist and pediatric GI specialist with Sandifer syndrome since birth. She experienced explosive bowel movements since birth but started to become constipated, going 3-5 days without a bowel movement. During her first week of life she would scream uncontrollably, experience or hiccups, was spitting up constantly and would not lay flat. At four-weeks old she experienced episodes of vomiting. A week prior to the office visit, laying supine would cause her hands to shake, eyes bounce, face to become red, and she would scream. Her hands were getting raw from chewing her hands, and she would constantly fidget. The patient was taking Zantac twice per day for reflux and was breastfed.

Prenatal History

During pregnancy the mom had nausea and heartburn for 40 weeks and experienced vomiting into the night for six weeks until 23rd week of pregnancy. The mother also experienced emotional stress from moving into a new home at 30 weeks. The baby experienced in-utero constraint and posterior presentation leading to a more difficult labor. The mother took prenatal vitamins during pregnancy and delivered the baby two hours after an epidural was given at the hospital.

Intervention

Every visit the patient had was recorded using SOAP notes. Subjective, objective, assessment and plan was used to assess the patient progress from visit to visit. On each visit an objective assessment was also recorded in terms of chiropractic findings such as restriction of joint motion at certain spinal levels.

Each visit would consist of a chiropractic evaluation and an adjustment. Static palpation of the bony and soft tissue components were used to determine the presence of edema.[11] Motion palpation is used to identify and find segments that are moving freely or are restricted in any of their normal ranges of motion.[12] It has been determined that motion palpation is a useful assessment tool for location of vertebral subluxation.[12] Dysfunction of the paraspinal musculature is generally accepted as a clinical manifestation of vertebral subluxation.[13]

Rolling paraspinal thermography was performed from C1 to S1 (Figures 1 a,b,c) on initial visit, mid-way through treatment plan and final exam. Due to the neurological control of skin vascularity, temperature differentials can be used as an outcome measure to assess vertebral subluxation.[14] Figure 1a shows severe temperature differences at C1, T3, T4, T5 and T6. Figure 1b shows moderate temperature differences were noted at T2 and T7. Figure 1c shows mild recordings were indicated at C7.

Torque Release Technique

Torque Release Technique (TRT) evolved out of a 1996 university-based study on the associated benefit of chiropractic care with residential addiction patients who were being investigated for signs of Reward Deficiency Syndrome (RDS).[15] This landmark study, co-developed by chiropractic researcher Dr. Jay Holder, required a truly reproducible adjustive thrust that emulated the directional forces of a specific adjustment.[15] This technique is considered to be "tonal" because it relies upon real-time interaction with the nervous system to determine where, when, and how to adjust in order to best reduce abnormal torsional and tensile forces affecting the neuro-skeleton as a whole.[16]

TRT recognizes that both tonal and segmental subluxations are possible, also known as cord pressure and cord tension as described in the Chiropractic Textbook by Stephenson.[17] Cord tension has been described early in chiropractic literature as being a major and distinct component of subluxation, as opposed to nerve pressure or compression.[15,16] These attachment points are the sphenoid, occiput, C1 (indirect attachment), C2, C5, sacrum and coccyx.[18] Holder contends that these locations are where subluxations most commonly occur.[16] Altered tension on the connections of the dura, connective tissue, dentate ligaments, the vertebrae and surrounding spinal structures are what creates this cord tension/tone.[18]

TRT begins with an analysis of the entire system, searching for patterns of subluxation.[15] The nervous system is viewed in its non-linear, non-mechanistic state.[15] The coalesced evaluation utilizes tissue and inter-segmental motion palpation, functional leg length inequality tests, abductor/adductor resistance analysis, foot flare (inversion, eversion, pronation, supination), heel tension, aberrant respiratory analysis, gait and postural

analysis, abnormal thermographic heat radiation, joint pain and tissue tenderness, and assessing for asymmetry of musculature and hypertonicity.[15] The intent of the examination is to rule in the potential location of the primary subluxation most needing to be adjusted at any given point in time.[15,19] The differential diagnosis of the site and vectors of the primary subluxation is then made utilizing advanced functional leg length reflex (FLLR) testing combined with skin contact pressure testing of vertebral contact points with suspected correctional vectors.[15]

In this technique the patient is analyzed in the prone position. TRT focuses on the entire spinal-neural relationship as its analysis identifies the dominant or major subluxation pattern.[15,16,19] In this way, the doctor rarely needs to adjust more than two segments per visit.[15] The detailed analysis also assists in determining the laterality and posteriority of subluxation patterns.[19]

The Integrator

In 1995 a 510K medical device designation was granted by the FDA for the Integrator, which specified that the Integrator was found safe and effective for the correction of vertebral subluxation.[19] The torque corrects the superior or inferior listing of the subluxation with the axial correcting the lateral and/or posterior rotational listing of the subluxation.[19]

It surpasses the specifications of the Toggle Recoil adjustment, delivering a pre-loaded thrust in 1/10,000th of a second at 64 Hz with recoil, and left or right torque included in the correctional thrust.[19,20] When the instrument has been loaded, placed on the appropriate contact point and the correct amount of pressure is applied, it is fired independently of the practitioner allowing for true inter-professional reproducibility, and signals to the chiropractor and patient that a specific adjustment has been delivered.[19]

Outcomes

After the 7th visit receiving chiropractic specific adjustments at each visit using TRT protocol, the patient showed more frequent bowel movements, decreased reflux, decreased gagging, decreased spitting up and was able to lay flat/supine. The following chiropractic listings were used to better define the subluxation pattern: C1 LA CCW (lateral atlas, counter-clockwise) and lateral sacrum counter-clockwise. A high velocity thrust with recoil using the Integrator was delivered to the patient.

On the 10th office visit it was noted that the patient began sleeping in her crib for six hours a night and having bowel movements daily. The patient was adjusted using TRT protocol and spinal palpation revealed areas of spasm and hypo-mobility indicative of subluxation at right C1 and right coccyx.

On the 12th visit a progressive evaluation was given asking the mother what changes she noticed in her child since beginning care, including physical, emotional, behavioral and immune changes. Results are as follows: less congested, no more episodes of shaking, better during tummy time, less fighting when eating, better sleeping and more frequent bowel movements. Patient exhibited more energy, elation and overall playfulness. The rolling thermal scan (Figure 2 a,b,c) was also performed indicating the following; severe temperature difference at T2, T3, T4, T6, T7; moderate temperature differences at C6; and mild at C7 and S1.

After the 30th office visit the patient was no longer throwing arms up when placed on her back, was not congested, nor having issues with laying and fidgety. Developmentally she was ahead where prior she was slightly behind. She was now rolling around and sitting up unassisted. Bowel function was now every three days and the mother temporarily switched to a dairy-free diet. Prior to chiropractic care and dietary changes she only went once a week. Behavioral changes included a happier mood and less agitation. Figure 3 (a,b,c) shows rolling thermal scan, with mild differences at C1, C2, T4, T5, S1 and with moderate temperature difference at T6.

Discussion

Conventional Treatment

Early recognition and understanding of this condition allows successful treatment and resolution of the symptoms and avoids costly and unnecessary neurologic evaluation.

In one case management of an infant with Sandifer syndrome, medication was given including Domperidon, Sodium Alginate and Lansoprasole.[21] The paroxysmal dystonic behaviors were dramatically diminished with treatment, but not completely resolved. Cow's milk was removed from her diet and after three days all of the movements disappeared.[21] Fifteen days later cows milk was introduced to the diet of the patient and the dystonic movements started again.[21] Cow's milk was removed once again from her diet and movements disappeared.[21] So, SS was diagnosed due to cow's milk protein allergy.[21] Improvement or disappearance of symptoms on a CMP-free diet adds substantial evidence to the diagnosis.[21]

If the reintroduction of CMP causes relapse of symptoms, the diagnosis seems established, because a challenge test is considered as the golden standard diagnostic test.[21,22] Corrado et al reported a Sandifer syndrome in a breast fed infant.[23] They proposed food allergy to dietary proteins ingested by a lactating mother may play a role.[23]

In another case a 2-year-old female was referred for otolaryngology evaluation.[24] Neonatal history was negative except for frequent irritability and 'spitting up'.[24] Gastro-intestinal consultation suggested a diagnosis of Sandifer syndrome and medical management was initiated with upright positioning after eating and frequent reduced volume feedings.[24] Medical management also included a brief course of Bethanecol at 8.7 mg/meter' per 24 h in two divided doses to increase upper sphincter tone and cimetidine at 20 mgIkg per day in four divided doses.[24]

After six months of no improvement, Nissen fundoplication was performed.[24] This procedure surgically wraps the upper end of the stomach around the lower esophagus and sutures it anteriorly creating a circular acute angle valve mechanism.[24] This also maintains the lower esophagus in the abdomen and the hiatus is closed. Reflux was controlled over a 3-6 month interval.[24]

Chiropractic Mechanism

There is no chiropractic literature on Sandifer syndrome. According to Kent's dysafferentation model, neurological dysfunction associated with the vertebral subluxation may take other forms.[25] The intervertebral motion segment is richly endowed by nociceptive and mechanoreceptive structures.[25] As a consequence, biomechanical dysfunction may result in an alteration in normal nociception and/or mechanoreception.[25]

Some evidence has emerged that the abnormal movements in this syndrome are due to any dysfunction of the basal ganglia.[1,26] It is known that the motor nerve supply of the diaphragm arises from the cervical 3, 4, and 5 roots and that its sensory nerve supply almost certainly comes from the same roots.[1,23] As these roots also supply muscles such as the trapezius, sternomastoid, and the scalenus medius which control movements of the head and neck, it may well be that abnormal stimuli caused by the hiatus hernia at the level of the diaphragm could reflexly cause abnormal movements.[1,23]

Conclusion

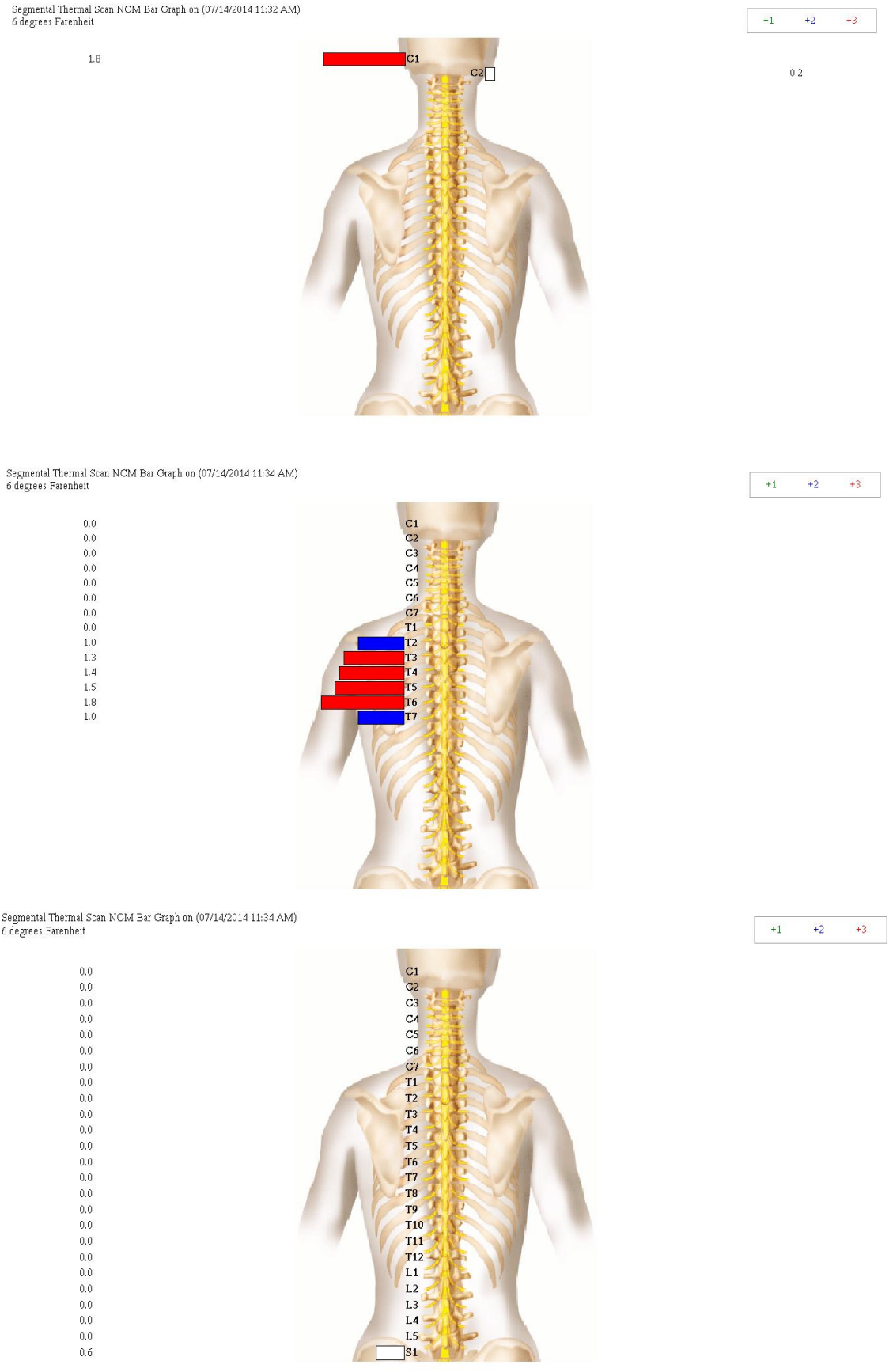
Sandifer syndrome has many different symptoms that should be looked at individually before any intervention is taken in order to give the patient the proper treatment needed. This is the first reported case in the literature on chiropractic, vertebral subluxation and Sandifer syndrome.

References

1. Gordon N. Sandifer's syndrome: Investigations and treatment. Journal Of Pediatric Neurology [serial on the Internet]. (2007, Oct), [cited December 9, 2014]; 5(4): 275.
2. S.L. Werlin, B.J. D'Souza, W.J. Hogan, W.J. Dodds and R.C. Arndorfer, Sandifer syndrome: an unappreciated clinical entity,Dev Med Child Neurol 22 (1980), 374–378.
3. Kinsbourne M, Oxon DM. Hiatus hernia with contortions of the neck. *Lancet* 1964; i: 1058-61.
4. Lehwald N, Krausch M, Franke C, et al. Sandifer syndrome -- a multidisciplinary diagnostic andtherapeutic challenge. *Eur J Pediatr Surg* 2007;17(3):203-6.
5. M. Olguner, F.M. Akgur, G. Hakguder and T. Aktug, Gastroesophageal reflux associated with dystonic movements: Sandifer's syndrome, Pediatr Int 41 (1999), 321–322.
6. Djamal D., Guy K., Blanchard E., Ammari M., Delanaud S., Bach V., Telliez F.,Involvement of Autonomic Nervous Activity Changes in Gastroesophageal Reflux in Neonates during Sleep and Wakefulness .2013. DOI: 10.1371/journal.pone.0083464Involvement
7. Dobrek L, Nowakowski M, Mazur M, Herman RM, Thor PJ (2004) Disturbances of the parasympathetic branch of the autonomic nervous system in patients with gastro esophageal reflux disease (GERD) estimated by short-term heart rate variability recordings. J Physiol Pharmacol 55: 77-90.
8. Tirosh E, Ariov-Antebi N, Cohen A (2010) Autonomic function, gastro esophageal reflux in apparent life threatening event. Clin Auton Res 20: 161-166.
9. H. Mandel, E. Tirosh and M. Berant, Sandifer syndrome reconsidered,Acta Paediatr Scand 78 (1989), 797–799.
10. M. Shahnawaz, L.R. van der Westhuizen and R.F. Gledhill,Episodic cervical dystonia associated with gastro-oesophageal reflux. A case of adult-onset Sandifer syndrome, *Clin Neurol Neurosurg* **103** (2001), 212–215.
11. Haneline MT, Young M. A review of intraexaminer and interexaminer reliability of static spinal palpation: A literature synthesis. J Manipulative Physiol Ther. 2009; 32(7)379-55.
12. Lakhani E, Nook B, Haas M, Docrat A. Motion palpation used as a post manipulation assessment tool for monitoring end-feel improvement: A randomized control trial of test responsiveness. J Manipulative Physiol Ther. 2009; 32(7):549-55.
13. Kent C. Surface electromyography in the assessment of changes in paraspinal muscle activity associated with vertebral subluxation: a review. J Vert Sublux Res 1997;1(3):15-22.
14. McCoy M, Campbell I, Stone P, Fedorchuk C, Wijayawardana S, Easley K. Intra-examiner and inter-examiner reproducibility of paraspinal thermography. PLoS One 2011;6(2):e16535.
15. Fletcher, D. (2004). A tonal solution for subluxation patterns. Torque Release Technique analyzes cranial-spinal-meningeal functional unit. Canadian Chiropractor. 2004 April; 9(2): 20-23
16. TRT Seminar notes 2002. Holder Research Institute, 2002.
17. Stephenson RW. Chiropractic Text-book. Davenport,IA: Palmer School of Chiropractic, 1927.
18. Humphreys B, Kenin S, Hubbard B, Cramer G. Investigation of connective tissue attachments to the cervical spinal dura mater. Clin Anat. 2002;15:182-185.
19. Shriner S. A review of torque release technique. A Vert Sublux Res. 2012 July; 12: 72-76
20. Hodgson NJ. Torque Release Technique Instruction Manual: Self Published. 2011.
21. Kabakus N, Kurt A. Sandifer Syndrome: a continuing problem of misdiagnosis. Pediatrics International [serial on the Internet]. (2006, Dec), [cited December 9, 2014]; 48(6): 622-625
22. Vandenplas Y, Gottrand F, Veereman-Wauters G, et al.Gastrointestinal manifestations of cow's milk protein allergy and gastrointestinal motility. Acta Pediatr 2012;101(11) 1105-9.
23. Corrado G, Cavaliere M, D'Eufemia P, et al. Sandifer's syndrome in a breast fed infant. Am J Perinatol 2000;17(3):147-150.
24. Deskin R. Sandifer syndrome: a cause of torticollis in infancy.International Journal of Pediatric Otorhinolaryngology ELSEVIER 32 (1995) 183-185
25. Kent C, Models of vertebral subluxation: A review. J Vertebral Subluxation Res 1996; 1(1):1-7.
26. A. Hadari, E. Azizi, O. Lernau and S. Nissan, Sandifer's syndrome-a rare complication of hiatal hernia. A case report,Z Kinderchir39(1984), 202–203.

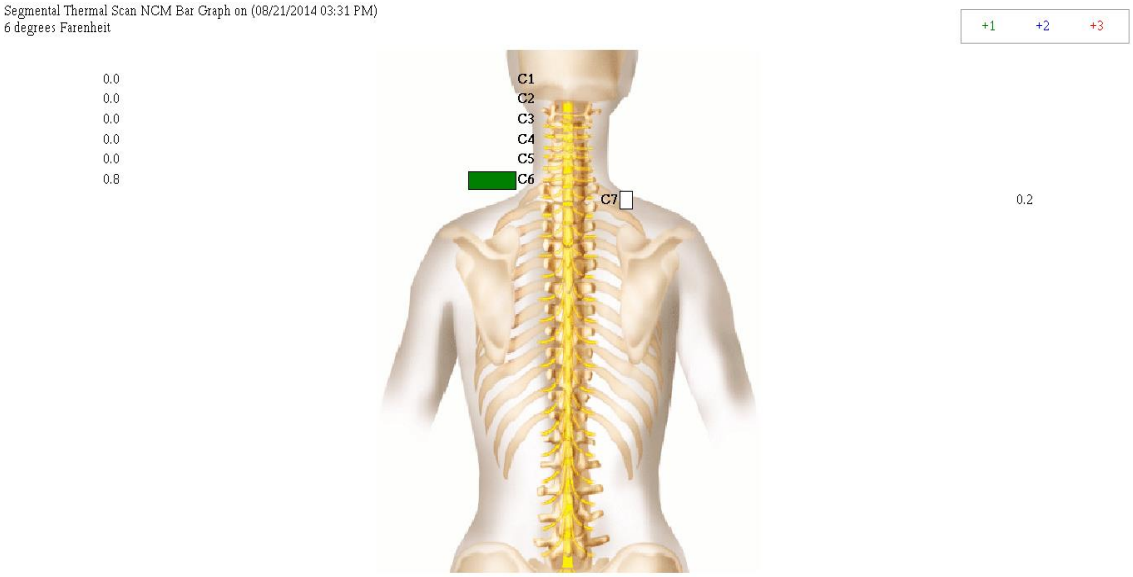
Figures 1 a,b,c - 1st examination

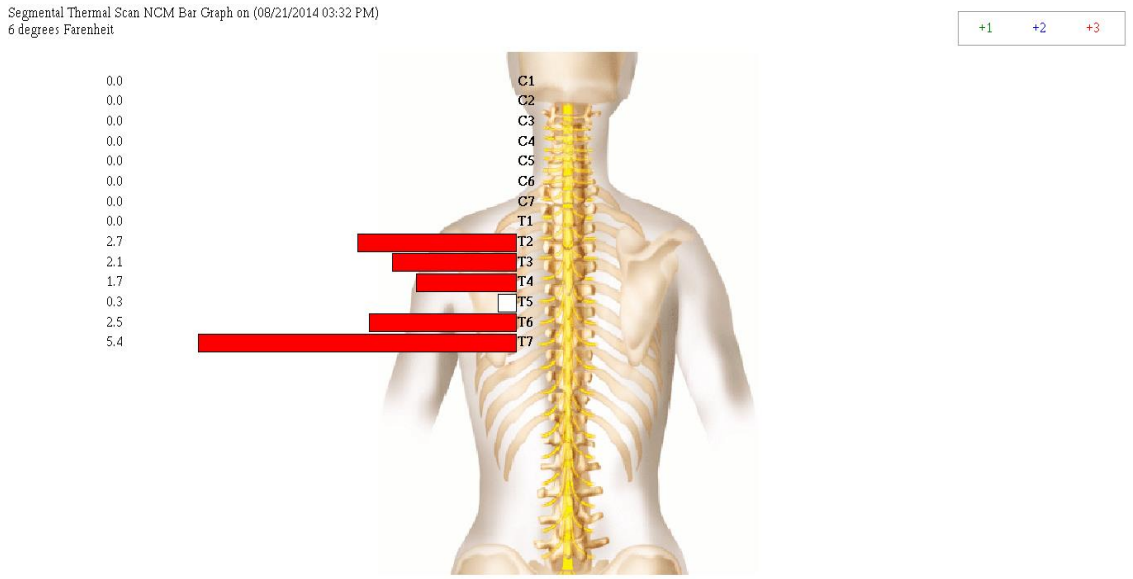
Segmental thermal scans of three different regions of the spine



Figures 2 a,b,c - re-examination

Segmental thermal scans of three different regions of the spine





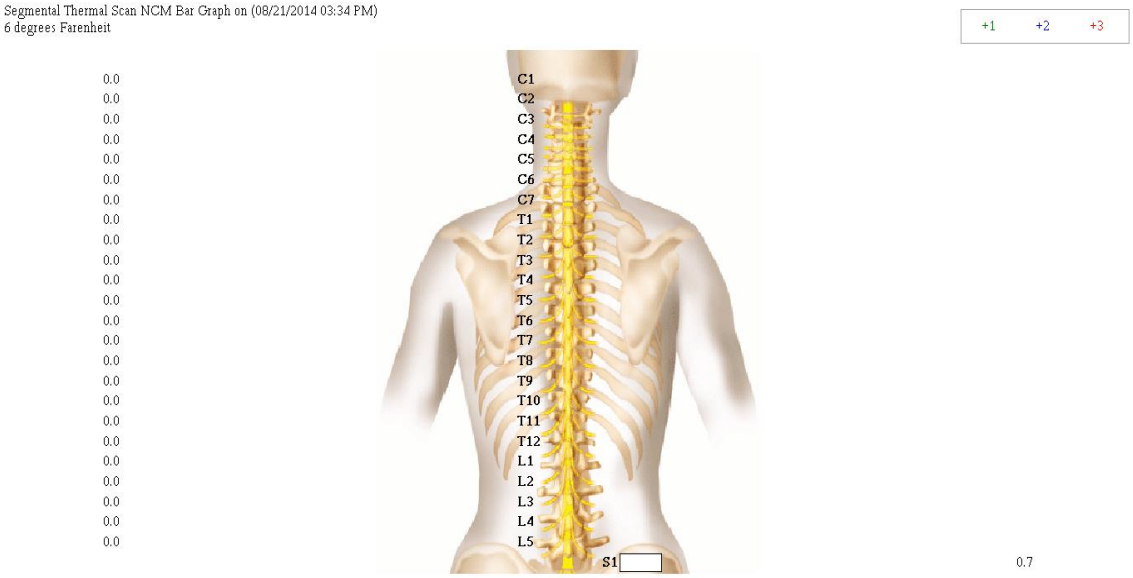


Figure 3 a,b,c - 2nd re-examination

Segmental thermal scans of three different regions of the spine