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Menkes kinky hair syndrome.

C S Houston and S R Harding

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LETTERS

Inadvertent Intrathecal Use of Ionic Contrast Media

I read with great interest the paper by Bohn et al (1) entitled "Inadvertent Intrathecal Use of Ionic Contrast Media for Myelography." The authors should be applauded for their excellent case descriptions and review the literature regarding the potentially devastating consequences of intrathecal injection of ionic contrast medium.

I would like to point out, however, that the authors neglected to cite a brief case description, which I reported, of inadvertent intrathecal administration of ionic contrast in a patient who developed a postoperative pelvic abscess complicated by subarachnoid fistula (2). In this case, the patient was recovering from a right hemipelvectomy for recurrent squamous cell carcinoma of the sacrum, when copious drainage of infected, yellow serous fluid from a Jackson-Pratt drain was noted. The clinical impression was that a postoperative abscess/urinoma developed, which prompted a request for sinography. Under fluoroscopy, 100 mL of 30% meglumine diatrizoate were slowly infused through the drain. This resulted in filling of a large pelvic abscess cavity and delayed, abrupt filling of the lumbosacral thecal sac. Upon filling the thecal sac, the patient complained of severe lower back pain with radiating dysesthesias in both legs. The contrast was immediately aspirated and the patient was placed in an upright position to enhance dependent drainage. The patient's symptoms resolved within minutes and no further neurologic sequelae ensued. The subarachnoid fistula was successfully treated with a lumbar spinal drain.

After reading the article by Bohn et al (1), I must conclude that I was very fortunate that my patient did not suffer serious complications from inadvertent infusion of ionic contrast media into the subarachnoid space. I am sure that the benign outcome of this case was at least partly due to my ability to recover promptly most of the infused contrast media and the favorable cerebrospinal fluid flow dynamics of the situation (in which there was dependent drainage of cerebrospinal fluid out of the thecal sac).

I wish to share my experience with others mainly to emphasize that inadvertent intrathecal injection of ionic contrast can also occur outside the usual setting of myelography and thus should always be taken into consideration when sinography is planned for pathologic conditions in the vicinity of the spine.

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2. Chaloupka JC. Pelvic abscess complicated by subarachnoid fistula. *AJR: Am J Roentgenol* 1988;152:433

Reply

I wish to thank Dr. John C. Chaloupka for drawing our attention to the case he described in 1989. My co-authors and I do find this case interesting and highly relevant to the problem under discussion.

We agree with Dr. Chaloupka that the benign outcome of this case may well have been due to the prompt removal of most of the contrast medium from the cerebrospinal fluid. From this, it is tempting to suggest that immediate removal may be the treatment of choice whenever ionic contrast media have inadvertently been injected intrathecally.

However, the case described by Dr. Chaloupka is unusual in that the inadvertent injection immediately was recognized at fluoroscopy and, as he points out, there was an unusual opportunity for effective and immediate cerebrospinal fluid drainage. It seems likely that timing may be a decisive factor in determining the effectiveness of contrast medium removal. If the problem is recognized early, contrast medium withdrawal may be highly effective. Unfortunately, it is our experience that most cases of inadvertent intrathecal injection of an ionic contrast medium occur during myelography, when no alarm is being raised by the mere presence of intrathecal contrast medium; the problem is recognized only after the patient has become severely ill and the contrast medium has had time to disperse within the cerebrospinal fluid and to be taken up into nervous tissue. In such cases, it may be too late to retrieve the contrast medium effectively; preference should be given to the patient's urgent need for seizure control and medical treatment.

Dr. Chaloupka's reminder that inadvertent intrathecal injection of an ionic contrast medium may also occur outside the usual setting of myelography is an important fact to be noted by all persons who are involved in the administration of intravenous radiopharmaceuticals for the enhancement of medical images.

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Menkes Kinky Hair Syndrome

Brian E. Kendall has presented an excellent review article of disorders of lysosomes, peroxisomes, and mitochondria (1). Regrettably, he has given Menkes kinky hair syndrome a new name, trichopoliodystrophy, without citing a reference, and reported that death occurs in 4 to 6 months. He failed to state that in at least two patients, daily copper histidinate therapy initiated within 2 months of birth has prevented the severe progressive brain damage characteristic of this disease. We wish to call attention to an important paper by G. Sherwood, an obscure publication (2), and three easily overlooked letters in *Lancet* (3–5). The junior author of this letter is the mother of one of Sherwood's patients, who at age 32 months was "a walking and talking bundle of happy mischief." In recent tests of verbal performance, at age 5 1/2 years, he was superior for his age.

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CANADA

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4. Danks D, Camakaris J, McArdle H, Mercer J. Menkes' disease: a disorder of zinc metabolism? *Lancet* 1989;1:1399
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Reply

I am grateful to Drs. C Stuart Houston and Sheila Rutledge Harding for drawing my attention to the successful use of parenteral copper histidinate therapy, commenced before 2 months of age in two unrelated boys, in preventing the progressive brain damage that characterizes Menkes disease. I am pleased to hear that Dr. Rutledge Harding's son continues to do so well on the therapy. I have been unable to find in the medical literature or standard texts any further references to the use of copper histidinate therapy, but hopefully the letter will draw attention to the importance of early diagnosis of Menkes disease and the early application of the therapy.

Trichopoliodystrophy is a descriptive pseudonym for Menkes kinky hair disease that has been used by many authors, including Menkes himself (1).

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