

Bringing Threat to the Fore: Participating in Lifelong Surveillance for Genetic Risk of Cancer

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Purpose/Objectives: To explain how patients with multiple endocrine neoplasia type 2a (MEN2a) and family members conceptualize participation in lifelong surveillance in genetic cancer care.

Design: Qualitative naturalistic inquiry.

Setting: Northeastern United States. Data were collected during interviews in informants' homes.

Sample: 12 adult patients and 9 family members were recruited purposively through endocrinology clinics at two East Coast medical centers.

Methods: A grounded theory design was used. Subjects completed a demographic questionnaire and were interviewed on two occasions. In-depth interviews were audiotaped. The core concept and process variables emerged through three levels of narrative content analysis, theoretical sampling, and constant comparison. The generated theory was presented to the participants for validation.

Main Research Variables: The basic psychological issue associated with participation in lifelong surveillance and the psychosocial processes used by individuals to deal with the issues.

Findings: MEN2a patients and family members experience surveillance as a (Re)Minding of a threat to health. Once threat is brought to the fore, participants interpret the meaning and negotiate control of the impact of the diagnosis, related events, and consequences. Meaningful information from surveillance activities is incorporated into participants' self-image and daily lives through a process of (Re)Integration. The genetic nature of MEN2a did not emerge as a significant subcategory in relation to the core variable.

Conclusions: Finding meaning in the outcomes of surveillance events is a psychosocial process that is central to participation in lifelong surveillance. Genetic predisposition to cancer was a peripheral concern to subjects as they dealt with surveillance issues.

Implications for Nursing: This investigation provides a model for the development of a grounded theory for understanding how people with other genetic cancer syndromes participate in lifelong surveillance.

Surveillance is a risk management strategy used in health care for the early detection of disease and for complications associated with medical treatment. Once a diagnosis or risk of disease is known, healthcare professionals advise patients to participate in lifelong surveillance activities to monitor for the expression of disease and the physical and psychological consequences of treatment. In the context of genetic cancer care, monitoring an individual who is predisposed to cancer is referred to as "enhanced surveillance." These activities occur more frequently than in the general population and continue over a lifetime. In social context, surveillance occurs within the relationship constructed be-

Key Points . . .

- Two types of lifelong surveillance activities exist: planned surveillance events and incidental surveillance events.
- Patients and family members conceptualize surveillance as a personal and family phenomena rather than the responsibility of healthcare professionals.
- Self-monitoring and observation of affected loved ones by family members dominate the surveillance phenomena.
- Incidental surveillance events may replace planned events as indicators of health and wellness.

tween the observer and the one who is observed. The social relationship may be impersonal or intimate, yet within this relationship, the observer and the one observed watch and wait for a threatening event (Giarelli, 2002).

How patients think about participating in lifelong surveillance will affect their ability to accept, adhere to, and adopt behaviors that promote health. This article describes how people in families with the genetic cancer syndrome multiple endocrine neoplasia type 2a (MEN2a) conceptualized their participation in lifelong surveillance.

Background

MEN2a is a rare genetic cancer syndrome that leads to abnormal cell growth in endocrine glands. Medical geneticists consider MEN2a a prototype for their clinical practice because patients have access to a reliable and accurate genetic

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test and effective treatments such as preventive surgery (Gimm & Dralle, 1999). Genetic testing is offered to people in families with a high incidence of thyroid cancer or adrenal tumors (Howe et al., 1992; Learoyd et al., 1997; Ledger, Khosla, Lindor, Thibodeau, & Gharib, 1995) or to members of families with a known gene mutation. Once diagnosed, people with MEN2a are advised to participate in lifelong surveillance activities.

MEN2a is caused by a mutation to the rearranged transfection (RET) gene on chromosome 10 (Mulligan et al., 1993). All people with the mutation eventually develop one or more different tumors and have a 50% chance of passing the genetic mutation to a child. The syndrome leads to the development of medullary thyroid carcinoma, pheochromocytoma (adrenal tumor) (Modigliani et al., 1995; Schimke & Hartmann, 1965), and parathyroid hyperplasia and adenomas (Keiser, Beaven, Doppman, Wells, & Buja, 1997; Pausova et al., 1996).

Approximately 20,700 cases of thyroid cancer were diagnosed in 2002 (Jemal, Thomas, Murray, & Thun, 2002). Medullary thyroid carcinoma represents about 10% of these cancers (i.e., nearly 2,000 cases) (Hundahl, Fleming, Fremgen, & Menck, 1998), and 25% are inherited (i.e., almost 200 cases) (Randolf & Maniar, 2000). In some patients who are genetically predisposed to MEN2a, thyroid cells that have precancerous changes may be present at birth.

Medullary thyroid carcinoma is the main cause of illness and death and develops in nearly 100% of patients with the RET mutation (Chi & Moley, 1998; Frank-Raue, Hoppner, Buhr, Herfarth, & Raue, 1997; Lairmore, Frisella, & Wells, 1996). Medullary thyroid carcinoma is slow growing, painless, and metastasized to local and distant sites before detectable (Gimm & Dralle, 1999). Patients with the RET mutation that causes MEN2a may elect to have prophylactic or curative thyroidectomy as early as age five (Lairmore et al.).

Another MEN2a-induced tumor is pheochromocytoma. This adrenal tumor may be multifocal and occurs in up to 57% of patients with the RET mutation (Correia et al., 2000). Hyperparathyroidism, a benign preneoplastic condition, is found in 15%–30% of people with MEN2a (Modigliani et al., 1995). Each of the possible neoplasias has respective surveillance guidelines.

Surveillance Activities

Patients with MEN2a are advised to participate in health surveillance activities (Gagel, 1997; Johnston et al., 2000; Raue, 1997) as soon as they become aware of their risk for the disease through genetic testing or family history. Lifelong surveillance activities are performed to detect the first signs of disease and to watch for health problems that result from thyroidectomy, parathyroidectomy, adrenalectomy (Szubin, Kacker, Kakani, Komisar, & Blaugrund, 1996), and hormone replacement therapy (Geiger & Thompson, 1996; Skinner, DeBenedetti, Moley, Norton, & Wells, 1996). Postoperative medical management requires considerable and careful follow-up (de Graaf, Dullaart, & Zwierstra, 1999) for evidence of hormone deficiency. Patients must undergo yearly (or more frequent) tests of (a) serum calcitonin for C-cell hyperplasia or carcinoma, (b) parathyroid hormone levels, (c) thyroid-stimulating hormone levels and thyroid hormone replacement effectiveness, (d) serum calcium levels, and (e) 24-hour urine collection for catecholamines associated with pheochromocytoma. Surveillance for MEN2a is complex, involves the moni-

toring of multiple organ systems over a lifetime, and is an important means of cancer control (Easton et al., 1989; Telander, Zimmerman, van Heerden, & Sizemore, 1986; Wells et al., 1982, 1994). Immediate and extended family members, especially those who live in a patient's home, also participate as observers in surveillance of their loved one. Although it is a wise healthcare option, participating in lifelong surveillance has psychosocial consequences.

Psychosocial Experiences of Risk Management

Early research exploring psychosocial issues associated with lifelong risk management revealed some of the problems experienced by patients and relatives in MEN2a families. Cleiren, Oskam, and Lips (1989) surveyed 32 patients and 27 relatives and reported that, although patients and family members both believed that yearly screening was a positive health-promoting behavior, they experienced frustration when encountering delays in receiving test results. The most problematic aspect of lifelong risk management of patients with MEN2a is postoperative medical management with hormone replacement therapy. Daily self-medication is cumbersome, and the physical side effects associated with missed medication and dose adjustment caused distress and reduced patients' quality of life. This was relevant especially for patients after adrenalectomy for pheochromocytoma (Telenius-Berg, Ponder, Berg, Ponder, & Werner, 1989). Even patients who choose preventive surgery may experience emotional distress and reduced quality of life. In one case study, an adolescent girl underwent preventive thyroidectomy; months after the surgery, her resulting distress manifested itself as self-destructive behavior (Giarelli, 1999). The few studies of the risk management strategy of surveillance offer limited and incomplete accounts of lifelong experiences of MEN2a families. The current study was undertaken to explore the deeper meaning of the social phenomena of risk management to generate a conceptual model of participation in lifelong surveillance for MEN2a.

Methods

Data Generation

The grounded theory method is a systematic way to generate theoretical constructs that represent a social phenomena of participation. This method is well suited to uncovering psychosocial structural components of a phenomena from the perspective of the participants. It captures the experiences of patients and family members who want to share equally in health-promoting activities (Cleiren et al., 1989).

The theoretical assumptions underlying grounded theory are derived from the premises of the social-psychological theoretical perspective of symbolic interactionism. First proposed by Mead (1962) and refined by Blumer (1969), symbolic interactionism has been used as a framework in grounded theory research in cancer nursing since the 1960s (Benoliel, 1967; Hutchinson, 2000). Symbolic interactionism describes a process by which people attribute meaning to events based on personal experience and interaction with others in their world by proposing that concepts of self, the world, social action, and the meaning of events are constructed cognitively and change through processes of social interaction. People develop shared meanings of objects and individuals in their lives, and the meanings are attached to situations, others, self, and things through a process of interpretation. The symbolic

interactionist perspective and the methodology of Strauss and Corbin (1998) presume that human experience may be understood as both a personal and a social phenomena.

Participants

The study was approved by the committee for scientific review at a university cancer center and the institutional review boards at two medical centers in the northeastern United States. Participants were recruited from an adult endocrinology and medical genetics clinic in a large medical center, from a pediatric endocrinology clinic in a regional children's hospital in the northeastern United States, and by patient referral. Patients diagnosed with MEN2a by molecular genetic testing or by a combination of personal and significant family history of thyroid or adrenal tumors were eligible to participate. Unaffected family members also were eligible to be informants because the social nature of the phenomena depends on the participation of patients and family members in monitoring activities. Additional inclusion criteria were English speaking, adolescent (14 years of age and older) or adult age, and ongoing participation in follow-up care. To ensure credibility, the researcher sought participants who had experience with a range of surveillance activities and the ability to communicate and expound thoughts, feelings, beliefs, and expectations. In addition, the investigator attempted to invite informants with a wide variety of ages, races, socioeconomic status, and relationships to patients with MEN2a. Recruitment ended when conceptual categories reached saturation (Glaser, 1978).

Procedures

To protect confidentiality, the attending physicians contacted patients diagnosed with MEN2a to describe the study. With verbal consent from patients, physicians notified the investigator who contacted each patient by telephone, described the study, and sent additional information by mail. All invited patients consented to enroll. Two invited family members refused to join the study. Each of these was a spouse (one male, one female) who wanted to keep personal information and feelings private.

A single question was first asked of all participants: "Tell me the story of your MEN2a [MEN2a in your family], and start at whatever point in time you like." Responses were encouraged with probes for additional and elaborated information (Morse & Field, 1995), and an interview guide was revised continually for theoretical sampling.

Informants completed a demographic survey and were interviewed on two occasions, spaced two to three months apart. Sequential interviews facilitated the capturing of the ongoing nature of participation in surveillance activities, helped to establish the trust necessary for sharing feelings (Fontana & Fry, 1994), and allowed the informants to explore their thoughts and experiences. The in-depth interviews took place in the informants' homes. One informant was interviewed only once because she was unable to elaborate beyond very brief responses and "yes" or "no." The informant's cursory responses, however, made a contribution to understanding refusal and resistance as dimensions of the psychosocial process of negotiating control.

Each interview lasted 60–90 minutes, was audiotaped and transcribed by a research assistant, and imported into NUD*IST (non-numerical unstructured data indexing, searching, and

theorizing) N4 Classic version software program for coding, categorizing, and theory development (Richards, 2000).

Theory Construction

Three levels of coding were used to analyze the content of transcribed interviews. First, the narratives were read as a whole, then microanalyzed line by line. At this first level of analysis, open coding generated many units of meaning in the language of the informants and resulted in a large set of basic concepts. During level II coding, these concepts were condensed and grouped into categories and subcategories. Level III coding linked categories to each other and to a main concept to describe an organizational psychosocial scheme for participation in lifelong surveillance. Constant comparison was used to concurrently collect and analyze empirical materials. Analytical strategies included theoretical sampling, memo writing, selective coding, and continual questioning of the emerging concepts and categories.

During the multilevel analysis, the central category of (Re)Minding emerged. This category is related to all other conceptual categories as the basic social-psychological problem experienced by people who participate in lifelong surveillance. Once this core variable was identified, theoretical sampling was used to develop and saturate related conceptual process categories (Glaser, 1978).

Credibility was enhanced by discussing the findings with colleagues while undergoing peer review and with study participants during the process of member checking (i.e., verifying with informants the accuracy of their statements and how closely their statements match their experiences). To ensure that the findings were derived consistently (dependability), the investigator attempted to control bias through bracketing and theoretical memo writing. To ensure that the empirical evidence corroborated the theoretical conclusions (confirmability), the author used data management software to document material collection, analysis procedures, and theory development and to archive empirical materials (Guba, 1981).

Findings

Participants

A purposive sample of 21 informants was interviewed. Of the 12 people with MEN2a, 7 were female and 5 were male, and their ages ranged from 19–74 years. Of the nine family members, five were female and four were male, and their ages ranged from 20–69 years. Family members were spouses, siblings, and sons and daughters of patients. Nearly all informants were Caucasian (except for one Hispanic family member), were from lower- to upper-middle class in socioeconomic status, and were educated from one year of high school to three years of graduate school. Informants were employed full time ($n = 9$) or part time ($n = 3$), unemployed ($n = 3$), retired ($n = 3$), disabled ($n = 1$), or students ($n = 2$). Only one patient did not have health insurance. Informants' occupations included homemaker, student, stagehand, cashier, church deacon, art teacher, physician's assistant, accountant, bank executive, and architect. Seventeen informants were Catholic, three did not follow a religion, and one was Presbyterian. Most attended religious services occasionally; five never attended, and five attended regularly.

All patients rated their health as good and either had their thyroids removed prophylactically or because of disease. In

addition, four of these patients had an adrenalectomy for pheochromocytoma. All patients were taking thyroid hormone replacement therapy. Only one of the patients with children did not have a child with MEN2a (see Table 1). Five family members were in excellent health, three were in good health, and one was in poor health.

The men and women in this study related, with ease and vivid clarity, their experiences of the event that started the journey to identify the family disease. Most confessed they rarely talked about it and that it is not at all a part of their lives, yet family members and patients recounted their feelings and actions in detail that occurred as recently as one day and up to 10 years prior to the interview. When asked to tell the story of MEN2a in her family, one woman recalled the following.

It's probably one of the strongest memories I have. . . . We were gathered around together. I can remember so clearly. . . . It was a sunny day; the kids were outside in the neighborhood where . . . my son was saying words to this other little kid in the neighborhood and all this thing was going on outside by the fence and I remember being inside dealing with . . . these fears and doom and wanting to cry when my brother told me [he had cancer] . . . everything but his words were just a blur.

The story of their MEN2a became part of a fixed family record with only minor variations in detail among members. When asked to describe the day-to-day involvement in lifelong care, a complex process materialized, including watching and waiting, looking for meaning behind minor observations, and seeking ways to negotiate control over a threat to health.

For the participants, learning their risk for cancer brought threat to the fore and marked the beginning of the phenomena of participation in lifelong surveillance. The threats were cancer and possible disability. No informant identified the genetic

mutation for MEN2a as the source of threat. The diagnosis of a genetic predisposition caused by a RET mutation received perfunctory attention. One participant's comment represents how patients and family members thought about the genetic nature of their condition: "It's not the MEN2a I'm thinking about—it's the neck that's bothering me today . . . this wind is cold."

Surveillance Events

People in MEN2a families conceptualized lifelong surveillance in broader and looser terms than professionals. Surveillance includes planned and incidental events. Planned events are observations scheduled in advance at intervals and involve the participation of a professional healthcare provider. For example, a planned surveillance event could be an annual visit to the endocrinologist or a blood draw for serum calcitonin level. Incidental events are ordinary daily observations that may occur at irregular intervals or several times a day. Mundane activities occupied the daily reality of lifelong surveillance. For example, a patient might check her face every morning for a twitch that may be a sign of calcium depletion and note the number of pills left in the prescription bottle of an affected child. Incidental and planned surveillance events are reminders of the threat to health. Patients and family members consciously but casually shared the monitoring tasks of checking expiration dates on medicine vials, keeping files of laboratory reports, confirming that children took their thyroid replacement, and following the experiences of strangers on the Internet for episodes or reactions similar to theirs (see Table 2).

(Re)Minding: The Core Concept

Participation in lifelong surveillance is characterized by a core psychosocial process of (Re)Minding. Surveillance events bring threat to the fore as patients and family members recall diagnosis and treatment. Once the threat of disease is brought to awareness by a surveillance event, the individual "minds" or "tends" to the perceived meaning and consequences. Minding involves apprehending and attending to the relationship between the surveillance event and the threat to health. For one patient, the threat to health was brought to the fore by leisure activities.

I exercise a lot, and I'm reminded constantly when I'm in class of what I can and cannot do. . . . I can't [do what I used to do] . . . because I'm missing so much structure in my neck muscles for one. . . . When I do a sit-up, I have to have my hand on my head because I can't pull my head up from the ground.

A twitch in the arm, a stiff neck, a feeling of anxiety, palpitations, a headache, or any medical problem that may be related or unrelated to MEN2a was acknowledged and suspected as a possible manifestation of recurrent cancer or complication from treatment. Although these may seem to be intrusive thoughts, the source was empirical and the thoughts occurred as a consequence of participation in a surveillance event. One patient said,

When I wake up in the morning and I have pins and needles in my arms . . . I think about it, and, when I'm uncomfortable at night, I think about it. . . . When I walk outside and the wind hits my neck and I feel cold on the left side . . . I think about it.

Table 1. Mutation Status of Patients' Offspring

Patient	Patient's Gender	Patient's Age (years)	Patient's Offspring	Offspring's Mutation Status
1	Female	74	Daughter	Positive
			Daughter	Negative
			Son	Positive
2	Female	34	No children	—
3	Male	54	Daughter	Positive
4	Female	20	No children	—
5	Male	40	Daughter	Negative
			Son	Negative
6	Female	45	Three sons	Unknown
7	Female	41	Son	Positive
			Son	Negative
8	Female	34	Daughter	Unknown
			Son	Unknown
9	Female	40	Daughter	Positive
			Son	Positive
10	Male	50	Daughter	Negative
			Son	Positive
11	Male	19	No children	—
12	Male	43	Son	Positive

Negative—does not have the mutation causing multiple endocrine neoplasia type 2a (MEN2a); Positive—has the mutation causing MEN2a

Table 2. Description and Examples of Health Surveillance Events

Event	Description	Event Examples
Planned surveillance	These events are scheduled in advance and involve the participation of a professional healthcare provider as the observer. They occur at infrequent intervals and may require several hours of the patient's and/or family member's time.	• Serum and urine biochemical analyses • Follow-up visits to endocrinologist • Follow-up visits to surgeon
Incidental surveillance	These events do not involve a professional healthcare provider as the observer. They occur at irregular intervals and may occur daily or several times during a day.	• Monitoring oneself or an affected family member for physical sequelae of thyroid hormone replacement therapy • Assessment of oneself or an affected family member for signs of serum calcium depletion • Monitoring oneself or an affected family member for functional or physical limitations • Overseeing status of pharmaceutical supplies of thyroxine and calcium

(Re)Minding is the central psychological process for family members also. They watch over their affected relatives. When thinking about their roles as caregivers and their other responsibilities, spouses or parents may become concerned about their loved ones' health. The mother of a high-school-aged boy said,

When other things are stressing me or him out, it's [reminder of the MEN2a] just one more thing . . . to worry about . . . but it just blossoms in my mind. . . . I think of how I could forget to take him for his checkup or renew his prescription.

The nature of lifelong surveillance is to be aware of and attentive to many kinds of subtle and conspicuous physical feelings and environmental cues that trigger a memory of the disease or treatment. Patients and family members are reminded of the need to watch for problems that are possible at any time and often beyond their control. Each incident of surveillance brings to the consciousness a sense of the threat of the disease that was presumed to be treated or cured.

Feeling Response

(Re)Minding generates a range of mostly unpleasant feelings, including worry, regret, sadness, fear, and anger. Family members and patients also may feel protective of their loved ones and relieved after completing planned surveillance events such as an annual visit to the endocrinologist (see Figure 1). Planned events typically generate feelings of anger, annoyance, irritation, fear, and anxiety, whereas incidental events may generate feelings of sadness, worry, self-doubt, and uncertainty. Family members reported anxiety and worry. The mother of an affected daughter said,

Prior to the test, I'm anxious because I know I could go there and they could say the calcitonin has increased. And they could say to her . . . that she needs another operation. So, I kind of get real anxious and real . . . I don't know, it's just like an anxiety to me.

The personal needs of family members may be placed second in importance to those of their affected relatives. A wife and mother of MEN2a patients said, "I think their issues are more important than my issues. . . . Theirs are always primary in my mind."

(Re)Minding also generates feelings of uncertainty that have a disintegrating effect.

I hesitate doing things now because I know, in the long haul, my body's going to react to it and be down. I'm cautious now in everything I do. It seems I'm almost disconnected from it [my body] a lot of times. It's like, uh, you know, ancient history, almost. It seems so long ago in a lot of respects. It's almost . . . I talk about it almost as a third person five years from now.

Routine, planned follow-up visits are especially likely to fracture feelings of well-being.

I don't want to go; why do I have to go? I'm sick and tired of this. I don't feel like waiting in the office for three hours. . . . But, I go and get it done with and then leave.

Feelings of certainty may be experienced when patients and family members receive information that is understandable and empirically substantiated. For example, when a patient received a laboratory report that his thyroid replacement level was abnormal and it matched his physical feelings of tiredness and mental clouding, he was certain that his assessment of his state of health was correct, repairable, and not indicative of cancer.

(Re)Minding is central to the experience of participation in lifelong surveillance for patients and family members who have to process their thoughts and feelings associated with monitoring predisposition to disease. (Re)Minding involves

- | | |
|---------------------------|----------------------------|
| • Uncertainty | • Abandonment or isolation |
| • Anger | • Weariness |
| • Irritation or annoyance | • Regret |
| • Fear | • Urgency |
| • Worry | • Vulnerability |
| • Resentment | • Gratitude |
| • Guilt | • Determination |
| • Sadness | • Courage |
| • Anxiety | • Relief |
| • Helplessness | • Hopefulness |
| • Self-doubt | • Certainty |

Figure 1. Feelings Generated During (Re)Minding by Surveillance Events

remembering and being attentive to thoughts about the threat of cancer and the effects of treatment. Once threat is brought to the fore by the surveillance event, participants begin the sequential, complex psychosocial processes of Interpreting the Object, Negotiating Control, and (Re)Integration.

Interpreting the Object

(Re)Minding impels the psychosocial process of Interpreting the Object. This process guides and forms the action or response to a surveillance event. The “object” is anything that can be indicated or referred to and is the product of symbolic interaction between the observer and the one observed (Blumer, 1969). An object may be tangible, such as a physical sensation of neck stiffness; social, such as the relationship between a patient and an endocrinologist; or abstract, such as an idea, a principle of behavior, or an expectation. For example, the object of a patient’s inspection of his or her neck incision line in the mirror could be an expectation that the scar is less visible. Three steps in the process of Interpreting the Object identified from the narratives were to (a) seek information, (b) validate the threat, and (c) assign meaning.

Seek information: Usually their first step, patients and family members seek information through expert, amateur, and unqualified sources. Information may be sought from relatives, Internet support groups or online chat rooms, and periodical literature, which may not be current. Amateur and unqualified sources may be consulted before physicians. Patients may avoid seeking information from experts when physical feelings are vague or sensations occur frequently. Family members process memories with observations to make judgments about the meaning of a surveillance event. For example, the wife of a man with fluctuating calcitonin levels attempted to relate her reaction to his laboratory values and symptoms.

Every time he gets a calcitonin level it’s different . . . sometimes much higher . . . sometimes lower. I can’t see a connection between the way he felt before the first surgery—when his calcitonin was through the roof—and this one that is way down. So, you see, I don’t place much stock in the lab work to tell me how he’s doing.

Validate the threat: After seeking information, a patient or family member validates the threat. The wife of a patient and mother of a child with MEN2a relied on past experience. She said,

Every time my daughter tells me she has a headache, I start to get nervous . . . because she has had only one adrenal gland out and they tell me that it’s a good chance the other will form a pheo [pheochromocytoma] because of the way that the disease goes . . . and to look for the symptoms that she had prior to that. She had the surgery when she was 17, but I know when she was 10, maybe 8 years old, when she would tell me she had headaches. When I think back. I know now.

This woman validated the threat when she recognized the possibility that the object (i.e., her daughter’s report of headache) could be linked to recurrent disease but also validated her own tacit knowledge that the headache was similar to past headaches and not a symptom of adrenal disease.

Assign meaning: During the third step of Interpreting the Object, individuals assign meaning to the outcome of the sur-

veillance event intuitively or empirically. One patient recalled the following.

When I was in the hospital after my car accident, the x-ray of my back showed a shadow around the adrenal area. I immediately thought it was a pheo [pheochromocytoma], even though they said it was probably a hematoma. I still thought it was related. I talked about it with my husband and my daughter when they came to visit me. A CT [computed tomography] scan showed that I was okay. I realized my blood pressure was not high, and I didn’t really feel like it was a pheo.

This patient had a physical experience in the context of a doctor-patient relationship that required her to seek additional information to determine the validity of the threat and how it related to her health. Patients and parents of affected children contact healthcare professionals when a threat is validated as real and only after observing persistent symptoms and discussions with family members and informal sources of information.

The process of Interpreting the Object requires individuals to seek information, compare the information with observations, and assign meaning based on collected information from scientific and professional sources and personal, physical, and emotional feelings. Experienced healthcare providers help to interpret the object during planned surveillance events. This rarely occurs after incidental surveillance events for which the opinions of people with specific experience with MEN2a are more valued. Patients may become experts within their family. This process is cyclical, not linear. People may need to seek information repeatedly before validating a threat. Once meaning is assigned, interpretation is complete and the psychosocial process shifts to Negotiating Control.

Negotiating Control

Negotiating Control is the process during which a person deals with meaning and arbitrates behaviors. The observer and the one observed reconcile their different interpretations of the meaning of the object. The patient typically has the more powerful position in the negotiations because he or she is the subject of care. Negotiating Control is rarely positional or adversarial. From patients’ point of view, negotiating is a process based on principles such as shared interests, best options and outcomes, and standards of care. Patients and family members confer over what is to be done with the meaning of a surveillance event.

The two steps in Negotiating Control are to manage effect and manage care. Effect is managed by mediating the impact of the surveillance event on the quality of life of the patient and family. Care is managed by attempting to preside over the way care is delivered by healthcare professionals and how it is accepted or followed by patients.

Manage effect: During the process of Negotiating Control, patients and family members attempt to take charge of the potential effect to the family. This is done by anticipating possible outcomes, identifying a best outcome, and naming standards against which to measure the value of the outcome. The effect of the surveillance event is managed through selective inattention. People will control the sources and flow of information. They selectively attend to some advice and disregard other information. For example, when a female patient

experienced cardiac palpitations, she described feeling panic. She recalled the signs of adrenal hyperplasia, the recent favorable screening test results, and her history of anxiety attacks. She interpreted the palpitations to be a consequence of anxiety unrelated to MEN2a and attempted to control the effect.

I try to convince myself, "You know it's just a panic attack. You know you're not going to drop dead. . . . Nothing's going to happen. You just went to the doctor a couple of months ago; all your tests were good." So, I talk myself out of it. I guess I do what people do. . . . They kind of put it in the back of their head and not think about it.

Her way of managing the effect was similar to a male patient's reaction to a slight change in his laboratory results.

There is very little that can be done about it. I really don't give it much thought. To give it thought is to set yourself into a state of panic. I have fear from it, but I try not to let it rule my life.

For some patients, selective inattention was extreme. One woman said,

I don't collect information, I don't read about it, and I don't care to know. I don't save my medical stuff. I just don't care. I probably don't want to know if something else is going wrong.

Ultimately, selective processing of information has a protective effect.

Manage care: Patients and family members negotiate control by acting on each interpretation of the meaning of an event. By managing care, patients exert a degree of control over an otherwise uncontrollable disease process. Patients actively and passively manage their own care. Patients may actively control their care by simply attending each scheduled follow-up visit and adhering to health-promoting guidelines or by refusing care. One patient controls her care in discrete and constructive ways.

With my own care . . . I'm kind of in charge. I find new studies or new tests; I fax them to my oncologist or bring them along. I don't know if he is aggressively looking for information, but I always am. It's a good control thing for me because I feel like I have a little bit of control. As I take care of my son, I talk to his doctor the same way I talk to my own oncologist. I don't think it's them looking for [information for] me. . . . It's me looking for them.

Another patient passively controlled his care by refusing advice and missing appointments.

I just act dumb, I guess. Basically, I mentally shut down and I just sit there and stare at my doctor who tells me all the good things and the bad things, and I just shake my head, yes. And, I quietly leave.

Patients control their care by manipulating medication schedules, dosages, and availability. A female patient and her husband interpreted world events as a threat to her supply of medicine. She and her husband negotiated control and managed her care accordingly.

Ever since the Y2K scare, I get a couple of prescriptions with refills when I go to Dr. K, and I have most of them refilled. My husband takes care of that for me. I want to

have it on hand. To this day, I have at least three months in advance and I always will get another prescription when I go. We think it is very important that a person should [refill prescriptions in advance]. . . . Their life depends on it.

Unrelated events may remind people of the threat, which proceeds to interpretation and leads to Negotiating Control. One man admitted,

Before 9/11, I didn't think that anything was going to happen to my [thyroid replacement medicine] supply. But there have been times when I think . . . and said half jesting . . . "Come the civil emergency, you can go to the shelter; I'm going to the drug store."

Patients may adjust their daily dosages of thyroid hormone replacement based on self-evaluation and perceived physical responses. Low energy, weight gain, increased sensitivity to cold, and irritability all may cause patients to "take a little more or take a little less [medication] today." Calcium also is used to self-medicate. When patients feel shaky, have muscle cramps, or experience tingling in their hands, they take more calcium.

Parents collaborate in managing the care of affected children. When one set of parents learned that their son had the genetic mutation for MEN2a, they selectively attended to information, used personal experience, and considered possible outcomes. They managed care through the process of principled negotiation with healthcare providers.

My son, at the age of seven, had his thyroid removed, much to the dismay of my and his endocrinologist. His thyroid indeed contained C-cell hyperplasia, definitely a precancerous form of medullary thyroid carcinoma. We made the right, but agonizing, decision [to have his thyroid removed]. . . . We made the decision after many visits to the World Wide Web and online medical databases and going back and forth with the doctors and family. We thought this [the surgery] was the best possible solution.

(Re)Integration

Participation in lifelong surveillance proceeds from Negotiating Control through the process of (Re)Integration. (Re)Minding has a disintegrating effect that may not be acknowledged but always is followed by efforts to restore the integrity of self-image, image of the affected family member, or integrity of the family. The process of reestablishing an intact image of self or other has two steps during which the patients and family members integrate the effect and move on.

Integrate the effect and move on: All surveillance events cause change, however slight, in perceived health and self-image. A patient's sense of self and a family member's sense of the wholeness of an affected loved one may be restored as a revised ideal. One woman integrated her physical feelings after surgery with follow-up care.

My whole perspective, everything's changed. I've become different because of it. I feel physically better now. It's like the way I felt after the adrenal tumor was removed. And, mentally, yeah, I think I'm a lot better. Who knows? Maybe this has all been a good thing. I am more health conscious in general.

Even the visual reminder from a radical neck incision line becomes integrated with daily life.

My scar? I sometimes don't notice it. I don't think I have scars anymore. It's weird; I'm not conscious of it on a daily basis. . . . I'm a salesman and in the public eye or in public view all day long, five days a week. I deal with and meet new people every day. And I think at first they look at me differently; I don't even notice that they look at me differently, but then after they get to know me, it's no big thing.

The young spouse of a man with MEN2a related how they (Re)Integrated after daily reminders.

After he goes to the doctor or even when he looks in the mirror, he's reminded that he has this disease, and maybe he doesn't realize how long he has. We know we have to build our relationship back up each time we get bad lab results.

Some patients (Re)Integrate by dismissing daily changes and lifelong follow-up. This was true for some affected family members who refused to participate in scheduled follow-up monitoring. They negotiated control through avoidance and maintained a sense of wholeness by perceiving the outcome of surveillance as irrelevant. One man confessed his indifference to the value of follow-up monitoring.

I look at it this way . . . it happened. I took care of it. It's over, it's done with, that's it. I have a bottle [for an annual 24-hour urine collection] in my back room, which I've had for about 10 years now. Unless I'm sick, I don't really bother with the [surveillance] regimen.

A health-promoting outcome of (Re)Integration is to make monitoring routine. Parents with affected children are especially conscious of the need to blend incidental surveillance events with activities of daily living and limit the stress associated with formal surveillance activities. The father of two teenage sons, one who is affected, said,

We kind of kept all of it in the background 'cause there's no need to alarm them. I think there's no real reason to alarm them with stark information because they listen pretty well.

Making surveillance activities routine is especially important for parents who struggle to prepare their children for lifelong monitoring. A mother speaking about her son said,

I worry [taking his pill and watching his health] may not be instilled in him yet to do on his own. I try to say, "Did you take your thyroid pill yet?," to kind of start. . . . You have to be responsible for yourself. I want to make that connection between himself and taking the pill . . . not just to take it because we put it on his plate or that I'm making him do it.

When the activities and outcomes of surveillance are changed from extraordinary to ordinary, patients and family members "move on." Participation is put behind them, and life goes on. As one female patient remarked,

I would rather be here with the MEN [MEN2a] than to not exist at all. I'm happy to be here. I do the right things. I don't steal, I don't cheat, I don't lie. I'm fine. I try to adapt to the changes that follow-up care brings. I do this

by just getting on with life. Despite any changes . . . life still goes on and the changes and challenges must be met. I mean, I try to get my life back to almost as normal as possible. There is nothing you can do about it, so just live your life happily if you can.

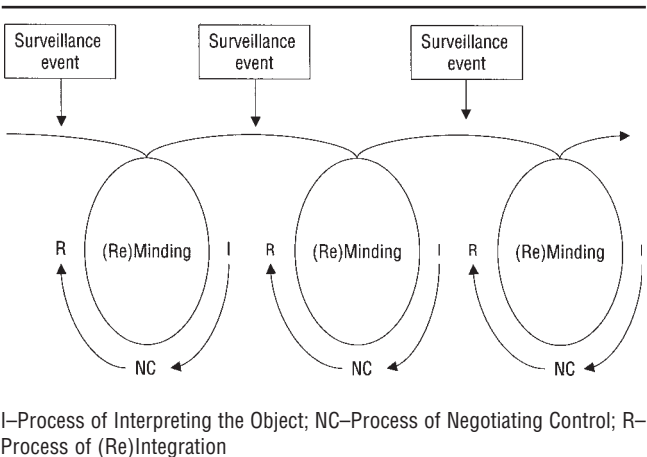
The impact of monitoring for MEN2a lessens, and thoughts diminish with time for patients and family members, until the next surveillance event reminds them of the threat to health and triggers the processes to deal with persistent threats.

Discussion

The core concept of (Re)Minding captures the rhythmic and repetitive nature of surveillance. Because of the temporal nature of recurrent threats to health with MEN2a, it may be termed "chronic." One researcher described a relapsing or episodic course of illness that alternates between stable periods of varying length and periods of flare-ups. Families with MEN2a are flexible because they respond to the strain created by the movement back and forth from crisis to noncrisis and the uncertainty created by not knowing when the threat of cancer will become a real danger (Rolland, 1987, 1990) (see Figure 2).

Finding meaning in the outcome of a surveillance event is a psychosocial process central to participation in surveillance. Like the experiences of patients with other cancers and their family members, finding meaning is important to maintaining an intact sense of self (Davis, Zinkand, & Fitch, 2000). Finding meaning helps patients to situate their diagnosis and ongoing care in a new life and cope with symptom distress and the possibility of an early death (Bowes, Tamlyn, & Butler, 2002; Lammers, Schaefer, Ladd, & Echenberg, 2000). Unlike the experiences of patients with other kinds of cancers, patients with MEN2a and family members are much less engaged in aggressive action to battle illness. The word "surveillance" engenders images of waiting, clandestine work, and inactivity; yet, they feel satisfaction from prevailing over adversity (Allchin-Petardi, 1998).

Rolland (1999) suggested that families maintain a mastery over daily events by acknowledging the possibility or inevi-



Note. Surveillance events may be planned or incidental and continue over a lifetime.

Figure 2. Surveillance Events, Basic Psychosocial Problems, and Participation in Lifelong Surveillance

tability of loss, sustaining hope, and being flexible and adaptable to change. Surveillance activities are most relevant to the presymptomatic chronic phase of genetic predisposition to cancer; however, patients and family members may have a heightened sense of the preciousness of life and relationships as a consequence of participation in incidental and planned surveillance events.

Symbols represent something else by association, resemblance, or convention; for example, a picture of a heart may symbolize love. Symbols are social and defined by interaction (Charon, 2001). People make them, and people agree about what they represent. Genetic predisposition to cancer is symbolized by the interaction of the observer and the one observed as they watch and wait for a threat to health. The observer and the one observed manifest the meaning in human action toward the symbol by creation of a social fabric in which the symbol is shared among members (i.e., affected children, professional caregivers, extended family) and contribute to the construction of individual life (Blumer, 1969; Charon).

In this social fabric of participation in lifelong surveillance, members of MEN2a families negotiate control over the symbolic meaning of a surveillance event. In part, this is achieved by engaging in self-management. Although Fox and Gruman (1999) suggested that this process was difficult to develop and hard to sustain, MEN2a patients were motivated by the desire to minimize negative effects. Rather than needing instruction in self-monitoring, as proposed by Von Korff, Gruman, Schaefer, Curry, and Wagner (1997), participants in this study easily, naturally, and intuitively monitored themselves and others, tailored health care, and sought support from peer experts.

Each surveillance event culminated with the process of (Re)Integration to restore an intact image of self or a loved one. Participants' accounts of their experiences began with descriptions of the crisis and concluded by elaborating how they had moved on with their lives. As for people with other chronic illnesses and cancer survivors, their aim was to normalize in the face of persistent symbols of a health threat (Anderson, Elfert, & Lai, 1989; Knafle & Deatrck, 1986; Rechner, 1990; Robinson, 1993) (see Figure 3).

MEN2a tumors are indolent and slow growing, and long stretches of time separate medical and surgical interventions. This time is filled with incidental surveillance. Incidental surveillance events give people in MEN2a families opportunities to take control of care. These opportunities may temper feelings of urgency, worry, and uncertainty. Engaging in incidental surveillance, therefore, may have a positive and protective effect. Over time, patients and families find it easier to interpret the object, negotiate control, and integrate the effect and move on. The cyclic, repetitive nature of incidental surveillance, however, may cause, for some, a decrease in worry and a decreased interest in formal follow-up. Planned surveillance events become bothersome and perfunctory, especially when annual checkups offer no new information, mistakes are made with laboratory results, or communication is careless.

The Genetic Nature of Multiple Endocrine Neoplasia Type 2a

When trying to explain human behavior, often that which is not observed or left unsaid is as important as visible acts and explicit remarks. The genetic nature of MEN2a was unexpectedly absent from the narratives and, therefore, is not

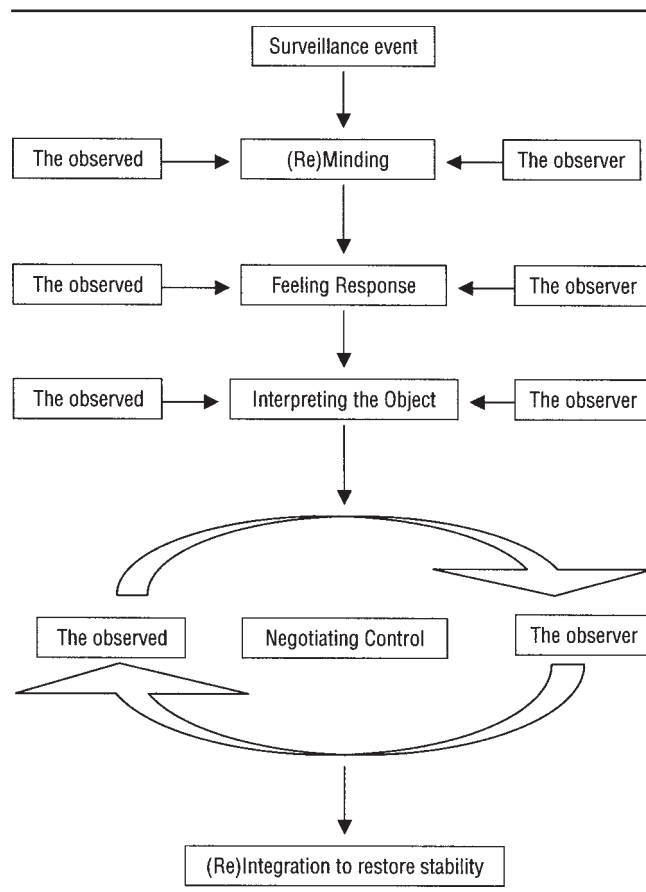


Figure 3. Relationship Among Core and Process Variables

named as part of the core or process concepts. Yet, when asked, patients and family members conveyed a sophisticated understanding of the relationship between genetic mutation and disease. The omission may represent a subconscious and an intuitive rejection of the notion of genetic determinism (Sober, 2000). If a patient with MEN2a or a family member had embraced the notion that “we are what our genes make us,” this belief would manifest itself in the process of interpretation. This did not emerge. An acceptance of genetic determinism would mean that nature could not be modified by nurture, so some efforts to Negotiate Control would be futile and (Re)Integration might be less likely. The implied rejection of the singular power of the RET mutation to affect the course of their disease and life with MEN2a may be essential to (Re)Integration (i.e., normalization, moving on). Also, rejection of the notion may herald the coming of a new standard of normalcy for which the absence of a disease-causing gene is not a criterion. People who carry a genetic mutation that predisposes them to cancer may see themselves as healthy even when clinical geneticists would instruct otherwise. Healthcare professionals may need to examine how patients and family members construct their beliefs about health and illness before they can offer a definition that is meaningful to patients with MEN2a.

Nursing Implications

People in MEN2a families conceptualize lifelong surveillance as a personal and family phenomena more than the re-

sponsibility of their healthcare professionals. To these families, surveillance is largely informal and infuses daily life. Ultimately, the benefit of enhanced surveillance depends on patients' and family members' willingness and ability to participate in these activities. Nurses can be key communicators of the importance of this relationship and positively influence patient participation at all process stages described in the model.

Findings point out that, although planned surveillance is professionally guided, most informal surveillance activities are not based on scientific evidence of usefulness. Patients' perceptions of the effectiveness of ongoing treatment, complexity of screening tests, and fear of results are the main determinants of compliance with screening (Becker, Kraft, Southwell, & Jorgenson, 2000; Blalock, DeVellis, & Sandler, 1987; Matthews & Hingson, 1977; Neilson & Whyne, 1995; Vernon et al., 1997). In a descriptive study by Berenson, Groshen, Miller, and DeCosse (1989) of patient compliance with chemoprevention trials, investigators proposed that long-term compliance may be markedly worse than short term, especially if extra effort is involved, such as frequent medical examinations, long travel and waiting room time for medical visits, and repeat blood work because of technical errors. The emotional tax of (Re)Minding and the psychosocial energy required for Interpreting, Negotiating Control, and (Re)Integration are not known. If the emotional tax and psychosocial energy are great, the psychosocial expense may be the chief reason for cases of nonadherence to surveillance guidelines by patients with MEN2a. They often perceived laboratory tests to have little meaning in relation to their feelings, and visits to specialists were burdensome. Patients in the study tended to postpone visits or lengthen the interval between visits. Nurses, therefore, must evaluate how patients and family members use findings from informal surveillance to modify participation in planned surveillance, manage disease, and live with the diagnosis. Nurses may use the proposed model to develop assessment and intervention strategies to optimize patient benefit from clinical genetics surveillance guidelines.

Limitations and Recommendations

When recruiting informants, the researcher aimed to include adolescents, aged 14 and older, who began surveillance activi-

ties early in life. This was not accomplished. In addition, the sample was racially homogenous. Only one family member was Hispanic; all other informants were Caucasian, and only one young man (age 19) had a prophylactic thyroidectomy at age 12. The theoretical explanations related to the core concept and process concepts may not capture the experiences of minors and, therefore, should be explored in depth with this cohort of patients and their family caregivers.

The study barely addressed the issues faced by parents who must transfer responsibilities for lifelong monitoring to their affected children. The unique aspect of parenting a child with MEN2a is an essential component of their experience and should be studied with a cohort of mixed ages, races, genders, and socioeconomic diversity. Because the condition is rare, a multisite and, possibly, multinational study will be necessary.

This study underscores the dynamic nature of health surveillance in genetic cancer care. It illustrates the social nature of the phenomena, the important role played by family members, and how self-surveillance and informal observation dominate the phenomena. Prior to each encounter, nurses should remember that surveillance activities are not limited only to the visit with the healthcare professional but also are an important part of daily life and involve significant others in the immediate and extended family.

Lastly, findings and the conceptual model are valid for members of MEN2a families and may be valid for families with genetic predisposition to other cancer syndromes, such as familial adenomatous polyposis. The author recommends that research be conducted with families with familial adenomatous polyposis, breast and ovarian cancers, malignant melanoma, and other cancer predisposition syndromes with enhanced lifelong surveillance guidelines.

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