

Osteoradionecrosis of External Auditory Canal in Nasopharyngeal Carcinoma

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Background: Osteoradionecrosis (ORN) is one of the most serious complications of radiotherapy of nasopharyngeal carcinoma (NPC). ORN rarely occurs in the external auditory canal.

Methods: This is a retrospective review of 11 NPC patients who had ORN in the external auditory canal. Exposed necrotic bone over the external auditory canal was the diagnostic hallmark. Three patients wore hearing aids and 2 had had previous otological surgery. ORN was detected between 2 and 15 years after radiation therapy (mean 8 years). The radiation dose ranged from 6480 to 8460 rads in 8 patients.

Results: The symptoms of external auditory canal ORN were crust (100%), otorrhea (91%), otalgia (91%), hearing impairment (73%), foul odor (45%) and retroauricular discharging fistula (9%), all of which mimicked those of chronic otitis media. After treatment, the diagnoses of 2 patients were rectified to external auditory canal malignancy and external auditory canal cholesteatoma, and the other 9 (82%) patients' diagnoses remained as ORN. After combined treatment with local cleansing, hyperbaric oxygen therapy, sequestrectomy and ear drops, resolution of symptoms was achieved in all the patients. Three patients who underwent sequestrectomy were free of disease.

Conclusions: External auditory canal ORN is rare in NPC patients. The clinical presentation mimics that of chronic otitis media and differentiation is difficult. The disease is prone to occur in patients who wear hearing aids or have had previous surgery. A high index of suspicion is mandatory for early diagnosis. The disease may lead to disastrous complications and should never be neglected by clinicians.

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Key words: osteoradionecrosis, external auditory canal, radiation therapy, hearing aid, nasopharyngeal carcinoma

Radiotherapy is the mainstay treatment of nasopharyngeal carcinoma (NPC). With the classic 3 H principle of hypoxia, hypovascularity and

hypocellularity, radiation therapy may actually impair normal collagen synthesis and cell production, leading to tissue breakdown and eventual

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radionecrosis.⁽¹⁾ When the necrotic process involves bone, osteoradionecrosis (ORN) occurs. ORN, though not commonly seen nowadays, is one of the most serious complications of radiotherapy in NPC.⁽²⁾ It may occur in the skull base,⁽²⁾ mandible,⁽³⁾ maxilla and external auditory canal in NPC patients. Signs of ORN include unhealed ulcer, exposed bone and accompanying granuloma, all of which closely mimic signs of malignancy and pose a significant challenge for differentiation.⁽³⁾

External auditory canal ORN is seldom reported in the literature.⁽⁴⁻⁷⁾ It is probably neglected by many clinicians because the initial symptomatology of crust, otorrhea, and otalgia may lead to the diagnosis of chronic otitis media instead. However, potentially disastrous complications, such as malignant otitis externa, meningitis or brain abscess, may occur and patients may die. The aim of this article is to explore the clinical symptomatology of external auditory canal ORN in NPC patients and bring this complicated disease to the clinician's attention.

METHODS

We made a retrospective review of 11 NPC cases initially diagnosed with external auditory canal

ORN at the clinic of the senior author (SP Hao) (Table 1). There were 8 male and 3 female patients. They were aged between 46 and 70 years (mean 54.2 years). The diagnostic criteria of external auditory canal ORN includes ulcer of the skin or exposed necrotic bone. The latency period between completion of radiotherapy and diagnosis of external auditory canal ORN was between 2 to 15 years (mean 8 years). After diagnosis, the patients were followed up in the clinic for 3 to 50 months. One patient had previous salvage nasopharyngectomy for local recurrence of NPC and right patch myringoplasty for perforated drum. One patient had skull base ORN and underwent endoscopic sequestrectomy. One patient was initially treated by mastoidectomy elsewhere and was referred to us with a persistent discharging fistula on the post-auricular wound. Three patients wore a hearing aid on the affected ear. The initial radiotherapy dose data for NPC was only available in 8 patients and ranged from 6480 to 8460 rads. These NPC patients underwent radiotherapy in a similar manner in our hospital.

RESULTS

The initial presenting symptoms of the 11

Table 1. Case Summary

Case	Age (Y/O)	Gender	Primary cancer	R/T dose (rads)	Latency period(yr)	Hearing aid	Intervention	Complication
1	58	F	NPC	6480	4	Nil	Local treatment + HBO	Nil
2	55	M	NPC	6400	2	Nil	Local treatment + HBO	Nil
3	46	F	NPC	7280	8	Right	Local treatment + HBO	Nil
4	70	M	NPC	‡	12	Left	Endoaural sequestrectomy + HBO	Nil
5	48	M	NPC	6660	3	Nil	Endoaural sequestrectomy + HBO	Nil
6*	64	M	NPC	6880	15	Nil	Temporal bone resection	Nil
7	53	M	NPC	8160	10	Nil	Local treatment + HBO	Nil
8	51	M	NPC	8220	11	Right	Local treatment + HBO	Malignant otitis externa
9	46	M	NPC	8460	5	Nil	Local treatment + HBO	Nil
10	53	M	NPC	‡	9	Nil	Open sequestrectomy & free flap	Facial palsy
11†	52	F	NPC	‡	9	Nil	Debridement	Nil
Mean	54			7317.5	8			

Abbreviations: 6*: final diagnosis of ear malignancy; 11†: final diagnosis of external auditory canal cholesteatoma; ‡: unknown; HBO: hyperbaric oxygen therapy.

patients with initial diagnosis of external auditory canal ORN included otorrhea (91%), otalgia (91%), foul odor (45%), hearing impairment (73%), crust formation (100%) and retroauricular discharging fistula (9%). One (9%) patient was actually diagnosed during regular check-up without objective symptoms and the disease was completely ignored by the patient himself. The treatment consisted of conservative treatment of aural hygiene improvement by frequent (weekly) aural local treatment, otic drops and antibiotics for otorrhea, avoidance of ear wax picking, hyperbaric oxygen therapy (HBO) and sequestrectomy (Table 1) (Fig. 1 and 2). Three patients underwent endoaural sequestrectomy and one patient had open radical sequestrectomy. Six patients had HBO. After treatment, the diagnoses of 2 patients were rectified to external auditory canal malignancy and external auditory canal cholesteatoma, and the other 9 (82%) patients' diagnoses remained as ORN (Table 1). Patient 6 had skull base ORN previously, which was successfully controlled with endoscopic sequestrectomy 2 years ago. Later, he had otorrhea, otalgia and granulation tissue over the external auditory canal, and had a tentative diagnosis of external auditory canal ORN. He subsequently underwent regular local treatment of the ear canal in the clinic but in vain, and had progressive tumor formation though the initial debride-

ment revealed only acute and chronic inflammation on pathology. The patient then underwent a limited endoaural sequestrectomy that revealed carcinoma on pathology and he subsequently underwent lateral temporal bone resection. He was excluded from the analysis. Patient 11 had local recurrence of NPC after initial radiation and underwent salvage nasopharyngectomy. She also had a perforated ear drum on the right side and underwent a patch myringoplasty. Twenty-eight months after myringoplasty, she was diagnosed with external auditory canal ORN but this was rectified to external auditory canal cholesteatoma, after endoaural debridement, as spontaneous mastoidectomy by cholesteatoma matrix was evident and there was no bare bone after removing the cholesteatoma matrix. Thus, there were 9 patients with true external auditory canal ORN. One patient initially presented to us with retro-auricular discharging fistula after previous mastoidectomy and later underwent an open radical sequestrectomy of the temporal bone, the resulting defect being reconstructed with a free anterior lateral thigh flap. However, he had facial paralysis on the affected side after surgery. Patient 8 had complications of 2 episodes of malignant otitis externa. Treatment results of the 9 patients with true external auditory canal ORN were 3 disease free without visible bare bone, and 6 with visible bare bone and crust accumu-

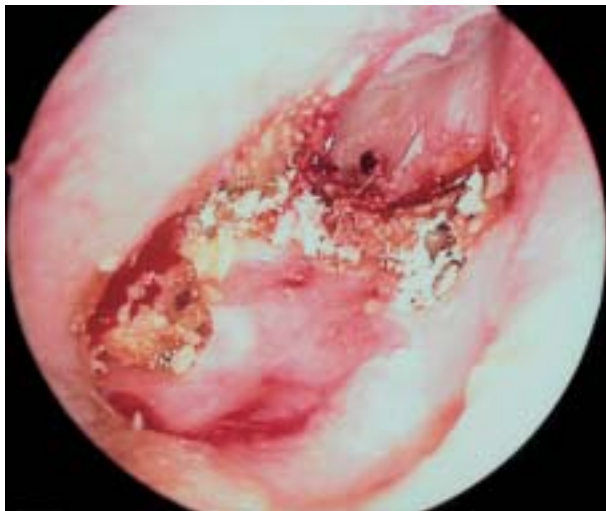


Fig. 1 Left ear, case 4, microscopic view (250 \times). Exposed necrotic bone over inferior portion of the external auditory canal.



Fig. 2 Left ear, case 4, microscopic view (250 \times). Immediately after endoaural sequestrectomy, the necrotic bone was thoroughly removed until bleeding Haversian canals appeared.

lation but without obvious otorrhea.

DISCUSSION

The external auditory canal is derived from the pharyngeal groove between the first and second branchial arches embryologically. Thus, the external auditory canal is derived from ectoderm and is lined by squamous epithelium. The skin outlining the external auditory canal is thin and delicate. The aural cartilage and tympanic bone meet at the narrowest part of the canal in the inner one third of the external auditory canal. These facts predispose this particular site to frequent iatrogenic trauma that may lead to ORN in irradiated patients. It is interesting to note that in the present series of 9 patients with true external auditory canal ORN, 3 wore hearing aids and 2 had had previous otological surgery. It is possible for a poorly fitting hearing aid mold to exert minor trauma over the external auditory canal and predispose to ORN. Iatrogenic trauma generated by surgery or picking the ears may also play a role in the pathogenesis of ORN. Radiation exerts the 3 Hs of hypoxia, hypovascularity and hypocellularity, impairs normal collagen synthesis and cell production, and leads to tissue breakdown and eventual ORN. ORN of the external auditory canal typically occurs in the lower portion of the canal, which can be explained by the downward pressure exerted by wearing a hearing aid or picking at ear wax. The pressure may also involve the perichondrium that carries the main blood supply to the cartilage, resulting in pressure necrosis. Then, microorganisms may penetrate through the pressure ulcer, leading to serious perichondritis and chondritis. The infected ORN may lead to malignant otitis externa as shown in case 9 in this series. The principle of treating ORN is basically surgical, as the necrotic substance cannot be revitalized and has to be surgically removed.⁽³⁾ The lack of vascularized tissue around the ORN site further complicates treatment. Prevention of infection with antibiotics and maintaining local hygiene by frequent cleansing are also mandatory in controlling the disease.

HBO, which reverses the 3 Hs and reinforces the tissue repairing mechanism, is a useful adjunct. However, in the preliminary results from our previous publication,⁽³⁾ we found that surgical sequestrectomy seemed inevitable and has been proved to yield

good results. In the current series, the 3 patients who were disease free were all in the surgery group, though their disease condition was worse compared to those who did not undergo surgery. In comparison, HBO only plays an adjunctive role as only stationary results are achieved with local treatment and HBO alone.

The symptomatology of external auditory canal ORN of purulent otorrhea, otalgia, exposed necrotic bone and granuloma, may mimic that of chronic otitis media, external auditory canal cholesteatoma or ear malignancy. The differentiation between ORN and recurrent cancer might be difficult: our previous article on head and neck ORN showed that an average of 2.4 sequestrectomy procedures were carried out before reaching the final correct diagnosis of recurrent cancer and 21% of cases initially diagnosed as ORN were rectified to cancer.⁽³⁾ The differentiation of ORN from external auditory canal cholesteatoma is even more complicated. The diagnosis of patient 11 was rectified to external auditory canal cholesteatoma because there was almost no bone necrosis but the bone remodeling from spontaneous mastoidectomy by cholesteatoma was quite evident.

NPC patients with external auditory canal ORN typically present with foul odor because of the sequestrum and thus may be socially repelled. The symptoms of otorrhea or otalgia, which mimic those of chronic otitis media, may be neglected by patients. The disease is also ignored by clinicians, probably because of unawareness of the disease entity. It is imperative that we, as health care providers, should be aware of the disease and keep a high index of suspicion regarding NPC patients who wear hearing aids or have had previous otological surgery.

Conclusions

ORN involving the external auditory canal is quite unusual in NPC patients. The clinical presentations of otorrhea, otalgia and crust formation are very similar to those of chronic otitis media, external auditory canal cholesteatoma or malignancy. The disease is prone to occur in patients wearing hearing aids or in those who have had previous surgery. A high suspicion index is mandatory for early diagnosis. The disease may lead to disastrous complications and should never be neglected by clinicians.

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鼻咽癌病人外耳道放射性骨壞死

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背景：放射性骨壞死是鼻咽癌病人因放射線照射引起的嚴重併發症之一，放射性骨壞死不常發生於外耳道。

方法：吾人回溯性整理 11 位罹患鼻咽癌，且同時被初步診斷為患有外耳道放射性骨壞死的病人。診斷的主要依據為外耳道曝露的腐蝕骨頭，這些病人分別於放射線照射治療後 2 至 15 年 (平均 8 年)，被診斷為外耳道放射性骨壞死。8 位可知放射線照射劑量的病人，其照射劑量從 6480 至 8460 雷得不等，病人中有 3 位配戴助聽器，兩位接受過同側耳科手術。

結果：外耳道放射性骨壞死的症狀分別為：耳癢 (100%)，耳痛 (91%)，聽障 (73%)，惡臭 (45%)，耳後膿漏瘻管 (9%)。這些症狀和慢性中耳炎症狀類似。治療後有兩位病人分別被更正診斷為耳癢及外耳道珍珠瘤，餘 9 位 (82%) 仍維持外耳道放射性骨壞死的診斷。治療方式是以下列方式單獨或合併治療，包括耳道局部治療，高壓氧治療，耳滴劑，抗生素及腐骨去除術。結果所有的病人症狀可獲緩解，而三位接受腐骨去除術的病人更可痊癒。

結論：鼻咽癌病人，其外耳道放射性骨壞死仍屬罕見，臨床症狀和中耳炎類似且兩者區分不易。此病好發於配戴助聽器或先前接受過耳科手術者，早期診斷則有賴醫師的高度警覺性，臨床醫師不應輕忽。
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關鍵詞：放射性骨壞死，鼻咽癌，外耳道

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