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ARTICLE

Limited Value of Plain Radiographs in Infant Torticollis

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ABSTRACT

OBJECTIVE. The purpose of this work was to assess the frequency of clinically relevant findings from plain films of infants evaluated for torticollis.

PATIENTS AND METHODS. After institutional review board approval, radiology records were searched for infants 0 to 12 months of age who underwent plain film study for torticollis or "head tilt." Infants evaluated for trauma or Down syndrome were excluded. All of the studies were reviewed, demographic data was recorded, and any additional imaging studies were examined.

RESULTS. A total of 502 patients (189 girls and 313 boys) were identified with an average age of 0.37 ± 0.2 years. Head tilt was to the left in two thirds of patients. Ten patients had abnormal findings reported. Six of these proved normal on subsequent studies (3 suspected occipital-C1 fusions, 2 suspected cervical fusions, and 1 suspected hemivertebra). Four patients had true bony vertebral abnormalities including absent left C7 pedicle, multiple fusion anomalies from C4 to T2, C3 hemivertebra and thoracic spine anomalies, and C4 hypoplasia. This last patient had abnormal kyphosis on physical examination and demonstrated instability with dynamic testing. Twenty-five additional patients with normal plain films underwent spine computed tomography or magnetic resonance imaging; all were normal.

CONCLUSIONS. The true-positive yield of plain films in nontraumatic infant torticollis was low (4 of 502). There were more false-positive than true-positive results. A common rationale for imaging is to exclude craniocervical or other unstable abnormalities that might contraindicate physical therapy, seen in only 1 of the 502 cases. Close physical examination could safely eliminate most patients sent for radiography.

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Key Words
radiology, evaluation, torticollis

Abbreviations
CMT— congenital muscular torticollis
SCM—sternocleidomastoid muscle
CT— computed tomography
CNS— central nervous system

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Torticollis is a deformity characterized by lateral inclination of the head, torsion of the neck, and deviation of the face.[1] Congenital muscular torticollis (CMT) is an idiopathic condition that begins in infancy as a rotation and flexion deformity of the neck caused by sternocleidomastoid muscle (SCM) shortening. More than 45 000 infants are born with congenital torticollis in the United States each year, and it has been estimated to affect ≤2% of newborn infants.[2–4] Clinical signs of CMT include restricted range of neck motion with a rotated chin, head tilt, a tight SCM muscle with or without a palpable mass, facial asymmetry, and plagiocephaly.[3,5]

Although many cases of CMT are diagnosed and treated clinically, many children are referred for plain radiographic assessment of the cervical spine to exclude osseous abnormalities.[6] Our anecdotal experience suggested that these studies were of limited use in infant torticollis, which prompted this study to clarify the value of radiographs in this condition.

PATIENTS AND METHODS

The study was approved by the Columbus Children's Hospital Institutional Review Board. Infants, ages 0–12 months, who had plain films of the cervical spine for an indication of torticollis or head tilt from July 1995 to June 2005 at Columbus Children's Hospital were identified through a search of the radiology information system database. Any infants evaluated for trauma or Down syndrome were excluded from the study. The examination for this condition consists of a frontal and lateral view of the cervical spine, with the head and neck held as straight as possible. All of the examinations and their reports were reviewed, and demographic data, the direction of head tilt, and the presence of any bony abnormalities were recorded. Any subsequent studies of the brain or spine performed on these patients were also reviewed to evaluate the accuracy of the plain film evaluation and to discern any other predisposing conditions to torticollis.

RESULTS

A total of 502 patients, 0–12 months of age, were identified during the 10 year time period. Most of the studies (85%) were performed in the most recent 5 years. The average age was 0.37 ± 0.2 years; 189 patients were girls, and 313 patients were boys. A head tilt to the left was found in two thirds of the patients. Of the 502 patients examined, 10 had reported abnormal findings. These patients underwent further testing with fluoroscopy, computed tomography (CT), MRI, and/or follow-up plain radiographs. The findings of these additional tests showed that 6 of the 10 patients with suspected abnormalities were actually normal and that the original interpretation was falsely positive. Of these 6 false-positives, 3 were suspected to have occipital-C1 fusions, 2 were suspected to have cervical fusions, and 1 was suspected to have multiple hemivertebrae. Four patients had true bony vertebral abnormalities identified on plain radiographs and subsequently confirmed with additional imaging: absent left C7 pedicle (Fig 1), multiple fusion anomalies from C4 to T2 (Fig 2), C3 hemivertebra and lower thoracic spine anomalies (and associated tetralogy of Fallot; Fig 3), and C4 hypoplasia (Fig 4). Only the last patient demonstrated instability with dynamic flexion and extension under fluoroscopy. In addition, 25 patients with normal plain films underwent spine CT or MRI because of continued clinical concern about an osseous abnormality. The additional testing did not detect any abnormalities, and all 25 of the patients had normal findings. Other diagnoses found among the study patients included gastroesophageal reflux disease, cerebellar abnormalities (infarct, Joubert's syndrome), cortical dysplasias, hypoxic-ischemic injury, and Chiari I malformation. There were no false-negative plain radiographs identified.

DISCUSSION

Torticollis is a combined head tilt and rotatory deformity. In older children, torticollis is a rather nonspecific sign and may result from a wide variety of abnormalities, including trauma, inflammation, muscular spasm, spinal cord and central nervous system (CNS) abnormalities, and gastroesophageal reflux.[7] In children <1 year of age, the most common cause of torticollis is related to injury of the SCM.[7] CMT is the third most common congenital

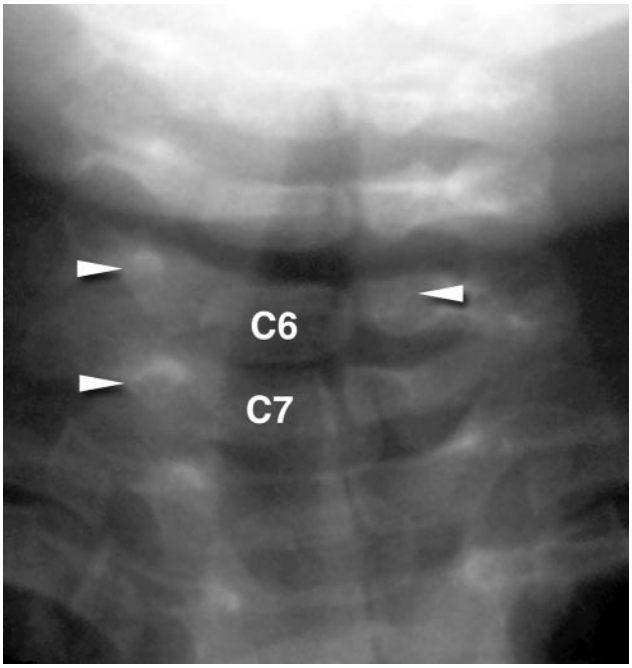

FIGURE 1
Frontal radiograph shows narrowing between C6 and C7. There are normal pedicles at C6 and a single right C7 pedicle (arrowheads). There is congenital absence of the left C7 pedicle. There was no instability on flexion/extension images.

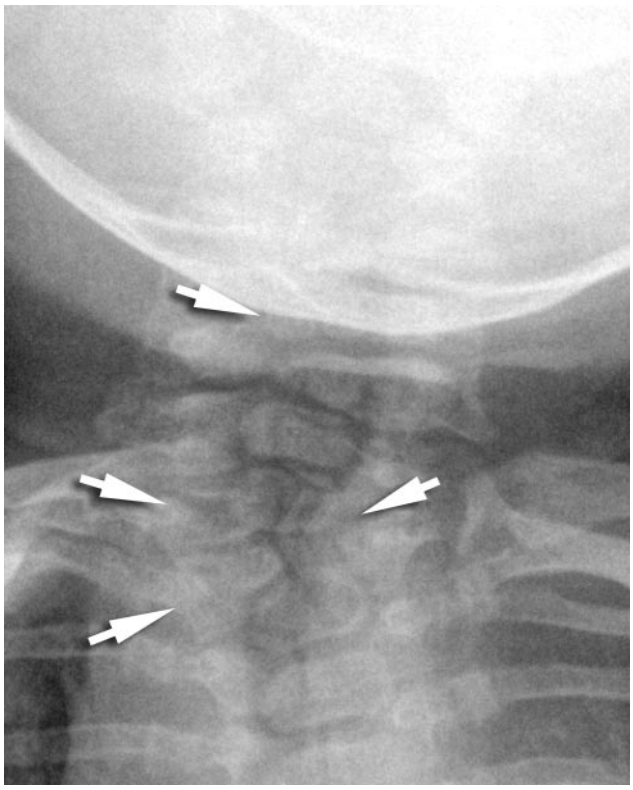

FIGURE 2
Frontal radiograph shows multiple segmentation abnormalities of the cervical and upper thoracic spine (arrows) producing torticollis.

problem of the musculoskeletal system in neonates and infants, having a reported incidence of 0.3–1.9%.[1,3,4] Infants with CMT have a higher incidence of breech presentation or difficult delivery (≤60%)[2,8,9] and lower extremity abnormalities (clubfoot and hip dysplasia), suggesting that in utero malposition and trauma are causative factors.[2,10,11] Regardless of predisposing factors, most authors suggest that some sort of ischemia or hemorrhage occurs within the SCM, which results in a muscular compartment syndrome[3] and eventual contracture of the SCM with secondary shortening.[4,10,12] On physical examination, there is restricted range of cervical motion with SCM tightness. The head is tilted toward the involved SCM, and the chin is often rotated to the contralateral side.[13] A firm palpable mass is often present, sometimes referred to as pseudotumor of infancy (or fibromatosis colli). Although some authors have regarded pseudotumor of infancy and CMT (without a palpable mass) as separate conditions, most consider them to be part of the same etiologic spectrum.[14]

Nonmuscular causes of torticollis may occur in children who have spondyloepiphyseal dysplasia, Klippel-Feil malformation (fusion of cervical vertebrae),[15] other vertebral body segmentation anomalies (such as hemivertebra), or subluxation of the atlanto-axial or atlanto-occipital joint.[16] Other causes include CNS abnormalities (cortical dysplasias, cerebellar tumors and malformations, or cerebral injury), spinal cord tumors, ocular abnormalities, and even severe gastroesophageal reflux (Sandifer syndrome).[16] Patients with skeletal or CNS abnormalities, however, usually have physical examination findings other than just torticollis. Patients with Klippel-Feil syndrome have a short neck, decreased range of motion in the cervical spine, and a low hair line.[15] These patients may also have a Sprengel anomaly, with elevation of the scapula. Children with CNS abnormalities often have other neurologic findings, and children with ocular torticollis generally have abnormal eye examinations. Those patients with structural abnormalities of the cervical spine (fusions and hemivertebrae) typically have fixed spinal curvatures (sometimes with abnormal kyphosis or lordosis) without SCM tightness on physical examination. Although it has been reported that ≤18% of children may have nonmuscular causes of torticollis,[4,17] it is uncommon in younger children.

If left untreated, CMT can lead to permanent limitation of neck range of motion. In addition, children often develop positional plagiocephaly and asymmetric growth of facial features. Treatment methods for CMT range from conservative to surgical. Conservative methods include active and passive stretching exercises, local heat, analgesics, and sensory biofeedback. Stretching therapies are the most common and have reported success rates as high as 90% if instituted early.[1,4,5,15,18] Early diagnosis is important, as Sonmez[8] showed that patients whose treatment began before 3 months of age did not need surgery, whereas 25% of those starting treatment later ultimately required surgical correction. If conservative methods are unsuccessful, then surgery is required to restore normal neck motion. The most common surgical treatment is SCM tenotomy, or muscle release, and is generally not performed until after the age of 1 year.

Children with bony abnormalities of the spine, however, will not respond to physical therapy. Moreover, stretching exercises could potentially be harmful to patients with bony anomalies if associated with instability of the neck. For this reason, plain radiographs of the cervical spine are generally recommended for evaluation of infant torticollis, especially before starting physical therapy.[1,3,11,19] Although the yield of such radiographs has not been investigated previously, other authors have remarked on the rare occurrence of skeletal anomalies in infant torticollis and their limited value in the evaluation of CMT.[14] Fixed vertebral structural abnormalities are not unimportant, but it is those malformations that predispose to cervical instability that are important to recognize. An unstable spine requires immobilization and neurosurgical consultation,[20,21] not physical therapy.

In our study of 502 cases, only 4 (0.8%) were classified as having true-positive findings. Three cases (absent left C7 pedicle, multiple fusion anomalies from C4 to T2, and C3 hemivertebra) exhibited no signs of instability. Because instability was not a factor in these 3 cases,

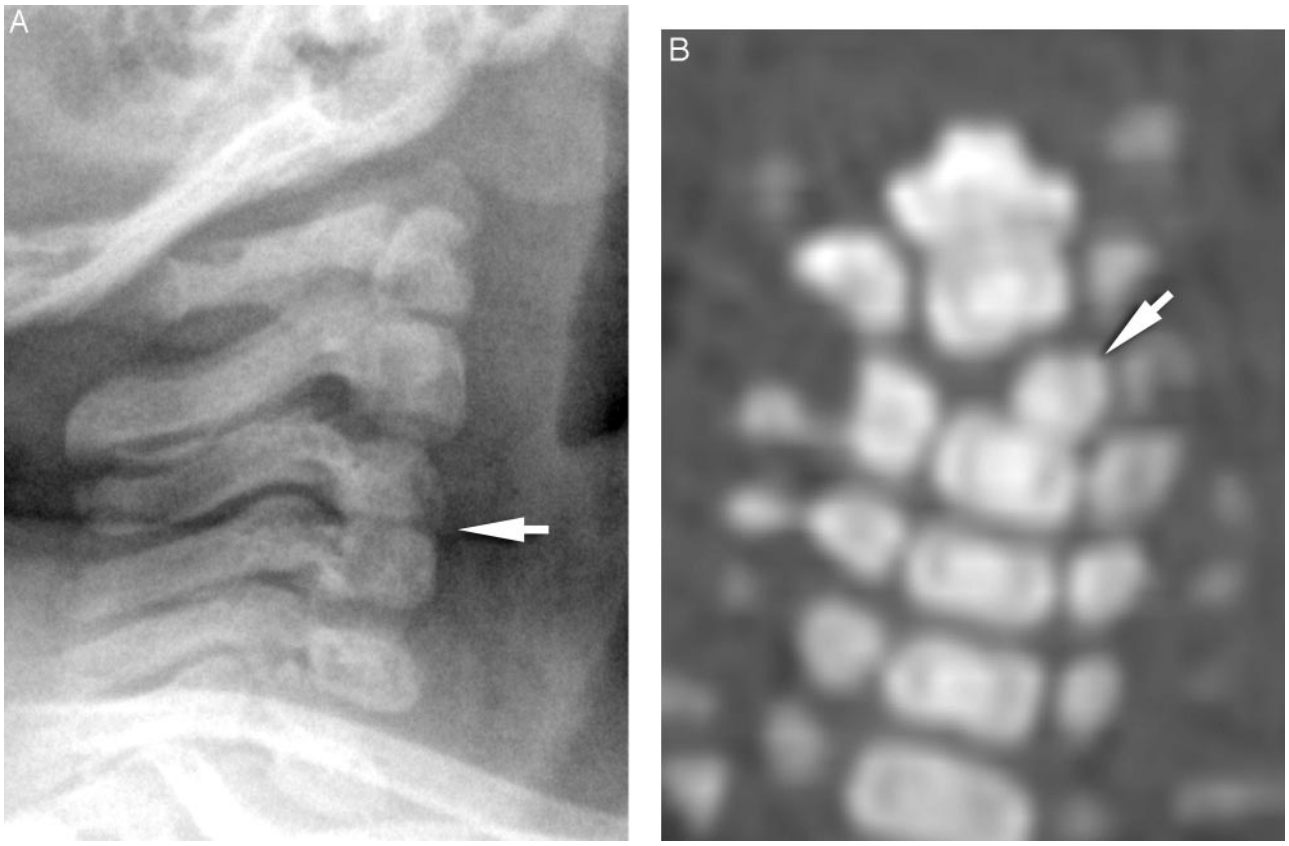

FIGURE 3
A, Lateral radiograph in an infant with a C3 hemivertebra shows abnormal narrowing of the C3-C4 disk space (arrow). B, Coronal reformatted CT image shows the left C3 hemivertebral body (arrow).

conservative treatment would have been harmless but ineffective. Only 1 of our true-positive cases had a relevant abnormality, characterized by C4 hypoplasia and instability. Along with torticollis, this child also had cervical kyphosis on physical examination, which led to the suspicion of a vertebral abnormality.

Abnormalities of the craniocervical junction are of potential concern, because 50% of cervical rotation occurs between the occiput and C2,[1,22,23] and the upper cervical region represents the most vulnerable part of the infant spine. Craniocervical instability could pose serious consequences for the child if vigorous physical therapy is to be instituted. The clinical appearance of children with atlanto-occipital anomalies is comparable to that of those with Klippel-Feil,[1,12] which may prompt radiologic investigation. Unfortunately, plain radiographs of the craniocervical junction are often difficult to interpret in infants and may be misleading.[12,20,23] This was confirmed in our study. Plain radiographs resulted in more false-positive readings than actual true-positive readings for upper cervical spine abnormalities. Four of our 6 false-positive results were related to suspected abnormalities from the occiput to C2, and all were shown to be normal on further imaging.

The issue of false-positive results needs to be emphasized. Aside from causing parental anxiety, false-positive results lead to additional procedures, such as CT and MRI. For infants, MRI usually requires sedation with its attendant potential complications. CT exposes patients to ionizing radiation, which is becoming increasingly recognized as a real source of future cancer, especially when performed in the very young.[24–27] Thus, one could argue that the plain radiographic evaluation of patients with apparent CMT, without other clinically concerning features, may lead to more potential harm than any benefit that it provides to patient treatment.

In assessing our findings, we are limited by the fact that the majority of patients with normal plain radiographs did not have further imaging to confirm the results. Our assumption is that there were no false-negative results, but this cannot be confirmed, and it is unreasonable to subject all infants to CT and MRI. However, given this assumption, our results show that for plain radiographs, there was 100% sensitivity and 98% specificity for detecting bony abnormalities in infants with torticollis, with a negative predictive value of 100% and a positive predictive value of 40%. Although these numbers indicate that plain radiography is a good screening test, the prevalence of true cervical spine abnormalities in our population is only 0.8% and of clin-

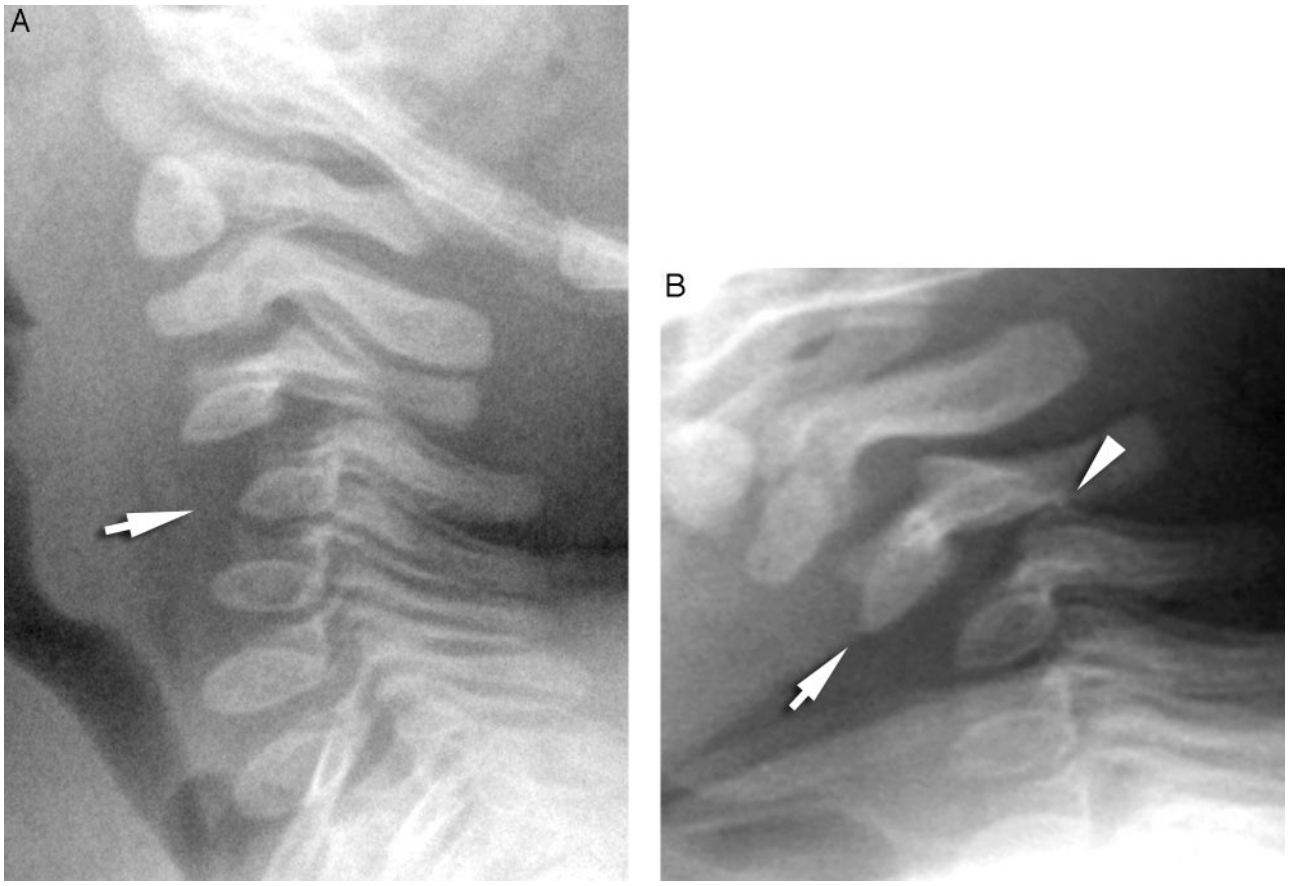

FIGURE 4
A, Lateral radiograph in an infant with torticollis and cervical kyphosis shows a hypoplastic C4 vertebral body (arrow). B, With flexion, there is marked anterior subluxation of C3 (arrow) and anterior displacement of the C3-C4 facet (arrowhead).

ically relevant abnormalities only 0.2% (1 in 502). This poor yield means that the pretest probability of an abnormality immediately relevant to instituting therapy is very small. This has been recognized by some authors who have recommended diagnostic algorithms relying on clinical history, physical examination, and response to therapy.[14,17]

CONCLUSIONS

Most infants with typical clinical findings of CMT are unlikely to benefit from plain radiography of the spine. In the absence of concerning clinical findings, the diagnostic yield is extremely low (98% normal), and the positive predictive value is only 40%, which leads to additional unnecessary testing. The vast majority of CMT patients will be successfully treated with conservative physical therapy. We would suggest that imaging be reserved for those children who fail to respond to physical therapy and those who have atypical clinical findings. This would save most patients from unnecessary radiation exposure and diminish parental anxiety over diagnostic testing, without compromising care of the child.

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