erative EA following surgery in medicare patients.[9] Out of 68,723 patients analyzed, 3,796 patients underwent thoracic surgery. Fifty percent of these patients had postoperative epidural usage following thoracic surgery (1,899 patients). EA has emerged as the preferred pain control. While EA has been used after thoracic surgery, it is debatable whether VATS patients would require EA because of the lower severity of postoperative pain in comparison with open thoracotomy.[1,4,6] However, it has not been clarified whether EA is necessary after VATS. To address this issue, we examined the pain score and side effects of EA in post-VATS patients using a randomized control study.

Patients and Methods

Patient entry and randomization

This study was approved by the ethics committee of Kumamoto University Hospital in August 1999. Patients who were scheduled to undergo lobectomy or partial lung resection by VATS at Kumamoto University Hospital from September 1999 to September 2001 were enrolled. Exclusion criteria included emergency cases, American Society of Anesthesiology (ASA) risk grade over III, and contraindication for EA.

At the time of entry, the patients received an explanation about the study by both an anesthesiologist and a surgeon before the operation, and a written informed consent was obtained from them. Patients who agreed to participate were randomly allocated using envelopes to one of 2 groups: an EA group and an NEA group.

Anesthesia and postoperative pain control

All patients received premedication with an intramuscular injection of atropine sulfate 0.25–0.5 mg and midazolam 0.06 mg/kg. In the operating room, epidural catheters of patients in the EA group were placed in Th5/6 or Th6/7 level before induction of general anesthesia. General anesthesia was induced with intravenous thiamylal sodium (4–6 mg/kg) and vecuronium bromide (0.1 mg/kg). Anesthesia was maintained with isoflurane, air and oxygen. Vecuronium bromide was occasionally given for muscle relaxation as required. Neither narcotics nor epidural anesthesia were used during surgery.

Immediately after surgery, patients in the EA group were given a single injection of 5 ml of 0.25% bupivacaine hydrochloride via the epidural catheter, followed by continuous infusion of 80 ml of 0.25% bupivacaine hydrochloride (90 ml in patients over 70 years old) and 1 mg of fentanyl citrate (0.5 mg in patients over 70 years old) using a balloon infuser at a rate of 2.0 ml/h. If adequate postoperative pain relief could not be achieved, additional analgesics were administered in both groups. The total dose of each additional analgesic was compared between the 2 groups.

Assessment of postoperative pain

Each patient was interviewed by anesthesiologists, who were not included by the authors in this study, every night from the day of surgery (0 POD) to the 2nd postoperative day (2 POD). The patients were asked to indicate their degree of postoperative pain on a visual analog scale (VAS) ranging from 0 mm (no pain) to 100 mm (extreme pain) on a 100-mm line drawing.[10] The verbal pain score at rest (VPS-R) was graded from 0 to 3 (0 = no pain, 1 = slight pain, 2 = moderate pain without analgesics, 3 = pain requiring analgesics). The verbal pain score on movement (VPS-M) was graded from 0 to 2 (0 = no pain on movement, 1 = pain on coughing and moving but controlled, 2 = pain on coughing or moving and not controlled).[11] The requirement for additional analgesics was examined from 0 to 2 POD. Postoperative symptoms, complications, and side effects related to EA were examined.

Operative procedure

VATS lobectomy was undertaken via one access port (12 mm) and one lateral utility thoracotomy (6 cm) without a rib retractor. Mediastinal lymph node dissection was performed routinely with VATS lobectomy. Partial lung resection was undertaken via 3 surgical ports (12 mm) for benign pulmonary nodules or bronchiolo-alveolar cell carcinomas less than 10 mm in diameter. A 28F intercostal drain was inserted through an access port incision in each patient. These drains were removed when the underlying lung was fully expanded with no residual air leak.

Statistical analysis

The χ^2 test and Fisher's exact test were used to compare discrete variables. Stepwise forward analysis was performed, with drop in VPS-R and VPS-M as dependent variables. Factors considered among the independent variables included EA, age, sex, ASA, the duration of surgery, and the duration of chest tube drainage. Statistical significance was assumed at $p<0.05$. All analysis was performed using StatView software (version 5.0, SAS Institute Inc., Cary, NC, USA). All values in the text and tables are given as mean±SD (standard deviation).

Table 1. Patient classification

	EA group	NEA group
Number	24	22
Age (years)	64.4±12.3	62.4±9.9
BMI (kg/m²)	22.9±3.2	22.9±2.7
Sex (M/F)	11/13	10/12
Duration of surgery (min)	170.7±94.8	172.0±96.5
Partial resection/lobectomy	10/14	10/12
ASA I/II/III	8/13/3	9/12/1
Chest tube drainage over 2 POD	13	12

BMI, body mass index; M, male; F, female; ASA, risk grade according to criteria set down by American Society of Anesthesiology; POD, postoperative day; EA, epidural anesthesia; NEA, non-EA.

Table 2. Number of patients required additional analgesia

Drug	Route of administration	Number of patients		p value
		EA group	NEA group	
Diclofenac sodium	Rectally	16	22	0.0040
Pentazocine	im	3	16	<0.0001
Loxoprofen sodium	Orally	6	9	0.3480

im, intramuscularly; EA, epidural anesthesia; NEA, non-EA.

Table 3. Total dose of diclofenac sodium and pentazocine per patient

Drug	EA group	NEA group	p value
Diclofenac sodium (mg)	44.8±44.2	109.1±93.7	0.0040
Pentazocine (mg)	1.9±5.1	20.5±18.7	<0.0001

EA, epidural anesthesia; NEA, non-EA.

Results

Forty-six patients were enrolled. There were 24 patients in the EA group and 22 patients in the NEA group (Table 1). There were no significant differences in age, body mass index (BMI), sex, duration of surgery, extent of surgery (partial resection or lobectomy) or ASA score between the 2 groups.

Most of the patients needed additional analgesics postoperatively until 2 POD. Rectal suppositories of diclofenac sodium, intramuscular injection of pentazocine, and oral loxoprofen sodium were commonly used in each group (Table 2). Rectal diclofenac sodium and intramuscular pentazocine were needed more frequently in the NEA group than in the EA group ($p<0.05$). Total doses of diclofenac sodium and pentazocine per patient were also higher in the NEA group than in the EA group ($p=0.0040$ and $p<0.0001$, respectively)(Table 3).

Pain scores assessed by VAS, VPS-R and VPS-M are shown in Figs. 1–3. The mean VAS scores in the NEA and EA groups were 35.3±28.6 and 18.7±25.5 at 0 POD, 29.6±20.5 and 21.6±19.1 at 1 POD, and 20.1±20.6 and 12.7±14.0 at 2 POD, respectively (Fig. 1). The mean VAS score in the EA group was significantly lower than that in the NEA group at 0 POD ($p=0.045$), but there was no significant difference at 1 and 2 POD. The mean VPS-R scores in the NEA and EA groups were 1.41±1.01 and 0.54±0.88 at 0 POD, 1.00±0.05 and 0.50±0.72 at 1 POD, and 0.73±0.83 and 0.38±0.58 at 2 POD, respectively (Fig. 2). The difference between the 2 groups was significant at 0 and 1 POD ($p=0.003$ and $p=0.046$, respectively). At 0 POD, VPS-R grade 0 (no pain at rest) was seen in 16 patients of the EA group (67%) and 4 of the NEA group (17%), while VPS-R grade 3 (pain requiring analgesics) was seen in one patient of the EA group (4%) and 4 of the NEA group (18%). At 1 POD, VPS-R grade 0 was seen in 15 patients of the EA group (63%) and 8 of the NEA group (36%), while grade 3 was

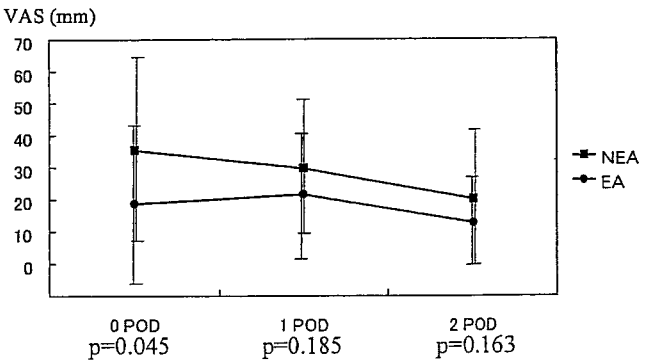

Fig. 1. Mean VAS score after VATS.
VAS, visual analogue scale; VATS, video-assisted thoracoscopic surgery; EA, epidural anesthesia; NEA, not EA; POD, postoperative day.

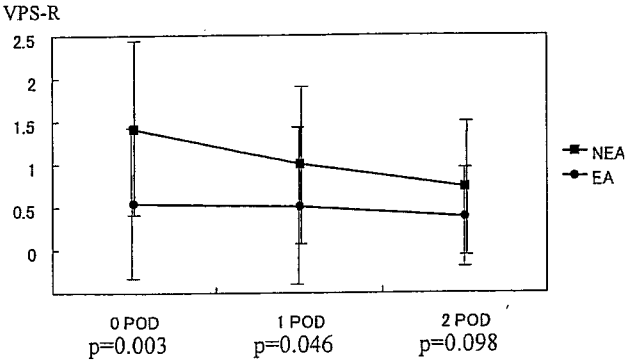

Fig. 2. Mean VPS-R score after VATS.
VPS-R, verbal pain score at rest; VATS, video-assisted thoracoscopic surgery; EA, epidural anesthesia; NEA, not EA; POD, postoperative day.

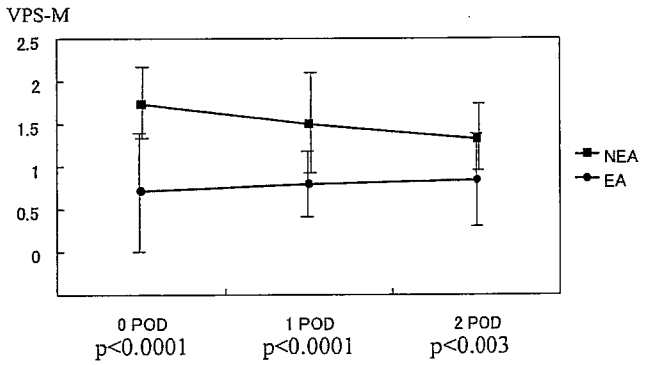

Fig. 3. Mean VPS-M score after VATS.
VPS-M, verbal pain score on movement; VATS, video-assisted thoracoscopic surgery; EA, epidural anesthesia; NEA, not EA; POD, postoperative day.

seen in none of the EA group (0%) and one of the NEA group (5%). The mean VPS-M scores in the NEA and EA groups were 1.73±0.46 and 0.71±0.69 at 0 POD, 1.50±0.60 and 0.79±0.42 at 1 POD, and 1.32±0.48 and 0.83±0.57 at 2 POD, respectively (Fig. 3). The difference between the 2 groups was significant from 0 to 2 POD ($p<0.0001$).

Table 4 presents the result of stepwise regression analysis, showing that EA was only one independent predictor of VPS-R at 0 and 1 POD ($p=0.0033$ and 0.0462). However, there was no predictor of VPS-R at 2 POD. With VPS-M, there were a few independent predictors at 0–2 POD, i.e. EA from 0 to 2 POD, ASA risk grade at 0 POD and the duration of surgery (DOS) at 2 POD ($p<0.0001$ and 0.0017).

Table 5 shows the incidence of EA-related side effects and complications after surgery. Seven patients in the EA group (29%) suffered nausea or vomiting, of which the frequency was higher than in the NEA group (1 patient, 5%)($p<0.05$). All patients who complained of nausea or vomiting were over 60 years old. While one patient in the NEA group had air leakage for more than 7 days after VATS, none of the other patients in both groups suffered any postoperative complications. EA was discontinued in 3 patients because of nausea or vomiting, but there were no incidences of marked hypotension in any of the patients in the EA group.

Discussion

In thoracic surgery, adequate control of acute postoperative pain is known to decrease postoperative morbidity and mortality and result in a lower incidence of chronic postsurgical pain.[12,13] For this reason, thoracic surgeons have pursued both minimally invasive surgery and better management of postoperative pain. VATS represents the ultimate minimally invasive surgery, and results in a significant reduction in postoperative pain.[14] EA and systemic opioids have been commonly used for postoperative analgesia. It is necessary to establish the most suitable type of pain management after VATS.[15]

There has been clinical debate about which analgesic techniques provides the most effective postoperative pain control, i.e., intravenous or epidural opioids, epidural or paravertebral local anesthetics with or without opioids, patient controlled analgesia (PCA) or continuous injection, cryoanalgesia, transcutaneous electrical nerve stimulation (TENS), or non-steroidal anti-inflammatory drugs

Table 4. Stepwise regression analysis

Dependent variable		Independent variable	Intercept	p value
VPS-R	0 POD	EA	1.409	0.0033
	1 POD	EA	1.000	0.0462
	2 POD	–	0.543	–
VPS-M	0 POD	EA, ASA	2.269	<0.0001
	1 POD	EA	1.500	<0.0001
	2 POD	EA, DOS	1.035	0.0017

VPS-R, verbal pain score at rest; VPS-M, verbal pain score on movement; POD, postoperative day; EA, epidural anesthesia; ASA, risk grade according to criteria set down by American Society of Anesthesiology; DOS, duration of surgery.

Table 5. Postoperative complication and side effect of EA

Complication	EA group	NEA group
Nausea/vomiting*	7	1
Pruritus	4	0
Vertigo	1	0
Air leakage (>7 POD)	0	1

POD, postoperative day; EA, epidural anesthesia; NEA, non-EA; *, $p<0.05$.

(NSAIDs), etc. It is important to find the best effect of each analgesic technique. Intravenous injection of narcotics is usually used for perioperative analgesia. However, we did not use intravenous narcotics in this study, because this would have made it difficult to assess the effect of EA due to the strong and long-lasting analgesic effect of narcotics.

Our first hypothesis was that there would be no need to use EA after VATS, because patients who undergo VATS suffer minimal postoperative pain. However, this study showed that patients in the EA group had less postoperative pain and needed less additional analgesics than those of the NEA group in the early postoperative period. While VAS evaluates global pain, VPS-R and VPS-M evaluate pain at rest and on movement, respectively. The incidences of pain between the 2 groups evaluated by VAS, VPS-R and VPS-M were significant at 0 POD, from 0 to 1 POD, and from 0 to 2 POD, respectively. From these results, we believe that EA is effective for control of postoperative pain until 1 POD, although other kinds of analgesics, such as NSAIDs, would be sufficient from 2 POD. It is notable that EA was effective for pain on movement from 0 to 2 POD. Controling pain on movement will improve the ability to deep breathe, cough and walk to prevent atelectasis and pneumonia. Using meta-analysis, Block et al. demonstrated that EA provided better postoperative analgesia than parenteral opioids during the early postoperative period, but not at the 4 POD.[16] Our study obtained similar results in that EA was effective until 1 POD, but that NSAIDs could control postoperaive pain from 2 POD. Nomori et al. stated that prolonged thoracic EA after limited thoracotomy significantly increased the severity of pain after withdrawal.[17] These studies demonstrated that EA should be withdrawn as soon as NSAIDs control postoperative pain.

Several complications of EA have been reported, such as nausea, vomiting, hypotension, pruritus and technical complications.[18] In this study, the patients in the EA group suffered nausea or vomiting more frequently than those in the NEA group. As all patients who complained of nausea or vomiting were over 60 years old, we consider it advisable to discontinue EA from 2 POD, especially in elderly patients.

In conclusion, results suggest that EA is recommended until 1 POD after VATS, and other kinds of analgesics should be employed from 2 POD. However, in patients who complain of side effects due to EA, such as nausea or vomiting, especially in elderly patients, EA should be discontinued within one day after surgery. Although developments have been made recently in VATS, the optimal postoperative analgesic methods have been controversial. We hope this study will help VATS surgeons to decide the optimal postoperative analgesic method.

References

1. Landreneau RJ, Hazelrigg SR, Mack MJ, et al. Postoperative pain-related morbidity: video-assisted thoracic surgery versus thoracotomy. *Ann Thorac Surg* 1993; **56**: 1285–9.

2. Waller DA, Forty J, Morritt GN. Video-assisted thoracoscopic surgery versus thoracotomy for spontaneous pneumothorax. *Ann Thorac Surg* 1994; **58**: 372–7.
3. Stammberger U, Steinacher C, Hillingger S, Schmid RA, Kinsbergen T, Weder W. Early and long-term complaints following video-assisted thoracoscopic surgery: evaluation in 173 patients. *Eur J Cardiothorac Surg* 2000; **18**: 7–11.
4. Nagahiro I, Andou A, Aoe M, Sano Y, Date H, Shimizu N. Pulmonary function, postoperative pain, and serum cytokine level after lobectomy: a comparison of VATS and conventional procedure. *Ann Thorac Surg* 2001; **72**: 362–5.
5. Macrae WA. Chronic pain after surgery. *Br J Anaesth* 2001; **87**: 88–98.
6. Li WW, Lee RL, Lee TW, et al. The impact of thoracic surgical access on early shoulder function: video-assisted thoracic surgery versus posterolateral thoracotomy. *Eur J Cardiothorac Surg* 2003; **23**: 390–6.
7. Benedetti F, Amanzio M, Casadio C, et al. Control of postoperative pain by transcutaneous electrical nerve stimulation after thoracic operations. *Ann Thorac Surg* 1997; **63**: 773–6.
8. Fernandez MI, Martin-Ucar AE, Lee HD, West KJ, Wyatt R, Waller DA. Does a thoracic epidural confer any additional benefit following video-assisted thoracoscopic pleurectomy for primary spontaneous pneumothorax? *Eur J Cardiothorac Surg* 2005; **27**: 671–4.
9. Wu CL, Hurley RW, Anderson GF, Herbert R, Rowlingson AJ, Fleisher LA. Effect of postoperative epidural analgesia on morbidity and mortality following surgery in medicare patients. *Reg Anesth Pain Med* 2004; **29**: 525–33.
10. Hazelrigg SR, Landreneau RJ, Boley TM, et al. The effect of muscle-sparing versus standard posterolateral thoracotomy on pulmonary function, muscle strength, and postoperative pain. *J Thorac Cardiovasc Surg* 1991; **101**: 394–401.
11. Richardson J, Sabanathan S. Pain management in video assisted thoracic surgery: evaluation of localised partial rib resection. A new technique. *J Cardiovasc Surg (Torino)* 1995; **36**: 505–9.
12. Landreneau RJ, Mack MJ, Hazelrigg SR, et al. Prevalence of chronic pain after pulmonary resection by thoracotomy or video-assisted thoracic surgery. *J Thorac Cardiovasc Surg* 1994; **107**: 1079–86.
13. Shuman RL, Peters RM. Epidural anesthesia following thoracotomy in patients with chronic obstructive airway disease. *J Thorac Cardiovasc Surg* 1976; **71**: 82–8.
14. Mulder DS. Pain management principles and anesthesia techniques for thoracoscopy. *Ann Thorac Surg* 1993; **56**: 630–2.
15. Horswell JL. Anesthetic techniques for thoracoscopy. *Ann Thorac Surg* 1993; **56**: 624–9.
16. Block BM, Liu SS, Rowlingson AJ, Cowan AR, Cowan JA Jr, Wu CL. Efficacy of postoperative epidural analgesia: a meta-analysis. *JAMA* 2003; **290**: 2455–63.
17. Nomori H, Horio H, Suemasu K. Comparison of short-term versus long-term epidural analgesia after limited thoracotomy with special reference to pain score, pulmonary function, and respiratory muscle strength. *Surg Today* 2001; **31**: 191–5.
18. Badner NH, Bhandari R, Komar WE. Bupivacaine 0.125% improves continuous postoperative epidural fentanyl analgesia after abdominal or thoracic surgery. *Can J Anaesth* 1994; **41**: 387–92.

Pathology International 2006; **56**: 674–677

doi:10.1111/j.1440-1827.2006.02028.x

Case Report

Sparganum mansoni parasitic infection in the lung showing a nodule

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Reported herein is a 57-year-old man infected by *Sparganum mansoni*, a kind of tapeworm, showing a solitary nodule of the middle lobe of the right lung. Because a transbronchial biopsy could not diagnose the nodule, a right middle lobectomy was performed on suspicion of malignant tumor. The lesion was diagnosed as sparganosis by histological and immuno-serological examinations. Histological examination revealed granulomatous inflammation with neutrophil and eosinophil infiltration around the worm and interstitial pneumonia surrounding the nodule. Moreover, vasculitis with foreign body giant cell was seen around the lesion. To the authors' knowledge this is the second case of sparganosis limited in the lung, and the current report presents the first detailed histological description of a pulmonary sparganosis case.

Key words: CT, ELISA, lung, sparganosis, surgery

Sparganum mansoni belongs to the cestoda (tapeworm). While it is usually parasitic in birds, snakes, frogs etc., it can also be parasitic in humans. More than 300 cases of human sparganosis have been reported worldwide, but it is the most common in East Asia. While sparganosis in humans usually appears as subcutaneous nodules all over the body, it rarely involves the internal organs such as eye, brain, and spinal cord.[1–3] On literature review there has been only one report of sparganosis limited in the lung, showing diffuse multiple pulmonary nodules, which was diagnosed by histological examination of the worm.[4] That report did not refer to histopathological changes surrounding the worm. Herein is presented a case of pulmonary sparganosis with a solitary nodule, which was diagnosed on both histological and immuno-serological examination. We report here unique histopathological changes around the worm.

CLINICAL SUMMARY

The patient was a 57-year-old man. He had unusual eating habits, and had often eaten raw chicken, cooked snakes, and cooked frogs since childhood. He was referred to Division of Thoracic Surgery, Minami Kyushu National Hospital because of an abnormal pulmonary shadow on a routine chest X-ray in June 2004. He had no symptoms. There were no lesions anywhere on the body. Chest X-ray and chest CT demonstrated a poorly defined mass 1.5 cm × 1.2 cm in size adjacent to the blood vessel, with ground-glass opacity around the mass in the right middle lobe (Fig. 1). Brain magnetic resonance imaging (MRI) and abdominal CT showed no abnormality. Hematological examination showed no abnormal findings, and peripheral eosinophilia was not detected (WBC 5300/mm^3, eosinophils 3.1%). While a transbronchial biopsy could not diagnose the lesion, we performed a right middle lobectomy because of a suspicion of lung cancer in June 2004. An intraoperative frozen section diagnosed the lesion as an infectious granuloma. The patient is free of disease without any further treatment 1 year after surgery.

PATHOLOGICAL FINDINGS

The tumor was grayish white on cut surface, and the outer margin of the tumor that measured approximately 1.7 × 1.2 cm was unclear. The core of the tumor, which measured 0.4 × 0.2 cm, contained some 0.1–0.2 cm oval structures (Fig. 2). Histological examination demonstrated that the

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Received 13 February 2006. Accepted for publication 17 July 2006.

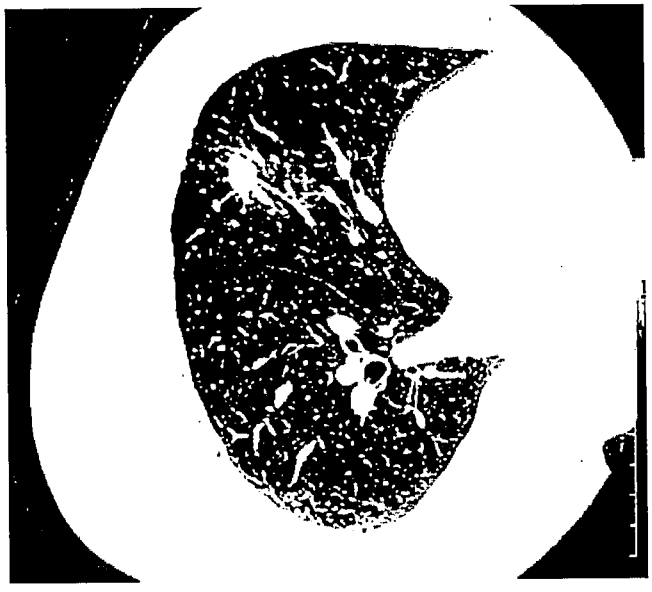

Figure 1 Computed tomography of the lung. A poorly defined mass is located in the middle lobe of the right lung (arrow).

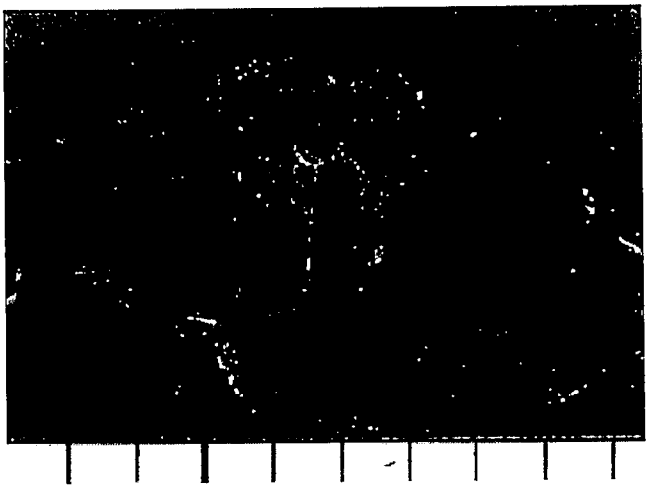

Figure 2 Tumor on cut section. The tumor is 1.7 × 1.2 cm in size. The core of the tumor surrounded by interstitial lung changes is composed of some 0.1–0.2 cm oval structures.

oval structures at the center of the tumor was a tapeworm 3.5 × 0.8 mm in size (Fig. 3a). The wall of the worm had thick eosinophilic tegument and subtegumental cells. The worm body contained numerous smooth muscle fiber bundles and characteristic calcareous corpuscles (Fig. 3b). The worm was surrounded by necrotic tissue with neutrophils and eosinophils (Fig. 3b) and granulomatous inflammation with multinucleated giant cells (Fig. 3c). In the lung tissue around the worm-containing tumor, interstitial inflammation occurred, and lymphocytic infiltrations with eosinophils and foreign body reactions were seen in the alveolar septum (Fig. 3d). Furthermore, the lymphocytic infiltrations with foreign body reactions involved the vessels and the tumor (Fig. 3e). An obstructive vasculitis was also seen.

The multiple-dot ELISA of the serum taken 8 days after the surgery was positive for antisparganum antibody.

Based on these findings, the lesion was diagnosed as intrapulmonary migration of *Sparganum*.

DISCUSSION

Sparganosis in human is a parasitic disease caused by plerocercoid larva (third stage of the worm) of the genus *Spirometra*. The adults can be parasitic in the definitive hosts (dogs, cats, and other carnivores), residing in their gastrointestinal tract. The eggs of *Spirometra*, which are approximately 60 μm in size, are excreted into stool of the definitive hosts, and then grow to the first stage (coracidium) in water. They grow up to the second stage (procercoid larvae, approximately 550 μm in length) in the first intermediate hosts, animal plankton (usually *Cyclops*). The first intermediate hosts infected by the procercoid larvae are eaten by the second intermediate hosts (usually birds, snakes, frogs, pigs etc.), in which the worms grow to the third stage (plerocercoid larvae, ≥20 mm in length) in subcutaneous tissue or muscles. Finally, they grow to adult *Spirometra*, up to approximately 60–100 cm in length, in the definitive hosts. Humans could be infected in three ways: (i) drinking water containing an infected animal plankton; (ii) eating raw or partially cooked flesh of the second intermediate hosts, such as birds, snakes, and frogs; and (iii) being directly infected via skin or mucous membrane by the application of a poultice of raw flesh to wounds (a folk remedy in some countries).[5] Because the present patient had eaten raw chicken, cooked snakes, and cooked frogs, he could have been infected by ingesting plerocercoid larvae, the third stage of the worm in these second intermediate hosts.

In humans the worm usually passes through the intestinal wall and the peritoneum, and finally reaches the subcutaneous tissues. However, it has been reported that the worm rarely migrates into the tissue around major vessels.[6] We therefore consider that, in the present patient without subcutaneous involvement, the worm might have penetrated through the intestinal wall, migrated into the tissue around the major vessel, and then reached the lung. The vasculitis observed in the present case was accompanied by foreign body reactions against the worm. This suggests that the worm can move into the lung along the vessels.

Definitive diagnosis can be established by microscopic identification of the worm in parasitic lesion. Histopathologically, the section of worm has non-cellular eosinophilic tegument, cellular subtegument, and parenchyma containing numerous bundles of muscle fibers, excretory canal, and calcareous corpuscles, findings of which are characteristic of

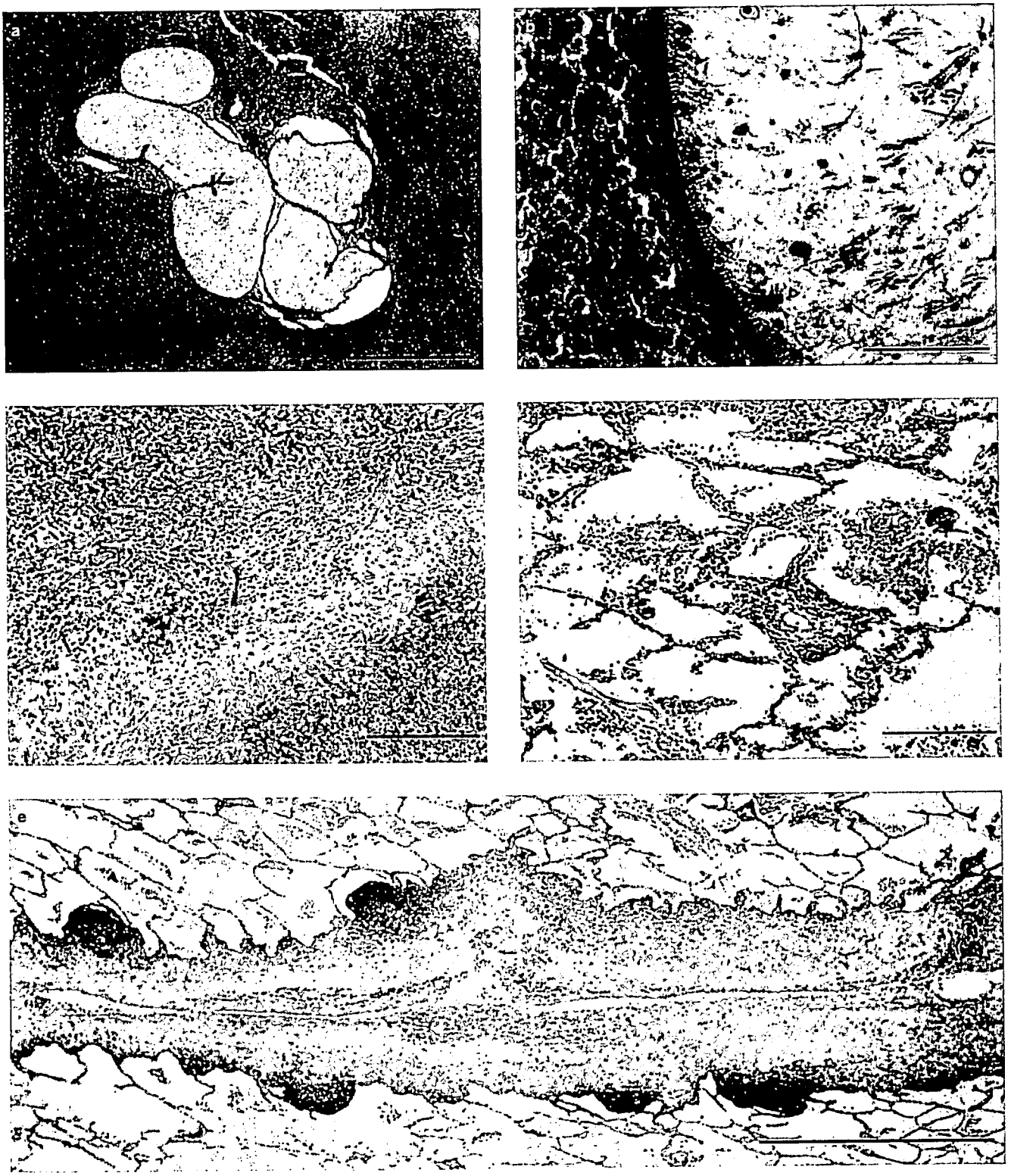

Figure 3 Microscopic appearance of the tumor. (**a**) The main feature of the tumor is a tapeworm. (**b**) The worm has non-cellular eosinophilic tegument and subtegumental cells in the wall. Parenchyma is composed of numerous bundles of smooth muscle fiber and calcareous corpuscles. The worm is surrounded by necrotic tissue, neutrophils and eosinophils. Eosinophilic infiltration is mild. (**c**) Granulomatous inflammation with multinucleated giant cells is seen around the worm. (**d**) Lymphocytic infiltration with foreign body reactions is seen in the alveolar and perivascular tissues around the tumor. (**e**) Inflammation with foreign body reactions is seen in the vessel and perivascular area. (**a**,**e**, bar, 1 mm; **b**, bar, 50 μm; **c**,**d**, bar, 200 μm).

cestodes. The absence of a protoscolex excludes larva of *Taenia solium* (cysticercosis). Cysticercosis often produces multiple nodules but sparganosis usually presents as a solitary nodule.[7] So the first reported case with multiple lung nodules may be cysticercosis instead of sparganosis.[4] Immunoserological examination for diagnosis of sparganosis has been demonstrated to be useful,[8] and this was used for diagnosis in the present patient.

The first choice of therapy is surgical removal. While the present patient needed resection of the right middle lobe due to suspicion of lung cancer, he is now free of disease 1 year after surgery.

REFERENCES

1 Mougeot G, Cambon M, Menerath JM *et al.* Human eye anterior chamber sparganosis. *Parasite* 1999; **6**: 365–7.

2 Jeong SC, Bae JC, Hwang SH *et al.* Cerebral sparganosis with intracerebral hemorrhage: A case report. *Neurology* 1998; **50**: 503–6.

3 Lo YK, Chao D, Yan SH *et al.* Spinal cord proliferative sparganosis in Taiwan: A case report. *Neurosurgery* 1987; **21**: 235–8.

4 Phunmanee A, Boonsawat W, Indharapoka B *et al.* Pulmonary sparganosis: A case report with five years follow-up. *J Med Assoc Thai* 2001; **84**: 130–35.

5 Griffin MP, Tompkins KJ, Ryan MT. Cutaneous sparganosis. *Am J Dermatopathol* 1996; **18**: 70–72.

6 Imamura Y, Akasaka T, Tomichi N. A case of *Sparganosis mansoni*. *Rinshouhihuka* 1992; **46**: 747–9 (in Japanese).

7 Tsutsumi Y. Cysticercosis, echinococcosis. In: Tsutsumi Y, ed. *Atlas of Infectious Disease Pathology*. Tokyo: Bunkodo, 2000; 252–5 (in Japanese).

8 Kanazawa T. Paragonimiasis and sparganosis mansoni. *Nihonkyouburinshou* 2002; **61**: 1045–52 (in Japanese).

Case Report

Obstructive Subglottic Granuloma after Removal of a Minitracheostomy Tube

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We report herein a patient with subglottic granuloma after removal of a minitracheostomy tube (Minitrach II, SIMS Portex Inc., Hythe, Kent, UK). The patient underwent pulmonary resection for lung cancer followed by insertion of the minitracheostomy tube for prevention of sputum retention. The tube was removed 4 days after insertion. Twelve weeks later, the patient developed severe dyspnea and stridor. Bronchoscopy showed an obstructive subglottic granuloma arising from the anterior wall. The granuloma was removed by coring out using a conventional tracheal tube, followed by local injection of methylprednisolone acetate. The patient is now asymptomatic without regrowth of the granulation tissue 12 weeks after the treatment. With complication in mind, attention should be paid to patients suffering dyspnea or stridor after removal of a minitracheostomy tube. (Ann Thorac Cardiovasc Surg 2006; 12: 265–6)

Key words: minitracheostomy, subglottic stenosis, subglottic granuloma

Introduction

The minitracheostomy (Minitrach II, SIMS Portex Inc., Hythe, Kent, UK) is a 4-mm diameter cricothyroidostomy tube that can be inserted percutaneously.[1)] A minitracheostomy device is sometimes inserted after thoracic surgery for prevention of sputum retention.[2)] Complications caused by minitracheostomy tubes reported up to now have included bleeding, local hematoma and hoarseness.[1,3)] Here we report on a patient with obstructive subglottic granuloma 12 weeks after removal of a minitracheostomy tube.

Case Report

A 58-year-old man underwent an uncomplicated left upper lobectomy for squamous cell carcinoma on November 27, 2004. The patient had a history of ischemic heart disease, and his preoperative forced expiratory volume in 1 second was 50% of the predicted value. He had failed to stop tobacco smoking before the operation. A mini-tracheostomy tube was inserted for prevention of sputum retention immediately after the operation, using the Seldinger technique through the cricothyroid membrane. The postoperative course was uneventful. The tube was removed on the 4th postoperative day, and the patient was discharged on the 8th postoperative day. Eight weeks after extubation, the patient developed stridor and gradually worsening dyspnea. Twelve weeks after extubation, he was admitted to our hospital because of his severe breathing difficulty. Bronchoscopy showed an obstructive granuloma arising from the anterior wall of the subglottic area (Fig. 1). Under local anesthesia, the granuloma was cored out using a conventional tracheal tube, resulting in removal of most of the granuloma tissue (Fig. 2). Twenty milligrams of methylprednisolone acetate was locally injected via the cricothyroid membrane. The patient's symptoms disappeared immediately after removal of the granuloma. Pathological examination showed inflammatory granulation without specific findings. The patient is asymptomatic without regrowth of the granuloma tissue 12 weeks after the treatment.

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Received July 28, 2005; accepted for publication February 1, 2006.
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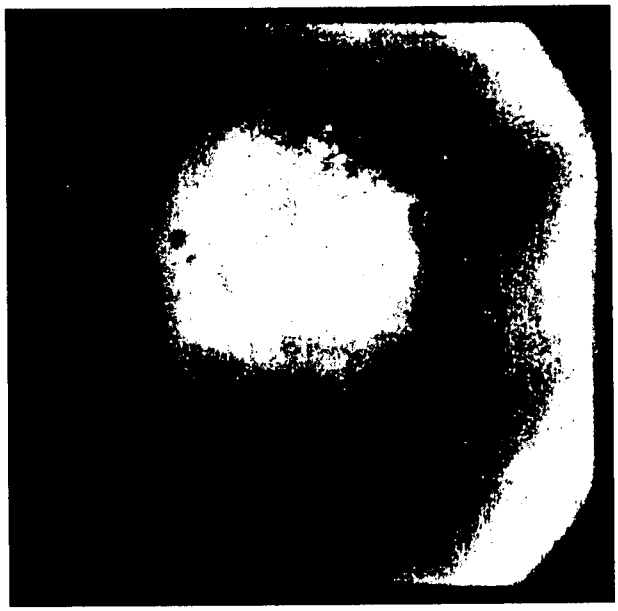

Fig. 1. Bronchoscopy showed subglottic granuloma from the anterior wall of the trachea obstructing the subglottic area.

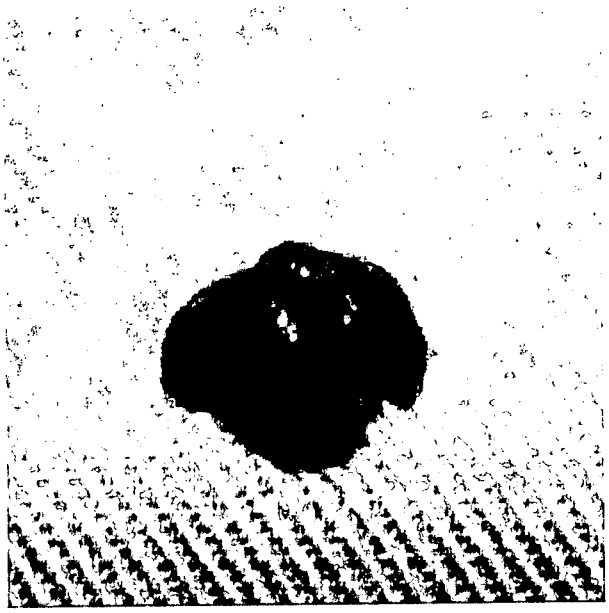

Fig. 2. Gross photograph of resected subglottic granuloma.

Discussion

Insertion of a minitracheostomy tube through the cricothyroid membrane is useful for removing airway secretions after thoracic surgery, especially in patients with a high risk of atelectasis and pneumonia. There have been several reports of complications associated with the use of a minitracheostomy tube, including bleeding, local hematoma and hoarseness.[1,3] However, to our knowledge, there have been no reports of obstructive granuloma after minitracheostomy. Subglottic stenosis usually occurs secondary to endotracheal intubation, surgical tracheostomy, trauma and systemic diseases such as amyloidosis, tuberculosis and Wegener's granulomatosis. Park et al. reported that injury to the subglottic area might result in stenosis because the cricoid cartilage is a complete ring lacking flexibility, the blood supply to the cricoid cartilage is limited, and the cricoid cartilage is susceptible to local infection, causing granulation.[4] Raghuraman et al. reported that fractures of the cricoid cartilage might also be responsible for subglottic stenosis following percutaneous dilatational tracheostomy.[5] While the present patient did not suffer fracture of the cricoid cartilage, minute injury to the cartilage might have occurred during insertion of the minitracheostomy tube, causing local infection which then led to obstructive granuloma long after extubation. This complication should be kept in mind and attention should be paid to patients who develop stridor or dyspnea after removal of a minitracheostomy tube.

References

1. Wain JC, Wilson DJ, Mathisen DJ. Clinical experience with minitracheostomy. *Ann Thorac Surg* 1990; **49**: 881–6.
2. Bonde P, Papachristos I, McCraith A, et al. Sputum retention after lung operation: prospective, randomized trial shows superiority of prophylactic minitracheostomy in high-risk patients. *Ann Thorac Surg* 2002; **74**: 196–203.
3. Browne J, McShane D, Donnelly M. An unusual complication of minitracheostomy. *Eur J Anaesthesiol* 1999; **16**: 571–3.
4. Park SS, Streitz JM Jr, Rebeiz EE, Shapshay SM. Idiopathic subglottic stenosis. *Arch Otolaryngol Head Neck Surg* 1995; **121**: 894–7.
5. Raghuraman G, Rajan S, Marzouk JK, Mullhi D, Smith FG. Is tracheal stenosis caused by percutaneous tracheostomy different from that by surgical tracheostomy? *Chest* 2005; **127**: 879–85.

Usefulness and complications of computed tomography-guided lipiodol marking for fluoroscopy-assisted thoracoscopic resection of small pulmonary nodules: Experience with 174 nodules

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Objective: Several techniques have been reported for the localization of small pulmonary nodules in thoracoscopic resection. In the present study we examined the usefulness and complications of computed tomography-guided lipiodol marking for thoracoscopic resection in our experience of 174 nodules.

Methods: Computed tomography-guided lipiodol marking was performed on 174 nodules less than 30 mm in size. Of these nodules, 45 showed ground-glass opacity images and 129 showed solid images on computed tomography. The mean size of the nodules was 10 ± 6 mm (range, 2-30 mm), and their mean depth from the pleural surface was 10 ± 7 mm (range, 0-30 mm). One to 7 days before thoracoscopy, all of the nodules were marked with 0.4 to 0.5 mL of lipiodol by using computed tomography. The marked nodules were grasped with a ring-shaped forceps during fluoroscopy and resected by means of thoracoscopy.

Results: All the nodules could be marked and localized by means of fluoroscopy as a clear spot during thoracoscopic surgery. Complications of the marking were chest pain requiring analgesia in 16 (11%) patients, hemosputum in 11 (6%) patients, pneumothorax in 30 (17%) patients, and hemopneumothorax in 1 (0.6%) patient. Eleven (6%) patients with pneumothorax required drainage, and the patient with hemopneumothorax required an emergency operation. No other complications were observed.

Conclusion: Lipiodol marking is a useful, safe, and inexpensive procedure for localizing ground-glass opacity lesions, small pulmonary nodules, or both for thoracoscopic resection.

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Received for publication Feb 22, 2006; revisions received April 2, 2006; accepted for publication April 20, 2006.

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J Thorac Cardiovasc Surg 2006;132:320-4

0022-5223/$32.00

doi:10.1016/j.jtcvs.2006.04.012

Recently, small pulmonary nodules have been frequently detected with high-resolution computed tomography (CT).[1,2] Performing a percutaneous or transbronchial biopsy for small pulmonary nodules is often difficult, and hence thoracoscopic surgical techniques have been used in diagnostic excisional biopsies, as well as in therapeutic resection. However, a major factor that limits the success of thoracoscopic resection in the case of small or deeply situated pulmonary nodules is the difficulty in localizing the target nodule because of the lack of digital palpation. Furthermore, a bronchioloalveolar carcinoma with a ground-glass opacity (GGO) finding on CT cannot be palpated or visualized frequently, even in the case of lesions that are located just beneath the visceral pleura. Therefore several techniques have been reported for localization of such small nodules or GGO lesions.[3-18] However, these techniques can result in complications, such as pneumothorax, hemorrhage, and air embolism. Since 1999, we have used CT-guided

Abbreviations and Acronyms
CT = computed tomography
GGO = ground-glass opacity
TTNA = transthoracic needle aspiration

lipiodol marking for the thoracoscopic resection of such lesions. The purpose of our study is to examine the usefulness and safety of this procedure.

Materials and Methods

Eligibility

The CT-guided lipiodol marking was approved by the ethics committee of Saiseikai Central Hospital in January 1999. Written informed consent was obtained from all patients after they discussed the risks and benefits of the procedure with the surgeons. The nodules that were thought to be difficult to localize during thoracoscopy, such as GGO lesions, nodules situated at a considerable depth from the pleural surface, and nodules smaller than 1 cm, were candidates for this procedure. The following nodules were excluded: (1) nodules larger than 30 mm in size; (2) nodules located within the inner two third of the lung; and (3) solid nodules that were larger than 10 mm in size and located within 10 mm from the pleural surface.

Patients

Between January 1999 and June 2005, CT-guided lipiodol marking was performed on 174 pulmonary nodules in 150 patients. The mean age of the patients was 62 ± 11 years (range, 35-84 years). Table 1 shows the characteristics of the nodules. The mean size of the nodules was 10 ± 6 mm (range, 2-30 mm). Their mean distance from the pleural surface was 10 ± 7 mm (range, 0-30 mm). There were 45 GGO lesions and 129 solid lesions. All the nodules were detected with chest CT; however, they could not be detected clearly with chest roentgenograms.

Marking Technique

The procedure used for marking was as follows. After 0.5 mg of atropine and 15 mg of pentazocine was injected, the patients were placed on the CT table in a suitable position (supine or prone). A scaled paper with metal wires was placed firmly on the patient, and the CT scan was performed. The shortest distance from the nodule to the thoracic wall was selected as the injection site (Figure 1, *A*). The site for marker injection was marked on the skin, and the angle and depth of the needle required to reach the nodule were determined. After local anesthesia was administered to the thoracic wall, a 22- or 23-gauge needle was introduced from the point marked on the skin to the nodule, in keeping with the angle and depth measured. The syringe was withdrawn to confirm that no blood had flowed backward, and 0.4 to 0.5 mL of lipiodol (Lipiodol Ultrafluid; Laboratoire Guerbet, Aulnay-Sous-Bois, France), which is generally used as a contrast medium for lymphatic vessels, was then injected in a single shot. The presence of the injected materials was confirmed by means of CT after the marking (Figure 1, *B*). Thoracoscopic surgery was performed 1 to 7 days (mean, 1.7 ± 1.1 days) after marking.

TABLE 1. Characteristics of the nodules

Mean size, mm (range)	10 ± 6 (2-30)
Mean distance from the pleura, mm (range)	10 ± 7 (0-30)
Location	
Right upper lobe	42
Right middle lobe	21
Right lower lobe	37
Left upper lobe	34
Left lower lobe	40
CT findings	
Ground-glass opacity	45
Solid	129

CT, Computed tomography.

Thoracoscopic Resection Technique

Thoracoscopy was performed during one-lung anesthesia by using a double-lumen tube. A C-arm–shaped fluoroscopic unit was used to detect the radiopaque nodules, and the radiopaque nodule was grasped with a ring-shaped forceps during fluoroscopy in multiple projections (Figure 1, *C*). The forceps was then moved in several directions to confirm that the nodule was grasped within a ring of the forceps. The grasped nodule was resected with an endostapler. The resected specimens were removed with a surgical bag. Successive resection of the nodules was confirmed by viewing the radiopaque nodule within the resected specimen during fluoroscopy. The nodules were histologically diagnosed by means of routine intraoperative pathologic examination, with the exception of the GGO lesions that were less than 10 mm in size; these were histologically diagnosed by a permanent section.

Results

All the nodules could be marked with lipiodol on the CT images. Even when the lipiodol could not mark within the nodules, it marked within 10 mm from the nodules, which caused no difficulty in their localization. All the nodules were successfully localized and resected during thoracoscopy without conversion to open thoracotomy. Even a nodule that was marked 7 days before the thoracoscopy could be detected during fluoroscopy as a clear spot. Of the 174 nodules, 107 (61%) were diagnosed as malignant, and 67 (39%) were diagnosed as benign (Table 2). For the 81 patients with primary lung cancers, 48 were followed with thoracoscopic lobectomy, and 10 were followed with thoracoscopic segmentectomy. The other 23 patients did not undergo the additional resection because of bronchioloalveolar cell carcinoma of the intraoperative frozen section, poor risk of the patients, or both. All of the surgical margins in patients treated with thoracoscopic wedge resection showed no malignancy.

Table 3 shows the complications accompanying lipiodol marking. Sixteen (11%) patients had chest pain that required analgesia. Eleven (6%) patients had a little hemosputum. Thirty (17%) patients had pneumothorax, and 11 (6%) of these patients required drainage. One (0.6%) patient had

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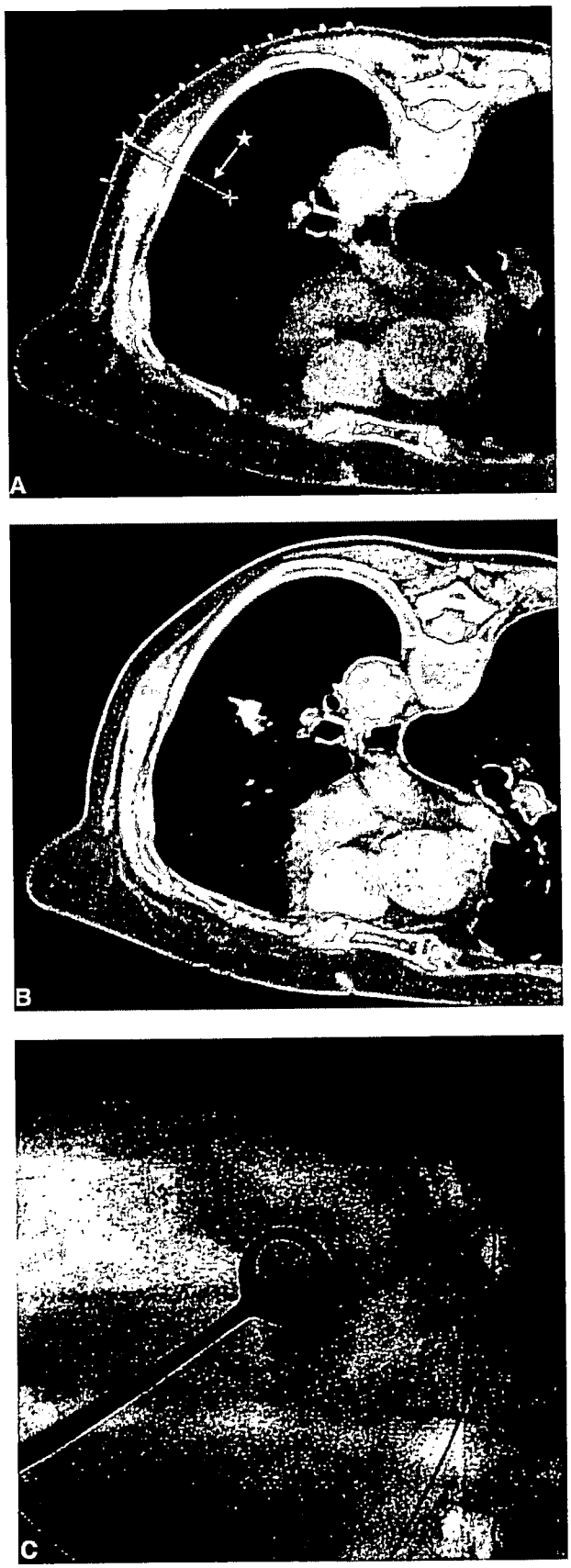

TABLE 2. Pathologic diagnosis of the nodules

Malignant (n = 107)	
Primary lung cancer	81
Metastatic lung cancer	26
Benign (n = 67)	
Tuberculosis	36
Old inflammation	11
Pulmonary lymphoid tissue	9
Atypical adenomatous hyperplasia	8
Hamartoma	3

hemopneumothorax caused by peeling off of the pleural adhesion, including blood vessels.

One patient had bilateral pulmonary nodules, both of which were marked by lipiodol; the patient was given a diagnosis of metastatic colon cancer by means of an ipsilateral thoracoscopy. Therefore the marked contralateral nodule was followed by CT, which had been marked by lipiodol for 3 months.

Discussion

Several methods have been reported for the localization of small pulmonary nodules. These include the hook-wire technique,[3-7] endoscopic ultrasonography,[8,9] barium marking through bronchoscopy,[10,11] and percutaneous injections of dyes,[12,13] colored collagen,[14] lipiodol,[15,16] agar,[17] and barium.[18] The original dye method, because of rapid diffusion around the lung tissue after injection, has the following drawbacks: (1) the marking must be performed within 3 hours before the thoracoscopy to enable dye detection, and therefore both CT and the operating room must be used simultaneously, and (2) the injection site appears blurred because of the diffusion. With the agar marking procedure, it is difficult to localize a deeply situated nodule because the palpation of the marked nodules is requisite. The ultrasound technique requires complete collapse of the lung, which is often impossible in patients with emphysema, resulting in a failure rate of 40% in localizing the nodules.[9] The hook-wire technique has been recently reported to cause massive air embolism,[19-23] which led to its prohibition in Japan. Barium marking with a CT-guided bronchoscopy is complicated because it requires simultaneous use of bronchoscopy and CT, and marking one nodule with this procedure

Figure 1. A, Computed tomographic *(CT)* scan showing a nodule with ground-glass opacity *(GGO)* in the left lower lobe. *The shortest distance from the nodule to the thoracic wall was selected as the injection site. The several lines on the chest wall reveal the metal wires of scaled-paper.* (B) CT showing the tumor marked with lipiodol. C, Intraoperative fluoroscopic imaging showing the radiopaque nodule grasped within a ring-shaped forceps. The nodule is indicated with an *arrow.*

Table 3. Complications of lipiodol marking

	No. of patients (%)
Chest pain requiring analgesia	16 (11)
Hemosputum	11 (6)
Pneumothorax	30 (17)
No treatment	19 (11)
Drainage	11 (6)
Hemopneumothorax	1 (0.6)

requires approximately 30 minutes (range, 15-60 minutes).[11] In addition, barium itself can be seen as a lesion in hematoxylin and eosin–stained sections and also can cause an inflammatory change of the lung tissue, which might make a histologic diagnosis difficult.

We previously marked a nodule with lipiodol and the pleural surface with colored collagen; this enabled a comparatively easier localization of the nodules than when only lipiodol was used.[16] However, 1 mL of collagen costs approximately $80.00. Therefore since March 2002, we started using only lipiodol to mark pulmonary nodules. Thus without using colored collagen, the nodules marked with lipiodol could be localized during fluoroscopy without great difficulty, resulting in a success rate of 100% for thoracoscopic biopsy. The marking procedure with lipiodol has the following advantages: (1) overresection of the normal lung tissue around the nodules was prevented because lipiodol marked the nodules as clear spots that were less than 1 cm in size during fluoroscopy; (2) the lipiodol remained for a long time, up to 3 months after the marking, which solves the problem of requiring both CT and the operating room simultaneously; (3) although the barium marking procedure affects pathologic findings caused by the inflammatory response and barium itself, lipiodol did not affect the pathologic findings; and (4) even in the case of deeply situated nodules (ie, up to 30 mm from the pleural surface in the present study), the lipiodol marking could easily localize the nodules as a clear spot because it diffused only to a small extent.

Transthoracic needle aspiration (TTNA) is also effective in the diagnosis of pulmonary nodules.[24-26] Although TTNA has been reported to have a false-negative rate of 3% to 11%,[24] Layfield and colleagues[25] reported that its diagnostic accuracy decreased to 60% for lesions smaller than 10 mm. Kashiwabara and associates[26] reported that the positive diagnostic rate for nonmalignant lesions by using TTNA was only 56%. All the nodules in the present study were GGO lesions or small nodules deeply situated within the lung, which were not only difficult to diagnose with TTNA but also difficult to locate by means of thoracoscopic inspection without marking. We therefore believe that GGO lesions or small nodules that are deeply situated should be diagnosed by means of thoracoscopic biopsy with preoperative marking.

Although the complications of lipiodol marking included temporary pain, a little hemosputum, pneumothorax, and hemopneumothorax, all of them arose because of the insertion of the needle into the lung and not because of the lipiodol itself. Although we did not encounter air embolism during the lipiodol marking, the frequency of massive air embolism during percutaneous needle insertion into the lung has been reported to be 0.02% to 0.07%.[21] There have been reports of 6 patients who experienced a massive air embolism during percutaneous marking procedures.[19-23] In 5 of the 6 patients, it was caused by the hook-wire technique and in 1 patient by the needle-marker procedure. The massive air embolisms could have occurred because of simultaneous injury of the bronchiole and the adjacent pulmonary vein.[23] Because all 6 patients with massive air embolism had nodules in the lower lobe, we believe that the lung tissue of the lower lobe could be injured more easily by the hook wire or needle marker than that of upper lobe because the former moves more with respiration during insertion of these apparatuses than the latter. Therefore we usually inject lipiodol immediately after needle insertion without confirming the location of the needle tip by means of CT scanning, enabling the time of placing the needle in the lung tissue to be less than 10 seconds, which could decrease the lung damage caused by the needle.

Lipiodol itself poses a potential risk of embolism because it is water insoluble. We therefore take the following precautions: (1) before injection of lipiodol, the syringe is withdrawn to confirm that blood has not flowed backward, and (2) a minimum amount of lipiodol, up to 0.5 mL, is injected.

Although the lipiodol marking procedure showed some complications caused by the needle insertion, they were not of a serious nature. Because it is a simple, safe, and inexpensive method for localizing GGO lesions, small and deeply situated pulmonary nodules, or both, we believe that lipiodol marking is one of the gold standard procedures for localization of these nodules during thoracoscopy.

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References

1. Kaneko M, Eguchi K, Ohmatsu H, et al. Peripheral lung cancer: screening and detection with low-dose spiral CT versus radiography. *Radiology.* 1996;201:798-802.
2. Swensen SJ, Jett JR, Hartman TE, et al. CT screening for lung cancer: five-year prospective experience. *Radiology.* 2005;235:259-65.
3. Dendo S, Kanazawa S, Ando A, et al. Preoperative localization of small pulmonary lesions with a short hook wire and suture system: experience with 168 procedures. *Radiology.* 2002;225:511-8.
4. Wicky S, Dusmet M, Doenz F, et al. Computed tomography-guided localization of small lung nodules before video-assisted resection: experience with an efficient hook-wire system. *J Thorac Cardiovasc Surg.* 2002;124:401-3.
5. Mack MJ, Gordon MJ, Postma TW, et al. Percutaneous localization of pulmonary nodules for thoracoscopic lung resection. *Ann Thorac Surg.* 1992;53:1123-4.

6. Plunkett MB, Peterson MS, Landreneau RJ, Ferson PF, Posner MC. Peripheral pulmonary nodules: preoperative percutaneous needle localization with CT guidance. *Radiology.* 1992;185:274-6.
7. Shah RM, Spirn PW, Salazar AM, et al. Localization of peripheral pulmonary nodules for thoracoscopic excision. *Am Roentgenol Surg.* 1992;161:1279-83.
8. Sortini D, Feo CV, Carcoforo P, et al. Thoracoscopic localization techniques for patients with solitary pulmonary nodule and history of malignancy. *Ann Thorac Surg.* 2005;79:258-62.
9. Shennib H, Bret P. Intraoperative transthoracic ultrasonographic localization of occult lung lesions. *Ann Thorac Surg.* 1993;55:67-9.
10. Asano F, Shindoh J, Shigemitsu K, et al. Ultrathin bronchoscopic barium marking with virtual bronchoscopic navigation for fluoroscopy-assisted thoracoscopic surgery. *Chest.* 2004;126:1687-93.
11. Okamura T, Kondo H, Suzuki K, et al. Fluoroscopy-assisted thoracoscopic surgery after computed tomography-guided bronchoscopic barium marking. *Ann Thorac Surg.* 2001;71:439-42.
12. Endo M, Kotani Y, Satouchi M, et al. CT fluoroscopy-guided bronchoscopic dye marking for resection of small peripheral pulmonary nodules. *Chest.* 2004;125:1747-52.
13. Kerrigan DC, Spence PA, Crittenden MD, Tripp MD. Methylene blue guidance for simplified resection of a lung lesion. *Ann Thorac Surg.* 1992;53:163-4.
14. Nomori H, Horio H. Colored collagen is a long-lasting point marker for small pulmonary nodules in thoracoscopic operations. *Ann Thorac Surg.* 1996;61:1070-3.
15. Moon SW, Wang YP, Jo KH, et al. Fluoroscopy-aided thoracoscopic resection of pulmonary nodule localized with contrast media. *Ann Thorac Surg.* 1999;68:1815-20.
16. Nomori H, Horio H, Naruke T, et al. Fluoroscopy-assisted thoracoscopic resection of lung nodules marked with lipiodol. *Ann Thorac Surg.* 2002;74:170-3.
17. Tsuchida M, Yamato Y, Aoki T, et al. CT-guided agar marking for localization of nonpalpable peripheral pulmonary lesions. *Chest.* 1999; 116:139-43.
18. Iwasaki Y, Nagata K, Yuba T, et al. Fluoroscopy-guided barium marking for localizing small pulmonary lesions before video-assisted thoracic surgery. *Respir Med.* 2005;99:285-9.
19. Kamiyoshihara M, Sakata K, Ishikawa S, et al. Cerebral arterial air embolism following CT-guided lung needle marking. Report of a case. *J Cardiovasc Surg (Torino).* 2001;42:699-700.
20. Horan TA, Pinheiro PM, Araujo LM, et al. Massive gas embolism during pulmonary nodule hook wire localization. *Ann Thorac Surg.* 2002;73:1647-9.
21. Sakiyama S, Kondo K, Matsuoka H, et al. Fatal air embolism during computed tomography-guided pulmonary marking with a hook-type marker. *J Thorac Cardiovasc Surg.* 2003;126:1207-9.
22. Sato K, Miyauchi K, Shikata F, et al. Arterial air embolism during percutaneous pulmonary marking under computed tomography guidance. *Jpn J Thorac Cardiovasc Surg.* 2005;53:404-6.
23. Ohi S, Itoh Y, Neyatani H, et al. [Air embolism following computed tomography-guided lung needle marking; report of a case]. *Kyobu Geka.* 2004;57:421-3.
24. Sisler GE. Malignant tumors of the lung: role of video-assisted thoracic surgery. *Chest Surg Clin North Am.* 1993;3:307-17.
25. Layfield LJ, Coogan A, Johnston WW, et al. Transthoracic fine needle aspiration biopsy: sensitivity in relation to guidance technique and lesion size and location. *Acta Cytol.* 1996;40:687-90.
26. Kashiwabara K, Nakamura H, Fukai Y, et al. Availability of diagnosis by percutaneous needle aspiration cytology of the lung in cases who showed a peripheral solitary tumorous shadow on chest Xp and diagnostic rate of transbroncial approach. *Jpn J Thorac Dis.* 1993;31: 1426-31.

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Surg Today (2006) 36:525–527
DOI 10.1007/s00595-006-3182-0

SURGERY TODAY

Case Reports

Isolated Unilateral Absence of a Pulmonary Artery Treated by Pneumonectomy in an Adult: Report of a Case

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Abstract
Congenital unilateral absence of a pulmonary artery (UAPA) is a rare anomaly usually diagnosed in childhood. We report a case of isolated UAPA in an adult without any other cardiovascular anomaly. The patient was admitted for repeated progressive hemoptysis, which we treated with embolization of the bronchial artery three times, despite which the hemoptysis kept recurring. Finally, the patient was treated successfully by right pneumonectomy. Thus, we think that surgical intervention is indicated for recurrent hemoptysis in patients with isolated UAPA.

Key words Unilateral absence of pulmonary artery · Pneumonectomy · Bronchial artery

Introduction

Congenital unilateral absence of a pulmonary artery (UAPA) is a rare anomaly commonly associated with cardiovascular abnormalities such as tetralogy of Fallot, septal defects, and patent ductus arteriosus.[1] Occasionally, UAPA occurs as an isolated anomaly without any other cardiovascular malformation.[1–3] We report the successful treatment of an isolated UAPA causing recurrent hemoptysis in an adult, by right pneumonectomy after embolization of the bronchial arteries.

Case Report

A 48-year-old man was admitted to our hospital for investigation and treatment of repeated progressive hemoptysis, which had started 3 years earlier. He underwent embolization of the bronchial artery three times during the course of almost 1 year, despite which the hemoptysis kept recurring. Finally, he was readmitted for surgical intervention. A chest roentgenogram showed absence of a right hilar shadow and volume loss in the right lung (Fig. 1A). Chest computed tomography showed a defect in the right main pulmonary artery and collateral vessels in the mediastinum (Fig. 1B). Pulmonary angiography showed complete absence of the right main pulmonary artery (Fig. 2). The left pulmonary vasculature was normal. Bronchial angiography showed lengthening and tortuosity of the right bronchial artery (Fig. 3A). A collateral artery from the superior phrenic artery was also identified (Fig. 3B). An echocardiogram demonstrated normal function and no other cardiovascular anomaly. Cardiac catheterization revealed that the pressure in the pulmonary artery, the pulmonary capillary wedge pressure, and the right atrium pressure were 36/16 (mean 23) mmHg, 11/7 (mean 8) mmHg, and 8/2 (mean 3) mmHg, respectively. The results of pulmonary function studies were: forced expiratory volume in 1 s ($FEV_{1.0}$) 2.14 l, and vital capacity (VC) 3.15 l. The arterial blood gas values while the patient breathed room air were as follows: PO_2, 66.7 mmHg; PCO_2, 39.1 mmHg; and pH, 7.41. The patient stated that he was a nonsmoker. To prevent bleeding, we performed embolization of right bronchial arteries 10 days before the patient underwent right pneumonectomy. During the operation, we observed highly developed bronchial arteries surrounding the right main bronchus, but no developed vessels in the pulmonary ligament. Pleural adhesion to the diaphragm with collateral arteries was also seen. There were three normally lobulated lobes. The pleural surface was covered with a developed network of dilated vessels. The right bronchus and pulmonary veins were in the normal position; however, there was no pulmonary artery (Fig. 4). The total operating time was 370 min, and the intraoperative blood loss was

Reprint requests to: T. Ohtsuka
Received: February 21, 2005 / Accepted: November 15, 2005

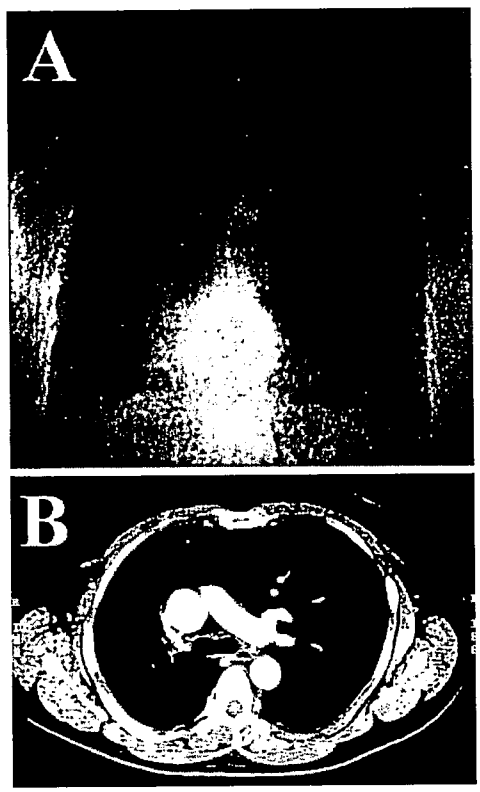

Fig. 1. **A** Chest X-ray film on admission showed absence of the right hilar shadow and volume loss of the right lung. **B** Chest computed tomography showed a defect in the right main pulmonary artery and collateral vessels in the mediastinum

Fig. 3. **A** Bronchial angiography showed a developed bronchial artery around the right main bronchus (*arrow*). **B** A collateral artery from the superior phrenic artery was identified (*arrowhead*)

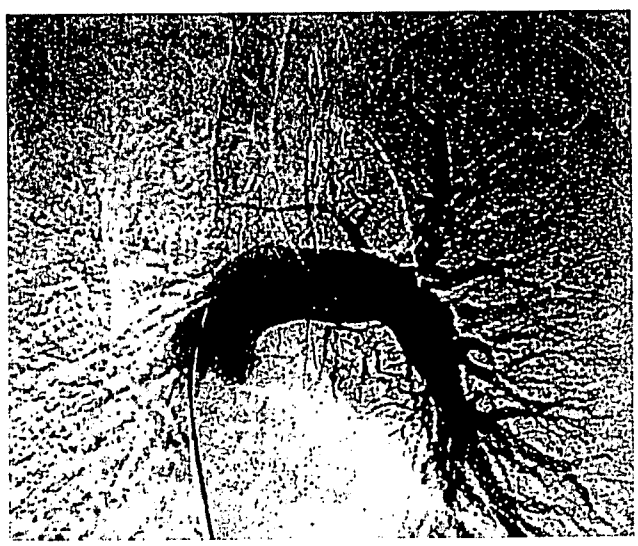

Fig. 2. Preoperative pulmonary angiography showed absence of the right pulmonary artery with only the left pulmonary artery originating from the main pulmonary artery

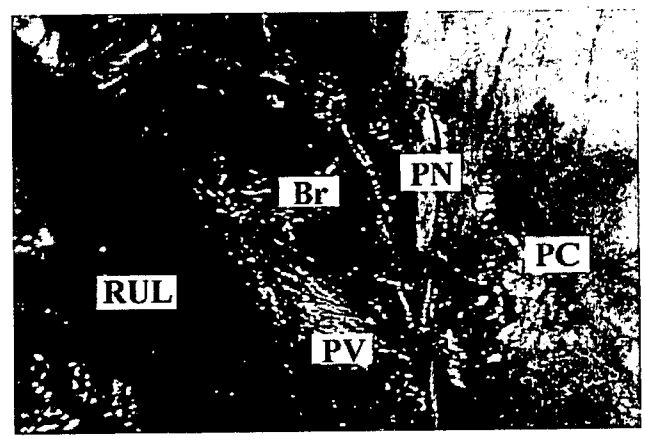

Fig. 4. Intraoperative photo of the right pulmonary hilum. The right main bronchus (*Br*) and superior pulmonary vein (*PV*) were in the proper position. There was no pulmonary artery. *RUL*, right upper lobe; *PC*, pericardium; *PN*, phrenic nerve

600 ml. Pathological examination of the resected specimen revealed hypoplasia of the bronchial cartilage and smooth muscle tissue in the peripheral bronchus. Inflammatory and emphysematous changes were observed in the resected lung. Thickening of the vascular wall, with congestion and dilatation of the lumen of the pulmonary vein, were also observed. The patient had an uneventful postoperative course and was discharged on the 9th postoperative day. Cardiac catheterization 1 month after the operation showed that the pressure in the pulmonary artery, the pulmonary capillary wedge pressure, and the right atrium pressure were 36/13 (mean 21) mmHg, 11/8 (mean 9) mmHg, and 16/9 (mean 13) mmHg, respectively, similar to the pressures measured preoperatively. The patient is now in good health, 6 months after surgery.

Discussion

Congenital UAPA is a rare anomaly, which commonly coexists with cardiovascular abnormalities. Although less frequent, UAPA may also occur as an isolated finding.[1–3] Unilateral absence of pulmonary artery is classically diagnosed and treated surgically during the first year of life.[1,4] Conversely, patients with isolated UAPA generally have a mild clinical course, although a few suffer pulmonary hypertension.[1]

To date, only sporadic case reports and reviews of isolated UAPA have been published, and very few patients have been treated by pulmonary resection.[5–7] Bekoe et al. reported the case of an adult with UAPA, who was treated with pneumonectomy followed by a good outcome. They recommended pneumonectomy for patients with recurrent hemoptysis caused by UAPA, because the lung affected by UAPA does not contribute to ventilation.[7] In our patient, the collateral arteries were highly developed and hemoptysis kept recurring despite embolization of the bronchial arteries. During the operation, we had to prevent bleeding from the collateral vessels originating from the bronchial, intercostal, subclavian, or subdiaphragmatic arteries.

The most common symptoms of UAPA are recurrent pulmonary infections, dyspnea and, less frequently, hemoptysis and signs of pulmonary hypertension.[1–8] The hemoptysis is caused by the extensive collateral circulation; however, the pulmonary hemorrhage and pulmonary hypertension in patients with isolated UAPA is usually not severe, because they have no other cardiovascular anomalies. However, if a patient with isolated UAPA complains of recurrent hemoptysis or recurrent pulmonary infections, surgical intervention should be considered.

References

1. Ten Harkel AD, Blom NA, Ottenkamp J. Isolated unilateral absence of a pulmonary artery: a case report and review of the literature. Chest 2002;122:1471–7.
2. Brassard JM, Johnson JE. Unilateral absence of a pulmonary artery. Data from cardiopulmonary exercise testing. Chest 1993; 103:293–5.
3. Hirata K, Kyushima M, Asato H. Two adult cases of unilateral absence of the right pulmonary artery with markedly different clinical presentations. Jpn Circ J 1995;59:574–8.
4. Shigenaga K, Mizuno T, Shibata K, Yamakawa Y, Nishida T, Masaoka A. Congenital absence of the right pulmonary artery — a case report (in Japanese). Kyobu Geka 1988;41:820–4.
5. Mehta AC, Livingston DR, Kawalek W, Golish JA, O'Donnell JK. Pulmonary artery agenesis presenting as massive hemoptysis — a case report. Angiology 1987;38:67–71.
6. Maeda S, Suzuki S, Moriya T, Suzuki T, Chida M, Suda H, et al. Isolated unilateral absence of a pulmonary artery: influence of systemic circulation on alveolar capillary vessels. Pathol Int 2001;51:649–53.
7. Bekoe S, Pellegrini RV, DiMarco RF, Jr., Grant KJ, Woelfel GF. Pneumonectomy for unremitting hemoptysis in unilateral absence of pulmonary artery. Ann Thorac Surg 1993;55:1553–4.
8. Muro K, Aoki M, Wada H, Hitomi S. Congenital absence of the right main pulmonary artery — a case report (in Japanese). Nihon Kyobu Shikkan Gakkai Zasshi 1992;30:1728–31.

Case Report

Kissing Pleural Metastases from Metastatic Osteosarcoma of the Lung

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Two patients with osteosarcoma lung metastases of which migrated to the parietal pleura due to contact are reported. The first patient was a 16-year-old male who had a pleural metastasis in the diaphragm within an area in contact with a single lung metastasis. Both of the tumors were resected, followed by systemic chemotherapy. Nine months after the resection of the first metastases, two other lung metastases were found which were resected after chemotherapy. The patient is alive without recurrence 84 months after the first resection of the metastases. The second patient was an 11-year-old female with a pleural metastasis of osteosarcoma which was within an area in contact with a single lung metastasis, which had been resected 4 months before. We concluded (1) that a lung metastasis of osteosarcoma occasionally metastasizes to the pleura due to contact; and (2) that because this kissing metastases of osteosarcoma could be cured by a complete resection, the intrathoracic cavity should be thoroughly observed. (Ann Thorac Cardiovasc Surg 2006; 12: 129–32)

Key words: metastasectomy, lung, pleura

Introduction

The lung is the most popular metastatic site of osteosarcoma. Jeffree et al. reported that osteosarcoma metastasized to the lung in over 90% of patients who died of osteosarcoma.[1] According to the Japan Autopsy Annual Database, 643 patients died of osteosarcoma between 1981 and 2002 in Japan.[2] Of the 643 patients, 78 (12.1%) had pleural metastases. We present two patients with metastases of parietal pleura which probably originated from a lung metastasis contacting the pleura, namely 'kissing metastases'.

Case Report

Case 1

A 16-year-old male with femoral osteosarcoma received preoperative chemotherapy with cisplatin (CDDP) and doxorubicin (DXR), followed by an extended resection of the right femur and reconstruction in July 1997. Adjuvant chemotherapy with CDDP and cyclophosphamide (CPA) was continued for a total of 10 courses until June 1998. In September 1998, a single lung metastasis in the right lower lobe was found with computed tomography (CT) (Fig. 1). A needle biopsy was not performed. Thoracoscopy showed that a lung metastasis was located in the right lower lobe, and that it was exposed on the surface of the visceral pleura. Additionally, a solitary pleural tumor, 3 mm in size, was found on the diaphragm (Fig. 2). Although these two tumors were separated from each other, the pleural tumor was in an area that was in contact with the pulmonary tumor during ordinary lung expansion in ventilation. Both the lung and pleural tumors were resected. Pathological examination showed that both of the tumors were metastatic osteosarcoma, and that they were covered with a fibrous capsule but not with mesothelial epithelium. Adjuvant chemotherapy with ifosfamide (IFO) and DXR was administered after the first metastasectomy. In July of 1999, two additional lung metastases were detected by CT in the right upper and middle lobes. After chemotherapy with CPA, both me-

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Received September 5, 2005; accepted for publication October 21, 2005.
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