Clinical Experience of Intraorbital Optic Nerve Sheath Meningioma
—Report of Eight Cases—

Ryotaro KURODA, Jiro NAKATANI, Akira YORIMAE, Yuzo NAKAO* and Toshifumi OHTORI*

Departments of Neurosurgery and *Ophthalmology, Kinki University School of Medicine, Osaka-sayama, Osaka

Abstract
Eight cases of primary optic nerve sheath meningioma were treated between 1980 and 1988. Five were females aged 37-61 years. The other three were two boys, one with neurofibromatosis, and an old male aged 71 years. They were first seen by the ophthalmologist with complaints of unilateral progressive visual loss or proptosis. Although blindness of the affected eye mainly occurred between 1 month and 3 years after the initial symptoms, the diagnosis tended to be made late in the adult cases. Intracranial extension was demonstrated in four of the six adult cases when contralateral visual loss or disturbance of consciousness presented. Larger intraorbital meningiomas were easily diagnosed by a combination of computed tomographic (CT) scanning, magnetic resonance (MR) imaging, and carotid angiography. MR imaging provided clear delineation of the optic nerve and its course through the tumor, and perioptic meningioma could be diagnosed. However, it was difficult to make a diagnosis without biopsy at the early stage, for example, when just the enlargement of the optic nerve was demonstrated by CT or MR imaging. Tumor removal was performed when blindness developed after definitive diagnosis by biopsy, intracranial extension was demonstrated, and advanced proptosis presented. The transcranial supraorbital approach or transcranial transorbital approach with resection of the supraorbital rim was used for these large intraorbital meningiomas. From our clinical experience, early diagnosis and early treatment should be emphasized.

Key words: optic nerve sheath meningioma, clinical course, diagnosis, surgical treatment, intracranial extension

Introduction
Optic nerve sheath meningioma or intraorbital meningioma is found in 3% of intraorbital expanding lesions,3 in 8.9% of intraorbital tumors,4 and in 17%7 of orbital diseases in patients visiting neurosurgical departments with chief complaints of unilateral proptosis. Neurosurgeons, then, do not frequently have the opportunity to treat such cases. There are, however, disagreements about therapeutic principles, especially operative timing in clinical consultation, when the literature is reviewed. On the other hand, the progress of imaging diagnosis allows the detection of abnormal conditions in the optic nerve at an earlier stage when vision can still be maintained which requires difficult decisions about differential diagnosis and therapeutic principles. We have treated eight cases of optic nerve sheath meningioma in the orbit since 1980. In this paper, we describe case reports and further investigations on therapeutic methods in addition to a discussion of the literature on clinical courses, diagnosis, and surgical treatment.

Case Reports

I. Severe proptosis
Case 1: A 61-year-old female visited our ophthalmological department in March, 1980, with a 1-year history of visual loss in the right lower half of the visual field. The right visual acuity was counting fingers at 20 cm. A computed tomographic (CT)
scan showed an enlargement of the right optic nerve at the orbital apex (Fig. 1A). In December, she lost sensitivity to light. Two years later, the tumor was demonstrated on CT scan (Fig. 1B) but she refused to undergo an operation. In September, 1983, she was transferred to our neurosurgical department because of marked proptosis.

On admission, the pupil was 5 mm in diameter, the light reflex disappeared, and proptosis by 2 mm, on the right side. A CT scan revealed tumor invasion into the ethmoidal sinus (Fig. 1C). The standard transcranial transorbital approach was taken through a frontotemporal craniotomy and opening of the orbital roof. The tumor was partially removed. However, postoperative CT scan revealed the remaining large tumor and the proptosis (Fig. 1D). Regrowth of the tumor was revealed on a follow-up CT scan (Fig. 1E).

She was admitted again in September, 1986, because her right eyeball had lateroinferior protrusion and there was severe tumor invasion into the ethmoidal sinus (Fig. 1F). Extraocular muscle paresis, severe eyelid edema, and chemosis were present. Cerebral angiography showed a tumor stain from the ophthalmic and maxillary arteries. Therefore, tumor removal was performed twice after artificial embolization of the maxillary artery. By adding a resection of the supraorbital rim to the previous craniotomy to locate the eyeball protruding beyond the orbital socket, the tumor was removed with the optic nerve. Because of the invasion, the external ocular muscles were also resected but tumor removal was discontinued when the mucous membrane in the paranasal sinus was exposed.

Histological diagnosis was fibroblastic meningioma. Postoperatively, the proptosis disappeared but two years later a localized recurrence was noticed.

Case 2: A 47-year-old female had experienced left proptosis from 1975. In 1982, she visited the ophthalmological department because of a blurred vision on the left side but she did not seek further medical attention. Complete visual loss occurred soon after. She was admitted for operation of a severe proptosis (18 mm) in 1987.

On admission, her left eye exhibited chemosis with congestion, loss of light reflex, and, funduscopically, atrophied optic disc, chorioretinal fold, and venous dilatation. T1- and T2-weighted spin echo magnetic resonance (MR) images revealed a slightly high-intensity mass surrounding the optic nerve in the orbit. Carotid angiography showed a marked tumor stain from the ophthalmic artery (Fig. 2). Biopsy was performed by the lateral transorbital approach.

Immediately after the biopsy, she was transferred to the neurosurgical service. A giant tumor was subtotally removed by the transcranial supraorbital approach (Fig. 3). During craniotomy, the frontal and ethmoidal sinuses were exposed but treated with the galea. The eyeball and the extraocular muscles were preserved. The optic nerve was removed from the retrobulbar region to the optic canal which was opened. Although blepharoptosis occurred postoperatively, she had no evidence of tumor recurrence 2 years after the disappearance of proptosis.

II. Intracranial extension

Case 3: A 46-year-old female presented with a 3-
year history of right visual loss. She was admitted to the ophthalmological department in 1978 with only light perception in the right eye. Thrombosis of the retinal vein in the right eye was found, which was treated with urokinase without achieving improvement in her vision. Loss of sight occurred soon after. A CT scan revealed proptosis by 5 mm and hyper trophy of the optic nerve.

In December, 1982, she noticed left visual loss and was readmitted to the department of ophthalmology with a diagnosis of thrombosis of the left retinal vein. For the first time right intraorbital meningioma with intracranial extension was diagnosed on a CT scan. She was transferred to our neurosurgical service.

In January, 1983, the tumor extending from the right sphenoidal rim to the sellar tubercle was excised, resulting in recovery of left vision. However, she refused a secondary operation to remove the intraorbital tumor because of right-sided blindness. Afterwards, her proptosis progressed (by 15 mm). She was admitted again to the ophthalmological department in 1987. An intraorbital meningioma was demonstrated by T1-weighted MR imaging and showed contrast enhancement after administration of gadolinium-diethylenetriaminepentaacetic acid (Gd-DTPA). The optic nerve running in the tumor was also demonstrated by MR imaging. A wider operative field was obtainable by the previous craniotomy and additional resection of the supraorbital rim. The tumor occupying the region from the retrobulbar pole to the optic canal was subtotally removed, although it had invaded to the extraocular muscles through the capsule. An inadvertent intraoperative eyeball injury was sutured but resulted in phthisis bulbi.

Case 4: A 71-year-old male had experienced right visual loss from 1969. Three years later, he became blind in the right eye. He received ophthalmological examinations in 1980 when an optic nerve sheath meningioma was suspected by CT but left untreated because of the blindness. Six years later, he was referred to our neurosurgical service because of left visual loss and an intracranial extension of the tumor.

After craniotomy, the tumor extending from the right anterior clinoid process to the sellar tubercle and the suprasellar region was removed. Remote hemorrhage was found in the cerebellar hemisphere postoperatively so that external decompression was performed on the suboccipital region. Right proptosis by 9 mm was recognizable but the intraorbital operation was not performed. He was transferred to another hospital after rehabilitation.

Case 5: A 45-year-old female visited a university hospital because of left visual blurring and noticeable proptosis in January, 1972. She had left visual acuity of 0.8, papilledema, and proptosis by 2 mm. In July, her vision was lost and the optic disc was pale so that decompression of the optic canal was performed by the neurosurgeon at the same hospital. Her vision was recovered for a while but completely lost 6 months later.

She visited the ophthalmological department of our hospital in 1975 when proptosis had evolved to 12 mm. On a brain scan with a radioisotope, no intracranial tumor was revealed. Partial removal of the intraorbital tumor and enucleation of the eyeball were conducted by the lateral transorbital approach on November 26, 1975.

She was readmitted after disturbance of consciousness appeared in 1986 and referred to our neurosurgical department. Both CT scans and MR images showed a right intraorbital meningioma extending into the ethmoidal sinus, the sphenoidal sinus, and the anterior and middle cranial bases; its posterior end was observed to surpass the tentorial notch and to directly compress the brainstem. The intracranial tumor was almost totally removed after three craniotomies. The intraorbital tumor is still to be excised.

Case 6: A 37-year-old female developed left visual loss in 1982. One month later, only light perception remained. In 1983, she visited an ophthalmologist when there was no light perception but proptosis by 4 mm. Papillitis or retrobulbar tumor was suspected from the CT finding of a hypertrophied optic nerve. In 1987, she was examined again because of right visual blurring. At this time, optic atrophy and opticocilliary vessels were observed in the left optic fundus. A CT scan showed a left intraorbital tumor with intracranial extension and she was admit-
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On admission, MR images revealed a left intraorbital tumor extending into the suprasellar region and the anterior cranial base through the left superior orbital fissure with isointensity on short spin echo and slightly high intensity on long spin echo. The optic nerve in the tumor was also revealed on the MR image. Intracranial tumor was removed and the right vision recovered postoperatively. Moreover, subtotal removal was performed for the left intraorbital meningioma, the eyeball and the extraocular muscles being well preserved.

III. Pediatrics

Case 7: A 6-year-old boy had left visual loss from autumn 1985. Next year, he visited our ophthalmologist because proptosis was found on physical examination at a kindergarten. A CT scan revealed a spindle-like tumor in the left orbit and kinking of the optic nerve immediately behind the eyeball (Fig. 4 upper). A negative shadow suggesting the optic nerve in the tumor and contrast-enhancement of the tumor were also demonstrated. An x-ray of the left optic canal showed a slight enlargement. Ophthalmological findings revealed papillary pallor, residual light reflex and light perception, and proptosis by 4 mm on the left side. Because of the suspicion of optic nerve glioma, a transcranial biopsy of the intraorbital tumor was carried out. Histological diagnosis was meningioma. One month later, the proptosis had progressed to 5 mm and left vision was lost so that the intraorbital meningioma was almost totally removed. The optic nerve was cut off from the retrobulbar pole but no abnormalities were observed in the intracranial optic nerve. A CT scan 1 year after the operation showed no recurrence (Fig. 4 lower).

Case 8: A 11-year-old boy was admitted to the ophthalmological department of our hospital in 1986 with left proptosis which began about 2 years prior to admission. According to his family history, his mother died of Recklinghausen's disease. On admission, he underwent a biopsy by the lateral transorbital approach because left proptosis had developed to 10 mm and an intraorbital tumor was identified. The tumor was diagnosed as meningioma. He was treated as an outpatient because of residual vision.

Seven months later, he was referred to our neurosurgical department because light perception had disappeared, when papilledema and proptosis by 11 mm were observed. MR images showed an intraorbital meningioma exhibiting a Gd-DTPA enhancement effect with isointensity on T1-weighted image and slightly high intensity on T2-weighted image, the intratumoral optic nerve being recognizable (Fig. 5). Artificial embolization was applied to the branches of the maxillary artery, one of the feeding arteries, because a marked tumor stain was recognized on carotid angiography. Then the tumor was removed with the optic nerve. He had Recklinghausen's disease and had two isolated tumors in the cranium.

Clinical findings of these eight cases are summarized in Tables 1-3.

Discussion

Intraorbital meningioma, especially optic nerve sheath meningioma, has been found in the arach-
noidal cap cells in the intraorbital optic nerve sheath and in all the regions of optic nerves, although mostly in the apical region in the orbit. It can be classified into the two categories of primary and secondary meningioma. The latter is reported to be about twice as frequent as the former. Intraorbital ectopic meningioma have rarely been reported. It is not always easy to differentiate primary intraorbital meningiomas from secondary ones. All our cases with intracranial extension, however, were followed-up by neuro-ophthalmologists from the first clinical examination and were regarded as having primary meningiomas. It is generally reported that primary meningioma is the meningothelial or mixed type, and others are difficult to classify as primary. Our four cases with intracranial extension were of the meningothelial type while the other two cases with intraorbital localization (Cases 1 and 8) were of fibroblastic type (Table 3).

Chief complaints at the first ophthalmological examinations were unilaterally progressive visual loss and proptosis (Table 1); both were complicated during the course in all cases. Initial diagnosis, such as optic nerve glioma, retinal artery thrombosis, papillitis, retrolobular optic neuritis, etc., tended to be made 6 months to 3 years after the onset of the initial symptom. The ophthalmologist will often hesitate to carry out a biopsy when a patient still preserves vision even if a tumor is suspected after recognition of enlargement of the optic nerve on CT scan. Recently, the CT-guided fine needle biopsy has been proposed. It is necessary to evaluate this method as diagnosis can be made in as many as 80% of cases although less reliable in cases of solid tumors in the orbital apex. In our cases, delay in diagnosis allowed intracranial extension in four. The disappearance of vision, i.e., blindness, was noticed in our cases with visual loss as the first symptom within 1 month to 3 years (average, 1.5 years) and those with proptosis as the first symptom within 2.4–7 years. This difference is considered to correspond to the difference between the intradural and extradural growth of the tumor in the orbit. In fact, in Cases 2 and 8 when the first symptom was proptosis, the optic nerves could be identified through the tumor on MR images and extradural (perioptic) tumor growth was suggested. Our adult cases except for one underwent surgical removal 4–12 years later (mean, 8 years); each of them was at the stage of intracranial extension or advanced proptosis. It is necessary to make an early diagnosis and persuade patients to undergo surgical removal as soon as possible even after becoming blind. Pediatric meningioma is reported to show aggressive growth. Our two cases were obliged to undergo early surgical removal.

Neuroradiological diagnosis has become easier to use since the development of CT and MR imaging. CT scan at the first examination frequently showed enlargement, hypertrophy or apical expansion of the optic nerve. Its typical growth pattern is shown in Fig. 1. Jakobiec et al. classified the CT findings of optic nerve meningioma into diffuse with apical expansion, diffuse and narrow expansion, calcified, diffuse and irregular expansion, and diffuse with anterior expansion. It must be added, however, that the appearance of Case 7 (Fig. 4 upper) showing kinking in the retrobulbar region agrees with optic

Table 1  Clinical course of optic nerve sheath meningiomas

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Age, Sex</th>
<th>Initial symptom</th>
<th>Interval from initial symptom</th>
<th>Degree of proptosis at operation</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td></td>
<td></td>
<td>Diagnosis</td>
<td>Blindness</td>
</tr>
<tr>
<td>1</td>
<td>61, F</td>
<td>visual loss</td>
<td>1 yr</td>
<td>1 yr 8 mos</td>
</tr>
<tr>
<td>2</td>
<td>47, F</td>
<td>proptosis</td>
<td>12 yrs</td>
<td>7 yrs</td>
</tr>
<tr>
<td>3</td>
<td>46, F</td>
<td>visual loss</td>
<td>3 yrs</td>
<td>2 yrs 8 mos</td>
</tr>
<tr>
<td>4</td>
<td>61, M</td>
<td>visual loss</td>
<td>11 yrs</td>
<td>3 yrs</td>
</tr>
<tr>
<td>5</td>
<td>54, F</td>
<td>visual loss</td>
<td>6 mos</td>
<td>1 yr</td>
</tr>
<tr>
<td>6</td>
<td>37, F</td>
<td>visual loss</td>
<td>1 yr</td>
<td>1 mo</td>
</tr>
<tr>
<td>7</td>
<td>6, M</td>
<td>visual loss</td>
<td>9 mos</td>
<td>10 mos</td>
</tr>
<tr>
<td>8</td>
<td>11, M</td>
<td>proptosis</td>
<td>2 yrs</td>
<td>2 yrs 5 mos</td>
</tr>
</tbody>
</table>

*Biopsy.
nerve glioma as they proposed. This case showed a slight tumor blush by angiography, which is indicative of meningioma.8 On MR images, our cases showed iso- or slightly high intensity than those from the cerebral parenchyma on T1- and T2-weighted images. Gd-DTPA administration showed the enhancement effect (Fig. 5 left). These characteristics are the same as those in intracranial meningioma.2)

The optic nerve could frequently be followed in the tumor on MR images, which could be identified as an isointense funicular structure with a surrounding low-intense ring in three cases (Fig. 5). The surrounding low-intense ring was thought to possibly be entrapped cerebrospinal fluid but this could not be confirmed during the operation. Carotid angiography revealed a tumor stain showing dilatation and stretching of the ophthalmic artery and its branches, and downward compression. Artificial embolization was applicable in some cases receiving blood flow from the maxillary artery in case of larger

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Initial CT findings</th>
<th>CT findings at operation</th>
<th>MR imaging finding</th>
<th>Angiographic finding</th>
</tr>
</thead>
</table>
| 1        | apical expansion of optic nerve | diffuse enlargement with apical expansion, CE (+) (at 1st op), replacement of orbital soft tissue, extension into paranasal cavity (at 2nd op) | slightly high intensity on T1- and T2-weighted spin echo images, optic nerve in tumor | tumor stain from OphA and MA*
| 2        | not available | bulbous mass with irregular margin, replacement of orbital soft tissue, CE (+) | | tumor stain from OphA |
| 3        | enlargement of optic nerve | diffuse enlargement with apical expansion and intracranial extension (at IC op) replacement of orbital soft tissue (at IO op) | isointensity as gray matter on T1-weighted spin echo image with Gd-DTPA, optic nerve in tumor | tumor stain from OphA and MA |
| 4        | diffuse enlargement of optic nerve with apical expansion | apical bulbous expansion with intracranial extension | | tumor stain on tuberculum sellae and intraorbital cavity |
| 5        | not available | large extensive tumor in orbital cavity, frontal base, middle fossa, and tentorial edge region | isointensity as gray matter on T2-weighted spin echo image | intraorbital tumor stain, intracranial mass |
| 6        | diffuse and narrow expansion | intracranial extension to tuberculum sellae and sphenoidal ridge | isointensity on T1-weighted and hyperintensity on T2-weighted spin echo images, optic nerve in tumor | intraorbital tumor stain, intracranial mass |
| 7        | fusiform expansion with kinking, CE (+) and negative shadow-casting of optic nerve | | isointensity on T1-weighted spin echo image | tumor stain from OphA |
| 8        | fusiform expansion, replacement of orbital soft tissue | | isointensity on T1-weighted and slightly high intensity on T2-weighted spin echo images with Gd-DTPA, optic nerve in tumor | tumor stain from OphA and MA* |

*Artificial embolization was performed. CE: contrast enhancement, Gd-DTPA: gadolinium-diethylenetriaminepentaacetic acid, IC: intracranial, IO: intraorbital, OphA: ophthalmic artery, MA: maxillary artery.
In general, the management of intraorbital meningioma ends in surgical removal but there is some disagreement about the timing. Surgical removal is generally used to prevent intracranial extension after the disappearance of visual function. However, there are reports on cases where vision was maintained after the tumor removal when a middle-aged or old patient had an extremely small tumor in the anterior or middle region of the optic nerve sheath. One patient with visual loss for 2 years recovered after removal of a periorbit meningioma by exploratory surgery for a possible meningioma which had resulted negative on radiological investigation. Early operation to conserve vision has often been tried. On the other hand, there are some patients who lost their vision after removal of a periorbit meningioma by exploratory surgery for a possible meningioma which had resulted negative on radiological investigation. 

Several approaches have been designed for operative procedures (Table 3). The lateral orbitotomy modified Kronlein method is superior for anteriorly located meningioma, with vision expected to be preserved. However, the transorbital approach after craniotomy is indicated for tumors in the apical region. In our cases with a large tumor, the retrobulbar pole was difficult to confirm because of marked proptosis with the standard transcranial transorbital approach so that supraorbital rim resection had to be added, which was used in five cases. Moreover, the transcranial supraorbital approach to remove the supraorbital rim and the orbital roof with the bone flap en bloc (Fig. 3) was so effective that we could expose the orbital content by minimum retraction of the frontal lobe extradurally. However, this had disadvantages such as the unexpected opening of the frontal sinus and/or the ethmoidal sinus or inability to forecast its degree.

Table 3: Intraorbital operative results and follow-up

<table>
<thead>
<tr>
<th>Case No.</th>
<th>Tumor removal</th>
<th>Operative approach</th>
<th>Histology</th>
<th>Postoperative proptosis</th>
<th>Complications</th>
<th>Recurrence</th>
<th>Duration of follow-up</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>partial</td>
<td>TCTO</td>
<td>FB</td>
<td>+</td>
<td>EOM, ptosis, EOM</td>
<td>+</td>
<td>3 yrs</td>
</tr>
<tr>
<td>2</td>
<td>partial</td>
<td>TCTO-SR</td>
<td>FB</td>
<td>-</td>
<td>ptosis, EOM, wound infection</td>
<td>-</td>
<td>2 yrs 3 mos</td>
</tr>
<tr>
<td>3</td>
<td>subtotal</td>
<td>TCS</td>
<td>mixed</td>
<td>-</td>
<td>ptosis, EOM, phthisis bulbi</td>
<td>-</td>
<td>1 yr 7 mos</td>
</tr>
<tr>
<td>4</td>
<td></td>
<td></td>
<td>MT</td>
<td>-</td>
<td></td>
<td>+</td>
<td>13 yrs</td>
</tr>
<tr>
<td>5</td>
<td>partial</td>
<td>lateral OT</td>
<td>MT</td>
<td>E</td>
<td></td>
<td>-</td>
<td>1 yr 6 mos</td>
</tr>
<tr>
<td>6</td>
<td>subtotal</td>
<td>TCTO-SR</td>
<td>MT</td>
<td>-</td>
<td>ptosis, EOM</td>
<td>-</td>
<td>1 mo</td>
</tr>
<tr>
<td>7</td>
<td>biopsy</td>
<td>TCTO-SR</td>
<td>MT</td>
<td>+</td>
<td>transient ptosis, partial EOM</td>
<td>-</td>
<td>2 yrs 6 mos</td>
</tr>
<tr>
<td>8</td>
<td>biopsy</td>
<td>lateral OT</td>
<td>FB</td>
<td>+</td>
<td>transient ptosis, EOM</td>
<td>-</td>
<td>7 mos</td>
</tr>
</tbody>
</table>

TCTO: transcranial transorbital approach, TCTO-SR: transcranial transorbital approach with supraorbital rim resection, TCS: transcranial supraorbital approach (according to Maroon et al.). OT: orbitotomy, MT: meningothelialomatous, FB: fibroblastic, EOM: disturbance of external ocular muscles, E: enucleation of the eyeball was also performed.
intraorbital tumor after about 2 years. In pediatrics, positive exenteration to remove the intraorbital content has been recommended, but we did not use this. Our two cases are in a state of psychological satisfaction as postoperative blephaloptosis is improving and eyeball movements have partially recovered. But it will be necessary to carry out the exenteration when the tumor recurs in pediatric cases. From our clinical experience in eight cases, future therapeutic principles are to be investigated as follows: It is optimal to carry out early surgical removal to preserve the vision in cases of anteriorly located optic nerve meningioma, but total removal when the vision is lost in cases of other tumors after diagnosis has been established by fine needle biopsy at the stage of intradural tumor; that is at the stage of CT-identified expansion or enlargement of the optic nerve, or the stage of small perioptic tumor.

Addendum

After presentation of this manuscript, we experienced a case of total removal of a optic nerve sheath meningioma at the stage of diffuse enlargement of the optic nerve on CT scan.

A 45-year-old female was first seen by our ophthalmologist because of unilateral visual loss beginning 14 months before. A CT scan revealed enlargement of the optic nerve and proptosis by 2 mm. Her vision was observed to be completely lost during hospitalization so that the intraorbital optic nerve was resected by craniotomy. The optic canal was also unroofed but the optic nerve in this region remained normal. Fine needle aspiration biopsy was tried during the operation and its safety and usefulness in pathological diagnosis confirmed. The resected optic nerve showed tumor cells covering the surroundings of the intrathecal region and a tumor cell nest in the fatty tissue adhering to the optic nerve as well as degeneration of the optic nerve fiber.

References


Address reprint requests to: R. Kuroda, M.D., Department of Neurosurgery, Kinki University School of Medicine, 377-2 Ohno-higashi, Osaka-sayama, Osaka 589, Japan.