An Office-Based Prospective Study of Deafness in Mumps

Hiromi Hashimoto, MD, Masashi Fujioka, MD,† Hiroshi Kinumaki, MD,‡ and Kinki Ambulatory Pediatrics Study Group*

Background: Deafness is a rare but important complication of mumps virus infection. Its incidence has been estimated at 0.5 to 5.0 per 100,000 cases of mumps, but recent reports from Japan, where mumps is endemic, suggest that the incidence might be higher.

Objective: Prospective office-based study to determine the incidence of hearing loss in children with mumps.

Methods: Forty pediatric practices participated in this survey. The study population consisted of patients ≤20 years old with mumps seen between January 2004 and December 2006. Clinical diagnosis of mumps was made by experienced pediatricians. Among those from whom written consent was obtained, parents were asked to conduct hearing screening tests by rubbing fingers near the ears twice daily for 2 weeks. Patients suspected with hearing loss were further examined by an otolaryngologist.

Results: Among 7400 children who underwent hearing ability assessment after clinical onset of mumps, 7 had confirmed hearing loss; none had been previously vaccinated against mumps. In all cases, hearing loss was unilateral but severe and did not improve over time.

Conclusions: The incidence of hearing loss in children due to mumps was 7/7400 (~1/1000 cases), which is higher than previously suggested. Prevention of deafness is another important reason for assuring universal immunization against mumps.

Key Words: mumps, hearing loss, incidence

(*Pediatr Infect Dis J* 2009;28: 173–175)

Mumps can cause sensorineural hearing loss, but the incidence has long been considered to be quite low (0.5–5.0/100,000 cases).[1–3] Deafness caused by mumps is rarely seen in most parts of the world, because mumps is uncommon in countries with extensive vaccination programs.[1,4] In Japan, however, mumps remains endemic because measles-rubella vaccination instead of measles-mumps-rubella vaccination is still used.

Recently, several reports[5–9] from Japan have suggested that the incidence of mumps-related deafness is much higher than had been previously believed, but the studies had methodological inaccuracies. In this current study, we determined the incidence of sudden acquired deafness in children with mumps based upon a population-based office survey. This is the first large-scale prospective study about mumps deafness.

METHODS

The study was a survey of 40 pediatric outpatient clinics (38 private clinics and 2 hospitals) where doctors were members of Kinki Ambulatory Pediatrics Study Group located in the western part of Japan. A total of 7831 patients were initially enrolled, but signed consent for research could not be obtained for 67 patients and hearing status could not be determined for 262 children (247 cases: patients too young, 15 cases: others), resulting in 7502 patients who participated in this investigation. Diagnosis of mumps was based on the following clinical case definition: acute onset of unilateral or bilateral tender, self-limited swelling of the parotid or other salivary gland, lasting 2 or more days, and without other apparent causes.

When the diagnosis of mumps was made, each patient was informed of the possibility of deafness and hearing screening was performed by the doctor rubbing the thumb and index finger beside each ear. Parents, whose written consent was obtained, were instructed to conduct finger-rubbing audibility tests twice daily for 2 weeks. For younger children, a sound toy might be used instead. After the testing period, the parents' records of hearing testing were submitted by each investigator immediately. When hearing loss was suspected during or after the 2 weeks testing period, patients were further referred to an otolaryngologist to confirm hearing ability by the use of audiometry test or auditory brainstem response measurement, and serologic tests for mumps-specific IgM antibodies were conducted to verify infection with the mumps virus.

RESULTS

We enrolled 7502 patients with clinically diagnosed mumps (male 4057, female 3445) and the parents' hearing testing records were obtained from 7400 (male 4009, female 3391); hearing test results could not be obtained in 102 subjects due to relocation or other reasons. Among the 7400 patients, the mean age was 5 years, the median age was 5, and the mode age was 4 years. In our study population, 607 (8%) had received 1 dose of mumps vaccine, 6788 (92%) had not received vaccine, and vaccination status was unknown in 5 cases.

Among the 7400 subjects with complete records, hearing loss was suspected in 24 subjects. Hearing loss was unilateral in all cases and no subjects developed aseptic meningitis. When examined by an otolaryngologist, 8 had no hearing loss, 8 had conductive hearing loss due to otitis media; one had conductive hearing loss from dysfunction of the Eustachian tube, and 7 were ultimately diagnosed with sensorineural hearing loss, 6 by audiometry and the youngest by auditory brainstem response (Table 1). The incidence of deafness among subjects with clinical mumps was 7/7400 cases, or 0.1% (95% confidence intervals 0.03%–0.18%).

Of the 7 subjects with deafness, all had confirmed mumps infection by positive serologic tests for mumps-specific IgM antibodies (EIA), and none had received mumps vaccination. Deafness among the 7 subjects was severe and remains unresolved compared with those of the other 17 subjects whose apparent deafness had resolved completely within the study period.

Deafness developed within 5 days after parotitis in 5 subjects. Prior normal hearing was confirmed by the finger-rubbing tests in 4 of 5 subjects; in 1 of the 2 subjects with early deafness, a normal hearing test by audiometry testing had been conducted several months before the onset of mumps. In both cases, there was no evidence to suggest congenital or other hearing loss.

Accepted for publication August 14, 2008.
From the *Hashimoto Pediatric Clinic; †Fujioka Pediatric Clinic; and ‡Kinumaki Pediatric Clinic, Osaka, Japan.
Supported partially by a grant from the Society of Ambulatory and General Pediatrics of Japan Research Foundation.
This study was approved and monitored by the Society of Ambulatory and General Pediatrics of Japan Ethics Committee.
Address for correspondence: Hiromi Hashimoto, MD, Hashimoto Pediatric Clinic, Oda 3-21-17, Ibaraki, Osaka, 567-0018, Japan. E-mail: hiromi@reasoning.org.

ISSN: 0891-3668/09/2803-0173
DOI: 10.1097/INF.0b013e31818a8ca8

TABLE 1. Patients With Mumps-Related Deafness in Our Study

Age	Sex	Recognition of Deafness	Affected Side	Vertigo	Diagnosis of Deafness	Therapy	Prognosis
7 yr and 1 mo	M	Day 3 finger test	L	−	Audiometer	Steroid	Lt total deafness
6 yr and 7 mo	F	Day 1 finger test	R	−	Audiometer	Steroid	Rt total deafness
3 yr and 8 mo	M	Day 5 finger test	L	+	ABR	Steroid, vitamin B_{12}	Lt ABR no response
7 yr and 5 mo	M	Day 30 audiometer*	L	−	Audiometer	None	Lt total deafness
4 yr and 10 mo	F	Day 36 telephone	L	−	Audiometer	Unknown	Lt total deafness
4 yr and 7 mo	M	Day 1 finger test	R	−	Audiometer	Steroid	Rt 50–70 dB
7 yr and 1 mo	M	Day 4 finger test	L	+	Audiometer	None	Lt total deafness

*Onset of hearing loss was Day 3.

In the remaining 2 subjects, the diagnosis of deafness was delayed because subjects did not complain of and parents did not identify hearing loss using the finger-rubbing test. In 1 (case 5), hearing loss was not noticed until the child held a telephone receiver to the affected ear 36 days after the onset of mumps. Deafness was overlooked in a 7-year-old-boy (case 4) despite his occasionally failing the hearing test from 3 days after mumps developed because he was suspected of faking the hearing loss. A health check at school indicated left-sided hearing loss and he was finally diagnosed with total deafness in his left ear 30 days after onset of parotitis.

DISCUSSION

In our prospective study, the incidence of deafness was approximately 1/1000 cases (95% confidence interval 1/549–1/3128), which is higher than previously considered.

Textbooks[1,2] describe the incidence of deafness to be from 0.5 to 5.0 per 100,000 cases of mumps. This rate was not based on a population-based prospective study but was an estimate calculated in 1957 by Everberg.[3] Everberg identified 5 cases of mumps deafness in Copenhagen during a period of 10 years, and calculated the incidence of mumps deafness using half of the population of Copenhagen, instead of the actual number of people infected with mumps. As a result, the estimated frequency of mumps deafness was about 0.05 per thousand. The result may not have been accurate because it used a rough estimate for the denominator and only a partial identification of the cases of mumps deafness.

Recently higher incidences have been reported in Japan where vaccination against mumps is not routine.[5–9] According to a survey of otolaryngologists, Kawashima et al[5] reported annual cases of mumps deafness as 650 in 2001, but they did not calculate an incidence rate. Japanese reports of high incidence (3 cases of deafness in 551 cases of mumps,[6] 5 cases of deafness in 1470 cases of mumps[7]) were based on retrospective studies limited to local outbreaks of mumps.

A prospective study is necessary to elucidate the incidence of mumps-related deafness. Vuori et al[10] reported a prospective study in 1962 of 298 armed forces members suffering from mumps, of whom 12 developed reversible hearing disorder and one had severe permanent deafness by audiometric testing. This investigation did not include children, and the sampling was too small to infer an incidence.

Our research was performed prospectively among a sufficiently large sample of children for the first time. Forty institutions widely distributed in western Japan provided the data. We believe that we were able to obtain more accurate results about the incidence of mumps deafness. Our study has several limitations, the most important being the lack of a control comparison group. Because we did not determine the incidence of deafness among children who did not have clinical mumps, we could not report the degree to which the incidence of mumps deafness differs from baseline deafness in a comparable child population. We can only assume that baseline incidence of acquired sensorineural deafness is quite rare.

It is difficult to know the actual number of mumps cases. Many subjects may not have symptoms severe enough to warrant medical attention and the clinical diagnosis of mumps infection may not be accurate. These are common problems in an office-based study that could not be effectively addressed in our study.

The number of cases of deafness is also uncertain. Because mumps-related deafness is mainly unilateral, it can be overlooked by parents and children. In our 7 cases of deafness, no one suspected hearing loss before actual assessment. If a patient develops deafness at a later time, the pathogenesis of deafness would be difficult to ascertain. This can lead to cases of mumps-related deafness being overlooked.

We investigated the hearing ability prospectively with a home parental hearing screening assessment. A more objective hearing test, such as otoacoustic emission or ABR, more accurate for younger children, was not used in this study. In our study, the youngest child diagnosed with hearing loss was 3 years and 8 months. The age range of deafness reported previously by Ishikawa et al[9] (14 cases) was 3 to 34 years old and by Kawaguchi et al[8] (12 cases) was 4 to 39 years old. In none of these studies was the otoacoustic emission or ABR tests used systematically. Together, these results suggest that identification of hearing loss in younger children might have been technically difficult, potentially underestimating the actual incidence of mumps-related hearing loss.

Our study provides the most accurate incidence of mumps-related deafness currently available in Japan. We believe that mumps deafness is not a rare complication. Deafness will likely persist regardless of treatment, is disabling, and affects the quality of life. Moreover, asymptomatic or mild mumps infection may also cause hearing disorders.[9,11] Universal vaccination against mumps could prevent such deafness.

ACKNOWLEDGMENTS

The authors thank KAPSG pediatric study group in Kinki district of Japan. The group members who completed this study are listed here: Hyogo, Dr. Takashi Fujita, Dr. Yoshikazu Harima, Dr. Toshiharu Hino, Dr. Kei Katayama, Dr. Naoki Kumagai, Dr. Hidenori Matsumoto, Dr. Shinro Matsuura, Dr. Oji Noma, Dr. Takao Okafuji, Dr. Teruo Okafuji, Dr. Kenji Sakuragi; Ishikawa, Dr. Shigeru Maruyama, Dr. Reiji Watanabe; Kyoto, Dr. Hidenori Funaki, Dr. Hajime Hanayasu, Dr. Naoki Kodo, Dr. Hidekazu Nakanoin; Nara, Dr. Takeshi Shimizu; Okayama, Dr. Kazunori Okamoto; Osaka, Dr. Shinji Katagiri, Dr. Yasuhiro Fukunaga, Dr. Naoki Nishida, Dr. Masanori Nishigaki, Dr. Tatsuo Nishimura, Dr. Fumiyoshi Yamaue; Saitama, Dr. Osamu Kakuta; Shiga, Dr. Takuro Kidowaki, Dr. Kyokazu Ono, Dr. Naruo Saito, Dr. Yoshiaki Takahashi; Wakayama, Dr. Kensaku Kashii, Dr. Shoji Kawarano, Dr. Yoichi Sato, Dr. Hiroyuki Negoro, Dr. Kazumasa

Otani, Dr. Takashi Yamairi; Yamaguchi, Dr. Shigeru Kaya, Dr. Satoshi Tanimura.

The authors also thank Dr. Yoshifumi Takahashi (a one-time Chief Doctor, Department of Otolaryngology, Toyonaka Municipal Hospital) for helpful advice and Dr. Kiyoshi Ichihara (Professor of Faculty of Health Sciences, Yamaguchi University School of Medicine) for helpful comments on statistical analyses. We are indebted to Dr. John Ichiro Takayama (Associate Clinical Professor, Department of Pediatrics, University of California San Francisco), Dr. Chiaki Miyazaki (Director of Fukuoka-West Rehabilitation Center for Children), Dr. Takeshi Minami (Minami Clinic) and Kiyota Hashimoto (Associate Professor of Linguistics, Osaka Prefecture University) for their invaluable help in revising the manuscript.

REFERENCES

1. Wilbert H. Mason Mumps. In: Robert MK, Richard EB, eds. *Nelson Textbook of Pediatrics*. 18th ed. Philadelphia, PA: Saunders Elsevier; 2007:1341–1344.
2. Cherry JD. Mumps virus. In: Feigin RD, Cherry JD, eds. *Textbook of Pediatric Infectious Diseases*. 5th ed. Philadelphia, PA: Lippincott Williams & Wilkins; 2004:2305–2314.
3. Everberg G. Deafness following mumps. *Acta Otolaryngol*. 1957;48:397–403.
4. Galazka AM, Robertson SE, Kraigher A. Mumps and mumps vaccine: a global review. *Bull WHO*. 1999;77:3–14.
5. Kawashima Y, Ihara K, Nakamura M, et al. Epidemiological study of mumps deafness in Japan. *Auris Nasus Larynx*. 2005;32:125–128.
6. Ishimaru Y, Nakano S, Nakano H, et al. Neurogical complication of mumps [in Japanese]. *Shonika Shinryo*. 1988;51:1421–1427.
7. Aoyagi N, Kodama A, Koike M, et al. Mumps deafness [in Japanese]. *Shonika*. 1996;37:1273–1279.
8. Kawaguchi S, Kawano A, Kanebayashi H, et al. Case reports of mumps deafness. *Pract Otorhinolaryngol*. 2003;96:865–869.
9. Ishikawa T, Ichimura K. An epidemiological study on deafness in patients with mumps. *Pract Otorhinolaryngol*. 2004;97:285–290.
10. Vuori M, Lahikainen EA, Peltonen T. Perceptive deafness in connection with mumps: a study of 298 servicemen suffering from mumps. *Acta Oto-laryng*. 1962;55:231–236.
11. Hall R, Richards H. Hearing loss due to mumps. *Arch Dis Child*. 1987;62: 189–191.