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At the Bedside

Throwing Jello: A Primer on Helping Patients

Edmund G. Howe

In this issue of *The Journal of Clinical Ethics,* a case from the Harvard Ethics Consortium discusses a patient named Lorraine who sometimes was so violent she drove careproviders from her room. Such situations are not uncommon, and the outcomes often are tragic. In this case, Lorraine eventually stopped treatment, suffered greatly, and died of infection from her decubitus ulcers.

In situations like this, staff are often split: some side with the patient and others side against. The latter may come to feel contempt for the patient, and, if they do, the situation typically worsens. The patient can feel increasingly alone, and become more aggressive, maybe because isolation is the most painful of all feelings. Staff may then feel even more helpless and enraged, and the vicious cycle intensifies.

This is apparently what happened with Lorraine. As Jennifer Repper-DeLisi and Susan M. Kilroy write in " 'We Need To Meet,' " the staff's "natural responses" to the patient were "frustration, anger, and rejection." Staff usually try to do the best they can, as in Lorraine's case, but we may not help patients as much as we hope. In reality, it may not be possible to help them — for example, in this case it is unlikely that Lorraine could have done better. But there are exceptional approaches that can be used when conventional approaches don't succeed. These approaches may help all patients, but, with patients like Lorraine, they may be lifesaving, so ethics consultants and other careproviders should at least know about them.

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PRELIMINARY CONSIDERATIONS

WHY WOULD A PATIENT ACT THIS WAY?

The approaches I'll describe can be used with any patients with whom we find it difficult to relate. Patients who become angry, as Lorraine did, pose greater problems when they express anger, which can evoke fear. We all are at greater risk of becoming inappropriately angry when we are ill — or even just feeling stressed. Some people, however, habitually

become inappropriately angry, and this is one hallmark of those who have borderline personality disorder.[1] Therefore, I will use this disorder as a paradigm for understanding patients like Lorraine.

The mood swings of people with this disorder are, characteristically, "stably unstable" — their moods are fragile, but the state of fragility is static.[2] Persons with this disorder are calm one minute and may feel enraged the next. As one clinician describes it, "The slightest perception of condemnation may result in a strong and unpredictable violent response."[3] Why?

One way to understand this behavior is that people with the disorder have great difficulty sustaining the feeling that they are "okay," and that almost any input can trigger a belief that they aren't okay. When the patient's mood plummets, she or he may feel worthless, and that may be unbearable. The patient's mind may immediately and unconsciously try to relieve this feeling by replacing it with another that is less painful. The "replacement feeling" might be fear, and the patient may become paranoid. Another feeling that may replace worthlessness is anger, because, for many people, it is less painful to feel anger. But, in the long run, anger may be worse for the patient, because it can frighten other people away, and the isolation that results may be more than the patient can stand.

The implication for careproviders is critical: they must avoid triggering patients' feelings that they aren't "okay." Recent advances in brain imaging provide additional support: they indicate that, physiologically, persons with this disorder may experience inappropriate and overly intense anger for two reasons. First, the parts of their brains that trigger aggression may be too sensitive, and their anger centers may be too easily triggered and "fire off." Second, the other parts of their brains whose "job it is" to inhibit this response may under-function.[4]

In any event, the result is the same. The "nerves for aggression" fire, and the patient "explodes." The aggressive behavior is virtually automatic.

One implication is the one just considered: careproviders should avoid inadvertently triggering patients' angry responses. A second implication is less self-evident: patients may be overly aggressive for another reason. They might intentionally be aggressive for personal gain.[5] This may be not because they are impaired, but because they choose to be aggressive. Such behavior is willful as well as exploitative.

Therefore, careproviders who want to help patients like Lorraine may feel they are in a bind. They can't know whether a patient's anger is fundamentally involuntary, and so is outside the patient's control, or is intentional — or maybe is both. Further, it's natural to assume such behavior is intentional, and to act in a way to best protect ourselves.

But if we do, it may cripple our efforts to help. If we misunderstood, patients may feel wronged and even betrayed. An example is when we respond to a patient who becomes angry and demanding and we reflexively set limits that are inappropriately strict. The patient may see these limits as arbitrary and lose trust in us.

WHAT SHOULD BE AVOIDED?

It's ideal for careproviders to not respond with inappropriately strict limits, but patients do test the limits set for them. This may be one of their underlying problems. As one careprovider noted, "Many of the most desperate patients refuse to play by our rules."[6] We may need to alter our usual limit-setting practices or risk failure with these patients. As another careprovider suggested, "Excessive technical rigidity limits receptivity to the client's style of problem solving."[7] What should we do instead? There are three ground rules. The first is the most important.

1. Allow others to set limits, while we help patients pursue their interests until the limits that are set are absolutely necessary. Stated another way: we should ally ourselves wholly with patients' needs and pursue meeting them as vigorously as we would our own.[8] This is the key. To any degree that we compromise our commitment to patients' interests, we lose

the capacity to help.[9] Other parties can and will, in time, set limits, and we can pursue patients' interests until this happens.

Here's an example. A father so distrusted surgeons that he would not consent to surgery for his six-year-old daughter unless he could watch while it was being done. He agreed to watch via video hook-up from another room, but the surgeons refused and threatened to go to court. An ethics consultant could have pursued the possibility of a video hook-up, and perhaps also arrange for a careprovider to describe what was being done. The rationale is that patients may not readily accept careproviders as their allies.

The remaining two ground rules are far less demanding, and follow from the first.

2. Careproviders must be wholly willing to ignore their own moral views when they hope to convince a patient that they serve only the patient's interests. This becomes more difficult because careproviders must retain the credibility of their colleagues when they do this.

3. The third ground rule, therefore, is that careproviders (beforehand, ideally) should explain to colleagues what they will do, their underlying rationale, and why it is not only justifiable, but morally obligatory. If they don't do this, in extreme cases, a patient may die unnecessarily. The approach of informing others that one will engage in an unusual therapeutic endeavor, and explaining why, has been carried out in other settings. For example, one psychiatrist I knew would routinely alert his colleagues that he wasn't initially going to give patients with schizophrenia who were psychotic antipsychotic medication. He believed that for them to be willing to take medication over the long run, they had to believe, themselves, that they needed it, but that sometimes they would only come to believe this if they could test it out for themselves.

When careproviders help patients to pursue what they believe they need and want, others may establish limits. When this happens, careproviders can say truthfully that they have done all that they can. Even when patients are greatly impaired, they are still likely to be able to fully understand and appreciate that, although their careproviders have some power, they aren't all-powerful. Careproviders should discuss this with patients in advance. It may allow them to remain the patients' allies.

WHAT CAN BE DONE?

To reach patients, above all else, careproviders must form a trusting relationship. If patients can acquire such a relationship with just one careprovider, it may be enough, because the patient will no longer feel alone. If this is possible, the patient may not feel alone, even if the careprovider is away.

When patients are severely impaired, it is easy to see how important it is for them to have a careprovider they feel they can trust. Even when patients are paranoid or floridly psychotic, a trusted careprovider still may be able to "get through" to them, even at the most difficult times. This is because it has been found that, even when patients are mostly out of touch with reality, they still may retain the capacity to respond to others in a normal way.[10]

To gain patients' trust, we may need to respond quite differently than we normally might. For example, we usually hold patients accountable when they act inappropriately. With a patient like Lorraine, in the Harvard case in this issue of *JCE,* we might do well to do the opposite. To gain a patient's trust, we might better respond as though we somehow provoked the inappropriate behavior, and ask the patient what *we* did.[11] In time, we may be able to be more fully honest with the patient; later, we may even choose to share how we and other staff feel toward the patient — and the feelings that we disclose may even include hate. To do this successfully, however, we must first earn patients' trust.

It is not wholly irrational to respond as if we somehow caused a patient to react as he or she did. As Mary Zanarini and Kenneth Silk, leading authorities on borderline personality disorders, state, "it is hard to imagine a borderline patient bothering to regress on a desert island."[12] That is, patients don't become

aggressive when they are alone. If we want to foster a trusting relationship with a patient, we may take this further: should a patient become aggressive, we may choose to respond paradoxically, as some of the careproviders in Lorraine's case did: we may choose to respond by being more caring and loving.

August Aichhorn used this approach when he treated involuntarily committed juvenile delinquents more than a half-century ago.[13] When, for example, these teenagers "acted badly," he invited them to join him and his wife for a special dinner in their home. Why did he find this approach effective? Above all else, doing this conveyed a feeling of all-overriding and unconditional love. By doing this, Aichhorn conveyed that he could distinguish at all times between what these delinquents did and who they were. He communicated clearly that he never forgot that they were still wholly lovable persons.

These delinquents may, like some patients, "act out" most when they hurt the most. Thus, when Aichhorn invited the delinquents to dinner, it may have also acknowledged his implicit understanding of their unexpressed hurt. We can indicate this too. If we were to bring a favorite meal to a patient like Lorraine after she "acts out," we might say, "I imagine that you may have been angry because you were hurting. Were you?" The key to reaching patients is best conceptualized as follows: we should act, in all circumstances, as if it is our relationship with patients that counts the most with us. For instance, after an ethics consult is finished, no matter what the outcome, our relationship with the patient should be intact.

SPECIFIC STEPS

1. CREATE AN ENVIRONMENT WHERE A PATIENT WILL MEET AND TALK

Getting a Patient to Meet

A patient may be unwilling to meet with careproviders; often this is due, in some way or other, to fear. When this happens, we can try to find ways that the patient will feel safe enough to meet with us. Here are three approaches.

Offer to go to the patient's home.[14] Some patients who won't come into the hospital will agree to be seen in their own home, on their "turf." In the patient's home, we are the outsider.

Suggest that the patient invite as many other family or friends as he or she wants, whether the meeting is inside or outside the hospital. The patient may see this as a demonstration of sensitivity to his or her emotional "plight," and it may have a profound positive effect.

This offer implicitly recognizes, and represents an effort to offset, the difference in power between patients and careproviders. If a patient invites others, it may help him or her to feel more secure. Would this cause us to feel intimidated? What if the patient invites 12 loved ones? Would we feel alone or anxious? The answer should be no, and the reason is important — as careproviders, we must be wholly devoted to serving only the patient's interests. If we instead try to facilitate a compromise between the patient and other parties, the patient may lose trust in us, and even feel betrayed. We probably can't meet a patient's needs and also facilitate a resolution between competing parties — even though this might be what we would otherwise usually try to do. So it makes no difference how many loved ones a patient invites to a meeting — we should never have a need to feel defensive, because our only goal is to further the patient's "agenda." If we do feel defensive, it may mean that, at some level, we are acting to further another party's needs. If we have a conflicting agenda, it should be acknowledged from the start. It may cause the patient to reject us altogether. Even if this happens, we have remained honest and forthright, and it may help to keep the relationship intact. Later, the patient may be able to reconsider and request that we again provide care.

Increase a patient's feelings of safety by saying, before meeting, that if the patient feels offended at any time during the meeting, she or he can leave the session immediately — no

questions asked. We can also assure the patient that, if she or he does leave, we won't feel angry, but instead appreciate that the patient agreed to meet at all. This provides the patient with an "escape route." With this reassurance, a patient may be willing to meet.

Getting Patients to Talk

If we can meet with a patient and can talk, it may allow a relationship to develop. Some patients, however, may not be willing to talk at all. In this situation, there are exceptional approaches that can be tried. The following approach may serve as a paradigm representing the kind of extra effort that we can make.

Some patients won't speak to careproviders because they are greatly impaired and withdrawn. They may literally be unable to look a careprovider in the eye. They may also find it extremely upsetting to have a careprovider speak directly to them. Since these patients find directness upsetting, we may be able to communicate with them only by looking away or at the floor, or by talking about them in the third person. It is conventionally thought in our society that we should look directly at patients and speak to them, not about them, and to do otherwise is implicitly demeaning. But some patients may not be able to speak to careproviders unless we act in a way that would usually seem demeaning. Why might this work? J.S. Gans notes, "This indirect method should be employed only when direct communication does not enable or facilitate therapeutic work . . . people do not have to be looked at or spoken to directly . . . to feel cared about. . . . With one patient, for example, when the therapist tried to make empathic statements the patient would wince."[15] This approach may assist careproviders. Gans also notes, "The truth is that we sometimes do need temporary, partial insulation. . . . [It can be] reassuring for both. [It communicates to the patient:] Since I know that you are doing the best you can, I will not burden you with expectations sometimes implied by eye contact."[16]

Almost all of us would feel helpless and inadequate when caring for a difficult patient like Lorraine. As a defense, we might come to feel very angry. Our jaw may clench when we are with a difficult patient, to the point we are nearly unable to speak. The potential benefits of being "indirect" when interacting with a difficult patient indicates an important point: Some of our difficulties in communicating may come from us.

Finally, the efficacy of speaking indirectly to a patient indicates another fact: to help patients like Lorraine, we may have to go "outside the box," or even against the values of society and the medical profession. Gans notes that talking with patients indirectly "deviates from what is accepted as normal behavior in our culture."[17] Recent data reports that there may be a scientific basis for taking this approach; brain imaging techniques indicate that it may increase the stress of those with autism when others relate with them directly.[18] Perhaps this is also true, to some degree, for patients like Lorraine.

2. ENGAGE PATIENTS

Careproviders must be able to communicate with and engage patients to help them. An important component in communicating with and reaching patients is the first time a patient meets his or her careprovider. If a patient seems upset at the first meeting, what should be done? If a patient is clearly upset and this is not addressed, the patient may find it degrading.[19] After the initial greetings have been made, a careprovider may engage a patient best by asking what is most important to him or her. We can make clear that it is our goal to try to help the patient to get whatever it is he or she wants most — and, over time, we have to deliver on this.

When we ask what a patient wants and explain our priorities before we do anything else, it conveys that what the patient wants is truly our primary concern. The need to be fully the patient's advocate is absolute. Unless we can do this, patients like Lorraine may not trust us. We may feel hesitant, for many reasons. We may feel that this isn't our role, or we may fear being criticized by colleagues, including colleagues who consult us. Regard-

less, this may be the only way that we can succeed.

Here is how this approach would work, using an extreme case from my own experience. A patient was "using up all of the blood" in the community. An ethics consultant was contacted, and was implicitly expected to help the patient and his family become more open to accepting that, at some time, the blood infusions would have to stop. The consultant worked it through in her head: "There's only so much blood, and other patients need it too. Sooner or later [the patient's] ongoing need for enormous amounts of blood will exhaust the supply. If we allow that to happen, others will die, so it's obvious that this can't go on indefinitely." Although she did not express herself to the patient and family in this way, they seemed to pick up on her dual allegiance. They felt she had come to persuade the patient to give up some days of his life. Rather than accept the reality that others' interests were at stake, they rejected the assistance of the consultant altogether.

The outcome might have been different if the consultant worked it out this way in her head: "I'll help the patient to continue to get blood in any way I can, and be frank about that with him. I'll leave it to others to decide when to say 'No more blood.' My goal is to help him achieve his ends. When it gets to the point that someone else says the infusions have to stop, he may trust me enough to let me continue to help him and his family deal with that news."

How could this same approach be used with a patient like Lorraine who "inappropriately" seeks to die? We could — although it might contradict our own values — side with the patient. How could we do this? We might offer to help the patient "die better." The end result could be paradoxical,[20] but obtaining a paradoxical result is not the goal. We should try to further patients' goals, as they see them, primarily because, if we do, patients won't be alone. They will have us as allies, and this may be enough to give patients a reason to live. Battin states this as follows: "there is a third alternative: . . . work with him, not against him, in planning [his suicide]. . . . My guess is that if [the patient] were really offered help in thinking through his plans for suicide in a straight-forward, non-disapproving, non-duplicitous way, he would be much less likely to kill himself, at least not right away."[21]

Patients might misconstrue such an unusual offer, as careproviders don't commonly offer to help patients to die. Patients may suspect that their careprovider wants them to die, but careproviders, by their words and actions, can convince patients that this isn't the case. The more difficult problem is that patients may suspect that careproviders who offer their help without limits are in some way "setting them up." For example, patients may fear that their careprovider will later use what is said, in some way, against them.

To defuse such fear, careproviders can explicitly acknowledge it. We can say that we can imagine that the patient, on the basis of prior experience, may not trust what a careprovider says, and we can understand that. Further, we can say that, even if this isn't the patient's past experience, it might make sense for the patient not to trust a careprovider, and it might be better for the patient not to try. We can then say that if the patient wants our advice, she or he should trust us only after trust has been earned, and we hope the patient will give us that chance.

How could this approach have been carried out with Lorraine? There are several ways, but this is one. Lorraine had pain, and she wanted greater doses of analgesics. This is one of the most difficult decisions careproviders encounter. The principle of pursuing Lorraine's needs, as she saw them, suggests that her careproviders should have tried to do all they could to give her the relief she sought, whether or not they personally agreed with this goal, and whether or not other careproviders opposed them. They could inform Lorraine that this is what they would do, and they could even say that if other careproviders refused, they would help her to the extent that they would bring her case to court. This might have caused others to give Lorraine greater pain relief, at least temporarily, although it

might have killed her. Thus, the result might have been that her careproviders would have given her analgesics or sedative meds until she had sufficient relief to say "enough," or until she was too obtunded to speak, or until she died. This approach is used by some hospices in situations when patients feel "existential" agony and request terminal sedation to gain relief. The patients report they feel agony, not because they are depressed, but because they feel emotional pain that is worse, knowing that they are waiting to die.[22]

Some patients who are given brief doses of extra sedative medication under these circumstances respond in the following way. After they have been sedated for a few days, the medications that "obtunded them" are slowly withdrawn, and they respond paradoxically by feeling more "alive." They no longer want terminal sedation for the rest of their lives, but rather cherish the remaining days they have to live. Why this happens is not yet known. It may be that after the patients have found a careprovider who is willing to meet their requests, even for a brief time, they feel more understood. Supported in their request, they may feel that they have an ally. This may be what they need, and indeed long for, more than anything else.

3. RELATE TO OUR OWN FEELINGS

We may presume that difficult patients realize how they affect others, but this may not be the case. This may be presumed because some truths seem self-evident. How could this person *not* know? It may be a great error, however, to attribute this knowledge to patients. For example, patients who attempt suicide may not know — or even imagine — at all accurately how their actions will and won't affect others. They may greatly overestimate or greatly underestimate others' pain. Once they attempt suicide and "fail," they may believe that others are able to respond to them in the same way that they did before. But others may not be able to — their loved ones may always fear that they will try this again.

If we can establish trust with a patient such that he or she can really hear us, we can give the patient accurate feedback about how she or he affects others. This may be critically important information to the patient, and may also have immense, hidden benefits for us, as well. Patients like Lorraine need feedback, first, to know what they are doing "wrong," so that they have the possibility of being able to change. They may want to change, so that they can better get whatever it is that they really most want. We may be able to help a patient by doing what careproviders rarely do: we can tell the patient about our own and other careproviders' negative feelings toward the patient. This feedback, and this alone, may enable the patient to acquire the new skills that he or she needs.

For example, Lorraine may have most needed better control of her anger.[23] After forming a bond of trust with Lorraine, a careprovider could have told her she needed this skill if she wanted to be on better terms with the staff. The real gain from such feedback may be even greater: honest feedback, even when it's negative, may help patients feel valued. It may affect them so much that they will feel that they are "among the living," rather than as bedridden, or as someone with another impairment who is "just waiting to die."

When we share with patients what we and others feel, it also helps patients to be able to grow. When a patient is abusive, as Lorraine was, we could say, for example, "I feel hurt when you get angry like this. I also feel afraid to come back into your room. Do you understand why you are doing this?"[24] This is another way that careproviders can help patients feel that they are "still living," rather than waiting to die. As the above phrasing suggests, we shouldn't presume that patients — or any of us, for that matter — know why they act as we do, because we may not. We can anticipate this and ask patients questions in ways that don't imply that they should know why they act as they do.

Or we could ask other questions that patients are more likely to know the answers to:

"Is there something I did that made you feel angry?" Asking in this way lessens the possibility that patients would feel shame, because they don't know why they act as they do. By confronting patients with their own and others' feelings, we can help them accept parts of themselves they feel shame about, such as envy and even hate. As one group of careproviders who did this reported: "By talking about [their] hate, the team helped the patient accept [this] part of herself. . . . Her envy needed to be . . . detoxified. . . . We [made sure that] she did not suffer a loss of human contact . . . [and] repeatedly emphasized our wish to talk to her . . . about these feelings . . . that made her life so difficult."[25]

We can also ask questions that are more confrontational but also show greater concern: "Is there something you are afraid could happen by working with me?" Since some patients may experience this question as accusatory, however, we can tell the patients our intent before we ask: "I know you have reasons for becoming angry. That is always the case, in some way. But it would help me greatly to understand you if I could know what your reasons are. When you become angry you drive me away. Do you want to? Is there something you are afraid could happen by working with me?"

Finally, we could share our own feelings of helplessness, which may help patients see that it is really they who are in control, and only they can try to bring about a better outcome. In most ways, these patients are helpless. Thus, when we share this information, it conveys a feeling that we may have in common, and "shared helplessness may provide a pathway to empathic connection."[26] Sharing in this way may also help to "level the playing field." More than anything else, such unexpected candor may reduce patients' feelings of being isolated, which may be much more painful than anything else.

4. TEACH PATIENTS NEW SKILLS

Once patients like Lorraine are sufficiently engaged to be able to really hear feedback, they may want to change. Even if they don't want to change for the sake of others, they may want to change for themselves. The task, then, is to help patients identify the skills they need, and to help patients find some way to attain them. Lorraine, like other patients who become angry, needed above all else to find a way to control her anger. She may have been able to use a practice called mindfulness,[27] which involves learning to distance oneself from one's immediate emotional responses, and then to continually monitor or even "grade" one's responses. This practice can allow us to not only control anger, it may help us bear even great pain.

Patients who work to learn a skill such as this should be told that each time they try, they will get better at it, and that the reason is that each time they try, the connections between the neurons in their brain will become greater.[28] Not only is this true, it can be a source of hope. But patients should also be given a "safety net," so that they have an alternative way to control their anger if the approach they are learning fails. They could, for example, acquire 24-hour access to someone, somewhere, whom they could call and talk to. This might, perhaps, be a someone that patients can emotionally accept as a "stand-in" for a careprovider they trust when the careprovider is absent. Patients may find surprisingly great feelings of relief in talking with another person whom they know beforehand is standing in the trusted careprovider's "stead." For example, careproviders' receptionists may fill this role. Although their primary duties may be to arrange schedules or answer phones, in some instances a receptionist's response to patients is as important to the patient as the careprovider's response. When a trusted careprovider can't be reached, receptionists have prevented patients from committing suicide. Even if a patient never makes any calls, having a "fall-back option" may prevent an outburst. Further, careproviders who arrange for a back-up may convey to patients how much they really care. Even if other interventions don't "get through," this one may.

Some patients may, on the other hand, have more than enough skills to accomplish what they need to do. When this is the case, our task is to help them identify the skills that they already have, so that they can "transfer" these skills to the situations in which they need them. Doing this may be preferable to helping patients acquire new skills for two reasons: first, patients' need to change may have considerable urgency, and, if they have skills already, they may be able to transfer those skills immediately. Second, if patients have the prerequisite skills, this is a ground on which they can feel genuine, greater self-esteem.

How we can help patients to identify and transfer skills that are already present is illustrated in the following case. An inmate had been repeatedly, impulsively violent before coming to prison. If he continued to be violent in prison, he wouldn't gain early parole, which he desperately wanted to do. He realized that he urgently needed to learn to control his anger, which he'd never been able to do before. He told his counselor in prison, "I can't help myself," and he meant it. His counselor said, "Sure you can. Tell me about when you feel angry, but choose not to beat someone up." The inmate replied, "When they have a gun." "Then what do you do?" the counselor asked. "Then, I walk away," the inmate said. The counselor told him that all he needed to do was to imagine that anyone he wanted to fight in prison had a gun, and he would be able to walk away. The prisoner was able to do this, and he earned early parole.

5. RESPOND TO FEELINGS OF COUNTER-TRANSFERENCE

It is essential that careproviders who interact with patients like Lorraine find some way to continue to care for them. When we can't do this, we may be better off not interacting with the patients at all, or possibly we should work in some other field. Sometimes we may care about a patient, but find that we have feelings like hate. When we do, the course we should take is well prescribed: we should first seek out other staff with whom we can share these feelings, if we don't have this outlet already. The staff we seek out must be people we know won't judge us for having such feelings. If this doesn't work, we should seek help from mental health careproviders who have special understanding of how the feelings of careproviders and patients work. They can explain what is otherwise hard to discern, or even imagine. For example, we may learn that some patients may act in a demeaning way because they need their careproviders to reject them.[29]

Just knowing this may greatly alleviate the intensity of our negative feelings. We may need to know of such possibilities to be able to believe that patients' abusive behavior isn't willful and/or that the underlying fault is actually theirs. An even more difficult emotional task may be to deal with negative feelings that we "can't" feel. It may be highly destructive when careproviders can't identify and manage their negative feelings.

Careproviders may not be able to experience these negative feelings consciously for many reasons. Chief among these may be that we think we shouldn't have them. Whatever the cause, we should try to infer the presence of these feelings from our own behavior. For instance, we should suspect we have negative feelings about a patient if we find that, in interacting with him or her, we atypically distance ourselves. Distancing may be more harmful than anger.[30] We may have negative feelings that come not from within ourselves, but from our professional culture. We may view patients like Lorraine, for example, with therapeutic nihilism. Many members of society and many careproviders are likely to believe that patients like Lorraine act only — or primarily — willfully, and then believe that the patients "deserve whatever they get."[31]

A further difficulty is one I have already mentioned: we may encounter problems with our colleagues when we try to help patients by taking exceptional measures, such as those

described above. There is an entrenched bias against helping difficult patients: the profession's cultural view that such patients *can't be helped*. It is unclear why some of us hold such pernicious views when, in so many other instances, we are able to transcend society's views. The proclivity of some patients to "act out" against their careproviders is well known.[32] This is generally understood to be an attempt by patients to have some sense of power. The particularly pernicious characteristic attributed to patients like Lorraine is that when they "act out" against staff, they do so willfully. What is increasingly clear over recent decades, but still has not yet been widely accepted, is that persons can lack the capacities that logic would suggest they have. The brain studies cited above exemplify this data.

This situation can be compounded when we erroneously think that some responses, such as setting very strict limits, will make patients better, when in fact they make patients worse.[33] What patients need most is now clear: it is not a show of power, which will only make them feel more deficient and alone; what they need is greater understanding and flexibility, which can enable them to feel and remain more in control. As Zanarini and Silk state, "After all, most of us learn more effectively sitting in a comfortable chair than trapped in a walk-in refrigerator."[34]

We may imagine that we can easily discard views such as therapeutic nihilism. But the beliefs that we acquire from our culture, whether from society or from the profession of medicine, may be much stronger than most of us imagine. We may have negative feelings that are outside of our awareness that can harm patients; we may have cultural beliefs, like therapeutic nihilism, that are outside our awareness that can harm patients. The best chance we have to recognize these hidden cultural biases may be to not only look at our own behavior for clues, but to listen carefully to what others say about such patients. If others say that they believe that trying to treat such patients is futile, we are all at risk of believing this to some extent, as well.

CONCLUSION

The key aspect in reaching patients such as Lorraine is to become their ally. This is less likely to occur when we act as negotiators who are trying to facilitate compromise solutions. The potential gains of the approaches that have been suggested here are profound, and their risks are slight.

The ways in which we should respond can be expressed by two stories. The first is well known; it involves Helen Keller and Ann Sullivan. Although Keller initially threw violent tantrums, Sullivan remained wholly loving toward her. This was also the genius of Aichhorn, as mentioned above, and both succeeded.[35] Once Keller "softened" to Sullivan's assistance, Sullivan found a way to give Keller what she needed: she helped her to learn to talk.[36]

When patients' situations are more dire, it won't suffice for us to take a "slower route" of showing total commitment and unconditional love. Immediate engagement is necessary. In this regard I think of a story I heard from a man who is probably the most skilled person in working with criminals I ever met, a psychologist named Joel Dvoskin. He was the one who taught the prisoner how to control his anger so he could gain early parole.

This is a tragic case with a tragic outcome. A man with emotional problems barricaded himself within his mobile home when police came to talk with him. His neighbors had complained to the police that he was playing music too loudly. He killed the two policemen at his door with a shotgun. He was sentenced to death, but died before he was executed.[37] The man had seen his psychiatrist the day before the shootings, and had had a "good session," which suggests that, at that time at least, he had been able to meaningfully relate to another person. I asked Dvoskin what he would have done if he had been at the man's mobile home before the shooting started. I said that I would have thought about calling the man's psychiatrist, who might, as an ally, have "gotten through" to him. Dvoskin responded with

what I can only describe as twinkle in his eye: "I might have thrown different colors of Jello at him, and kept throwing until one color happened to stick."

NOTES

1. It is estimated that at least 2 percent of persons between the ages of 19 and 55 in the U.S. have a borderline personality disorder. M. Swartz et al., "Estimating the Prevalence of Borderline Personality Disorder in the Community," *Journal of Personality Disorders* 4 (1990): 257-73.

2. M.C. Zanarini and K.R. Silk, "The Difficult-to-Treat Patient with Borderline Personality Disorder," in *The Difficult-to-Treat Psychiatric Patient,* ed. M.J. Dewan and R.W. Pies (Washington, D.C.: American Psychiatric Association, 2001), 179-208, p. 181.

3. M.A. Leibovich, "Difficulties in the Treatment of Patients with Borderline Personality, *The Psychotherapeutic Process* 29, no. 1-4 (1978): 250-3, p. 252.

4. R.J.R. Blair, "The Roles of Orbital Frontal Cortex in the Modulation of Antisocial Behavior," *Brain and Cognition* 55 (2004): 198-208.

5. R.J.R. Blair et al., "Reduced Sensitivity to Others' Fearful Expressions in Psychopathic Individuals," *Personality and Individual Differences* 37 (2004): 1111-22.

6. J.T. Maltsberger, "Treating the Suicidal Patient: Basic Principles," *Annals of the New York Academy of Science* 932 (April 2001): 158-65, 59.

7. D.T. Saposnek, "Aikido: a Model for Brief Strategic Therapy," *Family Process* 19, no. 3 (September 1980): 227-38, p. 233.

8. "If [the careprovider] functions in one or the other role, how can he then shift into the role of helper or ally?" M.H. Hollender and S.P. Hersh, "Impossible Consultation," *Archives of General Psychiatry* 23, no. 4 (October 1970): 343-5, p. 345.

9. J. Modestin, "Counter-Transference Reactions Contributing to Completed Suicide," *British Journal of Medical Psychology* 60, part 4 (December 1987): 379-85, p. 382.

10. A. Altman and M.A. Selzer, "Delusions in Transference: Psychotherapy with the Paranoid Patient," *Psychiatry Clinics of North America* 18, no. 2 (June 1995): 407-25, p. 410.

11. S. Shapiro, "The Provocative Masochistic Patient: An Intersubjective Approach to Treatment," *Bulletin of the Menninger Clinic* 53, no. 4 (July 1989): 319-30, p. 324.

12. Zanarini and Silk, see note 2 above, p. 192.

13. For an overview of Aichhorn's work and its influence today, see J.E. Schowalter, "Aichhorn Revisited," *Psychoanalytic Study of the Child* 55 (2000): 49-60.

14. See, for example, N. Muramatsu and T. Cornwell, "Needs for Physician Housecalls — Views from Health and Social Service Providers," *Home Health Care Service Quarterly* 22, no. 2 (2003): 17-29, and "Doctors' Group Cuts Readmissions by Examining Patients in Homes," *Clinical Resources Management* 1, no. 7 (July 2000): 105-7.

15. J.S. Gans, "Indirect Communication as a Therapeutic Technique: a Novel Use of Countertransference," *American Journal of Psychotherapy* 48, no. 1 (Winter 1994): 120-40, pp. 121-122, 124-125, 131, 134.

16. Ibid., pp. 126-128.

17. Ibid., p. 136.

18. K.M. Dalton et al., "Gaze Fixation and the Neural Circuitry of Face Processing in Autism," *Nature Neuroscience* (6 March 2005), *http://www.nature.com/nature/nature-neuroscience.*

19. H. Hendin et al., "Recognizing and Responding to a Suicide Crisis," *Suicide and Life-Threatening Behavior* 31, no. 2 (Summer 2001): 115-28, p. 125.

20. Such patients may have less reason to oppose others. The "part of their mind" whose "job it is" to be oppositional and above all "maintain control" may now "relax" more. It may allow "another part" that "wants to live" to "re-take the controls."

21. J.R. Maltsberger et al., "A Man Giving Up On Himself — Suicide & Life-Threatening Behavior," *American Association of Suicidology* 33, no. 3 (Fall 2003): 331-7, p. 334.

22. W.E. Milch, "Suicidal Patients' Psychological Attacks on the Therapist," *Bulletin of the Menninger Clinic* 54, no. 3 (Summer 1990): 384-90, p. 389.

23. R.A. Payne, *Relaxation Techniques,* 3rd

ed. (New York: Elsevier, 2003). Other approaches are presented in A.W. Kneier, "Coping with Melanoma — Ten Strategies That Promote Psychological Adjustment," *Surgical Clinics of North America* 83, no. 2 (April 2003): 417-30; L.N. Gruber, "Simple Techniques to Relieve Anxiety," *Journal of Family Practice* 5, no. 4 (October 1977): 641-4.

24. ee R. Lazar, "Presentness: An Intersubjective Dimension of the Therapeutic Act," *American Journal of Psychotherapy* 54, no. 3 (Summer 2000): 340-54.

25. R.G. Poggi and R. Ganzaain, "Countertransference Hate," *Bulletin of the Menninger Clinic* 47, no. 1 (January 1983): 15-35, p. 33.

26. T.G. Gutheil and D. Schetky, "A Date with Death: Management of Time-Based and Contingent Suicidal Intent," *American Journal of Psychiatry* 155 (1998): 1502-7, p. 1506.

27. Payne, see note 23 above, p. 423.

28. Ibid., p. 18.

29. S. Shapiro, "The Provocative Masochistic Patient: An Intersubjective Approach to Treatment," *Bulletin of the Menninger Clinic* 53, no. 4 (July 1989): 319-30, p. 322.

30. Payne, see note 23 above, p. 427.

31. F. Yeomans, "When a Therapist Overindulges a Demanding Borderline Patient," *Hospital and Community Psychiatry* 44, no. 4 (April 1993): 334-6.

32. E. Goffman, *Asylums* (New York: Anchor Books, 1961), 304-5.

33. How those with power mislead themselves in this way is developed by Antonio Gramsci: a key dimension of inequality for Gramsci is the inability of subaltern people to produce coherent accounts of the world they live in that have the potential to challenge the hegemonic account. K. Crehan, *Gramsci, Culture and Anthropology* (Berkeley, Calif.: University of California Press, 2002), 209.

34. Zanarini and Silk, see note 2, p. 189.

35. See H. Keller (and A. Sullivan), *The Story of My Life* (Garden City, N.Y.: Doubleday, 1954): Anne Sullivan: "My heart is singing for joy this morning. A miracle has happened. . . . This wild little creature of two weeks ago has been transformed into a gentle child," p. 252.

36. Helen Keller: "Sometimes I stood between two persons who were conversing. I moved my lips and gesticulated frantically without result. This made me so angry at times that I kicked and screamed until I was exhausted." Ibid., 27-8.

37. Personal conversation with Joel A. Dvoskin at a conference, "Mental Health Disability and the Law," 10 September 2004, Baltimore, Md. The person in the trailer was Francis Zito. "Zito Sentenced to Death," Associated Press, 30 May 2002, *http://mdcops.org/news/zito_sentenced_to_death.htm,* accessed 1 May 2005.

Features

Physicians' Legal Defensiveness in End-of-Life Treatment Decisions: Comparing Attitudes and Knowledge in States with Different Laws

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Despite years of discussion, research, and legislative and judicial activity, uncertainties and concerns persist in the medical community about legal implications when physicians and families consider decisions to terminate life-support for terminally ill patients. A study previously conducted in Texas by two of us (SVM and JWS) found that about 25 percent of surveyed physicians who regularly treat critically ill patients adopt a posture of extreme legal defensiveness about decision making for dying patients, and that the most defensive physicians are less knowledgeable about the laws that are relevant to such decisions than their less-defensive counterparts.[1] The same study also found evidence suggesting that legal defensiveness creates conflict between physicians and terminally ill patients or their family members, resulting in an adversarial relationship that is counterproductive and counter-therapeutic.[2] Moreover, legal defensiveness may significantly influence physicians' medical assessments of cases in ways that serve to foreclose the rights of patients and their family members to make their own healthcare decisions at the end of life.

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We have defined legal defensiveness as constituting more than "defensive medicine" as it is commonly understood, that is, providing diagnostic and treatment interventions largely for the purpose of avoiding possible legal claims. More broadly, our definition of "legal defensiveness" refers to physicians' general level of concern about the implications of both civil and criminal law with respect to their treatment decisions for terminally ill patients. Such defensiveness may be a detrimental factor in end-of-life decision making if it interferes with medical judgment that is based on respect for patients' autonomy and promotion of the patient's best interests.

For this comparative study conducted in New York State, we decided to replicate major portions of the Texas survey to generalize our findings and ascertain how the distinctive New York legal landscape might color physicians' analysis of decision making, perceptions of legal risk, and knowledge of relevant law regarding end-of-life treatment. New York physicians must contend with what is arguably the strictest judicial interpretation in the U.S. of the conditions under which life-sustaining interventions may be terminated, in the absence of a legally appointed surrogate decision maker. In the 1988 *O'Connor* decision, which we will discuss in greater detail below, the New York Court of Appeals established a standard of "clear and convincing" evidence, with a restriction that substantive evidence for decisions about life-sustaining treatment must be determined only by subjective criteria — that is, only explicit, precise, and firm statements from the patient himself or herself are legally acceptable for abatement of treatment.[3] Further, except for persons previously determined to be mentally disabled (for whom a best interests standard is allowed),[4] New York statutory law lacks important provisions that are found in the laws of most other states, including Texas. The Texas statute on advance directives contains detailed procedures to be employed when an incapacitated person has not executed a directive. These include: first, a familial hierarchy for identifying a surrogate decision maker who can then, with an attending physician, use a substituted judgment standard; and, second, if such family members are not available, a process to designate independent physicians or ethics committee members to participate in the decision process.[5] Because the statute does not specify a standard for the latter, and persons having knowledge sufficient for substituted judgment would not be available, it is reasonable to assume that a best interests standard would be applied to such cases in Texas. In contrast, New York law does not permit the use of a substituted judgment or best interests standard.

In this study, we hypothesized that New York physicians would be more legally defensive in general than their Texas counterparts, due to the unusual legal environment in New York, but that better knowledge of the law would have the same moderating effect on reported defensiveness in New York as it did in Texas. The data partially confirm and partially refute this hypothesis.

METHODS

BACKGROUND AND SAMPLE

The New York sample of physicians was selected from two tertiary care medical centers within the State University of New York (SUNY) system: Stony Brook Health Science Center, which includes Stony Brook University Hospital, and SUNY Upstate Medical University in Syracuse. The study populations were obtained from lists of all faculty and house staff in the Departments of Internal Medicine and Surgery (including a separate Department of Neurosurgery at Stony Brook). First-year residents were excluded to maintain consistency with the Texas sample, and because we anticipated they would lack sufficient experience to comment in a meaningful manner on the items of interest.

RESEARCH INSTRUMENT AND LEGAL DEFENSIVENESS SCALE

The New York questionnaire was a 37-item structured instrument with fixed-response categories. This instrument was a shortened

version of the 67-item instrument used in the Texas study, containing the most salient questions from the previous research, plus a quiz on New York law (described below). The study protocol was approved by the Stony Brook University Health Science Center and SUNY Upstate Institutional Review Boards. Potential respondents were informed that the purpose of the study was to assess external factors that affected treatment decisions for dying patients, including perceptions of legal risk. For the Texas study, we constructed a six-item scale to quantify the concept of physicians' legal defensiveness in their decisions to treat (or not to treat) terminally ill patients. For use in New York, based on factor analysis and face-validity considerations, we shortened the original Texas legal defensiveness scale (LD6) into a five-item scale (LD5), which is shown in figure 1. Four of these items comprise a subset of the LD6 scale: items 1 and 2 asked how often the physicians worried about (1) being sued for malpractice and (2) being criminally prosecuted regarding decisions to withdraw lifesupport. Items 4 and 5 assessed physicians' concerns about these issues — civil liability and criminal prosecution — in the context of a case vignette with legally relevant features. The revised scale included one additional item, a question that appeared in both surveys but was not part of the original LD6 scale (item 3 in figure 1): "How often do you perceive that the law requires you to perform medical procedures on terminally ill patients when you think that such procedures are medically inappropriate?" This question broadens our measure to include perceived conflict between medical and legal imperatives. The scale was re-coded to be equivalent in both site samples. Factor analysis showed this variable loaded heavily on a single factor with the other four items.

Just as in the Texas study, we incorporated a 10-item quiz into the larger instrument to assess physicians' knowledge of state laws regarding life-sustaining treatment and decision making for critically ill patients. Our intent was to assess factual knowledge about concepts derived directly from New York statutes or case law. Areas of law covered by the quiz included healthcare agents and proxies,[6] artificial hydration and nutrition, refusal of treatment, suicide and assisted suicide, statutory immunity from civil actions, and do-not-resuscitate (DNR) orders.[7] Examples of actual questions are shown in figure 2. The test of

Items to measure New York physicians' legal defensiveness in decisions to abate treatment for terminally ill patients (LD5)

1. When you consider a decision to withdraw life-sustaining treatment from a terminally ill patient, how often do you worry about being sued for malpractice?

Never	Sometimes	Often	Very often	Always
1	2	3	4	5

2. When you consider a decision to withdraw life-sustaining treatment from a terminally ill patient, how often do you worry about being criminally prosecuted? *[Response options same as for query 1.]*

3. How often do you perceive that the law requires you to perform medical procedures on terminally ill patients when you think that such procedures are medically inappropriate? *[Response options same as for query 1.]*

[This brief case vignette is followed by a series of questions.] A family member requests that you withdraw life-sustaining treatment from a terminally ill, mentally incompetent patient. While competent, this patient repeatedly expressed a wish that his life not be prolonged. *[Respondent is asked to rate the importance of several factors in considering whether to refuse the family members' request in the above case. Only the two factors listed below were included in the scale.]*

4. Possibility of malpractice suit

Not important	Somewhat important	Very important	Most important
1	2	3	4

5. Possibility of criminal prosecution *[Response options same as for query 4.]*

Figure 1.

legal knowledge in New York contained four general questions that were essentially identical in content to those in the Texas study, and six questions specific to New York laws on healthcare agents and proxies and DNR orders. We specifically designed question 2 in figure 2 to test New York physicians' knowledge of how the *O'Connor* case affects decisions about artificial hydration and nutrition. We constructed a "law score" based on the answers to the 10 questions, with each question answered correctly counting 10 points (for a total possible law score of 100). As in the Texas study, we designated a score of 70 or higher on the law quiz to be a "passing" score. In contrast to Texas, where all physicians are required to pass a medical jurisprudence examination before medical licensure, New York has no such requirement, which may limit predictability of any standardized knowledge of law among New York physicians. Additional detailed information regarding our research instrument and methodology has been published elsewhere.[8]

DISTRIBUTION AND RESPONSE

In the fall of 2002, we distributed 426 questionnaires at the respective schools (284 at Stony Brook and 142 at Upstate). By 30 January 2003, after two follow-up distributions, 180 completed questionnaires had been received, yielding an overall response rate of 42.3 percent: 117 physicians from Stony Brook (41.2 percent response) and 63 from Upstate (44.4 percent response). The overall response for the New York survey compares favorably to the previous response rate in Texas (40.7 percent). It is possible that New York respondents differed from nonrespondents on variables such as specialty, training, and experience, but the sample we obtained was of adequate size and diversity both to allow assessment of the statistical effect of such variables on the items of interest in New York — legal defensiveness and knowledge of medical law — and to permit statistical comparison with the Texas data. Our moderate response rate for New York is sufficient to allow an exploratory study of these items and is comparable to rates from some other such exploratory studies, but we cannot confidently generalize our results to all New York physicians. Other limitations of our study are: (1) it is a cross-sectional survey, so we cannot establish causal ordering between variables that are associated; and (2) the two studies were separated by a decade, and we cannot with certainty attribute changes to regional differences that might also be influenced by historical patterns.

Both women and non-White minority physicians were represented in higher proportions in the New York sample than in the Texas

Sample questions from the research instrument to measure New York physicians' knowledge of medical law

1. The New York Health Care Proxy law applies only to patients who are terminally ill. True False
2. A Health Care Agent's decision to refuse artificial nutrition and hydration for a patient may be honored only if the patient has indicated that he or she wishes to decline artificial nutrition and hydration. True False
3. A Health Care Proxy in New York is valid:
    1. For six months
    2. Until revoked in writing
    3. For one year
    4. Until revoked
4. Health care professionals are immune from civil liability and criminal prosecution for acts performed in good faith that comply with the New York statutes on Orders Not to Resuscitate and Health Care Agents and Proxies. True False

Answers: 1 = false; 2 = true; 3 = 4. Until revoked; 4 = true.

Figure 2.

sample. Of the New York participants, 33 percent were female and 29 percent were non-White, while 17 percent of the Texas participants were female and 13 percent were non-White. Regarding specialty, internists were the largest group in both samples, but the proportion of oncologists was higher in Texas than New York (New York = 8 percent; Texas = 26 percent), because one of the three Texas study sites was a dedicated cancer center. However, the proportion of surgeons was almost identical in both samples (New York = 21 percent; Texas = 22 percent). The average age of the respondents was comparable in both samples (41 years in New York, 40 in Texas) as was the average years of clinical experience (15 years in the New York sample, 14 years in Texas). The samples were also comparable in terms of the number of terminally ill patients treated during the preceding year, although slightly more of the physicians in Texas reported seeing at least 50 terminally ill patients per year (25 percent in the Texas sample versus 18 percent in the New York sample).

METHODS OF ANALYSIS

Frequency analysis, measures of central tendency, and dispersion were used to describe the performance of our sample of physicians on the test of medical law and on the indicator of legal defensiveness in their approach to terminally ill patients. Similarly, comparisons were made between subgroups of physicians as defined by demographic attributes, clinical experience, and medical specialty. *Chi*-square statistics and associated probabilities were calculated to assess the significance of categorical differences between subgroups of physicians and to determine whether such differences could reasonably have occurred by chance alone. Finally, we developed a multivariate regression model of the legal defensiveness among physicians in both states. We used multiple regression analysis to examine the net effect of knowledge of medical law on legal defensiveness, controlling for practice state (New York versus Texas), years of clinical experience, and medical specialty. We also tested the interaction effect of state by legal knowledge, that is, the extent to which legal knowledge affected legal defensiveness differently among physicians in New York versus physicians in Texas. In linear multiple regression analysis, the regression coefficient *b* expresses the net amount of change (either positive or negative) in the dependent variable *Y* associated with a unit of change in a given independent variable X_1, controlling for any shared association with other independent variables in the model (X_2, X_3, and so forth). A two-way interaction effect expresses the additional change in *Y* associated with the combination, or product, of two independent variables, that is, X_1 times X_2, controlling for the main effect of X_1 and X_2. The multiple regression model yields an overall coefficient R^2, which expresses the total proportion of variation in the dependent variable that is accounted for, or "explained" by, the predictor variables in the model.

RESULTS

KNOWLEDGE OF MEDICAL LAW

How well did physicians in New York know the laws relevant to treatment of terminally ill patients and how did they compare to physicians in Texas? New York physicians performed somewhat better than Texas respondents on the 10-item law quiz (passing rates: New York = 30.68 percent; Texas = 22.92 percent; approached significance at $p < 0.10$). However, there was no significant difference between the New York and Texas samples on the four quiz items that were identical in both jurisdictions (New York = 28.98 percent correct; Texas = 26.91 percent correct). Also, the mean law scores did not significantly differ between the two samples (New York mean = 55.6, standard deviation — sd = 17.1; Texas mean = 53.9, sd = 14.9).

Further, New York physicians' performance on the law quiz was not associated with their source of legal information. This differs from the results of the Texas study, in which physicians who obtained at least some of their knowledge about medical law from other physicians were significantly less likely to pass

the law quiz than physicians who got none of their legal information from other doctors (p = 0.006). Regarding performance on the law quiz by physicians with various medical specialties in New York, physicians practicing internal medicine were significantly more likely than surgeons to obtain a passing score (p = 0.0014); this finding was consistent with the Texas data.

LEGAL DEFENSIVENESS

How did New York physicians compare to Texas doctors in terms of general legal defensiveness? The comparative distributions of legal defensiveness scores in New York and Texas are shown in figure 3. Generally, there was no difference by state in levels of legal defensiveness without controlling for knowledge of medical law. When knowledge of the law is taken into account, however, the results indicate dramatic differences. In aggregate, the New York data show an inverse relationship to those from the Texas study. That is to say, in Texas, passing the law quiz was associated with *lower* legal defensiveness, while in New York passing the law quiz was associated with *higher* legal defensiveness (p < 0.01). This pattern of responses (shown in figure 4) appears generally consistent with the fact that New York physicians face potentially higher legal risk than their Texas counterparts due to greater conflict between legal and medical-ethical criteria for abating treatment. Recall that New York law has neither adopted a "best interests" standard for most persons, nor recognized a hierarchy of persons who are authorized to use substituted judgment for patients with no appointed healthcare agent. The *O'Connor* decision makes it difficult for physicians to apply their best clinical judgment without concern for legal jeopardy in such cases. Thus, it may be rational for physicians in New York who are the most knowledgeable about the actual dilemmas and risks engendered by the law to be more defensive.

To further examine the specific impact of New York physicians' knowledge of the *O'Connor* decision on their degree of legal defensiveness, we isolated a group of respondents, *post hoc,* who both passed the law quiz, and correctly answered the *O'Connor* question (item 2 in figure 2). As shown in figure 5, this subgroup of respondents, the "legal experts," were found to have the highest degree of legal defensiveness, and, indeed, to account entirely for the contrasting pattern of association between legal knowledge and defensiveness in New York compared to Texas. When the "legal experts" are removed from the analysis, the association between knowledge and defensiveness in the remainder of the New

Distribution of legal defensiveness scores in Texas and New York physician surveys

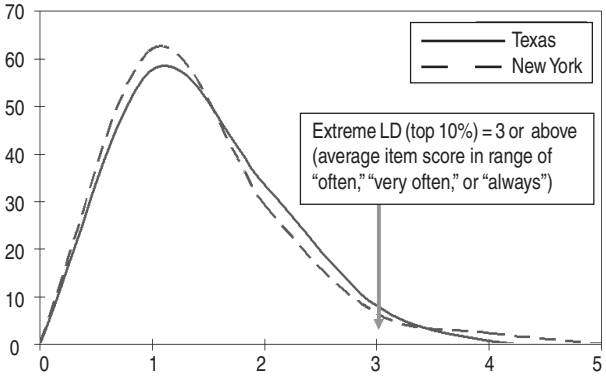


Figure 3

Physicians' legal defensiveness by knowledge of medical law regarding end-of-life treatment in two states

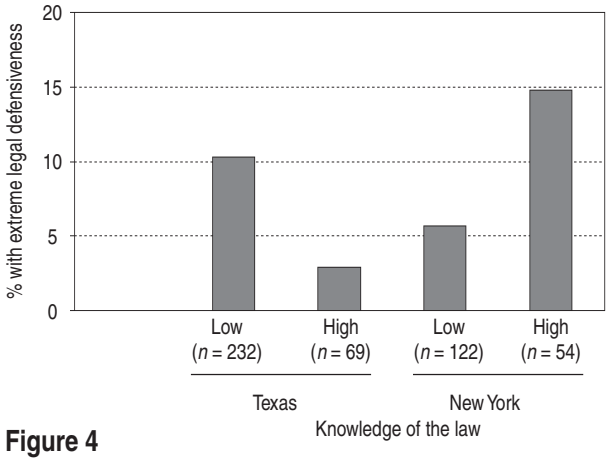
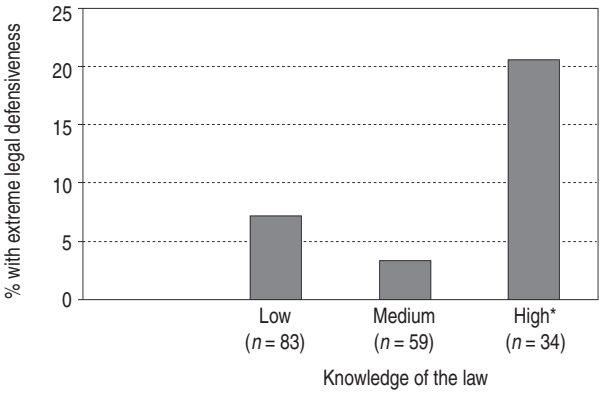


Figure 4

York sample is consistent with that found in the Texas sample: that is, as knowledge of law increases, legal defensiveness decreases. That is to say, the significant overall interaction effect between state and legal knowledge on legal defensiveness is completely attributable to a subgroup of respondents who possessed accurate knowledge of the law including the specific implications of the *O'Connor* case.

Table 1 presents a multiple linear regression analysis estimating the effects of practice state (New York versus Texas) and legal knowledge (law score) on legal defensiveness, controlling for years of experience and practice specialty. Model 1 shows that practice state and law score are not significant as main effects; however, Model 2 shows a significant interaction effect between practice state and law score on legal defensiveness. In the interaction model, there is also a significant main effect for practice state (New York physicians are more defensive) and law score (greater legal knowledge generally lowers defensiveness, except among New York physicians with the most knowledge, who are actually the *most* defensive). These effects are significant, controlling for years of experience and practice specialty.

New York physicians' legal defensiveness by knowledge of medical law regarding end-of-life treatment



*Score of 70% or higher and correct answer to *O'Connor* question (item 2 in figure 2).

Figure 5

Attitudes of extreme legal defensiveness, measured in terms of demographic and other physician characteristics, are presented in table 2. In addition to the associations between defensiveness and legal knowledge discussed above, the findings suggest that New York physicians who have 20 or more years of clinical experience are significantly less likely to report extreme legal defensiveness than less-experienced physicians ($p < 0.05$). This corresponds with the findings in the Texas study.

RESPONSES TO CASE VIGNETTES

An additional difference between New York and Texas physicians is illustrated by responses to a brief case vignette used in both studies: "A family member requests that you withdraw life-sustaining treatment from a terminally ill, mentally incompetent patient. While competent, this patient repeatedly expressed a wish that his life not be prolonged. Are there circumstances in which you would consider refusing this family's request that you withdraw treatment?" (See figure 1.) A total of 51 New York respondents answered "yes" (30.2 percent, compared to 45.2 percent in Texas; $p < 0.05$). When asked to rate the factors (other than hospital policy) that would influence their decision in this case, New York physicians selected "Disagreement among family members" as a "Very important" or "Most important" factor, significantly more often than did Texas physicians (61.67 percent versus 44.15 percent; $p < 0.001$). In contrast, Texas physicians selected "Disagreement between family and physician" as a "Very important" or "Most important" factor significantly more often than their New York counterparts did (67.56 percent versus 57.32 percent; $p < 0.05$).

DISCUSSION

Our results bring into stark relief the potential detrimental effects of the *O'Connor* decision on clinical practice in New York. In general, New York and Texas physicians report similar attitudes and practices when considering how state laws affect end-of-life de-

cisions for incompetent patients. Namely, as general knowledge of relevant law increases, concern about the general legal impact of end-of-life decisions decreases. Yet, simultaneously, our finding about the reported perceptions of the physicians we call "legal experts" suggests that the subgroup of New York physicians who are aware of the specific legal restrictions imposed by the *O'Connor* case react to life-support decisions in a much more defensive way than other New York physicians. This finding has crucial implications for the care of patients who lack both capacity and an advance directive.

IMPLICATIONS FOR PHYSICIANS OF THE NEW YORK EVIDENTIARY STANDARD

Through the process of informed consent, competent patients have the right to refuse treatment, including life-sustaining treatment when seriously ill and dying, and healthcare professionals and families have correlative obligations to respect the treatment refusals. National consensus about decisions near the end of life extends this same principle to decision making for incompetent patients, recognizing that all competent adults may write advance directives for healthcare (proxy directives, living wills, or a combination of the two) and that, in the absence of a written document, family members (and sometimes close friends) should be entrusted and empowered to make healthcare decisions, in keeping with the patient's wishes and best interests.

Governing law in the vast majority of states, including Texas, is highly deferential to families who must bear the burdens of end-of-life decisions. Typically, a family member may authorize forgoing of life support for a terminally ill or permanently unconscious loved one based upon personal knowledge of the patient's wishes and values and in accordance with the patient's best interests. Numerous cases have carved out this decisional authority for families by holding that there must be a "preponderance of evidence" of the patient's wish to refuse life support, or "some trustworthy evidence," or simply that families may exercise "substituted judgment" without specific reference to an evidentiary standard. Statutes in more than half of states, often referred to as "consent statutes" or "surrogate decision making laws," give further voice to family authority.[9] While some states carve out special rules for forgoing artificial fluids and nutrition, such as requiring specific evidence of the patient's refusal of this form of medical treatment, the overarching intent of this body of law is to establish a "zone" of private decision making for families and physicians that recognizes the proper role of families in making end-of-life decisions.

However, in a handful of states — most notably New York — law departs from this national consensus. A number of cases adopt the more demanding standard that there must be "clear and convincing evidence" of the patient's refusal of life support. This standard

TABLE 1 Multivariable regression analysis of effects on legal defensiveness

	Model 1: main effects				Model 2: interaction effect			
	df	*b*	se	*t*	df	*b*	se	*t*
New York compared to Texas	1	-0.12	0.07	-1.78	1	-0.06	0.23	-2.59**
Law score (number correct)	1	0.00	0.00	0.20	1	-0.01	0.01	-1.98*
New York *x* law score interaction	--	--	--	--	1	0.01	0.00	2.17*
	Adjusted R^2 = 0.04				Adjusted R^2 = 0.05			

The models are controlled for years of experience and by medical specialty.
Statistical significance: *$p < 0.05$; **$p < 0.01$.

was first adopted by the New York Court of Appeals in *Matter of Storar,*[10] (1981) and later re-affirmed in *O'Connor* (1988).[11] In *O'Connor,* the 77-year-old patient suffered multi-infarct dementia following a series of strokes and was unable to swallow food or water. She had substantial cognitive impairment and clearly lacked decisional capacity, although her consciousness waxed and waned, ranging from being completely unresponsive to being reasonably alert and sometimes able to follow simple commands. Her condition was considered irreversible with no hope of substantial neurologic recovery. When physicians sought permission to insert a nasogastric (NG) feeding tube, Mrs. O'Connor's daughters, acting as her surrogates, but not formally appointed as healthcare agent, refused to consent. The hospital sought a court order to insert the NG tube. At trial, her two daughters testified that, while competent, their mother had repeatedly stated her wish to refuse medical treatment, saying for example that it was "monstrous" to keep someone alive by using "machinery and things like that" when they are "not going to get better."[12] Reversing the two opinions below, in which courts had refused the hospital's request, the New York Court of Appeals found that the evidence of the patient's wishes was not clear and convincing, and ordered insertion of the NG tube. Reaffirming the standard it had adopted seven years earlier in *Storar,* the *O'Connor* court embraced a particularly narrow subjective intent interpretation of the clear and convincing evidence standard, stating that "nothing less than unequivocal proof will suffice when the decision to terminate life support is at issue."[13] Offering further definition of the standard, the opinion also states that there must be proof

TABLE 2 New York physicians' characteristics related to attitudes of extreme legal defensiveness in treating terminally ill patients

Independent variable	*n*	% of group with extreme LD5 score	Statistical significance
Gender			
Male	116	8.62	--
Female	58	8.33	n.s.
Racial/ethnic group status			
White non-Hispanic	118	7.63	--
Non-White minority	58	10.34	n.s.
Specialty			
Internal medicine	114	8.05	--
Surgery	35	0.00	--
Oncology	13	7.69	--
Critical Care	12	0.00	n.s.
Clinical experience			
1 - 4 years	39	15.38	--
5 - 9 years	26	11.54	--
10 - 19 years	43	6.98	--
20 or more years	65	4.62	$p < 0.05$
Number of terminally ill patients treated last year			
Less than 50 patients	144	9.03	--
50 or more patients	32	6.25	n.s.
Objective knowledge of medical law			
Failing score (< 70%)	122	5.74	--
Passing score (70% +)	54	14.81	$p < 0.05$

"that the patient held a firm and settled commitment to the termination of life supports under the circumstances like those presented."[14] Under such a standard, if the patient is unable to predict with a high degree of accuracy her future medical circumstances, then the law dictates that her general wishes be ignored.

New York's strict standard is permissible, under the U.S. Supreme Court's *Cruzan*[15] decision, which upheld state authority to establish reasonable guidelines for end-of-life decisions, but which has been severely criticized by legal scholars. In fact, New York courts have not uniformly embraced the *O'Connor* rule. Some lower court decisions have authorized forgoing of life support for incompetent patients based on evidence of the patient's prior expressions and values, which plainly does not meet the *O'Connor* test.[16] And other law rejects the idea that life support may only be terminated based on the patient's own, competently expressed wishes and values. For example, New York law recognizes parental authority to direct termination of life support for young children,[17] and establishes a guardianship process for decisions to forgo life support for terminally ill, mentally disabled patients.[18] Still, this body of law does not disturb the decision of the state's highest court. A recently published *Legal Manual for Physicians,*[19] jointly authored by the state's Bar Association and Medical Society, recognizes that *O'Connor* is the governing law for incompetent patients without healthcare proxies (unless the decision concerns a DNR order, in which case New York's DNR statute controls). The *Manual* goes on to state the prevailing view of the meaning of this rule at the bedside of dying patients: "When there is no clear and convincing evidence of the patient's wishes and no healthcare agent, nobody may authorize the withdrawal or withholding of life-sustaining treatment — not the family, not the physician or hospital, not even the court or a court-appointed guardian (except for guardians of mentally retarded persons . . .)." This rigid rule likely explains the finding of our study that New York physicians who are aware of how the *O'Connor* case affects end-of-life decisions are disproportionately likely to practice defensively.

Many New York physicians are also aware that more than a decade has passed since the New York State Task Force on Life and the Law issued a report and proposed legislation addressing issues of surrogate decision making for patients lacking capacity, and that no action has been taken on it by the state legislature.[20] The Task Force was a 25-member, multi-disciplinary group charged with developing recommendations for responsible public policies on a host of issues generated by emerging medical technologies. Among its many recommendations, the Task Force advocated the legislative establishment of a process for surrogate decision making on behalf of patients lacking capacity, which would supersede the *O'Connor* rule.[21] The Task Force's model legislation became the foundation of the Family Healthcare Decisions Act, which has been filed as a bill in every session of the New York State Assembly since 1992, but has never passed.

IMPACT OF NEW YORK LAW ON PATIENTS AND FAMILIES

Our findings support a widely held view that New York's standard of clear and convincing evidence is a substantial barrier to family decision making and compassionate care for dying patients who lack capacity and have not selected a healthcare agent.[22] Taken literally, the *O'Connor* court's pure subjective standard for substantive evidence means that abatement of treatment (other than a DNR order) is only permitted for incompetent persons who do not have healthcare proxies in cases when decision makers can be virtually certain of the wishes of the patient in the same medical circumstance in which the patient finds him- or herself. The result is that abatement will rarely be possible within the law. Anecdotal reports suggest that one unintended consequence of this vexing legal situation is that, in some healthcare facilities in various parts of New York state, families of dying patients who have

not formally appointed a healthcare agent possibly are sometimes allowed to make decisions to abate treatment without meeting the standard set forth in *O'Connor,* a practice known colloquially as "flying under the radar."[23] It is unclear from these reports whether healthcare professionals in such cases are aware that their actions may not be in compliance with New York law. If they are aware, circumventing the law suggests physicians' commitment to the ethical imperative to provide compassionate care for the seriously ill, despite concern regarding potentially increased risk of legal liability for such decisions in light of *O'Connor.* Our finding that New York physicians were more concerned with possible family discord than with disagreement between families and physicians also illuminates this point. Family agreement reduces the risk of legal entanglements, and thus will be central to the effective implementation of such a practice. In our collective experience as physicians and as ethics consultants, New York physicians (who are often unsupported by the law in withdrawal of treatment) are acutely sensitive to the possibility a dissenting family member could publicly disclose such practices or file a lawsuit.

Because a literal interpretation of the *O'Connor* decision essentially requires clairvoyance on the part of patients for whom abatement of treatment is sought in the absence of a healthcare proxy (in terms of predicting in advance their precise medical condition and treatment preferences in that event), this legal standard imposes a heavy burden on families who seek to assist their loved ones, and may exact a heavy toll on patients in terms of unnecessary suffering. Especially relevant here is the continuing low percentage of persons in the U.S. who have executed advance directives — currently estimated at about 15 to 20 percent of the general population.[24] There are scant data regarding the completion of healthcare proxies in New York. One study at several Manhattan senior citizen centers found that 35 percent of participants reported that they had completed a healthcare proxy document.[25] While this may suggest that more New Yorkers avail themselves of this legal mechanism than in some other parts of the country, a majority remain at risk for confronting an *O'Connor*-type situation at the bedside or in court. As Joseph Fins and Ralph Nachman have noted, commenting on the *O'Connor* case and the New York legislature's ongoing failure since 1992 to pass a statute that would enable surrogate decision making in the absence of a healthcare proxy, "The inadequacy of our state's end-of-life decision-making laws will not be apparent to most New Yorkers until it is too late."[26] These authors also note that because most New Yorkers have not executed a proxy or left other clear evidence of their wishes (such as a living will that clearly contemplates the patient's current situation), "their family members will be powerless to withdraw burdensome interventions and their physicians will be obliged to provide them."[27]

CONCLUSION

The results of this comparative study, while demonstrating the negative effects of the *O'Connor* decision, are encouraging on one point. In general, as physicians in both New York and Texas acquire additional years of clinical experience in assisting families with treatment decisions, they report being less defensive in their perceptions and practices. Further, if New York modifies its laws through legislation to bring them closer to the national mainstream, it is reasonable to assume that legal education for physicians (providing updates for them about these specific changes) may, over time, reduce legal defensiveness and defensive practice in New York, as was the case in our study of physicians in Texas. Nonetheless, our study suggests that the combined effects of the *O'Connor* decision and the New York legislature's inaction currently have a significant effect on some New York physicians' analysis of treatment decisions in ways that are discordant with national trends and likely narrow the perceived range of treatment options. In our view, such a situation has potential for serious harmful consequences for

the well-being of both critically ill patients and their families, and should provide an impetus for persons seeking to bring an end to the decade-long period of legislative inaction on this important issue. If, in the future, the New York legislature acts to change the law on this topic, such revisions will be a matter worthy of further research.

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NOTES

1. S.V. McCrary et al., "Treatment Decisions for Terminally Ill Patients: Physicians' Legal Defensiveness and Knowledge of Medical Law," *Law, Medicine and Healthcare* 20, no. 4 (Winter 1992): 364-76.

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3. *In re Westchester County Medical Center ex rel. O'Connor,* 72 N.Y.2d 517, 534 N.Y.S.2d 886, 531 N.E.2d 607 (1988).

4. N.Y. Mental Hyg. Law §§ 80.01-80.13 (Consol. 2004).

5. Tex. Health & Safety Code Ann. § 166.039 (West 2004).

6. N.Y. Pub. Health Law §§ 2980-2994 (Consol. 2003).

7. N.Y. Pub. Health Law §§ 2960-2979 (Consol. 2003).

8. McCrary et al., see note 1 above, 365-8; Swanson and McCrary, see note 2 above, 126-9.

9. See generally A. Meisel and K.L. Cerminara, *The Right to Die: The Law of End-of-Life Decisionmaking,* 3rd ed. (New York: Aspen Publishers, 2004).

10. *In re Storar,* 52 N.Y.2d 363, 438 N.Y.S.2d 266 (1981).

11. *O'Connor,* see note 3 above.

12. Ibid., 611.

13. Ibid., 612.

14. Ibid., 613.

15. D. Gindes, "Judicial Postponement of Death Recognition: The Tragic Case of Mary O'Connor," *American Journal of Law and Medicine* 15, no. 2-3 (1989): 301-31; G.J. Annas, "Precatory Prediction and Mindless Mimicry: The Case of Mary O'Connor," *Hastings Center Report* 18, no. 6 (December 1988): 31-3; *Cruzan v. Director, Missouri Department of Health,* 497 U.S. 261 (1990).

16. *See, e.g., In re Christopher,* 177 Misc.2d 352, 675 N.Y.S.2d 807 (S.Ct., Queens Cty., 1988); *In re Beth Israel Medical Center,* 136 Misc.2d 931, 519 N.Y.S.2d 511 (S.Ct., N.Y. Cty., 1987) (decided in light of *Storar*).

17. *In re A.B.,* 196 Misc.2d 940, 768 N.Y.S.2d 256 (Sup. Ct., 2003).

18. See note 4 above.

19. New York State Bar Association and Medical Society of the State of New York, *Legal Manual for New York Physicians* (New York: New York State Bar Association, 2003), 337.

20. New York State Task Force on Life and the Law, *When Others Must Choose: Deciding for Patients Without Capacity* (Albany, N.Y.: New York State Task Force: March 1992).

21. Ibid., 170-1 and 247-68.

22. J.J. Fins and R.L. Nachman, "Needs of Dying Should be Freed of Red Tape, *Albany Times Union* (4 April 2004): C1; Gindes, see note 15 above; Annas, see note 15 above.

23. This point was made by several participants in a New York State summit of bioethicists, physicians, lawyers, health policy experts, clergy, and patient advocates, entitled "Bridging the Gap: From Sound Recommendations to Improved Realities," held 29-31 May 2002, in Syracuse, New York.

24. C.E. Schwartz et al., "Early Intervention in Planning End-of-Life Care with Ambulatory Geriatric Patients," *Archives of Internal Medicine* 162 (2002): 1611-8; Last Acts, *Means to a Better End: A Report on Dying in America Today,* November 2002, *http://www.rwjf.org/files/publications/other/meansbetterend.pdf,* p. 9.

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Interest in Physician-Assisted Suicide among Oregon Cancer Patients

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INTRODUCTION

Legalization of physician-assisted suicide (PAS) and euthanasia continues to be debated in medical, ethical, and political arenas. Even where illegal, physicians assist patients in suicide.[1] In the United States, only Oregon has legalized PAS, limiting this option to competent, terminally ill patients who request it. Since enactment of the Oregon Death with Dignity Act (ODDA) in 1997, 208 patients have died by PAS. Cancer is the most common terminal illness associated with PAS in all jurisdictions in which this option is legalized.[2]

When faced with a patient who requests PAS, clinicians in Oregon are advised that their obligation is to search for and treat remediable factors that may address the patient's suffering, rendering PAS an option of last resort.[3] Studies among patients with advanced cancer outside of Oregon report that those who endorse on a survey that they might request PAS or prefer a hastened death are often depressed, hopeless, or psychosocially distressed. Less consistently, these studies identify poor social support, feeling unappreciated, spiritual distress, poor quality of life, declining functional status, concern with being a burden to others, and physical symptoms as factors.[4]

In studies outside of Oregon, approximately 10 percent of terminally ill cancer patients endorse a desire for death to come sooner or an interest in PAS.[5] Yet only one in 1,000 deaths in Oregon are by PAS.[6] Surveys of physicians and hospice professionals in Oregon paint a different picture of patients

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who actually receive lethal prescriptions, emphasizing the role of desire for control, independence, and avoiding institutionalization; and existential concerns such as lack of meaning and readiness to die. Depression and poor social support were not considered important reasons for actual PAS requests.[7] Reconciling these somewhat disparate views of why patients request PAS would facilitate the development of interventions that render PAS an option of last resort. The goals of this study were to measure:

1. The level of interest in and actions around PAS among cancer patients in Oregon,
2. How this interest changes over time,
3. The association between psychosocial and medical factors and interest in PAS, and
4. The relationship between expressing an interest in PAS on a survey and actually requesting legalized PAS from a physician.

PATIENTS AND METHODS

Cognitively intact patients with advanced cancer were recruited from the oncology clinics of the Portland Veterans Affairs Medical Center (VAMC) and the Oregon Health & Science University (OHSU) (acronyms are listed in table 1). Subject entry began in 1998 and was completed in 2001. During the study, participants who said that they were potentially interested in PAS were assessed every three months for up to two years. The patients who were considered eligible to be recruited for participation in the study included:

- Patients who had cancer, with 50 percent probability of dying within two years.
- Patients who could communicate in English.
- Patients who were negative for human immunodeficiency syndrome (HIV).
- Patients who had a Folstein Mini-Mental State Examination (MMSE) score of greater than 23.[8]
- Patients who received ongoing care at OHSU or Portland VAMC.

Each potential subject received a letter of invitation to participate from his or her primary oncologist. The study was approved by the institutional review boards at both OHSU and the Portland VAMC. After the study was described to all potential participants, subjects gave their written informed consent.

Demographic information was obtained from each participant. Pain was measured

TABLE 1 Acronyms and definitions

alpha	Indicates the probability of rejecting the hypothesis tested, when the hypothesis tested is true.
chi-square	test that compares the frequency of an observation in the collected data and those that would be expected if there were no relationships between the variables
CI	confidence interval: a range of values around the mean where it is expected that the true population mean is located
df	degrees of freedom: in tests of statistical significance, refers to the number of values in the sample that cannot be mathematically calculated from knowing other values and a calculated statistic
DIS	Diagnostic Interview Schedule
ECOG	Eastern Cooperative Oncology Group
GDS	Geriatric Depression Scale
HIV	human immunodeficiency syndrome
MMSE	(Folstein) Mini-Mental State Examination
n	number of subjects in a group
N	number of subjects in the study
ODDA	Oregon Death with Dignity Act
OHSU	Oregon Health & Science University
p	probability value
PAS	physician-assisted suicide
SD	standard deviation: one of several indices of variability that are used to characterize the dispersion among the measures in a given population
se	standard error: the estimated standard deviation of a statistic
Spearman's *rho*	measure of the linear relationship between two values
two-tailed test	test of a statistical hypothesis in which the value of the statistic that is either too small or too large will lead to rejection of the hypothesis
VAMC	Veterans Affairs Medical Center

with three items from the Wisconsin Brief Pain Questionnaire, rating pain on average and at its worst in the previous two weeks, and pain at the time of the interview, all on 0 - 10 scales.[9] Standardized instruments were used to measure functional status (Eastern Cooperative Oncology Group — ECOG[10]), social support (Duke-University of North Carolina Functional Support Scale[11]), depression (Geriatric Depression Scale — GDS[12] and Diagnostic Interview Schedule — DIS[13]), and satisfaction with advanced cancer care.[14] Four items were completed from the Beck Hopelessness Scale, which had been previously reported to be statistically different in amyotrophic lateral sclerosis patients who were interested in PAS and those who were not.[15] Three items from the Zarit Burden Inventory were revised to measure patients' perception of how often their medical condition stressed or strained or caused hardship to their families or how often they felt that they were a burden to their families.[16] Suffering, the importance of religion, and the degree to which health problems limited quality of life were each measured on 0 - 10 Likert scales. Each subject indicated whether she or he had ever seen a mental health professional or had ever made a suicide attempt.

Subjects indicated their overall position on legalized PAS, whether they had contacted any organizations that assist patients in obtaining legal lethal prescriptions, and whether they discussed their interest in PAS with their family, friends, or physician. They were asked whether they would ever consider requesting a lethal prescription; those who indicated they might consider or were planning to request PAS were followed every three months for up to two years, to understand variability in interest in PAS over time. Patients who indicated that they would never make such a request or were unsure were not evaluated further. At initial and follow-up appointments, participants rated their interest in requesting a lethal prescription in the previous two weeks (0 = I am very *unlikely* to request a lethal prescription, 10 = I am very *likely* to request a lethal prescription), as well as their desire for death to come sooner in the previous two weeks, also on a 0 - 10 Likert scale. At each visit, participants who rated their interest in requesting a lethal prescription as "0" were considered to have no interest, "1 - 2" minimal interest, "3 - 4" mild interest, "5" or greater a serious interest. Once referred to hospice, patients were followed monthly, as tolerated. After the patient's death, each oncologist was asked if the patient had initiated discussion about PAS and any details of the discussion.

STATISTICAL ANALYSIS

Summary statistics included proportions for categorical variables, means with standard deviations (SDs) for continuous variables, and Spearman's *rho* to examine correlations. The main dependent variable of interest, likelihood of requesting a lethal prescription in the previous two weeks, was an ordinal variable ranging from 0 to 10. We used proportional odds models[17] to identify potential predictors for the likelihood of requesting PAS. The proportional odds model is commonly used for the analysis of ordinal data, and it assumes that the odds ratio of a predictor variable (for example, hopelessness) is equivalent for all logistic regressions defined by various cutpoints (for example, PAS > 4 versus PAS $\leq$ 4, PAS > 5 versus PAS $\leq$5). The odds ratio is referred to as the cumulative odds ratio. Predictor variables were examined in a stepwise procedure, using a forward Wald procedure with an *alpha* to enter of 0.1 and *alpha* to remove of 0.15. All tests were two-tailed, and a p value of < 0.05 was considered to be statistically significant.

For a subset of subjects who were followed longitudinally, we evaluated how interest in obtaining a lethal prescription (0 - 10 scale) changed over time in response to other time-varying factors (that is, an ECOG score, support scale, depression, hopelessness, family burden, religiousness, quality of life, suffering, pain, satisfaction with medical care, and hospice care). We fit a random effects model, treating each subject as a random effect,[18] and estimated a change in an interest in PAS as a

function of each time-varying factor. Since the residual analysis using the original PAS interest data revealed non-normality, we transformed the data using a square-root transformation and repeated the analysis. The *p* values from both analyses were almost identical, and did not change statistically significant factors. Because the interpretation of the slope estimate is more intuitive for untransformed data, we presented the results from the original data. The mixed model analysis was performed using SAS PROC MIXED procedure.[19]

RESULTS

PARTICIPANTS

We located 417 potential subjects from patients with advanced cancer at the Portland VAMC and at OHSU; of these, 53 patients were ineligible for the following reasons:

- The patient had a low MMSE score (*n* = 26),
- The patient did not speak English (*n* = 15)
- The patient did not have ongoing care (*n* = 12).

Among the remaining 364 potentially eligible participants, 93 (26 percent) were not approved by their oncologist or primary physician: for 69 patients, the oncologist or primary physician gave no reason for disapproval. Other reasons physicians gave for disapproval included:

- The patient had other mental or cognitive disorders,
- The patient was too sick or not aware of diagnosis,
- The oncologist or physician had not met the patient.

Of the 31 referring physicians, 10 referred 10 or more patients. Eight of these 10 physicians disapproved fewer than 10 percent of potential referrals. Two oncologists disapproved substantial numbers of referrals without explanation: one disapproved 17 of 52 patients (33 percent), and the second disapproved 51 of 132 patients (39 percent). Because these were otherwise potentially eligible patients, they are included in the non-respondent proportion.

Of the 364 remaining potentially eligible subjects, 110 declined participation (30 percent); thus, the 161 subjects who agreed to participate comprised 44 percent of the 364 potential subjects who were initially identified. Of the 161, almost three-quarters were men, 93 percent were White, and the mean age was 62 years; 60 percent were VAMC patients, with 40 percent enrolled at the OHSU site. The most frequent cancer types were lymphoma/hematologic (23 percent) and lung (22 percent). Of these 161 patients, 38 (24 percent) met criteria for major depressive disorder on the DIS, and 32 (20 percent) were probably "depressed" as measured by the GDS, because they endorsed 14 or more depressive symptoms (see table 2).

Of our 161 subjects, 31 percent indicated at initial evaluation that they might or planned to request PAS, and 9 percent said that they had had a serious interest in obtaining a lethal prescription in the previous two weeks. We followed 42 of the subjects longitudinally; of these subjects, 19 developed a serious interest in obtaining a lethal prescription at some point, eight of the 19 (42 percent) informed their oncologist of this interest, and two made an explicit request for PAS, one of whom never expressed a strong interest at any point in the study.

SUPPORT FOR PAS AND INTEREST IN PAS

Of the 161 participants, 57 percent supported or strongly supported legalization of PAS (see table 2); 47 percent stated they would never consider PAS (*n* = 76); 14 percent were unsure (*n* = 23); 34 percent might consider it in the future (*n* = 55); and 4 percent were planning to request PAS (*n* = 7). No patient had made a request for PAS to a physician at the first evaluation. Of the 161 participants, 15 (9 percent) indicated a serious interest in PAS in the previous two weeks. Of these 15 sub-

TABLE 2 Characteristics and views on assisted suicide of cancer patients at first evaluation ($N = 161$)

Characteristic	Mean	SD
Age in years	61.6	11.7
Years of education	13.5	2.9
Characteristic	***n***	**%**
Sex		
Male	118	73
Female	43	27
Race		
White	150	93
African-American	4	2
Asian-American	2	1
Hispanic	1	1
Other	4	2
Marital status		
Married/long-term partner	94	58
Single/widowed/divorced	67	42
Healthcare affiliation		
VAMC	97	60
OHSU	64	40
Functional status		
Fully active	35	22
Restricted in strenuous activities	88	55
Ambulant, but unable to work	24	15
Limited self care or bed ridden	14	9
History of suicide attempt	11	7
Ever seen by a mental health provider	71	44
Current major depressive disorder*	38	24
GDS Score ≥14 **	32	20
Patients' view	***n***	**%**
Support for legalized assisted suicide		
Strongly oppose	35	212
Oppose	13	8
Undecided	21	13
Support	39	24
Strongly support	53	33
Interest in obtaining lethal prescription in 2 weeks previous		
None (0)	117	723
Minimal (1 - 2)	24	15
Mild (3 - 4)	5	3
Serious (5 - 10)	15	9

* As determined by Diagnostic Interview Schedule.
** As determined by Geriatric Depression Scale.

jects, one had discussed PAS with his physician, and one had contacted a PAS advocacy organization.

Of all of the 161 participants, 57 percent had discussed their attitudes about PAS with friends or family before receiving the letter to participate in the study (see table 3); 81 percent (50 of 62) of those who either might consider PAS or who had planned to request PAS had discussed their views with friends or family, whereas only 48 percent (47 of 99) of those who would never consider PAS or who were "unsure" had discussed their views (*chi*-square = 17.5, df = 1, $p < 0.001$). Among those who might consider PAS ($n = 62$), 55 percent ($n = 34$) thought that most friends and family would be supportive, 10 percent ($n = 6$) thought that friends and family would talk them out of it, and 35 percent ($n = 22$) were unsure whether family and friends would support or oppose this decision.

FACTORS ASSOCIATED WITH INTEREST IN REQUESTING PAS

At initial evaluation, increasing interest in requesting a lethal prescription in the previous two weeks was statistically associated with greater suffering, increasing depression scores measured by GDS, increasing hopelessness, more limitations on quality of life from health problems, more pain, sense of burden to others, and less satisfaction with medical care, but not with any demographic variables, social support, or functional status (see table 4). The correlation between desire to die and interest in PAS was 0.48 ($p < 0.001$). Five of the 15 patients with a serious interest in PAS (33 percent) met criteria for major depressive disorder on DIS; 33 of the 146 patients without a serious interest in PAS (23 percent) were also depressed (*chi*-square = 0.839, df = 1, $p = 0.36$). In the multivariate analysis, only the degree to which health problems limited quality of life remained a significant predictor of interest in PAS. There was no difference between subjects with and without a serious interest in PAS on whether they had ever seen a mental health provider (eight of the 15 with

serious interest in PAS, 53 percent versus 63 of the 146 without a serious interest in PAS, 43 percent; *chi*-square = 0.752, df = 1, $p = 0.45$) or who had a previous suicide attempt (two of the 15 with a serious interest in PAS, 13 percent versus nine of the 146 participants without a serious interest in PAS, 6 percent; Fishers exact test = 0.27).

LONGITUDINAL INTEREST IN PAS

Of the 62 subjects who expressed a potential future interest in PAS (who had marked that they were planning to request a lethal prescription or might consider it in the future), 42 agreed to participate in a longitudinal evaluation to help understand how their views might change over time. Persons who declined follow-up or who died before follow-up were more depressed (seven of the 20 who declined to participate, 35 percent versus four of the 42 who agreed to participate, 10 percent; *chi*-square = 6.03, df = 1, $p = 0.01$), but did not differ in the other factors including demographic factors, interest in PAS, mean GDS, desire for death, or satisfaction with medical care, compared to those who participated in the longitudinal study. Of the 42 subjects, 29 completed three or more evaluations, 21 completed four or more evaluations, and 15 completed five or more evaluations. Among those who were followed longitudinally, 19 had a serious interest in obtaining a lethal prescription at some point, eight had a serious interest in obtaining a lethal prescription at the first evaluation, but never subsequently; and 11 had a serious interest in PAS at some point after the first evaluation. Figure 1 shows the variation over time in these 11 respondents. Four of the 19 (21 percent) who had ever indicated a serious interest in obtaining a lethal prescription at any point in the longitudinal part of the study requested information from a PAS advocacy organization, and eight of these 19 (42 percent) had discussed their interest with their oncologist.

For the 42 subjects who were followed longitudinally, we evaluated how interest in obtaining PAS changed over time (on a 0 - 10 scale) in response to other time-varying factors. Religiousness and hospice status were not found to influence their interest. Factors that were associated with increasing interest in obtaining a lethal prescription included:

- Declining functional status (estimate = 0.735, se = 0.247, $p = 0.004$);
- Increasing social support (estimate = -0.119, se = 0.037, $p = 0.002$);
- Increasing depression measured by GDS (estimate = 0.148, se = 0.031, $p < 0.001$);
- Increasing hopelessness (estimate = 0.377, se = 0.155, $p = 0.02$);
- Increasing sense of burden to family (estimate = 0.229, se = 0.080, $p = 0.005$);
- Degree to which poor health limited quality of life (estimate = 0.362, se = 0.075, $p < 0.001$);
- Increasing suffering (estimate = 0.398, se = 0.075, $p < 0.001$);

TABLE 3 Actions of patients around PAS at first evaluation

Characteristic	*n*	%
Who would be told about PAS plans* (*n* = 85)		
Only physician	4	5
Only one other person	15	18
Several people	26	31
Many people	23	27
Not sure	17	20
Perception of family/friends' response to patient's request for PAS* (*n* = 85)		
Would be supportive	41	48
Try to talk out of assisted suicide	8	9
Not sure	36	42
Contacted PAS organization (*n* = 161)		
Yes, contacted	11	7
Discussed assisted suicide w/ family/friends (*n* = 161)		
Before study contact	92	57
After study contact	4	2
Never discussed	64	40
Missing	1	1
Discussed assisted suicide with physician (*n* = 161)		
Yes	9	6
No	111	69
Missing**	41	25

* Only asked of 85 participants who would consider PAS or who were not sure.

** Question inadvertently deleted from 41 questionnaires.

- Increasing pain (estimate = 0.282, se = 0.097, $p = 0.004$); and
- Decreasing satisfaction with medical care (estimate = -0.0773, se = 0.016, $p < 0.001$).

REQUESTS FOR PAS

Two subjects made an explicit request to their oncologists for a lethal prescription under the ODDA. Both had been followed longitudinally. One patient, who completed four research assessments over one year, was diagnosed with depression by DIS at each assessment; his GDS scores, which ranged from 12 to 24, indicated minimal to severe depression, and his level of hopelessness was high. He received mental health treatment in the last year of life, but he was not enrolled in hospice care before the final study evaluation. He had very little pain, rated his social support as good, with minimal burden to others, but his satisfaction with his medical care decreased from the top quartile to the 25 percent percentile at the final evaluation. At no study assessment did he indicate that he had contacted a PAS advocacy organization or that he planned to request a lethal prescription.

TABLE 4 Proportional odds model: predictors of interest in requesting a lethal prescription

	Univariate analysis			Multivariate analysis		
	Odds Ratio[1]	95% CI	*P* Value	Odds Ratio	95% CI	*P* Value
Age (years)	1.00	(0.97, 1.03)	.79	--	--	--
Education (years)	1.05	(0.93, 1.18)	.45	--	--	--
Functional status[2]	1.26	(0.86, 1.86)	.24	--	--	--
Social support[3]	0.96	(0.92, 1.00)	.06	--	--	--
Depression (DIS)[4]	1.92	(0.90, 4.09)	.09	--	--	--
Depression (GDS)[5]	1.08	(1.02, 1.13)	.01	--	--	--
Hopelessness[6]	1.46	(1.14, 1.86)	.003	1.28	(0.98, 1.66)	0.07
Sense of burden[7]	1.13	(1.02, 1.27)	.03	--	--	--
Importance of religion[8]	0.94	(0.84, 1.04)	.19	0.90	(0.80, 1.01)	0.07
Quality of life[9]	1.25	(1.10, 1.42)	.001	1.26	(1.10, 1.45)	0.001
Suffering[10]	1.17	(1.04, 1.31)	.01	--	--	--
Satisfaction with medical care[11]	0.96	(0.93, 0.99)	.002	--	--	--
Pain[12]	1.15	(1.01, 1.31)	.03	--	--	--

NOTES

1. Odds ratio associated with one unit change of the predictor variable. E.g., the odds ratio for hopelessness is 1.46 in the univariate analysis. This means that there is a 1.46-fold increase in the likelihood of requesting PAS when the hopelessness score increases by 1 (e.g., 2 to 3).
2. Eastern Cooperative Oncology Scale.
3. Duke-University of North Carolina Functional Support Scale.
4. Diagnostic Interview Schedule.
5. Geriatric Depression Scale.
6. Beck Hopelessness Scale – 4 items.
7. Three items revised from Zarit Burden Inventory.
8. 0 - 10 Likert scale.
9. 0 - 10 Likert scale.
10. 0 - 10 Likert scale.
11. FAMCARE.
12. Wisconsin Brief Pain Questionnaire.

He rated his interest in obtaining a lethal prescription and desire for death as 0 at each evaluation. He rated the importance of religion very high on the first three evaluations, but low at the last, at which time he told the examiner he was "resisting his religion." He died two months after his final study assessment, during which time, as reported by his oncologist, he made two explicit requests for a lethal prescription. His physician did not prescribe a lethal medication.

The second subject was assessed six times over nine months, including twice while enrolled in hospice. He discussed his interest in PAS with his physician over time, contacted a PAS advocacy organization, and indicated that he intended to request PAS on all study assessments. He rated his interest in obtaining a lethal prescription as serious at all but one evaluation, although his desire for death was always low. He met criteria for major depressive disorder by DIS on one of six visits (but not his final visit) and he endorsed many hopeless statements, but his GDS scores never indicated depression. His pain was moderate, his social support good, but he felt he was somewhat of a burden to his family. His satisfaction with medical care was low, ranging from the 12th to the 42nd percentile. His physician reported that he made an explicit request for PAS just before his final study assessment, and he died nine days after this assessment. His physician did not prescribe a lethal medication.

DISCUSSION

This study was conducted in Oregon after enactment of the ODDA, which legalized PAS. The main findings include:

1. Approximately one in 10 cancer patients expressed a serious interest in obtaining a lethal prescription at the initial evaluation.
2. Medical and psychosocial concerns were important in subjects' interest in PAS.
3. Among those with interest in PAS, there was a high degree of variability in subjects' interest, when examined over time.
4. Fewer than half of the patients who were followed longitudinally who endorsed a

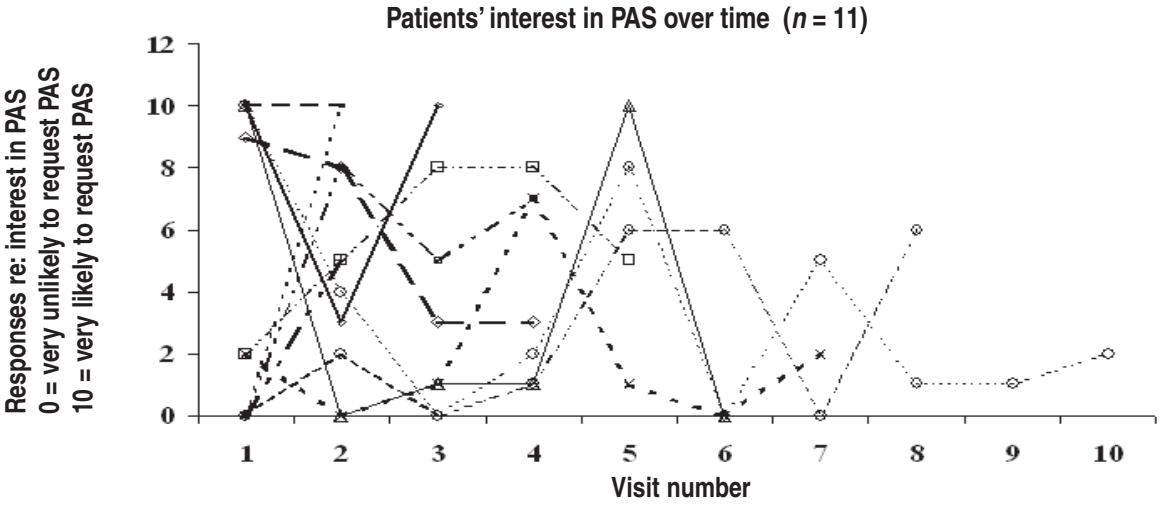

Figure 1.
This figure tracks the responses of 11 patients in the longitudinal portion of the study who expressed a serious interest in PAS at some point after the first evaluation. Each patient is represented by a unique symbol; the lines between the symbols track the patients' responses, over time, to a query about their interest in PAS. Patients were asked about their interest in obtaining a lethal prescription, in which 0 = very unlikely to request PAS, and 10 = very likely to request PAS. The patients in the longitudinal portion of the study were seen approximately every three months.

serious interest in obtaining a lethal prescription discussed this issue with their oncologist.

5. Only two study participants who were followed over time ultimately made an explicit request for PAS, one of whom never indicated interest in PAS on any study assessment.

Although the majority of patients in the study supported legalization of PAS, only 9 percent indicated a serious interest at initial evaluation in obtaining a lethal prescription. This level of interest in pursuing PAS, and wanting to hasten death, is similar to that reported in studies in North America outside Oregon. For example, Emanuel and colleagues, in a study of 988 terminally ill patients, reported that 10.6 percent indicated they had seriously considered euthanasia or PAS for themselves.[20] Chochinov and colleagues, in a study of 200 terminally ill cancer in-patients, reported that 8.5 percent had a serious and pervasive interest in hastened death.[21] Rosenfeld and colleagues developed an instrument to measure desire for hastened death among terminally ill patients — 16.3 percent of patients were found to have a high desire for death.[22] Wilson and colleagues, in a study of 70 patients with advanced cancer, reported that 12 percent would have made a request for PAS or euthanasia at the time of the study interview.[23] As such, despite differences in how this construct is measured, there is increasing agreement across studies that approximately one in 10 patients with advanced cancer will, when queried, endorse a serious interest in PAS or hastened death. Despite legal availability and heightened public awareness, Oregon cancer patients do not appear to have a greater interest in pursuing PAS for themselves.

Surveys of cancer patients have found that, when examining desire for death to come sooner (including having serious thoughts about PAS or endorsing that one would request PAS if it were available), associated factors found in some, although not all studies, include depression, hopelessness, psychological distress, pain, poor social support, spiritual well-being, religiousness, quality of life, and overall symptom distress.[24] Although we found many of these factors associated in univariate analysis, only the degree to which health problems limited quality of life predicted interest in PAS in the multivariate analysis. Over time, depression, hopelessness, sense of burden to others, pain, the impact of health on quality of life, and suffering, proved to be important.

Our study is the first to measure satisfaction with medical care, and we found that dissatisfaction with care was associated with interest in PAS over time. Further exploring the specific sources of dissatisfaction would be key to developing interventions. Moving palliative care approaches earlier into the medical encounter may be important in addressing this level of distress. Since completion of this study, both institutions at which the study was performed have developed palliative care consultation teams. Patients in the study were evaluated for the most part before entering hospice, which we previously demonstrated was associated with patients changing their mind about PAS.[25] Nevertheless, 86 percent of all PAS deaths in Oregon have occurred after hospice enrollment.[26]

We found that many patients with serious interest in PAS neither bring this up with their physician, nor, at times, with their friends and family, despite the legal availability of this option. Clinicians in Oregon have been advised not to initiate discussions about PAS, as there are concerns that patients may misinterpret inquiries, believing that the clinician is promoting assisted suicide. Clinician apprehension about this may be reflected in the large number of cases in which the oncologist withheld approval for participation in the survey. Although we do not have information about why physicians blocked referral, it is possible that clinicians are concerned that even referring patients to the study might be a detrimental communication or increase patients' interest in PAS. Notably, eight patients who had a very strong interest in PAS at first study evaluation never expressed interest in

the remaining months of the survey, rebutting concerns that asking about PAS may be misinterpreted by the patient as support.

Sensitive inquiry by the physician may be needed to elicit this important information. Hesitance to bring up the issue appears to leave many patients struggling in isolation. Endorsing interest in PAS may not predict actual requests very well, but does indicate psychosocial distress. Discussion of PAS presents an opportunity to explore fears and worries regarding the future. For example, an oncologist might probe further at the transition point between care focused on life prolongation to care focused on palliation. Questions about mood or limitation of life-saving care could be followed by queries about thoughts of wanting death to come sooner. An affirmative answer should be followed by thorough exploration of the nature of these thoughts. Within psychiatry, the belief that asking about suicide plants a previously unconsidered idea in the patient's mind has been thoroughly rebutted.

We found a high degree of variability over time in interest in PAS among patients who were predisposed to consider it. Similarly, Emanuel and colleagues report that, among 988 patients who were seen at two points in time, half of those who initially considered PAS had changed their mind, whereas an equal number had began to consider this intervention.[27] In our study, among 19 patients followed longitudinally who expressed a serious interest in obtaining a lethal prescription, only one patient actually made a request for PAS to his physician, despite the fact that PAS was legalized throughout this time period. A second patient, who at no point in the study expressed a serious interest in PAS, also made an explicit request to his oncologist. As such, our ongoing inquires regarding interest in PAS were neither sensitive nor specific for actual requests.

There are several shortcomings and limitations to our study. It is possible that the wording of our questions was flawed, resulting in failure to elicit actual intent to pursue PAS. However, most studies show a much higher rate of endorsement of interest in hastening death than actual requests or suicide. Our study may support that it is inappropriate to extrapolate information about patients who actually pursue PAS from information about patients who endorse interest on a survey. This may be one reason for the discrepant findings regarding characteristics of persons who request PAS and actually pursue it in Oregon. Referral biases by physicians and acceptance biases may exist among patients who ultimately participated in the study — only 44 percent of eligible patients participated — threatening generalizability. We did not ask about physical symptoms other than pain, or the adequacy of their treatment, which have been important in explaining interest in PAS. Finally, this study was performed in the only U.S. state in which PAS has been legalized. This may limit generalizability, although our data suggests that Oregon patients are more similar than different to other cancer patients in North America who are interested in this option.

In summary, although the interest of Oregon patients in PAS may be similar to interest in terminally ill patients from other jurisdictions, Oregon patients bring this up with their physician less than half of the time. Other novel findings of our study include the role of dissatisfaction with healthcare in a patient's interest in PAS, and the variability and mutability of interest over time. Endorsing an interest in PAS on a survey appears to be a marker of psychological and symptom distress, however, it may neither be sensitive nor specific to patients who actually request PAS under legalized conditions. This information is important in interpreting data from studies that purport to apply information derived from surveys of terminally ill to those who actually pursue PAS.

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"Physician-Assisted Suicide among Oregon Cancer Patients": A Fading Issue

Colleen C. Denny and Ezekiel J. Emanuel

Articles like Ganzini and colleagues' "Physician-Assisted Suicide Among Oregon Cancer Patients" are increasingly infrequent. Public and academic discussion of physician-assisted suicide (PAS) and voluntary euthanasia reached a dramatic highpoint in 1994 with the passage of Oregon's Death with Dignity Act.[1] Since the Supreme Court's 1997 ruling that there is neither a constitutional right nor constitutional prohibition to PAS, however, interest in the subject has declined significantly. Physicians and lawmakers alike have gradually come to the realization that the furor over PAS affects only a very small minority of U.S. patients. The percentage of terminally ill patients who are mentally competent and would actually take advantage of legalized PAS leaves only a very few individuals directly impacted by PAS legislation — a population much smaller than perhaps is suggested by the attention focused on the topic. Ganzini and colleagues' findings illustrate some of the factors that make the legal status of PAS a relatively marginal issue for the American public in its efforts to improve patients' experiences at the end of life.

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PROBLEMS OF METHODOLOGY

Before considering the implications of Ganzini and colleagues' findings, we must consider whether the data are reliable and generalizable. Overall, several aspects of the study cast doubt on the soundness of the final conclusions. First, the sample population from which the researchers glean most of their results is biased by a number of factors. A significant part of this bias cannot be attributed to any fault of the researchers: a large number of physicians declined to refer their patients to the study, often without explanation, and more than 30 percent of eligible participants declined to enroll. However, additional bias occurred when 80 subjects who indicated initially that they were uncertain or uninterested in requesting PAS were never re-evaluated. As the authors' and others' previous findings indicate, patients' interest in PAS can vary significantly over time, increasing as well as de-

creasing.[2] Thus, those patients with no initial inclination towards PAS might have subsequently expressed interest. Excluding these subjects — more than 60 percent of those enrolled — from further data collection not only reduced the sample size unnecessarily, but distorted the final results by eliminating patients whose PAS interest may have developed over time, rather than presenting at the first assessment.

Another problematic aspect of the study design concerns whether the assessment questionnaires accurately gauged PAS interest and its correlates. The decision to classify any interest greater than 4 on a 10-point scale as "serious interest" in PAS, for example, may confound data interpretation. It seems quite possible that a patient's choice of a number in the middle of the scale, such as 5, could indicate "mild" rather than "serious" interest. Accordingly, there seems to be significant potential for a disconnect between patients' expressed level of PAS interest and researchers' interpretation of this self-report. The decision to ask doctors, rather than subjects, about the occurrence of physician-patient PAS discussions may also confuse the authors' interpretation of the factors associated with PAS interest. As all the other possible correlates of PAS interest were self-reported by the patients, gathering this one piece of information from an external source — especially a physician who treats many terminally ill patients and may have a faulty recollection — may undermine the final data analyses. The assessments' questionable sensitivity and specificity seems best evidenced by the fact that one of the two patients who explicitly requested PAS failed to indicate any PAS interest whatsoever on the questionnaire, while other subjects expressing "serious" levels of interest never even brought up the subject with their physicians. These methodology concerns indicate that the results should be interpreted very cautiously.

LONGITUDINAL INTEREST IN PAS

The most critical finding in Ganzini and colleagues' work is that terminally ill patients' interest in PAS fluctuates significantly over time. No individual subject had the same self-reported level of interest for more than two evaluations in a row (unless that level is zero), nor consistently expressed a high level of interest in obtaining a lethal prescription. Additionally, the data displayed in figure 1 are from the subgroup of participants who are probably most interested in PAS; 40 percent of patients who were assessed multiple times expressed zero interest on all assessments subsequent to the initial evaluation and were not included in this graph. These longitudinal data indicate that an individual patient's self-reported PAS interest is typically highly unstable, moving up and down over the course of a few months.

DEPRESSION, PAIN, DIGNITY, AND INTEREST IN PAS

There is a widely held public perception that uncontrollable pain plays a pivotal role in a terminally ill patient's desire for PAS, and is the primary justification for legalizing the prescription of lethal medications.[3] Contrary to this belief, however, almost all previous studies on attitudes toward PAS have found that depression and other indicators of psychological distress are more strongly correlated with elevated interest and desire for PAS among terminally ill patients, eclipsing pain level or even the wish for a dignified death, another rationale often championed by PAS advocates.[4]

Ganzini and colleagues confirm these findings, concluding that feelings of depression, hopelessness, being a burden to one's friends and family, and dissatisfaction with medical care all played a statistically significant role in heightened interest in PAS among patients. It is important that the two patients in the study who made explicit requests for PAS both showed signs of clinical depression. In contrast, neither man felt that he was in unbearable pain. Although some proponents argue that the legal status of PAS allows those with unbearable suffering to end their lives with dignity, empirical studies like those of Ganzini

and colleagues consistently show that depression and general psychological distress play a more significant role in increasing patients' interest in PAS.[5]

IMPORTANCE FOR PHYSICIANS AND POLICY MAKERS

The themes reiterated by Ganzini and colleagues have important indications for future action, both at the local level of health professionals as well as the state and national levels of government. The most significant implication of this study for physicians dealing with terminally ill patients is that an individual patient's interest in PAS often fluctuates considerably and rapidly. Those patients who are initially interested in PAS may decide against it in the coming weeks or months, just as patients who are opposed may eventually gain interest. Rather than taking quick action or assuming that a patient who indicates interest in PAS will soon move to make an explicit request, physicians should understand that such interest is particularly volatile over time, and make a concerted effort to understand the motives behind a particular patient's interest.

In addition to illuminating the instability of PAS interest, Ganzini and colleagues' data regarding the significant role that depression and poor mental health play in PAS interest re-emphasize to physicians the importance of mental health assessments and treatment for terminally ill patients. Physicians should maintain a low threshold for signs of depression in such patients and should be sure to request psychological assessment and counseling for the terminally ill. This applies equally to terminally ill patients who make no PAS requests; as Ganzini and colleagues and others' data indicate, approximately one in five terminally ill patients suffers from major depressive disorder, including those uninterested in obtaining a lethal prescription.

The authors' one suggestion for physicians — that they initiate more conversations with their patients about PAS — seems misguided. They interpret their finding that only half of patients expressing PAS interest actually discuss the option with physicians to signify poor communication between doctors and their patients. Rather than indicating a lack of communication, however, this finding seems more likely to reflect patients' feelings that such a conversation implies a much more serious level of intent than expressing interest on an anonymous questionnaire. Furthermore, such physician-initiated discussion might be potentially harmful to patients' decision making. In their conclusions, the authors dismiss the widespread apprehension among physicians that bringing up the subject of PAS may be understood by the patient as an endorsement. In support of this dismissal, the authors cite both psychology literature and their own data, claiming psychiatric patients are not more likely to have suicidal thoughts after being asked about suicidal intentions, and pointing to their eight subjects who were assessed multiple times, but never expressed any PAS interest subsequent to the initial evaluation. These arguments alone, however, are insufficient to discount the concerns of a perceived endorsement of PAS by a physician.

First, the psychiatry literature may not be an appropriate comparison: while a psychiatrist will never advocate suicide as an option for a mentally ill patient, a physician might legally approve PAS for a terminally ill patient. Second, the eight subjects mentioned by the authors already had an interest in PAS, by their own report. To counter worries of perceived endorsement, it would be important to know whether patients who communicated no initial interest in PAS came to express interest after repeated evaluation. Unfortunately, in this study design, these individuals were not followed-up after the first assessment. Thus, without further data, the authors' dismissal of the concern that patients will read endorsement into a physician's initiations of PAS discussion seems premature and unsupported by empirical data.

For legislators and policy makers, the implications of Ganzini and colleagues' findings are twofold. First, the percentage of terminally ill individuals who would actually take ad-

vantage of the PAS option is markedly small, estimated at significantly less than 1 percent of the general patient population.[6] The legal status of PAS simply does not impact the lives of the great majority of Americans, hospital patients, or even terminally ill patients as a genuine personal option. Policy makers should accordingly be in no rush to legalize PAS or euthanasia before sufficient research can examine the full range of implications of such legislation, as the few lives directly impacted by the legal status of PAS are more than counterbalanced by the potential dangers and pitfalls of legalization. Second, with the legislation that already exists, policy makers should be sure to institute and enforce mandatory waiting periods in recognition of the fluctuation of PAS interest among terminally ill patients. In light of Ganzini and colleagues' evidence that patients' interest in PAS is unstable over short periods of time, mandatory waiting periods will help to assure that patients are consistent in their desire for PAS, rather than making an explicit request during an upswing of their oscillating interest that will soon diminish.

CONCLUSION

Ganzini and colleagues' findings reiterate the instability of PAS interest among terminally ill patients, as well as the significant role played by depression and psychological distress in elevated PAS interest levels. Physicians should accordingly be cautious in taking initial interest in PAS seriously among the terminally ill as an indication of stable preference. The trends reemphasized in this study – instability of interest, disinclination to act on interest, and the strong role of poor mental health in interest – indicate the exceedingly small population of individuals that the legal status of PAS directly affects, a conclusion with important implications for policy makers faced with questions of legalizing PAS. Rather than debating over PAS questions that affect only a minute fraction of patients, doctors and policy makers should focus on the broader-impact issue of enriching end-of-life care for the 2.4 million Americans who die each year by improving pain management, mental health treatment, and options for hospice and palliative care. [7]

DISCLAIMER

The opinions expressed are the authors' own. They do not reflect any position or policy of the National Institutes of Health, the Public Health Services, or the Department of Health and Human Services.

NOTES

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Response to Denny and Emanuel

Linda Ganzini

This response to Denny and Emanuel focuses on two areas: their critique of the methods used in our study, and their trivialization of the study of PAS.

First, Denny and Emanuel express concern about possible bias in our results. Low rates of participation may bias results, if the study group does not represent the broader population from which the participants were sampled. "Bias" suggests a distortion of the relationship between a risk factor and the outcome, but excluding those thought to be at low risk for an outcome in the prospective study — in this case, those who indicated they would never request PAS — does not result in scientific bias. In prospective studies, researchers frequently and intentionally limit members of the inception cohort to those who are anticipated to be at most risk for an outcome. Prospective studies are very labor intensive and inefficient, and the financial costs of following a large group of subjects who have a low likelihood of the outcomes of interest can be prohibitive. The more valid methodological problem in our study was the rarity of actual PAS requests, which limited our statistical power to detect predictors of this very infrequent outcome.

Yet if our measure failed to predict actual PAS requests, then experts should hesitate to extrapolate from studies about the interest in hastened death to requests for PAS, as the relationship between attitudes and requests for PAS in most other studies remains untested. Taken in total, Oregon data suggest that about 10 percent of terminally ill patients seriously consider PAS.[1] When we set the cut-off point for serious interest in PAS at 5 on our 1 - 10 scale, we obtained a similar proportion. Yet only 1 percent of Oregon decedents have made an actual request for a lethal prescription to a physician, and only 0.1 percent reportedly die by this method. Studies outside Oregon similarly support that about 10 percent of terminally ill patients who were surveyed had some type of interest in PAS or hastened death.[2] But one cannot conclude that because a set of factors such as depression or psychosocial distress are associated with endorsing interest in PAS on a survey, that these factors are necessarily associated with actual requested or completed PAS. Such assumptions often prove to be incorrect, as they fail to recognize a varyingly large gap between attitudes and behaviors. Only prospective and case control studies of patients who have actually made requests for PAS should affect policy, as they are the only studies that are relevant to actual legalization of PAS.

Second, Denny and Emanuel question our ascertainment of the outcome, a request for PAS, and suggest physicians may have forgotten their patients' requests. Our evidence suggests physicians find such requests to be quite

memorable.[3] Further, the physicians who cared for patients in our study knew they were in the study, and that they would be eventually asked about their patients' requests, if any.

Denny and Emanuel assert that academic discussion of PAS peaked in 1994, and then declined, reflecting diminished interest. A PubMed search for *physician-assisted suicide* (narrowed to articles relevant to humans, in English) shows that interest, measured by published articles, increased in 1993 to 219, crested in 1997 at 449, and gradually declined through 2005. (Adding *euthanasia* changed the peak to 1993, but this includes a large proportion of studies on passive euthanasia, such as withholding/withdrawing treatment and advance directives). Most of the articles are ethical and legal analyses or opinion pieces. Another interpretation might be that it is increasingly difficult to write ethical and legal formulations novel enough to be published. The development, funding, execution, analysis, and publication of data-based studies often takes years. They rarely can be produced as quickly as ethical or legal analyses. In the five years from 1995 to 1999, the 101 articles published on PAS were empirical — mostly surveys of the public, patients, families, and clinicians. In the five years from 2000 to 2004, 125 data-based studies were published. Studies such as ours are not "increasingly infrequent," but rather are becoming a larger proportion of publications over time. This represents a natural history of scientific progress, and the evolution of knowledge in medicine.[4]

The comment that PAS, or research on it, may be marginal or fading merits consideration. As media coverage about Terri Schiavo suggests, autonomy and decision making at the end of life capture the imaginations of many Americans, even when their own risk of a similar fate, such as persistent vegitative state, remains low. A 2005 Pew Research Center survey of 1,500 adults reports that Americans remained divided on PAS: 46 percent approved of PAS laws and 45 percent disapproved; 57 percent, who said they gave "great deal of thought" to PAS, approved of legalization, a view not shared by 35 percent, who said they gave "little" or "no thought" to end-of-life issues; 53 percent said that people have the moral right to end their lives if they have a chronic disease, up from 49 percent in 1990.[5] This does not support a statement that there is decreasing support for the issue of PAS based on a realization of its lack of importance.

Only one in 1,000 deaths a year in Oregon are caused by legal PAS, a rate much smaller than predicted when the law was passed. Oregon, with about 1 percent of the U.S. population, is the only state in which PAS is licit. From a public health perspective, PAS does not rank with heart disease, cancer, or stroke as a cause of death; however, it is a more common cause of death than sickle cell anemia, Tay-Sachs disease, tetanus, or rabies. None of these are trivial, and all are disorders that are important enough to warrant current funding from the National Institutes of Health. "Marginal" and "fading" are dismissive terms that communicate that trying to understand why people pursue PAS is not worth our effort.

There are many ways to judge the importance of studying PAS, and the most important benefits are not related to political advocacy for or against the issue: it may improve patient care. Information from PAS studies may help refine conceptual models of suicide, which most would agree is a public health problem, especially among medically ill elderly. Open study of PAS is facilitated in Oregon because it is legal, which allows the collection of information to guide clinical practice. Findings may help clinicians who struggle with PAS requests in other states, where PAS is illegal, and secret — clinicians are understandably reticent to discuss cases with peers. If one in 10 terminally ill patients, at some point, wish to hasten death, then, nationally, thousands of patients are affected each year. Clinicians err in avoiding discussion of such difficult issues. As underscored by Fallowfield and coauthors in their studies of cancer patients, silence can result in heightened fear and anxiety, not in increased calm and equanimity.[6] Because interest in PAS is a proxy for potentially remediable suffering, research is crucial and systematic and thor-

ough exploration by physicians in clinical settings is mandatory.

The dominant characteristics of those who request PAS are a desire to be in control of one's life, to minimize dependence on others, and to maintain self-sufficiency.[7] Conceptual models developed to explain why people request PAS cannot ignore this data. Currently, palliative care focuses on symptom management, spirituality, and family needs, not on how to leave this world in the "driver's seat." Highlighting a group of people with these values underscores the need for diversification of treatment in palliative care and development of individualized approaches reflecting the values of this minority. Even with the best care at the end of life, not all suffering can be assuaged, and some patients may be comforted to learn they have some choices.

Study of legal PAS can be considered a cautionary tale. During the peak of speculation about Oregon's law in the mid-1990s, experts predicted that legalized PAS would undermine palliative care; lead to legalization of other types of euthanasia; prey on the poor, the underprivileged, women, and on minorities; and lead to PAS clinics in Oregon for individuals from other states.[8] None of this has happened. Studies on PAS cannot overcome moral objections to it, but can serve as humbling reminders that our expertise alone cannot guarantee accurate prediction.

DISCLAIMER

The views expressed are the author's and do not represent the views of the Department of Veterans Affairs or the U.S. government.

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Clinical Practice and Challenges

The Duty to Re-Contact for Newly Appreciated Risk Factors: Fragile X Premutation

Gregory F. Guzauskas and Robert Roger Lebel

INTRODUCTION

The most common *heritable* form of mental retardation is fragile X syndrome (FXS), which causes 2 to 5 percent of mental retardation in males[1] and 30 to 40 percent of all cases of X-linked mental retardation.[2] FXS is caused by a change or mutation in a gene on the X chromosome, the *FMR1* (fragile X mental retardation 1) gene.[3] The gene appears in three forms, which are defined by the number of times it is repeated in a pattern of DNA, called "CGG repeats."[4] In the normal population, the number of CGG repeats varies between seven and 55; the most common number is 30. From one generation to the next, CGG repeats in the "normal" range will be transmitted stably, with no variation. Persons who have repeats of CGG in the range of 55 to about 200 are considered to have a *premutation:* while they do not exhibit FXS, the number of repeats may be unstable during passage through female oocyte production (the creation of an ovum.) Passage through male spermatocyte production is stable.[5] More than 230 repeats is considered a full mutation; this causes a shut down of a region of the *FMR1* gene that would normally produce the protein FMR (*FMRP*); a lack of *FMRP* causes FXS.

Males have only one copy of the X chromosome to depend on, and so if they have full mutation of the *FMR1* gene, they will produce no *FMRP* and will develop full FXS.[6] Most females with full mutation function normally, since they have a normal copy of the *FMR1* gene on their other X chromosome. However, 30 to 50 percent of women with full mutation will have a learning disability, and some have outright mental retardation.

The risk that premutation will expand during oogenesis and result in offspring with a full mutation will vary, based on the number of CGG repeats the woman has; women who have small premutations (55 to 70 CGG repeats) have a 10 to 17 percent risk that the premutation will expand in their offspring; slightly larger repeats (70 to 90) have a 50 to 80 percent risk that the premutation will ex-

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pand, and repeats greater than 90 will have a nearly 100 percent risk of expansion.[7] These figures pertain to only 50 percent of the carriers' pregnancies, because women may transmit their other (normal) X chromosome instead. Males with a premutation transmit them stably to their daughters, who in turn may transmit the gene to their children in either its premutation state (stably or further expanded) or expanded to full mutation state.

Historically, individuals found to carry FXS premutations have been informed of the reproductive risks to their progeny, but *have been told not to worry about their own health.*

FXTAS

Recent studies have suggested that there may be an association between fragile X premutation and a late-onset disorder, fragile-X-associated tremor-ataxia syndrome (FXTAS). While the mechanism for FXTAS is not yet fully delineated, one prominent group of experts on FXS has reported evidence that its cause is this premutation.[8] Signs and symptoms of the disease are said to include tremor, ataxia, and intellectual deterioration. Onset is reportedly during late middle age; pathologic changes have been seen on MRI (magnetic resonance imaging) and at autopsy.[9] Thus far, primarily males have been reported as fully symptomatic, as in the fragile X phenotype.[10] Only a few carrier females appear to show a FXTAS phenotype.[11] Laboratory models employing fruit flies[12] and mice[13] have been reported to provide supportive evidence, and are helping to explore the molecular mechanism of disease.

While they are clinically distinct conditions, FXTAS and the early stages of Huntington disease (HD) have similarities, although the latter has virtually 100 percent penetrance and carries a well-recognized poor prognosis. ("Penetrance" is the likelihood that an individual who inherits a mutation will manifest clinical evidence of its presence.) Both are late-onset degenerative genetic conditions for which pre-symptomatic testing is available. Studies of families with HD report that the revelation of test results can be complex and fraught with anxiety, particularly for those closely related to an affected individual. Some people who are presumed to be at risk want to know their status more precisely through genetic testing; others prefer not to address the issue. An individual's result has implications for other family members. Physicians are discouraged from contacting relatives of a positive HD patient without prior consent from both the patient and the relative.

FXTAS presents different problems than HD because it is not thought to have 100 percent penetrance or relatively early onset of symptoms. Its penetrance may not, however, be trivial; one study suggests that it may be in excess of 75 percent by age 80.[14] Whereas HD is well known, FXTAS is slowly working its way into the lexicon of public health. It is included in the materials available to the general public on the website of the fragile X support group, which could, in time, prove to be a premature elevation of concern among interested parties. To provide families with at least the option of knowing the risks, physicians would have to re-contact current and former patients who are known (from previous workup) to carry FXS premutations.

The received wisdom, in the form of a carefully defined protocol for presymptomatic HD testing, may be useful in considering how to disseminate information about FXTAS. Presymptomatic testing for HD has a well-defined and relatively clear meaning when a pathologic mutation is identified; the results of presymptomatic testing for FXTAS remain ill-defined and uncertain. Thus, counseling for persons at risk for FXTAS should acknowledge current limitations of knowledge about personal prognosis (although familial reproductive aspects are clearly established).

ETHICS BACKGROUND

The *duty to re-contact* is a subset of the *duty to warn;* it is the notion that there may be an ethical and/or legal obligation for physicians to re-contact patients about advances in knowledge that may be of relevance to a previously diagnosed and/or treated condi-

tion.[15] This notion is well-known; for example, if a physician learns in reading medical literature that a medication she prescribed last week has been found to precipitate adverse events in 30 percent of patients, she should seek re-contact promptly and offer to change the regimen. The question of the temporal extent of this obligation remains moot.

If the complication is sudden death, re-contact is an urgent priority. If the complication is relatively minor, the physician may feel satisfied with informing the patient at the next scheduled office visit. It will often be sufficient to establish an office practice that identifies charts of those at newly appreciated risk, so that it is broached at the next visit. Most offices will not have a computerized database to facilitate prospective identification of such charts. Regardless of the severity of the complication, it is reasonable to assume that most people will want to know about their risks.

The American College of Medical Genetics concluded that consulting geneticists should not be expected to convey new information to past consultants, but that primary care physicians hold the responsibility to maintain or renew communications with sources of specialized information.[16] Therefore, the primary care physician is also responsible for recognizing that there may be new information *and* for informing patients of the relevant changes in their prognoses and/or treatments. This has led many clinical geneticists to add a statement to that effect at the end of consultation letters.

But practicing geneticists lack consensus on this issue.[17] Sharpe helped introduce and explore the implications of expanding the notion of duty to re-contact,[18] but did not actually advocate this expansion.[19] Knoppers proposed involving the patient in the discussion of follow-up as part of the initial consultation.[20] Lebel and colleagues studied re-contact with new information about identifiable risks that were not part of the initial consultation, and found patients were unlikely to respond to an invitation for a new consultation.[21]

Bernard and colleagues advocated a duty to re-contact persons at risk for carrier status of *FMR1* mutations, once molecular testing had become available, if these persons had first been seen in the era before the availability of such testing.[22] Such a duty can exist, of course, only after the materials in question have been shown to be firmly established in the scientific literature, a condition not as yet achieved by the researchers of FXTAS. We wish to explore the ethical problems posed by new discoveries such as this proposed clinical entity, in part to prepare the way for applying the existing notion of duty to re-contact to new situations.

PROBLEMS:
BENEFICENCE, AUTONOMY

Individuals found to carry the premutation, after learning of their family history, were originally counseled that the finding had no personal clinical impact. They were informed of the risk of expansion in their offspring, but told their own health was not jeopardized. Re-contacting these individuals to inform them of the discovery of FXTAS seems, *prima facie,* to be the ethical thing to do.

However, with substantially less than 100 percent penetrance for FXTAS, as already noted, re-contact may cause unnecessary stress and anxiety for those who may never express the trait. They may spend the rest of their lives preparing for an illness that never materializes; the added psychological stress may in itself aggravate other latent stress-affected conditions. Consequently, we think that dissemination to the general public of an alert for this entity should be avoided.

Some persons may perceive re-contact as an invasion of privacy and autonomy if they do not wish to know about such risks, to avoid fear of developing symptoms. In genetics consulting, it is well-established that persons have a *right not to know* about factors that may predispose them to unpleasant future developments.[23] This is observed in some of those who seek consultation regarding HD or familial carcinoma, but who elect not to follow-up with laboratory testing, and also in those who choose not to utilize medically indicated genetic testing during pregnancy.

In simply opening the conversation by re-contact, we may reveal a possibility of a matter for concern. Conversely, failure to contact these people could be interpreted as a breach of the duty to re-contact with important new information. They may have implied consent to re-contact with new information, given the fact that they elected to be tested, although they may not have been clearly aware of the possibility of premutation status (if they were part of an extended family analysis after diagnosis of an affected relative). This is notwithstanding the complex psychosocial aspects of knowing one's carrier status.[24]

PROBLEMS: TECHNICAL

The infrastructure of a clinical genetics practice may not support a labor-intensive search for persons that we might wish to re-contact, given technologic advances. Lebel and colleagues searched a computerized database into which were entered, at the time of initial consultation, all of the family history health problems identified in a four-generation family history for some 3,000 consultants.[25] Most had been seen for either prenatal or pediatric questions, but the database included such items as the occurrence of cancers in their first-, second-, and third-degree relatives on both sides of the family.

A couple being seen for consultation because their newborn had Down syndrome, or because of abnormal maternal serum screen results during pregnancy, or because of a history of recurrent miscarriages, would have information recorded that might include the death of a grandfather with stroke, the illness of a cousin with multiple sclerosis, or loss of two paternal aunts with breast cancer.

Contacting such people five to 10 years later (to advise them of the development of predisposition testing to identify persons at risk for breast, ovarian, and colon cancer) proved to be unwieldy, and ultimately futile. Half of those identified as potential beneficiaries from such new information could not be found for re-contact, even when the original referring physician was asked for updated telephone and address information. Not one of those contacted followed through with a consultation.

A complete report is being prepared for publication,[26] but, in summary, it is evident that the doubts expressed by Hunter and colleagues seem to be well founded when the topic for re-contact is not the same topic that drove the initial consultation.[27] If re-contact is for *the same reason* as addressed in the initial consultation, and existing databases make re-contact reasonably easy to accomplish, and the material is of potentially grave concern, then re-contact may in fact be highly useful and appropriate. When and if fragile X premutation carrier status will meet these three criteria remains to be seen.

PROBLEMS: LEGAL

Most patients previously tested for fragile X carrier status were not tested because of concern for FXTAS (an entity only recently delineated, and unknown to anyone when most of the samples were obtained). The purpose of the testing was clinical: to generate information about mutation or premutation carrier status, and to guide further testing in the family. It was not diagnostic or therapeutic. The Health Insurance Portability and Accountability Act (HIPAA) of 1996 states that any "protected health information," including genetic information, must not be disclosed without the specific authorization by the individual.[28]

Does this mean that an individual's *own* genetic information may not be used to inform *him or her* of a possible, newly appreciated health risk without expressed consent? We doubt that this was the intent of the legislation, and think that this information may, and indeed should, be so used. FXTAS should be brought to a patient's awareness only by a primary care physician. We submit that it is appropriate for the reference laboratory, when there is knowledge of FXTAS, to find ways to alert primary care physicians of its potential importance. This opinion rests simply on the following: (1) the information already exists in others' hands, (2) it is relevant to the health of the person who was tested, and (3) the per-

son tested has a right to be warned of the possible health hazards involved.

THE POPULATIONS

Overlapping but distinct problems arise in the ethical analysis for the three populations who must be considered in light of emerging information about newly appreciated significance of *FMR1* premutations.

1. Persons previously shown to carry FMR1 premutations. They are numerous. They often are relatives of individuals who presented clinically with FXS, but who were/are asymptomatic themselves. The effort to identify the incidence of movement disorder in this population has only begun. Some of the major laboratories have databases that may be able to identify these people easily and prospectively.

2. Persons previously identified as at-risk for carrier status who declined to be studied at first contact. These people may wish to reconsider investigating carrier status if they are advised of the new potential significance of being a carrier. They may have declined at the first contact because they did not plan to have children, and so considered the question to be theoretical and uninteresting. But now this information pertains not only to children, but to their own health. These are people who will be difficult to locate, since they elected not to enter the database. Some of them, if contacted, may still elect not to pursue the issue.

3. Persons with movement disorders of uncertain etiology whose previous workups did not include this test. If the movement disorder in this group is similar to, or at least sometimes overlaps with the features of HD and/or Parkinson disease (PD), and/or cerebellar ataxia (CA), then the laboratories that study these clinical conditions for diagnosis or presymptomatic assessment must have seen numerous samples, many of which may still be in storage, from persons whose study was unproductive of a final molecular diagnosis. Identifying the samples would not be difficult. The research under way for the genetic basis of Parkinsonism has probably produced substantial numbers of samples for which no genetic lesion has thus far been identified, but probably *FMR1* has not been investigated for these people. Certainly it is less morally ambiguous for treating physicians to re-contact these individuals, as the individuals must already recognize signs of neurologic deterioration and have begun to seek a diagnosis.

Another population not discussed here consists of families in which a new diagnosis of FXS is made, whether based on clinical assessment of an affected male, or the results of newly introduced neonatal (presymptomatic) screening. These will be families subject not to *re*-contact, but to full provision of pertinent information at the time of new diagnosis. It is clear that geneticists who approach these families for investigation must include what is known of FXTAS in the consenting process for laboratory tests that involve *FMR1* mutation and premutation status.

CONCLUSIONS

As FXTAS is being confirmed in other centers of FXS study, re-contact of populations 1 and 3 above is warranted. It is not clear, however, exactly what risk these populations should be given for the personal health implications from carrier status. Until this deficiency is met, re-contact is probably premature (at least for those in population 2). We run the risk of invading the privacy of these individuals if they prefer not to know, and we run the risk of causing them psychosocial damage if we open the possibility of insurance and/or employment discrimination on the basis of such recognition. Yet we should advise people who had an interest in the initial testing, once we know something more about the implications of their results. It may be difficult to honor their autonomy, since raising the question is a *de facto* disclosure of previously unrecognized risk. In balance, the genetics community will be derelict if we fail to make the effort if/when the specificity of this problem has been established.

As FXTAS becomes better defined and delineated, it should become standard practice to disclose the risks involved (prior to

consent) to all of those tested for FXS carrier status. The uncertainty about re-contact of past patients will be clarified when the impact this information has on patients who will be forced to grapple with it prospectively is better undersood. If it is clear that knowledge about FXTAS has mental and social health implications, our expertise will increase.

One aspect of FXTAS gives physicians and ethicists a distinct advantage: time. The disorder is understood to be late in onset, and some of those at risk will never develop the phenotype at all; that proportion, and the extent to which the premutation might account for signs and symptoms interpreted as other diseases, is as yet not fully defined. Patients in the middle to late age range can be targeted first, to mitigate the initial (potentially overwhelming) breadth of the project. Patients not yet within this range have a much larger window of opportunity for re-contact. Some known premutation carriers are relatively young, as they are siblings of FXS patients, or are siblings of the children's parents, who have been diagnosed by the relatively recent practice of genetic testing.

Once a sufficient amount of information is available, how many years of files should be opened is open to question. We propose no less than five years, but no more than 10 years, on the basis of findings in a long-term re-contact study, which reports that mobility interferes with re-contact after a decade.[29] How vigorously we should seek updated contact information is another problem.

We propose that a single effort with the existing records suffices to discharge a reasonable duty. In regard to population 1, there are two approaches. Individual physicians may send a letter, or make a telephone call, to attempt re-contact. Reference laboratories may alert referring physicians to the new information pertaining to previously ordered and reported studies. These actions should be adequate, when performed in good faith.

Population 2 does not warrant a re-contact effort since it consists of persons who, at first contact, elected not to participate, and whose contact information is presumably not available. Persons in population 1, when advised of the newly appreciated risks they face, may be encouraged to re-invite their relatives who constitute population 2. In the event that such persons re-enter the circle of discussion by asking a new question or consulting about plans for initiating pregnancy, then full disclosure regarding FXTAS (as well as FXS), obviously is obligatory.

In regard to population 3, geneticists already in contact with people who have had nondiagnostic study of genes associated with HD, PD, CA, et cetera, may re-contact them with the option to extend the laboratory effort to include *FMR1* premutations. Presumably, an appropriate provision of genetic counseling has prepared those patients for the DNA analysis that was undertaken in their workups thus far. Professionals should be prepared to extend genetic counseling to include implications for the patients' children and grandchildren. In work that is already under way on seeking genetic markers for Parkinson families, revisiting samples to identify *FMR1* premutations will open up an entirely different avenue of concern and discussion for those families. This is well worth the effort.

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Child-to-Parent Bone Marrow Donation for Treatment of Sickle Cell Disease

Lisa Anderson-Shaw and Kristina Orfali

Natalie Johnson (not her real name) was a 25-year-old woman who had endured the chronic symptoms of sickle cell disease (SCD) since shortly after her birth. The symptoms of her SCD had progressed over her short lifetime from joint pain and swelling, in early childhood, to more severe pain that was difficult to manage as a teenager, with organ involvement as a young adult.

Natalie had always been under a doctor's care for her SCD. The hematology staff at a local hospital followed her consistently since she was a child. Between the ages of 11 and 14, she experienced sickle cell crises for which she was hospitalized each year; however, after age 14, her crises increased in frequency and she required hospitalizations every three months until age 24, when she was hospitalized approximately three weeks out of every month within that year. During this time of frequent crises, her hematologist began to discuss the possibility of bone marrow transplantation (BMT) with her. She was told that if a BMT was successful, she might be cured of her SCD. At that time she was very positive about this option, and began to pursue this with her mother and her doctors. Before the search for a potential donor took place, Natalie found out she was pregnant.

After a very complicated pregnancy, Natalie delivered a healthy baby girl named Faith (not her real name), and cord blood was collected and stored. Faith's biological father was not present for the birth nor was he involved with the baby after her birth. Natalie had not been in contact with him since before Faith's birth. Soon after the birth, Natalie began pretransplant therapy, but could not tolerate the treatments due to severe pain and fatigue. At that time, unfortunately, her right hip joint began to show signs of deterioration from her disease, and she had hip replacement surgery. She recovered well from this surgery, but continued to have severe and frequent

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pain crises, requiring her to take daily pain medication and resulting in frequent admissions to the hospital. She was not physically strong enough to continue with her transplant at age 25.

Her symptoms continued to progress to the point that her quality of life was severely compromised. Her hematologist believed that BMT was her best chance to improve the quality and length of her life. Her physician also believed that, without this intervention, Natalie had a life expectancy of 10 years or less, with progressive degeneration of her joints, severe organ involvement, and intractable pain. Natalie agreed to undergo BMT. Her mother, father, and brother agreed to be tested for donation, but her physicians wanted to test Faith's cord blood first. Should Faith be a close enough match, Natalie would have less chance of rejection and other related complications. Surprisingly, her daughter's human leucocyte antigen (HLA) was found to be identical to her own, and therefore Faith might be an excellent donor. In addition to stem cells taken from Faith's cord blood, the transplant team wanted to harvest her bone marrow to have an adequate volume of the cells that would be needed for Natalie. Natalie agreed to allow her three-year-old daughter to donate bone marrow for her own transplant.

It is at this point that the hematology staff contacted the institutional clinical ethics consult service so the ethical issues surrounding the use of a child-to-parent BMT, as a means to cure the parent's SCD, could be explored. Children have been used as bone marrow donors in siblings for many years, and BMT in children with SCD has been done for the past 20 years. In this case, however, the child would not be a donor for a sibling, but for a parent, which the team had done only one other time.

This article will explore several of the ethical issues raised by the clinical ethics consult service, medical and legal staff, and the patient and her family including the following: Should a mother be allowed to consent for her young daughter's participation in a procedure that is solely for the mother's own physical benefit, which includes a risk of harm to the small child? Whose best interest is being served by going through with this procedure? Is there a conflict of interest involved, and, if so, should it impede the BMT process? What obligations exist on the part of the transplant team to Natalie and her daughter in preparation for this procedure?

We will also explore some of the less obvious ethical concerns related to the emotional and social transition that Natalie encountered as a result of her experience before and after her BMT.

INTRODUCTION

BRIEF OVERVIEW OF SCD

SCD is a fairly common genetic disorder, affecting 50,000 Americans, and seen most often in the United States within the African-American population. This disease affects the synthesis of hemoglobin, causing various symptoms throughout the body, including "hemolytic anemia, splenic dysfunction, pain crises and bacterial infections."[1] SCD continues to run a progressive course of organ destruction in those afflicted, for which there is currently no conclusive cure.

Although screening for the early detection of persons with SCD is routinely done, treatments often do not successfully manage symptoms or halt progression of the disease. It is estimated that the average lifespan for a person with SCD is between 42 and 46 years of age.[2] Throughout the course of this disease, the person suffers much physiological and psychological stress. Pain and symptom management regimes often cause reliance and dependence on healthcare systems. Infection, which tends to "occur in previously damaged areas such as the lungs, kidneys, and bones,"[3] threatens people with SCD throughout their lifetime. The frequency of pain crises peaks in young adults with SCD, and is accompanied by musculoskeletal strain and painful joint swelling.[4] Traditionally, standard therapy focused on preventative action and symptom-based treatment in the acute phase. Symptoms of SCD may vary from mild to severe in indi-

viduals;[5] in this case, Natalie suffered from very severe clinical symptoms. Only recently has BMT been offered to adult patients as a treatment for SCD, and it is reported that BMT may offer the only curative therapy.[6]

DISCUSSION

BMT FOR SCD

BMT for SCD in children has been performed since the mid-1980s. Studies report a cure rate among children who have had BMT to be greater than 80 percent, with rates of transplant-related morbidity (such as graft-versus-host disease, infection, adverse drug affects from the medications associated with the transplant itself) and mortality at about 10 percent. This research reports that children in the early stages of SCD have better outcomes from BMT than those in more advanced stages.[7] The donor pool is often restricted, because bone marrow from a related HLA-matched donor is therapeutically preferable to bone marrow from a nonrelated donor. A familial match is optimal, but is only rarely available. Future research on unrelated HLA-matched donation from adult donors or from newborn stem cells contained in umbilical cords may improve the chances of successful BMT for those in need.[8] In a recent review of 127 pediatric patients who received HLA-identical sibling donor BMT, 77 percent (98 patients) were free of SCD at the three-year mark.[9] Hoppe and Walters note, "since the first allogeneic transplant for SCD in 1984, approximately 175 children with SCD have received matched sibling-donor HCT [hematopoietic cell transplantation] . . . with overall 82 percent survival free from sickle cell anemia."[10] Davies and Roberts report an overall graft rejection rate of around 10 to 15 percent.[11]

Given all of the morbidity and early mortality otherwise associated with SCD, it would appear that BMT/HCT can significantly alter the course of this disease. The evidence appears favorable for children, but what about adults with SCD — can BMT/HCT help adults suffering from SCD? Unfortunately, adult patients with SCD have not usually been considered appropriate candidates for the therapy. This may be because, by the time a person with SCD reaches adulthood, significant end-organ damage may have occurred, which would prohibit the toxic conditioning regimes required for HCT.[12] However, should the SCD be cured in adult patients, they may be able to continue a life free of the debilitating symptoms of this disease. In fact, van Besien and colleagues surveyed adult patients with SCD, and found that many patients would be interested in HCT as a curative measure and would be willing to accept considerable risk to undergo it.[13]

Given that BMT/HCT for SCD has been performed for over 20 years in children, should it be considered experimental for adults in whom BMT has rarely been used for this purpose? Perhaps not, since BMTs/HCTs have been performed on hundreds of adults through the years for other diseases, for which the outcome data are widely available. Research is designed to systematically investigate a particular hypothesis, and is thus subject to various federal regulations designed to protect human subjects. BMT for SCD has been adequately researched in pediatric populations. As Kodish and colleagues noted in 1990, the use of BMT in this case would simply be for a new indication; therefore, it might be accepted as a nonexperimental therapy.[14] Although the new indication for an old therapy may not be considered experimental in nature, there would seem to be a strong obligation on the part of physicians and institutions that the proposed indication be well studied and reviewed by known experts in the field. And, since performing BMT/HCT in adults may not be considered an urgent procedure, perhaps review by an institutional review board would be helpful to clarify any patient-protection issues that may not have been initially obvious.

Should a mother be allowed to consent for her young daughter to participate in a procedure that is therapeutic for the mother that includes nontherapeutic risk for the child? This discussion must take into account that

children have long participated in research and nontherapeutic interventions for which they receive little or no benefit. An example of a nontherapeutic intervention would include a minor sibling donating bone marrow to his or her sibling who has cancer. The ethical issues surrounding the participation of children in research has been debated for years. In reality, however, curative treatments for many childhood diseases would not have been possible but for the participation of children in research. Cancer therapy research trials, for example, are often offered for children who may not be of the age of consent or assent, as the case may be. As children reach teenage years, they are often asked to participate in the decision-making process and may be asked to give their assent for participation in research trials. Even in cases when assent from the child is sought, it is the parent or guardian who must consent for a minor child to participate, in hopes that the child will benefit from the therapy, or, at the very least, other children might benefit.

In Natalie's case, however, the therapy in question is not to treat her child's illness, but rather to treat her own illness. In this case, Faith is a three-year-old potential bone marrow donor who is not able to understand the personal health risks associated with this donation. Faith may understand that she will be experiencing the pain of the procedure to help her mother, but it is difficult to know how much of this she will truly understand. But, as with most healthcare decisions made for small children, her mother and the healthcare team will probably have her best interest in mind, as they proceed with the treatment plan. One could argue that it is in the toddler's best interest to have her mother healthy — to be free of SCD so that she may live longer and be able to give Faith the physical and emotional care that she needs without the interruptions that SCD causes. One could also argue that it is in the toddler's best interest to know, down the road, that she was allowed to help her mother. If not given this opportunity by those who are in the position to do so, she may feel a great sense of sadness and anguish over not being given the chance to help her mother when she needed her. McCormick argues that parental consent in such cases would be morally valid "insofar as it is a reasonable presumption of the child's wishes." In addition, human beings — including children — are social in nature, and through our social interaction we wish to support our own values and the values of those around us.[15] It is reasonable, then, to presume that Faith's wishes would be to donate her own bone marrow to help her mother. Lantos discusses the concept of "involuntary altruism" as it relates to children as organ donors. He states, "in certain situations, parents may allow or encourage their children to act in ways that are detrimental to their interests in order to benefit another person. In other words, parents can choose, on behalf of their children, to have the children act altruistically."[16] Delany agrees that children should be allowed to exercise altruism and be given the right to be bone marrow donors through the consent of their parents.[17]

MORAL DECISION MAKING AND THE FAMILY

Ethical analysis involves looking at the moral decision-making process within the context of this family as well as the related ethical principles of respect for autonomy, beneficence, and nonmaleficence.[18] The burden of legal and moral consent for Natalie and Faith in this case is Natalie's. She must consent for herself and for her daughter so that these procedures can proceed. She is the autonomous agent for herself and for her young child. However, we suggest that the moral nature of this consent decision be taken within the context of the family, which may be defined as "a set of people who are indefinitely committed to care for each other and for those dependent on them."[19] The members of this immediate family include Natalie, Natalie's mother (with whom Natalie lives), and Faith. All three members are stakeholders in this decision and in its ultimate outcome. Family

members have a crucial role in moral decision making for children.[20] This is consistent with a family-based model of moral reasoning. In this situation, the decision for both Natalie and Faith involves the context of illness and potential cure for Natalie as well as the interrelatedness of Natalie, Faith, and Natalie's mother, all within this same context. As such, then, the moral decision to allow Faith to be a donor for Natalie is consistent with respect for a family decision-making model. In Natalie's case, her three-year-old daughter is the only one who can provide her with the treatment that can enhance her life, which in turn enhances her own life, and the life of Natalie's mother.

CONFLICT OF INTEREST

A common complaint when a proxy decision has been made that directly benefits the proxy is that a conflict of interest is present. When a conflict of interest is present, red flags often go up regarding a hidden motive that may lurk behind the otherwise accepted proxy decision. In Natalie's case, we suggest that a bilateral/collateral conflict of interest is present: that is, Natalie's decision to allow Faith to be a BMT donor directly benefits both. Therefore the principle of beneficence is clear, although on the surface one might argue that the principle of nonmaleficence may be violated, as Faith will sustain some physical harm during the procedure. The role of mother and the role of the child are complimentary here, because they have a joint interest unique to their family. It would be impossible to separate the interest of the mother from the interest of the daughter. Further, it may be argued that Faith has a moral obligation to be a donor to her mother, based "on their degree of emotional relatedness."[21] This obligation is directly related to the risks of BMT.

Nonetheless, the autonomy of Natalie to make decisions for her daughter and to act as her agent must be viewed within the familial context. Ross discusses the rights and duties of parents to children, within the context of the family unit, which would allow a parent to make a decision for a child to serve the interests of the family as a unit: "Parents must have the freedom to consider their own needs and interests provided that they have ensured for the provision of their child's basic needs."[22] For Natalie, her mother, and her daughter, a viable family goal is for Natalie to be free of debilitating SCD, which would allow her to better meet the needs of her daughter and to be less of a burden on her mother. In such a familial context it is impossible to separate individual interests from familial interests. However, it would not be acceptable for Natalie to consent for Faith to undergo a procedure for which the potential harm to Faith was so great, or greater than the potential benefits to Natalie, unless a proportionate familial goal or interest would then be attained.

What are the potential harms of this procedure for Faith? The harvesting of bone marrow from a donor has been described as simple and generally safe — with pain as the most common complication.[23] In addition, there are risks associated with general anesthesia. However, given that the overall risk of bone marrow donation from a three-year-old child is relatively low, Faith's interest in helping her mother, based on her emotional relatedness and notwithstanding her status as a child, should be taken into account.

To better quantify this conflict of interest, the known risks to Natalie's daughter should be weighed against the known benefits to herself. The risk to the child should be justified by the expected benefits to Natalie. As previously discussed, both Natalie and her daughter have a substantial interest in Natalie's good health: Natalie would be free from the progressively debilitating disease that has caused her pain and loss of mobility, and Faith would have her mother around to raise her and to be her mommy.

Others may be interested in seeing this procedure take place. For example, the physicians may wish to do this procedure because it not only helps Natalie and subsequently her daughter, but it might also promote their reputation, their program, and ultimately, their

institution. None of the interests described would be wrong in and of themselves; however, the interests of the physicians and their institution should weigh less than the interests of Natalie and her daughter.

What obligations exist on the part of the transplant team to Natalie and Faith in preparation for the procedure? The obligations on the part of the transplant team appear to be several. First, the team should be honest and open with Natalie about the treatment options available to her, BMT being one. She should be given information on this procedure as it relates to children, and be informed that it has only been performed on a very small number of adults for the treatment of SCD. Natalie's physicians felt that her disease would progress rapidly without BMT, so the option to wait until Faith was old enough to fully participate in the consent process was not thought to be clinically appropriate. Without the transplant, Natalie's other clinical options would be to treat her symptoms and complications as they arose, knowing that her condition would be progressive and debilitating, and ultimately lethal.

The team should provide Natalie with anticipatory counseling about this procedure related to (1) the risks to her daughter, (2) the expected benefits to herself, and (3) the postoperative phase of BMT, including medication regime and their side-effects.

She should meet with a social worker and be encouraged to provide an advance directive to guide clinicians, should she lack decisional capacity after the procedure. She might also need to explore childcare options for her daughter, in case she has serious complications, and perhaps have a legal guardian, such as Natalie's mother, named in advance, as the child's father is not involved. Perhaps a child advocate should be appointed to assist when a parent must consent for the use of her child as a bone marrow donor. This would allow for a somewhat objective review of the situation and may help to insure adequately informed and voluntary consent.[24]

ETHICS CONSULT FOR A FAMILY/TEAM MEETING

Natalie's medical team requested that the clinical ethics consult team meet with the team, family, and institutional legal representative to discuss the ethical issues prior to the proposed BMT. All of the issues previously described were discussed in detail, and all of the known risks and benefits to donor and recipient were reviewed. The authors, the clinical ethics consultants, further explored the need for Natalie to fill out some form of advance medical directive and for Natalie and her mother to look into guardianship for Faith, should Natalie die during the treatment. Natalie and her mother followed-up with the institutional legal affairs office to complete the appropriate legal paperwork that made Natalie's mother Faith's legal guardian, if needed.

At the conclusion of the meeting, all present believed it was appropriate for Natalie to consent for her daughter. All believed that appropriate information had been presented to allow Natalie to make an informed decision. The ethics consultants made themselves available to the team and to the family for follow-up.

Critical ethical analysis of this case is not simply an individual risk/benefit evaluation, but a more intimate analysis of the individuals in the context of the family unit. Within a family-based model of moral decision making, respect for the interests of each member within the family is taken into account. The principles of beneficence, nonmaleficence, and respect for autonomy are also evaluated within the familial context. Concurrently, we believe that there is a stronger obligation on the part of Faith to assume the risk of this procedure on behalf of her mother, even though it is her mother who ultimately allows Faith to fulfill this obligation.

Based upon post-procedural interactions with Natalie and her medical team, we realize that we should have suggested in advance that Natalie be assisted by a counselor who has experience in chronic illness and in the

issues that may come up should Natalie be sickle-cell free after the BMT.[25] For instance, would Natalie be able to function in her new context? Would she have psychological addictions related to her role as a SCD "patient"? Did she have an addiction to pain medication, and, if she did, how would she deal with addiction when she no longer needed SCD-related medication? What should Natalie expect after the procedure? Would she be seen in the clinic often? Would she still need pain medicine?

TRANSPLANT OUTCOME AND FOLLOW-UP

At two-year follow-up, Natalie felt "like a new person," and five-year-old Faith had adjusted well, and was a typical kindergarten student. However, the two years after transplant required a series of adjustments for Natalie and her family. Natalie recalls that her childhood was not easy because of her SCD. She missed too many days of school to track, missed field trips with her school friends, and could not participate in most sports for fear she would have a crisis. She missed teenage dances and other social events and was hospitalized frequently for long periods, even spending several holidays in the hospital. Her life and the life of her mother revolved around her illness. Her mother was dedicated to her, not experiencing the independence that her friends had. She often missed work to care for Natalie and sacrificed to make sure that Natalie received the healthcare she required. The first year of Faith's life, Natalie was in the hospital 36 weeks out of the year.

After she made the decision that she would have the BMT, Natalie began to daydream about what it would be like to be "normal": she wondered what it would be like not to go to the hospital every month or not experience the frequent pain that was so much a part of her life; she dreamed of running outside and playing in the snow without the fear of triggering a crisis. Natalie was very optimistic and was confident in her physicians. She had read all she could find on the topic, and when the time came for her transplant, she and her family were ready.

When the transplant was over, she was told that it was a success and neither she nor her daughter experienced any complications from their procedures. She was cured of SCD. She was normal. She was discharged and expected to do very well.

Although her physicians believed her to be medically cured of her SCD, her transition from years of SCD to being physically cured of the disease was not easy. After several visits to her doctors still complaining of intense pain, her doctors became frustrated that she continued to ask for prescription pain medication, and even suggested that she had addiction issues for which she was referred to a therapist and addiction specialists. She began to feel abandoned by her physicians, who had always been so attentive prior to the BMT, but now appeared frustrated that she continued to behave as though she still had SCD. Her first year post-transplant was emotionally difficult and filled with frustration. She had not been prepared for the intense emotional transition that was taking place. She dreamed about being cured, but was not prepared to leave the only world she had known — the world which evolved around pain, doctor visits, hospitals, and medication. We explored elsewhere the difficult transition from chronic illness to cure that Natalie experienced.[26]

At two and one-half years post-transplant, Natalie and her family were finally able to adjust emotionally to her new physical health. She came to realize that she had not been prepared for the major life changes that occurred after BMT. Her healthcare team did not anticipate the psychosocial adjustments that might have prompted pretransplant therapy, as BMT in adults for SCD had rarely been done.

Natalie is forever grateful for the success of her treatment and is letting her guard down a bit and moving on with her life, enjoying her new life with her daughter and mother.

CONCLUSION

In specific circumstances when an adult BMT/HCT match cannot be found, we believe that it is acceptable for a child to serve as bone marrow donor to his or her parent. It is always preferable to use matched related donors when possible, rather than a nonrelated donor via a bone marrow registry. BMT/HCT has been shown to be successful in curing children of SCD, enhancing quality and length of life, while the risk of treatment-related mortality appears reasonable. It has not, however, been used for treatment of SCD in many adults.

No long-term data exist on whether the risk to a child donor is justified by the potential benefits to a parental recipient. Preliminary data suggest that this procedure may be helpful to adult patients, but is less helpful than this procedure has been in their pediatric cohort.

There is room for future research related to the use of children as living organ/tissue donors to their parents, or even to other adult family members. Should there be an age limit on when a child can be involved as a living organ donor, specifically BMT or HCT donation? Should the institutional ethics committee be systematically involved when a child is sought as a living organ donor for a minor sibling or an adult family member? How involved should the child be in the decision to become a living donor? Clearly, the older the child is, the better understanding he or she might have regarding the procedure. Should a child advocate be assigned to each child who is being considered for bone marrow donation, and, if so, what are the parameters of the role as advocate?

In addition, close psychosocial follow-up should be provided to these patients to better address their specific post-transplant needs.

Finally, new models of ethical decision making on behalf of vulnerable populations, as with minor children, may need further exploration.

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Diagnosing PVS and Minimally Conscious State: The Role of Tacit Knowledge and Intuition

Mary Terrell White

INTRODUCTION: THE CASE OF TERRI SCHIAVO

Diagnosis of the cognitive capacity of profoundly brain-injured patients is frequently ambiguous and subject to change, potentially leading to charges of diagnostic error, patient mismanagement, and conflicts between family members, healthcare personnel, and the courts. This has been vividly illustrated in the case of Terri Schiavo, a young Florida resident who died a year ago, 15 years after the night her heart temporarily stopped beating. In 1990, Ms. Schiavo suffered cardiac arrest, possibly due to a potassium imbalance, which left her with significant brain impairment. She was diagnosed as in a persistent vegetative state and placed in a nursing home, where she received food and hydration artificially, as she could not swallow. In 1998, her husband, Michael Schiavo, first petitioned a Florida court to determine whether the tube feeding could be withdrawn. While the judge ruled in his favor, Ms. Schiavo's parents objected and appealed the judge's decision. For patients in a persistent or permanent vegetative state, withdrawal of life supports may be permitted as long as there is clear and convincing evidence that is what the patient would want, and which Mr. Schiavo claimed he had. But Ms. Schiavo's parents were not ready to give up hope, perhaps responding to the spontaneous movements she continued to make.

In the years that followed, the conflict between Mr. Schiavo and his wife's parents continued in the courts, Mr. Schiavo pressing for the right to withdraw treatment, Ms. Schiavo's parents claiming she possessed residual consciousness and with proper therapy could improve. Even though the judges consistently ruled that Ms. Schiavo was in a persistent vegetative state, the conflict eventually led to intervention by the Florida legislature and governor to maintain her life supports through a hastily written law that ordered the reinsertion of her feeding tube. The Florida Supreme Court ruled the law unconstitutional a year later. Then, in early 2005, after the U.S. Supreme Court declined to hear the case, a trial court judge ordered that the feeding tube be removed in 30 days. Two days after the tube

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was removed, a few U.S. senators, acting on behalf of the entire Senate, passed a law entitling a U.S. District Court in Florida to hear the case again. This legislation was approved by a majority of the House of Representatives and signed by the President of the United States. The following day, a U.S. District Court judge denied the parents' request for a restraining order that would mandate the reinsertion of the tube. Ms Schiavo died 31 March 2005.[1]

This case raises many questions, perhaps most provocatively having to do with the proper role of state and federal legislatures in medical decision making. The current political climate has also made it difficult to separate the case from the political agendas of some of the players involved. Nonetheless, Terri Schiavo's ordeal raises legitimate questions of how cognitive capacity is assessed in severely brain-injured patients and how accurate such diagnoses can be. Clearly, misdiagnoses may result in denial of appropriate care. In this discussion, I propose that an additional, if unorthodox, approach to cognitive assessment may improve diagnostic accuracy. Following a brief review of the clinical criteria for persistent vegetative state and minimally conscious state, I examine the incidence and sources of error in diagnosing cognitive capacity in brain-injured patients. I then propose that other kinds of knowledge, namely tacit knowledge and intuition, may provide valuable diagnostic evidence, and conclude by outlining steps necessary to integrate these forms of knowledge into current practices. I focus exclusively on the diagnostic process; I do not address moral questions involving the rights or interests of persons in a minimally conscious or persistent vegetative state, or whether severely brain-injured patients are owed any specific forms of treatment.

DIAGNOSIS OF PVS

At present, the diagnosis of persistent vegetative state (PVS) is made primarily by clinical observation, drawing on a number of neurological and behavioral factors. Functional neuroimaging via PET (positron emission tomography) and fMRI (functional magnetic resonance imaging) may offer confirmation of residual brain function, but at present, these technologies are too complex and their interpretation too uncertain for them to be routinely used in cognitive assessments of profoundly brain-injured patients.[2] Until these or other diagnostic technologies are better developed and understood, physicians will rely on their clinical judgment in assessing the cognitive capacity of severely brain-injured patients.

The current clinical guidelines for the diagnosis of PVS were established in 1994 by a Multi-Society Task Force comprised of a range of experts from diverse medical specialties. In these guidelines, the vegetative state is defined as,

> a clinical condition of complete unawareness of the self and the environment, accompanied by sleep-wake cycles with either complete or partial preservation of hypothalamic and brain-stem autonomic functions. The condition may be transient, marking a stage in the recovery from severe acute or chronic brain damage, or permanent, as a consequence of the failure to recover from such injuries. The vegetative state can also occur as a result of the relentless progression of degenerative or metabolic neurological diseases or from developmental malformations of the nervous system.[3]

Diagnosis is usually made after a person is in a vegetative state for a month, regardless of whether the cause of brain injury is acute traumatic or non-traumatic brain injury, neurodegenerative disease, a metabolic disorder, or congenital malformation. Diagnosis is based primarily on the following criteria.

1. No evidence of awareness of self or environment and an inability to interact with others.
2. No evidence of sustained, reproducible, purposeful, or voluntary behavioral responses to visual, auditory, tactile, or noxious stimuli.

3. No evidence of language comprehension or expression.
4. Intermittent wakefulness manifested by the presence of sleep-wake cycles.
5. Sufficiently preserved hypothalamic and brain stem autonomic functions to permit survival with medical and nursing care.
6. Bowel and bladder incontinence.
7. Variably preserved cranial-nerve reflexes (pupillary, oculocephalic, corneal, vestibulo-ocular, and gag) and spinal reflexes.[4]

The most distinguishing characteristic of PVS patients is the presence of sleep-wake cycles coupled with a seeming lack of any degree of self-awareness, awareness of environment, response to stimuli, or intentional behavior. Patients in this condition may move their limbs and occasionally smile, cry, or scream, but their movements and vocalizations appear to be meaningless. Some patients have startle responses; these appear to be reflexive responses to external stimuli rather than prompted by conscious awareness. Patients in this condition also do not track objects with their eyes or react to threatening gestures. An early sign of recovery from PVS is that the eyes begin to track.[5]

DIAGNOSIS OF MINIMALLY CONSCIOUS STATE

The minimally conscious state (MCS) is distinguishable from PVS by evidence that a patient possesses some degree of self-awareness, ability to act volitionally, and awareness of the environment. The presence of one or more of the following behaviors is considered evidence of minimal consciousness.

1. Following simple commands
2. Gestural or verbal yes/no responses (regardless of accuracy)
3. Intelligible verbalization
4. Purposeful behavior, including movements or affective behaviors that occur in contingent relation to relevant environmental stimuli and are not due to reflexive activity. Some examples of qualifying purposive behavior include:
   - Appropriate smiling or crying in response to the linguistic or visual content of emotional but not to neutral topics or stimuli,
   - Vocalizations or gestures that occur in direct response to the linguistic content of questions,
   - Reaching for objects that demonstrates a clear relation between object location and direction of reach,
   - Touching and holding objects in a manner that accommodates the size and shape of the object,
   - Pursuit eye movement or sustained fixation that occurs in direct response to moving or salient stimuli.[6]

These behaviors may be inconsistent but must be reproducible or sustained long enough to be considered indicative of cognition, not just random or reflexive behavior. For some minimally conscious patients, sensory deficits or motor dysfunction may limit their responsiveness such that evidence of consciousness may be overlooked or may take additional time to be observed.

DIAGNOSTIC AMBIGUITY AND RATES OF ERROR

Several studies suggest that errors may be made in diagnosis of PVS such that some patients diagnosed as vegetative may in fact possess some minimal degree of consciousness or awareness. This is not surprising, for patients for whom improvement is possible, changes may be so minimal or gradual they may not immediately be detected. In one study, 193 severely brain-injured patients admitted for in-patient neurorehabilitation in a Texas facility were reviewed for diagnostic accuracy; 49 of the patients carried a pre-admission diagnosis of PVS or coma after more than one month post injury. The patients' ages ranged from 11 to 62, with an average age of 28; gender distribution was roughly equivalent. Of these 49 patients, 18 (37 percent) received a change in diagnosis at the time of admission or shortly thereafter. Of these 18,

50 percent were recognized as possessing some degree of awareness the first day. Of the patients whose brain injuries were trauma-induced, 41 percent (14 of 34) were found to have been diagnosed inaccurately prior to admission. Misdiagnosis was more likely in patients who were over three months post-injury (48 percent, or 11 of 23), than those who were only one to three months post-injury (27 percent, or three of 11). Of patients whose brain injuries were due to non-traumatic circumstances, 27 percent (four of 15) were misdiagnosed. The time elapsed since the injury was not significant for this group.[7]

The authors of this above study suspect that part of the reason for the frequent misdiagnosis is the ambiguity of the language used to describe variations and changes in levels of consciousness that could lead to interchanging diagnoses of coma with PVS. Moreover, the various scales used to classify cognitive capacity are not consistent, and none is considered to be a "gold standard." The authors of the study also noted that discrepancies in diagnosis may be due to lack of careful, extended observation, especially as improvements in cognitive capacity can be slow in coming, erratic, and very difficult to discern.

A second study conducted at a rehabilitation center in London examined 97 adult patients admitted for severe brain damage; 40 of these patients were diagnosed as in PVS due to acute-onset brain damage. Of these, 43 percent (17) were found to be misdiagnosed on admission, including seven who had been considered vegetative for over a year. Of the remaining 23 patients, 25 percent (10) remained vegetative, and 33 percent (13) slowly gained minimal degrees of function over the course of the rehabilitation period. The main methods of assessing awareness were the ability to follow a simple command to press a buzzer switch or to look at a named object. Evidence of cognitive function was confirmed by at least two members of the medical team.[8]

Of the 17 patients who had been misdiagnosed, 10 suffered from traumatic brain damage, four from anoxia, two from vascular causes, and one from encephalitis. No limits were placed on the time elapsed since the brain damage. Of note in this study is that all of the misdiagnosed patients suffered from severe physical disabilities, 11 being blind or severely visually impaired. These deficits may well have diminished their responsiveness, further complicating the diagnostic process.[9]

Lastly, a study of patients with Alzheimer's disease sought to determine whether the diagnostic criteria for PVS could be applied to these patients;[10] 12 patients with advanced Alzheimer's disease were evaluated by three neurologists on two occasions, two months apart. Diagnostic criteria included the criteria for PVS developed by the Multi-Society Task Force, several tests of cognitive function, and an interview with a staff nurse who knew the patients well. No consistency was found between the positive findings of the three neurologists; moreover, some physicians changed their initial diagnoses at the second visit. The diagnostic discrepancies were explained as likely due to subjective factors as well as patients' fluctuating cognitive status, limited time available to neurologists, and dismissal of staff reports of responsiveness. The authors conclude that, given this disagreement, it is likely that Alzheimer's disease only rarely progresses to the point of PVS.

SOURCES OF ERROR

Changes in status are to be expected among the severely brain damaged, particularly in the first months after traumatic brain injury. Some degree of recovery is possible even after one year in PVS. In general, the younger the person is, the greater the likelihood of recovery. Recovery is also more likely if the cause of brain injury is traumatic rather than nontraumatic.[11] Thus, some changes in diagnosis should be assumed as reflecting accurate observations. However, that so many patients in these studies were found to be misdiagnosed long after their injuries suggests that diagnosis of PVS is prone to error.

Some errors may be due to confusion in terminology. As was noted in one of the studies, "the medical and legal literature does not

fully adhere to the distinction between coma and PVS and frequently uses the terms interchangeably."[12] Even more problematically, we lack a consensual definition of consciousness or language with which to describe it. The terms *consciousness* and *awareness* may be used either synonymously or to refer to distinctly different states of being, while *arousal, orienting, alertness,* and *wakefulness* are frequently used interchangeably to describe various stages between sleep and wakefulness.[13]

Part of the problem may also be the kinds of tests used to establish consciousness. Many of the tests for MCS seem to assume that the patient both wishes to respond (that is, is not apathetic or depressed) and is able to hear and see. These expectations may be unrealistic, setting the bar too high. For example, the tests used in the British study — of following commands to press a switch or to look at particular objects — imply or assume that brain injury has not affected volitional movement, hearing, or sight. Most of the patients wrongly diagnosed as PVS in this study were discovered to be blind.[14]

Errors in clinical judgment are another source of error. It is common knowledge that medical judgments are influenced by various heuristic strategies common to cognition. While these heuristic devices enable rapid decision making, they may lead to biased diagnosis. A few heuristics relevant to diagnoses of PVS include the following. *Confirmation bias*: a tendency to look for evidence that can support (or "confirm") a diagnosis, rather than evidence that refutes it. *Diagnosis momentum:* once a diagnosis is made and associated with a patient, with time, it becomes more and more difficult to change it. *Overconfidence bias*: a tendency to think we know more than we do, leading us to act on incomplete information; it can also lead us to place our faith in opinion rather than evidence. *Premature closure*: a tendency to accept a diagnosis as valid before fully verified, as in the maxim, "When the diagnosis is made, the thinking stops."[15]

Perhaps needless to say, diagnostic error may also result from the influence of unconscious emotional reactions. The physician who thinks he would rather die than have his or her life prolonged in a PVS or MSC may be more willing to establish grounds for withholding care, while the physician who believes life is a good to be preserved at all costs may be reluctant to concede that a patient is likely to be permanently vegetative. The strength of this sort of emotional influence, however, is difficult to verify. Finally, as Childs, Mercer, and Childs note, errors may result from

> a lack of extended observation for behavioral evidence of cognitive awareness by qualified personnel. Changes may be "slow and subtle, involving increasing consistency of responses which may initially appear random or coincidental." . . . Responses in these low-level patients are typically erratic, and health care providers may discount the reports of responses seen by family members or other caregivers. If untutored in assessment of consciousness, the physician examining the patient briefly on rounds may not see signs of awareness.[16]

Clearly, part of the problem in diagnosing the cognitive capacity of severely brain-injured patients is that volitional behaviors may be very subtle and sporadic. Physicians who only have time for a brief exam may not see these behaviors, and yet they alone are responsible for making diagnoses. But if some degree of consciousness remains in some patients, how can that consciousness be perceived, by whom, and how can that information be integrated into the diagnostic process?

SOURCES OF EXPERTISE: TACIT KNOWLEDGE AND INTUITION

While rarely taken seriously as medical authorities, family members and caregivers who are present with patients for continuous periods of time are obvious potential sources of patient information. Like Terri Schiavo's parents, they often claim to be able to interpret brain-injured patients' behaviors when

strangers to the patient only see random activity. For example, in a study of 13 children diagnosed as in a PVS, 82 percent of parents (12 of 13 cases) felt their children could recognize their voices, and 62 percent of parents (eight cases) felt the children could make their likes and dislikes known.[17] But, as noted in this study and studies cited above, physicians tend to discount the testimony of family members and nursing staff, under the assumption that their perceptions will be distorted by their attachments to the patient.[18]

Families and caregivers may gain some standing if they are recognized as possessing valid expertise germane to the diagnosis. But in what ways can family members be considered medical experts? Physicians undergo years of study to learn to detect and interpret symptoms of diverse diseases in a broad range of patients. By contrast, the expertise family and caregivers have is at best limited to behavioral observations of a single patient. This is not generalizable knowledge, but subjective interpretations of very small gestures that may be meaningful only to persons intimate with the patient. Since these behavioral responses may be difficult to identify or describe to others who are less familiar with the patient, they are difficult to confirm and easy to dismiss. In what follows, I propose that people with intimate knowledge of a particular individual also possess a kind of expertise that may be helpful when caring for patients who cannot speak for themselves.

This expertise is not something most of us are aware we possess; rather, it is a subconscious form of knowledge, born of experience, enabling one person to understand another, based on the smallest of cues. This kind of knowledge was described by Polanyi, a scientist and philosopher, as "subsidiary awareness" or "tacit knowledge."[19] He uses the example of a face to illustrate his point. How do we recognize a face? We can only hint at the features of the face through language, and yet we can unerringly recognize a face we know in a crowd. Similarly, we can recognize a person's mood from the expression on her or his face, but we usually cannot verbally identify or describe all the cues that lead us to our conclusions. Some features are very subtle or elude description, such that it is impossible to articulate how it is exactly that we make our judgments. Yet, without knowing how, we recognize and respond to these cues. This ability is what leads Polanyi to claim, "We know more than we can tell."[20]

Our subsidiary awareness emerges primarily from our experiences as physical beings, which is informed through our senses. What we see, hear, smell, touch, and taste, all provide information that we may not consciously assimilate, but that plays a significant role in how we respond to our circumstances or environment. The cues we pick up from someone with whom we are in conversation tell us a lot about the person and how we might most effectively respond. "Street smarts" help those who have them navigate complex neighborhoods safely. Those who are attuned to the subtleties of animal behaviors can work with them more successfully than those who are not. But we are not neutral recipients of sensory input. What we perceive and how we interpret sensory stimuli are shaped by our cultural environments. Whether and how we look into someone's eyes can mean different things, depending on the circumstances, social rank, and cultural context of the people involved. How dangerous a neighborhood is perceived to be depends on one's familiarity with its inhabitants and their customs. The degree to which a person is sensitive to animals will be dependent on the kinds of capacities they believe animals possess. In other words, how we perceive and respond to stimuli is dependent on the values we bring to our interpretation.[21]

We tend to think of knowledge as information we can communicate to others. But tacit knowledge, by definition, is knowledge we usually do not verbalize because we are not aware we have it. For example, in the medical arena, Polanyi observes that physicians cannot identify or describe many of the cues they take in when they diagnose disease. Although they can identify a disease by its typical appearance, they cannot describe it

completely because they are unaware of a great deal of the subsidiary information on which they rely. As a result, these details are not included in standard descriptions of disease.[22] Undoubtedly part of the reason disease cannot be fully described is because the concept of disease is itself culturally constructed — the models, categories, and language we use to describe disease shapes and limits the kinds of questions that physicians ask, as well as the details to which they attend. Indeed, as we have seen above, the limitations of language, the difficulty of distinguishing between terms such as "consciousness" and "awareness," or "alertness" and "wakefulness," may impede accurate diagnosis of brain-injured patients. But despite their inability to fully describe it, experienced physicians draw on their tacit knowledge routinely. This level of skill and understanding is sometimes referred to as the "art" of medicine.

Polanyi cites several experiments to illustrate that tacit knowledge is both learned and influences our behaviors. In one of these, a person experienced a slight shock whenever he uttered certain words. After a time, the person subconsciously learned to avoid saying those words. However, when questioned afterwards, he could not say that he was aware that he had done so. In developing his tacit knowledge, by learning how to avoid being shocked, he was drawing on what Polanyi calls "subception," or "learning without awareness."[23] That people can learn without awareness has been supported by more-recent research in neuroscience. In one such experiment, subjects were given four decks of cards and asked to flip the cards in a way that would maximize profits of play money. The subjects were fitted with sensors that measured skin conductance responses (SCRs), while the decks of cards, unbeknownst to the subjects, were rigged. After subjects turned about 10 cards, the sensors began to register anticipatory SCRs when they reached for a losing deck. But subjects needed to turn about 50 cards before they could verbalize hunches that two of the decks were "riskier" than the others, and they needed to flip 30 more cards before they could explain why their hunch was correct. Yet their bodies, through their senses, had registered a pattern long before the subjects were conscious of it.[24] These and related studies suggest that people are adept at perceiving patterns and at storing data in ways that permit a pattern to be recognized and behavioral responses to be elicited, even when they are unaware that this is happening. This ability to unconsciously perceive and respond to patterns of cues potentially offers a new source of diagnostic information for the severely brain-injured patients. Conceivably, some people who are very familiar with these individuals will be sensitive to any cues or patterns that are exhibited by the patient. Their intuition as to the patient's condition, based in part on their emotional responses to the patient, may constitute valid insights into the patient's condition. But how reliable can such claims be? How can such hunches be verified?

Gary Klein, a cognitive psychologist who has devoted his career to studying decision making, claims the ability to recognize patterns is what constitutes intuition. Klein has studied the role of intuition in decision making by experienced firefighters, military personnel, commercial pilots, and business executives. He has found that in the heat of battle or during a fire, experts do not analyze the risks and benefits of several options prior to making a decision; instead, they choose a single course of action that seems to them to be "good enough." They then run a mental simulation based on an assumed model of the problem, and revise the decision, if necessary. Based on this research, Klein concludes that intuitive judgments involve several steps: (1) perception of cues leads to recognition of a pattern; (2) pattern recognition signals a potential course of action or response; (3) the potential response is assessed and modified using a mental simulation based on a mental model of the problem, which is followed by an action.[25] All of this can happen so fast that sometimes decision makers are not even aware of making a choice — they just act. Even in retrospect, they may not be able to articulate why they made the decision they did.

Klein illustrates how intuition can effectively guide medical decisions with a case involving a neonate. In this case, an experienced neonatal intensive care nurse and a novice were both looking after a newborn infant. One day, the novice noticed that the infant was a bit lethargic, found her temperature a little low, and saw that a routine needlestick had left a little blood at the site. But none of these findings caused her any worry, as all were within range of normal. By contrast, when the experienced nurse arrived, she had a gut feeling the baby "looked funny," noticed that the heel stick was still bleeding, that the baby's flesh was a little mottled, and that her belly was slightly rounded. When she found that the baby's temperature had fallen constantly over the course of the previous shift, she instantly realized the baby was battling a serious infection, called the physician, and saved the baby's life. In this example, both nurses had noticed essentially the same set of symptoms, but the experienced nurse saw additional, very subtle cues, and recognized the totality as a pattern that indicated infection was probably present. Her expertise, which included her tacit knowledge of patterns, enabled her to draw the conclusion that the less experienced person missed.[26]

Klein concludes that intuition is developed through a person's accumulated experience in which similar constellations of cues are commonly associated. When one or more cues are present, an experienced person can infer that the others are likely to be also. With time and experience, it becomes possible to quickly recognize patterns of cues that suggest what to expect, what goals to anticipate, and what actions are required. In this way, Klein claims that pattern recognition, understood as intuition, is a valid and valuable source of information.[27]

TACIT KNOWLEDGE IN DIAGNOSIS OF PVS/MCS

As described by Polanyi and Klein, both tacit knowledge and intuition rely on subconscious pattern recognition, a skill that is derived through experience. While Klein focuses on professional expertise in a narrow range of decisions, Polanyi's tacit information is something we all possess and use in the most ordinary of activities. Both rely on experience in the form of stored data patterns. With regard to patients diagnosed with PVS or MCS, physicians clearly have specialized knowledge, experience, and clear mental models of the pathology of the injury, which will, in some cases, permit them to make the kind of intuitive judgments Klein describes. But astute family members and caregivers may also be considered to be experts if they have prolonged experience with a particular patient. If they are sensitive to the patient's needs, they will have tacit knowledge of the patient's patterns: how the patient reacts when touched, when bathed, when moved, or when spoken to. They will sense if a smile or a grimace is spontaneous or made in response to an external event, or if the patient's mood shifts in the presence of certain people. They will know the patient's likes and dislikes — if there are any — and whether certain music, tastes, or types of touch are pleasurable or not. And they will know when the patient is so injured that no signs of awareness are evident.

If one accepts tacit knowledge and pattern recognition as valid sources of information, when family members and caregivers voice their belief that their loved one is responsive, one may consider that they may be correct. But how can their perceptions be incorporated into the diagnostic process? Without knowing more about the sources and validity of tacit knowledge, it is reasonable only to ask that the healthcare team recognize that when dealing with cognitively impaired patients, the perceptions of family and caregivers may be valid. Even if they do not volunteer it, caregivers should be asked for their impressions of the patient and what they think the patient is experiencing. If a patient is claimed to be in any way responsive, physicians should proceed with a more careful and refined assessment. In this process, physicians

may wish to spend more time with the patient, ask the caregivers to show them what they are observing, and explore more sensitive means of assessing the patient's cognitive capacities. They may or may not change their conclusions as a result, but will have at least made an effort to use all the information at their disposal. In addition to improving diagnostic accuracy, the benefits may include greater respect and trust between the medical staff and family and a willingness on the part of the family to accept the diagnosis once it is made.

But if this sounds relatively simple, in practice it may not be. Physicians may be reluctant to accept that family members have significant information to offer or assume that caregivers' perceptions are influenced by their emotional attachments. Even if the physician is open to family input, he or she may not be present when the patient is responsive, may lack the experience to understand the patient's behaviors, or may fail to appreciate that very small signals can be behaviorally meaningful. If caregivers cannot put into words the cues that inform their intuitions or show others what they are seeing, they may have difficulty convincing others of their perceptions. Moreover, some family members may not be sufficiently attuned to the patient to have this knowledge, or their hopes and fears may indeed be shaping their perceptions. Part of the job of the healthcare team is to become sufficiently acquainted with each family to gain a sense of whether their contributions would be significant and when not.

Klein observes that the use of intuition in the workplace may also be impeded by institutional and environmental characteristics. These include organizational policies that restrict individual decision making, rapid turnover of personnel that limits the development of expertise, the pace of change in many environments, excessive reliance on procedures that reduce perceptions of more subtle cues and nuances, and increasing use of information technologies such that humans are being taken out of the decision-making process.[28] Each of these factors is a reality in modern medical practice, and may further limit opportunities for incorporating tacit knowledge in cognitive assessments of brain-injured patients.

Despite the interpersonal and institutional challenges to incorporating tacit knowledge in the diagnostic process, the anguish caused by diagnostic ambiguity and error is too great to ignore. Recognizing tacit knowledge and intuition as important sources of insight offers a possible approach toward alleviating some of the uncertainty and emotional distress that surrounds these patients and their families. When family members say, "I know she knows when I am here," "I know he is trying to communicate," or "I believe she is hungry and in pain," physicians should realize that these remarks may signal a need for further assessment, as well as the need to help the family accept that the person they knew is most likely gone forever. While many physicians may feel that attending to elusive or imperceptible cues claimed by nonmedical professionals is a waste of time, sincere attention to perceptions of family members may nonetheless contribute positively to the family's grieving process and their sense that everything possible has been done. Given the protracted legal battles that may ensue if a diagnosis cannot be satisfactorily established, this gesture alone may be well worth the time and effort.

CONCLUSION

There is clearly a great deal to learn about how we acquire our tacit knowledge and intuition and how they may best be used in medical practice. Conceivably, this subconscious knowledge offers a new source of diagnostic and therapeutic insight that could prove beneficial across all fields of healthcare. However, because tacit knowledge appears to be subjective, nonrational, and difficult to verify, it is unlikely to be readily embraced as a valid source of information by the medical community. Moreover, with regard to brain-

injured patients, critics will contend that including families in the diagnostic process will create additional problems — such as denial of the reality of the cognitive deficits, demands for futile rehabilitative services, or excessive grieving — that could lead to protracted delays and burdensome costs. This may or may not be an accurate perception. While some of these issues may arise with families, they may pose no greater difficulties than they do now, given our present diagnostic methods. And, as is evident in the Schiavo case, our current policies for resolving conflicts or lessening costs offer few constructive solutions. But it is also conceivable that, by making greater efforts to involve family members and caregivers as partners in the medical team, and by attending seriously and sincerely to their observations and concerns, they will be more willing to accept the diagnosis and treatment decisions that are eventually made. This is not an unreasonable expectation, and can only improve the quality of patient care.

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Advance Health Planning and Treatment Preferences among Recipients of Implantable Cardioverter Defibrillators: An Exploratory Study

Jeffrey T. Berger, Matthew Gorski, and Todd Cohen

BACKGROUND

Implantable cardioverter defibrillators (ICD) are devices designed to detect and treat lethal cardiac arrhythmias through electrical leads embedded into heart muscle. Cardiac electrophysiologists typically insert ICDs and these devices are placed using only local anesthesia. The use of ICDs is rapidly increasing in response to recent medical research that has demonstrated improved survival for patients with several common, but serious, cardiac conditions.[1] Despite this treatment, recipients of ICDs often have progressive noncardiac diseases in addition to their cardiovascular maladies, and mortality rates among these patients remain significant.[2] Some ill and debilitated ICD recipients may come to no longer value the additional survival provided by the ICD, and some patients may even determine that their ICD is a barrier to a timely death.[3] Near the end of life, a repeatedly discharging defibrillator may cause significant discomfort and thwart symptom palliation.[4] Moreover, sudden onset of ventricular tachycardia, the cardiac arrhythmia that ICDs are designed to treat, is a relatively painless manner of dying. The procedure for deactivating an ICD is simple and painless and involves placing a magnetic device over the area of the chest corresponding to the implanted defibrillator.[5]

Although physicians caring for patients with ICDs are likely to encounter situations in which deactivation of an ICD may be considered, the medical literature has been slow to address the ethical implications of this recently developed technology.[6] Professional organizations such as the American College

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of Cardiology and the American Heart Association offer guidelines for implanting ICDs, but not for deactivating already implanted defibrillators.[7] Furthermore, few physicians discuss disabling ICDs with recipients who are dying.[8] Little is known of patients' preferences for disabling their ICD. However, based on studies of patients' preferences for the use of other life-sustaining interventions, it is likely that many patients have concerns about limiting the use of ICDs under some circumstances.[9] Unfortunately, patients have little assistance in developing and documenting these preferences. Standard living will documents, used by only a minority of patients, do not yet anticipate decisions involving these devices.[10] Nevertheless, patients receiving ICDs should prepare for decision-making incapacity and consider using treatment directives to guide their surrogates regarding ICD use.[11]

There is little empirical study of ICD recipients' treatment preferences or of their use of advance directives. This exploratory study was designed to assess (1) whether recipients of ICDs have considered or developed preferences for non-use of the ICD at the time of its implantation and after living with the device, and (2) the prevalence of advance directives among ICD recipients and whether these advance directives address ICD use.

TABLE1 Acronyms and definitions

EP	electrophysiology
ICD	implantable cardioverter defibrillator
n	number of subjects in a group
N	number of subjects in the study
NS	not significant
p	probability value
SD	standard deviation: one of several indices of variability that are used to characterize the dispersion among the measures in a given population
t-test	a test to determine whether the mean for a given group exceeds a certain standard; used in comparing the means of two different groups

METHODS

The study was conducted in an out-patient cardiac electrophysiology (EP) office of a tertiary care hospital and was approved by its institutional review board. Subjects were limited to capacitated adults who had received an ICD one or more months prior to entry into the study. Persons with acute cardiologic conditions requiring urgent management or hospitalization were excluded.

A clinical staff member of the cardiology office secured permission from each prospective subject to receive information about the study. The researcher then provided prospective subjects with a verbal and written description of the study. Voluntary participation represented informed consent. Subjects completed a self-administered survey expressly developed for this study. The survey included open- and closed-end questions assessing subjects' understanding of the medical indications for the device, their health-related expectations for the device, their advance health planning both generally and with respect to the ICD, including any preferences for disabling the ICD. We refined and coded participants' subjective responses into categories.

RESULTS

Over a five-week period, all of the 59 eligible patients presenting to the EP office for medical follow-up of their ICD were approached regarding participation in the study, and 57 (97 percent) were enrolled and completed the study. Table 2 describes this population. These mostly White, largely Roman Catholic, and well-educated subjects had their devices implanted from one to 91 months prior to the study (mean 25 months, median 30 months). Subjects' self-assessed health status was poor for 5 percent, fair for 32 percent, good for 39 percent, and excellent for 21 percent; 55 subjects had good functional status, as evidenced by their ability to ambulate independently; two subjects were wheelchair-bound.

Subjects were asked to describe, to the best of their knowledge, the medical indication(s) for their ICD (see table 3). The 57 subjects offered 72 reasons, the vast majority of which were pertinent cardiac conditions or symptoms; two subjects indicated "physician recommendation" as the sole reason for receiving the device. We asked the subjects to describe the way in which they expected the ICD to affect their lives (see table 4). These responses centered on greater survival and increased quality of life.

Subjects were asked about their preferences for ICD deactivation: both currently held preferences and preferences they may have had at the time the device was implanted. Of the 57 subjects, 53 did not recall formulating preferences for disabling their ICD at the time the device was implanted. Of the four subjects that had done so, two had wanted the device disabled only for improvements in their cardiac condition that caused the ICD to be no longer needed.

When asked about currently held preferences for ICD use, 36 subjects had not developed preferences. However, 21 (38 percent) subjects described situations or conditions for which they would want the ICD deactivated (see table 5); 10 of these 21 subjects wished to have the device deactivated for impaired quality of life, and three subjects did not want the ICD used in the event of terminal illness.

An advance directive document was completed by 35 (61 percent) of the subjects. Of

TABLE 2 Demographics (*N* = 57)

Age		
Characteristic	Mean	Median
Years (range = 40 - 86)	70	72
***n* and % of Subjects by Characteristic**		
Characteristic	*n*	%
Gender		
Female	10	17
Male	47	82
Ethnicity		
Non-Hispanic White	52	91
African-American	3	5
Hispanic	2	4
Religion		
Roman Catholic	38	67
Protestant	9	16
Jewish	9	16
No preference	1	2
Education		
Less than high school graduate	12	21
High school graduate	30	53
College graduate	15	26

TABLE 3 Subject-Described Reason for ICD Placement

Reason	%
Cardiac condition (nonspecific)	44
Arrhythmia	44
Nonspecific symptoms	14
Syncope	11
Physician recommendation	9
Congestive heart failure	5

Percentages add to >100% because the 57 subjects offered 72 reasons.

TABLE 4 Subjects' Expectations of ICD-Derived Benefit

Expectation	%
Increased survival	45
Improved quality of life	42
Arrhythmia control	23
No particular effect or expectations*	11

* None of these subjects offered secondary expectations.

TABLE 5 Reasons for Disabling ICD in 21 Respondents

Reason	%
Impaired quality of life	48
Physician recommendation	19
Terminal illness	14
No benefit	10
Availability of better treatment/technology	10

these documents, 60 percent were living wills, 31 percent were healthcare proxy documents, and 9 percent were combined proxy-living will directives. Of the 35 advance directives, six (18 percent) were completed after the ICD was placed, and these documents were equally divided between living wills and proxy documents. No advance directive (executed either before or after the ICD was placed) addressed the use of an ICD.

Analysis of Variance (ANOVA) performed on the data revealed no relationship between subjects' anticipated ICD-related health benefits and their age, level of education, or self-reported health status. Paired *T*-tests revealed no relationship between self-reported health status and current preferences to disable the ICD. A relationship between education level and completion of an advance directive approached significance ($p = 0.056$) on the *T*-test procedure.

As mentioned, none of the advance directives addressed the use of an ICD, yet 15 (44 percent) of 34 subjects who had completed an advance directive, when asked by the researchers, indicated preferences for disabling the ICD. In contrast, only six (27 percent) of 22 of the subjects who had no advance directive considered disablement. Of the six subjects who completed an advance directive after ICD insertion, four (80 percent) communicated their preferences regarding disablement of their ICD to the researchers, compared to only 11 (39 percent) of 28 subjects whose directive was written prior to ICD insertion (NS, $p = 0.152$). It is interesting that none of the five subjects whose reason for having an ICD was "physician recommendation" would consider disabling the device. However, this finding was not statistically significant ($p = 0.145$). None of the six subjects who had an ICD implanted for syncope had a preference for disabling the device, compared to 42 percent of subjects whose ICD was placed for other indications (NS, $p = .074$). That approximately two-thirds of women (67 percent), compared to one-third of the men (32 percent), indicated a preference to disable the ICD (trend towards significance at $p = .066$).

DISCUSSION

Approximately 4 million Americans currently meet medical criteria for the implantation of an ICD, and it is expected that at least 300,000 will join them each year.[12] Undoubtedly, the prevalence of ICDs among hospitalized patients, and the likelihood that physicians, patients, and families will confront ethical dilemmas involving ICDs will increase. Among our study population, a significant minority (38 percent) indicated a preference for limiting the use of the ICD under some specified circumstance. Yet none communicated this preference, despite the apparent sophistication of this group as evidenced by a high level of education, a high prevalence of advance directives, and a good grasp of the medical indications for the ICD. It is surprising that the subjects who had completed an advance directive after they received an ICD neglected to mention use of their ICD in their formal health planning. Another study similarly reports no association between having completed an advance directive and discussing end-of-life preferences for ICD deactivation among dying patients who had ICDs.[13] Similar inattention to advance planning is evident among users of chronic hemodialysis.[14] Perhaps these findings parallel the public's reticence to consider and plan for disability and death. Perhaps patients are simply unaware of the discomfort an ICD may impose, through repeated electrical shocks, on the process of dying, and are similarly unaware of the advantages of health planning. Perhaps patients and their physicians mirror each other's inattention to the implications of having an ICD in states of poor quality of life or approaching end of life. Certainly, there is little guidance regarding the ethical implications of ICDs available to physicians who are, of course, best situated to advise these patients.

Modifying advance directives to address ICD-related decision making may have little clinical impact, since advance directives generally have not been proven to be of great value to patients and clinicians.[15] This may reflect

the need to reconceptualize advance health planning, rather than to discard it.[16] Regardless, some patients, surrogates, and physicians may find having clearly documented preferences for ICD use to be valuable. Provisions for commenting on ICDs can be easily incorporated into standardized living will, do-not-resuscitate forms, as well as into the informed consent process that precedes ICD placement. Patients whose directives indicate a preference to discontinue life-sustaining interventions should strongly consider a statement for deactivating the ICD in anticipation of inevitable fatal arrhythmias. Patients who wish to appoint an agent for healthcare should, of course, inform these proxies of their preferences for ICD use. Clinicians should routinely discuss the use of ICDs with patients when hospice services begin.

It is prudent for cardiac electrophysiologists and other cardiologists to ask these patients to consider parameters for ICD use at the time the device is implanted, or, if the patient is too ill, at some regular time intervals thereafter. Many patients prefer to discuss health planning when in the office, rather than during an illness requiring hospitalization.[17] These discussions, as well as discussions about limiting other treatments, are best embedded within conversations of broad health goals and general preferences for treatment. The care of patients may be improved by understanding their care goals for the ICD. For example, a patient with a recurrent primary brain tumor is expected to live less than four months. The tumor has impaired her decision-making capacity. During these last months, the ICD may be viewed as obstructing a timely, arrhythmic death and enabling her to live only to die of the malignancy, or, alternatively, as supporting her remaining life, however it may be valued. Another patient, persistently vegetative, may have his life prolonged indefinitely by a functioning ICD. In each of these examples, the identification of the patient's treatment goals and preferences for ICD use could assist health professionals in providing the patient with optimally appropriate care.

Despite entering nearly all potential subjects into the study, this data is limited by the lack of diversity in subjects' gender, ethnicity, and religion. Although the ages of the participants were fairly well clustered around 70 years, the duration of ICD use ranged from one month to over seven years. Since subjects' initial preferences regarding ICD use were based on recollection rather than obtained contemporaneously, the large range of time since ICD placement may have affected these results. Certainly, a more accurate method for identifying the evolution of preferences would be an assessment of preferences contemporaneous with ICD insertion and again at a point in the future.

The number of subjects enrolled into the study limited the statistical power of our results. However, this exploratory investigation identifies themes worthy of further research. Although a majority of subjects (61 percent) had advance directives, none of these addressed ICD use. Additionally, most ICD recipients had not considered or developed preferences for limiting ICD use. These two findings suggest a greater role for informational counseling and health planning around the time of ICD placement. Our finding that 38 percent of subjects indicated some preference for ICD deactivation suggests that we can better serve our ICD recipients by actively assisting them in developing, communicating, and documenting these preferences.

Additional study is needed to determine whether patients want this assistance, whether they want these preferences documented, and whether they want their family members bound by these preferences in surrogate decisions. Further study is needed to better identify and specify conditions under which patients want these devices deactivated. An interesting finding to be confirmed and studied in future research is the fact that most of the ICD recipients who had completed an advance directive after receiving the device had formulated preferences for deactivation, yet none had communicated these preferences.

We note, incidentally, that women and members of minority groups appeared to be

under-represented in our sample. This finding has been reported in other research on ICDs.[18] The question of whether ICDs represent yet another of several cardiac technologies associated with gender- or ethnicity-related disparity is one that should be investigated.

CONCLUSIONS

ICDs are another life-sustaining technology whose widespread use precedes careful thought about planning for treatment withdrawal. While our preliminary data should be confirmed by additional study, our investigation raises concerns that preferences of ICD recipients for limiting ICD use remain unidentified. Furthermore, typical treatment directives do not facilitate the recording of patients' preferences for ICD use. Physicians and other health professionals should assist recipients of ICDs in developing and communicating preferences for ICD deactivation.

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Cases from the Harvard Ethics Consortium

When a Village is Not Enough

Christine Mitchell and Robert Truog

There were at least 14 specialty services involved in Lorraine's hospital care: plastic surgery, nutrition support, gastroenterology, infectious disease, orthopedics, the wound care team, addiction services, psychiatry, forensic psych, the pain team, general surgery, oncology, gynecology, and palliative care; not to mention chaplains, the social worker, the case manager, rotating physicians, the nurses who took care of her every day — whether they wanted to or not — and the ethics committee. Lorriane's medical problems were formidable but treatable — if she would accept treatment.

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Partially paralyzed from an accident several years earlier and now in her early forties, Lorraine had chronically infected wounds so deep one could see her wasted muscle and bone. She was inconsistent (at best) in following prescriptions for her wound care, diet, medications, and therapies, and regularly refused dressing changes, antibiotics, blood tests, and other medical procedures. Worse, she had a history of depression, multiple attempted suicides, and drug addiction, and was frequently rude to the point of abusiveness.

Most of all, Lorraine was sick of being sick. She had exhausted her family and was burning through a large share of hospital resources much more rapidly than she was getting better. The nurses asked her healthcare team, and her healthcare team asked the ethics committee, whether they should do more to make Lorraine accept treatment, or should they support her refusals, which would bring about a slow spiral toward death? And, by the way, were they obligated to take care of a patient who is abusive and offensive?

Sick to Death

Grace Good

Lorraine was a 42-year-old White female admitted to Massachusetts General Hospital for care of a deteriorating sacral decubitus ulcer. Lorraine's visiting nurse and her primary care physician felt the decubitus ulcer was getting worse because of the lack of help at home to perform dressing changes and Lorraine's worsening depression.

Lorraine's past medical history was long and complex. Some of her other diagnoses were microcytic anemia, chronic obstructive pulmonary disease, deep vein thrombosis, left above-the-knee amputation, pulmonary embolism [a blood clot in the lung], chronic abdominal pain, diarrhea and vomiting, obesity, gastric bypass surgery, paraplegia, depression, intravenous-drug abuse, polypharmacy [when a patient takes many drugs, some of which may not be needed], bacterial endocariditis [infection of the inner surface of the heart], colostomy, urostomy, and suicidal ideation and attempt. While this is not a complete list, it does exemplify how complex her medical history was. I am not sure anyone had a complete and accurate list of her diagnoses, as Lorraine went to multiple physicians, hospitals, clinics, and hospitals. She would only disclose certain aspects of her history to certain caregivers. However, as time went by during her last admission, she did disclose what seemed to be all, or at least most, of her history.

Lorraine had been on the medical service for about a month when I became involved with her care. I was asked to see her because I work as nurse practitioner on the medical team Lorraine was assigned to, and I follow the more complex patients with chronic illnesses and long lengths of stay in the hospital.

The day I went to review Lorraine's chart, she was in the hallway in her wheelchair. I quickly realized I knew her from an admission a year earlier, which was also for her decubitus ulcer and bacteremia [presence of bacteria in the blood]. I remembered that her ulcer was in fact one of the worst I had ever seen, and that she was not compliant with her

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care. She had been discharged home following that hospitalization and apparently had been "getting by" until this admission.

I also remembered some of the behavioral guidelines that had been put in place for her on that last admission. It was no surprise that the nursing staff on the unit now had difficulty managing Lorraine's behavior. That all being said, something happened to all of us during this, Lorraine's last admission, which changed at least my way of approaching and caring for a chronically ill patient who has behavioral issues.

Nutrition was a major focus of this hospitalization. Lorraine had chronic nausea and vomiting without, it seemed, relief from any type of medication. Since her decubitus ulcer could not heal without adequate nutrition, a gastrointestinal (GI) workup was initiated. Lorraine would not consent to the last test that was to be performed, which probably would not have changed the treatment plan, even if had been abnormal. Even without the final test, she'd had an extensive GI workup and nothing abnormal was found. Total parenteral nutrition, a specially mixed intravenous infusion of essential nutrients and calories, was tried, but Lorraine found it disagreeable, saying it gave her a bad taste in her mouth. At times she shut it off. Lorraine was getting more frustrated with everything and everyone.

Staff on the plastic surgery service knew Lorraine from taking care of her in the outpatient clinic. They thought surgical flaps for her wound might help, but Lorraine refused to stop smoking, refused to stop sitting on her wound for prolonged periods of time, and refused to use a special bed in the hospital that would relieve pressure on her wound. The plastic surgery team did not want to do surgery if she was going to be noncompliant, which she readily said she would be. Lorraine knew throughout this last hospitalization that she could change her mind at any time and discuss surgery with the team. She opted not to change her mind, although she did see the plastic surgeons several times, which she said she did to make us happy.

As Lorraine's decubitus ulcer became even worse, it was clear we were going to reach a point where there would be no turning back. Her nutrition, even with many attempts, did not improve. Pain became a major issue, and the pain service followed her. They eventually took over control of her pain medication regime, as it became clear that we needed firm guidelines on when to increase or decrease doses of her medications.

Because of the bacterial infection in her blood, with the likely source being her decubitus ulcer, Lorraine was on a long course of intravenous antibiotics, and specialists in infectious disease (ID) were following her. Once her antibiotic course was complete, she still continued to have fevers, and questions arose about whether to start antibiotics again.

Lorraine spoke with her primary care physician, and, after thoughtful discussion, decided that if she was bacteremic again, she did not want antibiotics. She also did not want aggressive wound care. She decided she did not want cardiopulmonary resuscitation (CPR) if her heart stopped, and she did not want to be intubated if her breathing stopped.

Her decision, which on one hand I could understand, was still difficult for me to accept. Lorraine could have made different choices that would have made her life, in my opinion, easier, and perhaps would have kept her in better health. Since Lorraine had a long psychiatric history and the question of depression came up, a psychiatry consult was obtained. Lorraine declined to be interviewed in any real detail; a second consult and a third were obtained. The last consultant, after reviewing her history and speaking with her, determined Lorraine was capable of making her own decisions, including a decision to refuse treatment for bacteremia if it recurred. So, DNR (do-not-resuscitate) and DNI (do-not-intubate) orders were written.

I never doubted that Lorraine was capable of making decisions or that she knew she would be bacteremic in a matter of time. At times Lorraine seemed to want me to feel uncomfortable with the situation, at other times

she seemed to want approval. I told her more than once that I couldn't make this decision for her, but since she made it, I would try to be sure she was comfortable and cared for.

Lorraine's behavior became worse before it became better. She tested everyone caring for her. At times she took her frustration out in verbal abuse, yelling, or simply not talking. At other times, she "acted out" by demanding more and more pain medication.

Every day brought a new experience for me when I went to see her. There were multiple meetings of the healthcare team about how to approach each situation. I never felt I was alone, especially since there was always an attending physician assigned to Lorraine who knew her case well. While I certainly was not running things, I became identified by the staff, Lorraine, and her family as the person who knew the most current plan.

Lorraine refused to let me talk about her condition and care in any detail with her family. While her mother and sister knew Lorraine was not doing well, that she was refusing treatment, and that she had made herself DNR/DNI, it was not easy to see them visit and not be able to talk with them about her. Lorraine said she did not want to worry them more than she already had, and that her mother was to be spared details that would upset her. Lorraine had a sister who had died a few years earlier, and she said that if she died she did not want it to be as hard on her mother.

While Lorraine had a new issue almost every day, I feel that each new thing was given serious consideration. Nothing was ever dismissed because of her prior behavior. The pain service, in particular, treated Lorraine with respect, as did everyone else involved with her care. I developed a relationship with Lorraine in which I was the disciplinarian at times, the advocate at times, and just someone for her to vent to at other times. While it wasn't easy to learn why Lorraine acted the way she did, once I gained some understanding and especially once I gained her trust, I could see the person Lorraine was. She had interests and a good sense of humor, which was apparent when she was trying to give me a hard time about something and could not keep from laughing herself, and a love for her family that was always her focus, although it was not always apparent.

More than once I was the bearer of bad news for her, but at other times I was able to allow her to have some particular request met. Her family were strong advocates for her, and I must say, at the end, that while they may have had questions, they focused on her comfort and spirit more than on details.

After at least 14 consulting teams, numerous tests, many team meetings, tears, and laughter, Lorraine died quietly and in peace. She taught me to use all of the resources available to me, to never try to handle this type of situation alone, and that there does need to be someone involved with a complex patient who stays with the case and keeps things in perspective. I was fortunate to have a strong team of physicians working with me, teaching me how to interpret test results and challenging me to look at things in more than one way. I was also fortunate to have a nursing staff caring for Lorraine who were willing to discuss and contribute to her plan of care.

I feel I developed a relationship with her family that was built on trust and caring. It was not only my relationship with them, but the entire team, that enabled us all to care for Lorraine in a way that enabled her to die with peace and dignity.

The Case Manager's View

Suzanne M. Burke

Case management at our hospital was designed, from its conception, as a patient-focused program with goals to improve patients' satisfaction, decrease length of stay, and decrease overall costs. I never found myself at odds with this philosophy until the year I was the case manager for Lorraine.

Lorraine was admitted in May 2004. Her primary care physician, who was overburdened and frustrated by Lorraine's constant paging and calling, admitted her in the hope that a last effort at wound management and pain management would be successful.

I knew Lorraine from an admission the year before. She'd been admitted then with similar issues and signed herself out "against medical advice" after several days. As a result of that admission, I was aware of several problems that would be barriers to ultimately discharging Lorraine.

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First, she had a long history of substance abuse and noncompliance with any medical plan. Lorraine had been discharged from services from several homecare agencies because of her behavior. Also, she lived with a significant other who was not available to assist with any of her care. Her closest living relatives, although supportive and involved, were unable to provide physical support. On admission Lorraine clearly informed the healthcare team that she would not consider being discharged to a skilled nursing facility. She was knowledgeable regarding Medicaid and disability benefits. Although these things would make discharge difficult, the greatest challenge to me, as well as the care team, was to formulate a clinical plan and engage Lorraine's participation and agreement with it. Throughout Lorraine's stay, all of the skills and talents that are typically identified with case management were put to the test: accessing resources (MassHealth, community providers, discharge experts), negotiating with the patient and potential caregivers, collaborating with multiple caregivers on a daily basis, and,

most importantly, advocating for the patient.

Case managers frequently ask questions of others and ourselves about ethics — questions about the decisions we make and the paths we follow as we try to find people and places to continue caring for the patient. Before and after Lorraine decided to withdraw from any further medical treatment, I struggled with her unwillingness to follow a medical plan that could make a difference for her well-being and the quality of her life. I had great difficulty with my urge to push Lorraine to follow the plan that we, the care team, knew was best for her. In plain words, I wanted to control the process. I was frustrated with her unwillingness to understand that home was not an option because she required too much care and was not considered trustworthy. At times I was stunned by the anger I felt toward her because she had done nothing, that I could see, to keep herself from ending up in this spot and because she would not help us to help her. It was in those moments that I was reminded of the importance of the care team; for at each hurdle that each of us faced during her stay, the support and guidance from our fellow team members ultimately made the difference for Lorraine and ensured that her wishes and decisions were respected.

My plan was to support Lorraine's plan. She wanted to die at home or at the hospital. Unfortunately there were no resources or agencies in the community that were able to support a transfer home. Over time it became clearer and clearer that Lorraine would die in our hospital. I found those months leading up to her death difficult. I frequently experienced a sense of failure, a sadness that I had been unable to "make it happen" for her. I had referred Lorraine to approximately 40 skilled nursing facilities/acute rehabs, in-patient hospice, several home health agencies, and hospice programs. Several times, especially as the holidays came closer, other members of the team would ask me, if I was able to, would I send Lorraine out before Christmas.

Lorraine died on the weekend just before Christmas. When I arrived on Monday morning and noted her name was no longer on the census, I felt an emptiness that I had not expected. This woman, who had such difficulty with trust, had developed a bond with us. When she died at night, she was not alone. Two of the nurses who had cared for her over the many months of her stay were with her, holding her hand as she slipped away.

It wasn't until I went to her wake, and then had a chance much later to speak with her mother and sister, that it became clear to me the difference we all had made for Lorraine and her family. The plan had worked.

"We Need to Meet"

Jennifer Repper-DeLisi and Susan M. Kilroy

More than four months into Lorraine's admission, the call from the unit had become a familiar one: "We need to meet." We had many meetings about many things: Lorraine's refusal of treatments, her abusive language, the weekend she threw her food tray, her demands to see the physician who was on call during the night. This time, at least one of the problems was that staff members were not sure how to deal with a patient who had been caught trying to inject herself with hoarded pain medication. It was one of many issues to be addressed in the care of a very complicated woman whose struggles seeped into every possible domain.

Our meeting of the minds included a physician (Robin Dauterive), nurse practitioner (Grace Good), case manager (Suzanne M. Burke), the clinical nurse specialist on Lorraine's unit (Susan M. Kilroy), a psychiatric nurse consultant (Jennifer Repper-DeLisi), several specialists, and staff nurses. There were 31 people at such meetings of Lorraine's care team. By the end of this meeting, everyone had had a say, and a two-page document of problems and proposed interventions was the result. In spite of the general frustration, there was the familiar infusion of concern and care as we communicated on the interventions necessary to maintain Lorraine's safety and security as a patient on the unit.

We had come a long way from the early days of Lorraine's admission, when she took the unit by storm. "She's moving in," we thought, when she showed up with what appeared to be a large proportion of her belongings from home, including clothes and a collection of dolls. Because of her care needs, she was moved into a single room. Then she sent the staff reeling as she persisted in chasing us out of the room. The chase was sometimes a literal "Get out!" Other times it was in her commands, threats, swearing, and, from the beginning, her refusal to accept our care. Her

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refusals were evenly matched by her unremitting requests and demands. Molly, her primary nurse, remembers, "It was like she had arrived with her own agenda, and what all of us wanted to do for her was completely different than what she had in mind."

Jennie (the first author of this article) recalls that when she was first called to see Lorraine early in her admission, she hoped she wouldn't be there too long. While Jennie knew that the staff were skilled in dealing with many of the psychiatric concerns of their patients, Lorraine's capacity to intimidate, to engage staff in destructive conversations about their peers, to criticize every aspect of her care, was formidable. Her primary nurse, Molly, described her first meeting: "The room was pitch black. It was a very, very dark space, a dark cave. She was a 'Being' in this bed who would boss you around. I knew what she had put others through. I was very intimidated by her." Many of her nurses were novices in the profession. As one new nurse put it, "She knew enough about her own care needs that she often tried to use her knowledge and experiences to intimidate, compelling me to frequently check and recheck my skills and knowledge level."

We have seen that patients with personality disorders have a tendency to divide staff and to lead them to feel demoralized and burned out in their efforts to provide care that is never "good enough." Molly highlighted this experience when, after a series of particularly grueling days, she said, "I don't like myself in the room." Lorraine had burned many bridges with healthcare agencies. We knew that the nurses on White 10 could be added to a long list.

We believed that frequent interdisciplinary huddles, with the understanding that one person's need to meet was everyone's need to meet, were essential to help both Lorraine and the staff rise above the natural responses of frustration, anger, and rejection. Meetings were a time to share ideas, bolster one another, discuss, and plan. Team cohesion became the glue of Lorraine's care. As Molly put it, "You got that extra emotional support from everyone else. Each day there was someone to ask how she was doing. There was always an outlet or a pow-wow." Additionally, we needed to give staff the interpersonal tools not only to help them survive eight hours of caring for Lorraine, but also to work productively toward a favorable outcome for her. Courtney, another nurse, said, "We had to stay checked in. We didn't have the choice to check out."

Our priorities became, first, how to help the staff stick together; second, how to balance hope for change and participation in Lorraine's care with an acceptance of her limitations; and third, how to foster therapeutic communication between Lorraine and the people taking care of her.

Caring for the nurse who was assigned to Lorraine for an eight-to-12-hour shift was our top priority. If the nurse didn't feel supported, Lorraine's care would be compromised. Primary nursing was next to impossible. No single nurse felt able to manage the burden of Lorraine's care day after day with unending responsibility for leading the nursing team. Taking care of Lorraine on an everyday basis was just too draining. Staff nurses were overwhelmed. Many reported thinking about her outside of work. Others dreamed about her.

Sue, a clinical nurse specialist and the second author of this article, knew it was crucial for the patient to have consistency among those planning and providing her daily care. Besides, our unit is small; most of the staff knew Lorraine. We agreed to rotate nurses, giving each nurse the opportunity to decide how long she or he could care for her. For instance, a certain nurse might be able to care for her for three days in a row, if Lorraine was in control of herself. Another nurse would need a break after one difficult 12-hour shift. We felt committed to the notion that staff would feel free to let us and each other know their limits. As a group, the staff felt a greater sense of control over their practice and confident in a consistent approach.

Caring for Lorraine required a thoughtful flexibility. Going where the "Lorraine wind"

blew was wearing and counter-therapeutic. But our efforts at holding rigidly to rules were ineffective as well. Developing reasonable expectations that accounted for Lorraine's maneuvers and changing capacities was an ongoing challenge that required a great deal of dialogue and "processing." There was almost a daily search to find the balance between a nurse's personal capacities and what was necessary to provide care to Lorraine.

Staff were vulnerable when they were in Lorraine's room, and she knew how to "get" to them. She was a master at "pushing buttons." Staff needed to learn how to preserve themselves and defuse the tension created by Lorraine's anger. Providing scripts for the most common and tension-producing situations helped staff to have a sense of control and to "get back in the driver's seat."

It became easier for staff to leave anger at the door if they entered the room with some tools to manage Lorraine's behavior. They began to provide her with constructive feedback, limiting their response to those behaviors that she used to push others away: "Lorraine, I can't be with you when you're calling me names. I'll come back in 10 minutes." They also made clear what behaviors could not go on and set reasonable consequences. For example, smoking in her room led to taking away her cigarettes and calling security for a room search. Eventually nurses were able to let Lorraine know when she was pushing the limits of what they could offer. Courtney remembers, "When she was found crushing her meds, a lot of us who had gotten close to her felt betrayed. When I first heard about it, I went into her room and let her know how disappointed I was in her."

Probably the most trying and anxiety-provoking aspect of Lorraine's care was pain management. With Lorraine, pain was a sticky subject. We knew from her primary care physician in the community that Lorraine had trouble resisting the temptation to overmedicate herself. This had been an out-patient concern for a long time. But now she wanted to overmedicate herself with our assistance. As nurses, we're taught that pain is pain and needs to be treated; but no matter what Lorraine was given, it was never enough. She would open her eyes in a sedated state and demand more. As a result, it was no surprise that nurses would often return with medication to find Lorraine sound asleep. Nurses became very uncomfortable at the thought of administering large doses of narcotics to someone with slurred speech and somnolence. When blood pressure and oxygen saturation parameters for giving pain medications were set, Lorraine would purposefully hyperventilate in an attempt to meet the requirements. Nurses were perpetually worrying that giving narcotics would further depress her mental and respiratory status. Yet if the nurse did not give Lorraine pain medication because she was asleep or had depressed vital signs, she reacted badly.

Nurses faced this quandary on a daily basis. What was Lorraine's physical pain and what was the pain of her addiction? With multiple narcotics ordered, both standing and as needed, and with Lorraine's constant requests for more, were we just reinforcing her addictive behaviors? According to Molly, the ethical dilemma became, "Am I medicating her because she's in pain, or am I medicating her to get her out of my hair?" The pain team had begun following Lorraine. Jennie hoped that the pain team would take on a decision-making role about Lorraine's pain medications to take something substantial off the nurses' very full plate. The day that Dr. Acguardro agreed to take on the decision making, ordering, and to be the "bad guy" with Lorraine was a day of enormous relief for the nursing staff.

These new boundaries around managing Lorraine's pain and her demands for medications seemed much more therapeutic for her and made it possible for us to work effectively in concert.

Then, about three months into her hospital stay, Lorraine decided to let herself die. No antibiotics for infection. No more efforts to heal her huge decubitus wound. Medically,

there was hope for Lorraine, but she was choosing a path that would lead to her death. Found competent by the psychiatrists, the choice was left in her hands. She was an adult, capable of making her own decisions, not actively suicidal, so she had to be allowed to make bad choices if she so desired, so long as she was informed of the risks. We were taken aback by this. Here we have this feisty woman who has struggled with us every step of the way. Although she was locking horns, she was engaged. Once again, she had pulled the rug out from under our feet. We felt shocked, confused, angry, sad. What should we do? Did she really mean this? Or was this just more acting-out behavior? All of us were disquieted. Were we going to just watch her die?

Initially, caring for Lorraine in this context increased the general stress of the staff. Not only were they struggling with the interpersonal stress of caring for her day after day, they were now in the position of dealing with additional ethical concerns and obligations. Should we try to change her mind? Do the doses of pain medications still matter? In our experience, sick patients came to the hospital for care. Those patients whose illness was not treatable went to hospice, or sometimes died on the unit. But they died because there were no other options. And usually they were older than Lorraine. They didn't die when there were successful treatment options available.

As a psychiatric nurse, Jennie struggled along with the staff through this crossroad in Lorraine's care. The chronicity of her dysphoria and wish to die were not acute, but were an underlying factor. No, she was not "actively" suicidal, did not have a plan or imminent intent, but she had expressed a longstanding wish to die and had at least one past suicide attempt. Were we aiding in her suicide? The legal response seemed clear after many psychiatric consultations and multiple team discussions — Lorraine has the right to refuse treatment even if her refusal results in her death — but the ethical one did not. Besides respecting Lorraine's right to decide, what constituted beneficent, faithful, and fair caring for Lorraine?

Then she didn't die, not for a very long time. We all waited for infection to overtake her, then stood back in amazement as Lorraine's own body fought her choice. Her hopelessness was clear when she said, "I can't even do this right." We had thought that she would quickly become septic and die. Now what? This added to the ambivalence and distress we all felt. When she periodically decided to have labs drawn, and at one point requested a blood transfusion, this added to the angst. Was she trying to slow the process down? Did she not want to die? As we approached the winter, she wanted to go shopping for a new winter coat. That was definitely a mixed message. Our big question was: is this the "normal" ambivalence that anyone would experience while dying, or was this Lorraine's way of doing this, or was she trying to tell us she didn't want to die? We continued to be uncomfortable, and we would meet to talk and to think about what exactly we should do on any given day.

The scope of Lorraine's care would have to be broadened, we thought, beyond what we usually provide in an acute-care setting. First, we tried to accept her decision to die, and to accept that she would not be leaving the hospital until she had died. We needed to transition from a position of vigorously trying to help Lorraine accept care and recover, to one where we would now see her through dying to her death. We had to help the staff take a more hospice-like approach — which meant a substantive change in values — to help Lorraine have a dignified and comfortable end to her life.

Nurses banded together to share the responsibility of caring consistently for Lorraine. This woman who had difficulty making positive relationships now found herself surrounded by individuals willing to care for her. A transformation occurred. For the first time, we all, Lorraine included, agreed on a plan of care. This was a bit of a turning point.

Most of the struggle was gone. We could focus on providing Lorraine with what she needed without arguing about it. Several nurses took the time to search for the inroad, to find common ground. Interactions became more relaxed. Staff found they could "chat" with Lorraine about music, or about her children, or clothes. They could let her talk about her life. While it was still critical for staff to support and care for those who were doing the difficult work of caring for Lorraine, this new goal enabled staff to rally, to re-engage, and to help Lorraine in a way she experienced as caring at least some of the time. In this way the nurses were able to build relationships with a patient who was skilled at chasing people away.

The one point we continued to have some difficulty around was Lorraine's dressing changes. She often refused the changes or wanted to put them off. The dressings became saturated and foul smelling. So a compromise was reached: nurses figured out a way to change the dressing as quickly as possible, with a minimal amount of turning. So it essentially became a win-win situation.

Courtney said, "I got a picture of what her life had been like, anguish, heartache, and disappointment. She had this yearning in her eyes for someone to really like her. But she was really scared to have someone like her. Some days I would go home and rip my hair out, but I was finally able to develop a trust with her." Courtney repeatedly questioned her own caring. "It's my job to take care of you, and I get paid to come and take care of you. It's my job to care. How much do I care as a person, and how much do I care because I'm here? It was confusing to me. But I knew I cared about how I affected her, especially when I knew she was benefiting."

Molly said, "I opened my eyes to the fact that she wasn't just a patient, she was a person. I would almost hate to care for her some days, but she let me see past all that. I thought, you just had a really, really bad life handed to you. She let me in. Then we found the happy medium of what she could tolerate and what she was requiring. Each day when I did her dressing was when we connected the most. We would turn on the stereo, put her favorite CD on, and we would either talk or sing. On the days when she was too sick or too tired, she would let me put on my favorite radio station and I would sing by myself."

Courtney was at her side when she died. "The day into the night that she died, it was very calm and peaceful, the opposite of her life. Several members of the nursing staff stood by her bed through her last moments, holding her hand, telling her who was there, that they were there for her — she deserved a little peace in her life. That was all she wanted."

Was My Patient Fortunate or Forsaken?

Robin Dauterive

As I teach the first year medical students in the course, "Patient-Doctor Interviewing, I am often asked, "How do you care for patients who do not care for themselves?" One particular patient comes to mind when I respond to the students.

A couple years ago, I became involved in the care of a woman who challenged the ideas and ideals of her healthcare team each day. Lorraine was a paraplegic woman who had extensive decubitus ulcers and was admitted to Massachusetts General Hospital (MGH) to improve care of these wounds. Apparently, the visiting nurses would no longer agree to provide home care because she had been difficult to manage and noncompliant with her treatment. Her boyfriend and family were unable to provide the aggressive care she needed.

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First, she had been refusing to turn in bed to relieve pressure from her wounds, and she had refused to quit smoking. The plastic surgery service said that they would be unable to provide potentially curative therapy involving debridement and flap placement because these treatments depend upon a patient's cooperation and compliance. As a physician, I knew the wounds were severe. Any treatment options were heroic and would likely fail, even with a cooperative patient. The most reasonable expected medical outcome would be a prolongation of the patient's life for a few years. I was certain that her difficulty complying with medical treatment would worsen her medical prognosis.

I believe that noncompliance with medical treatment is complex and stems from deeply rooted instinctual desires and needs that are in conflict with one another. These behaviors are deeply seated and guided by feelings such as loss of control of one's environment, denial of and rebellion toward illness, mistrust of others, and inability to com-

prehend the long-term effects of refusing treatment. Lorraine's behavior revealed that she was a master at finding ways to feel "in control."

Lorraine was physically and emotionally destitute. She tried to fill her insatiable needs by manipulating the people around her. Unfortunately, such lifelong, ingrained internal depravity cannot be erased by the late arrival of valiant effort. It was from this point in Lorraine's life that our healthcare team tried to "pick up the pieces."

Lorraine was immature and fought each day to get what she wanted without regard for the consequences. Most of the time, she wanted more pain medication or to go outside and smoke. In the opinion of the healthcare team, both of these requests were contraindicated in any plan to improve her medical condition. The questions the team asked themselves were: How much pain medication is too much? When the nursing staff is uncomfortable with the care they are being asked to provide, how do you go about deciding which care is appropriate? What may healthcare workers justifiably refuse to do? Should a patient like Lorraine, who exhibits self-destructive behavior, make her own choices, and should we accept them? I am not sure we found answers to all of these questions, but we did learn to work together and to support each other.

Lorraine exhibited such masterful and cunning drug-seeking behavior, I knew she must have spent a lifetime perfecting her approach. When we decided not to increase her pain medication any further, she decided otherwise. She would wait until late at night and insist on seeing the covering nighttime physician about her pain. Initially, this approach was successful, until the physicians caught on and signed out to each other that she was not to receive any additional pain medication overnight. Her next ploy was to say she had vomited up her meds (often at night) so she could receive an extra dose. After a while we detected this scheme and stopped giving the extra pain medication dose. Finally, she was found with her pain pills broken down in a spoon. She was confronted, and her pain medications were rotated and changed to liquid form. We required that she be observed when she ingested each dose of pain medication. I can imagine that this regimented program provided daily discomfort for the nursing staff, whose main goal is to advocate for and to build a trusting relationship with patients.

When I think about patient advocacy and the physician's role in a patient's care, I again reflect on the sessions with my medical students. During the last year, we had a very intense but productive session about taking care of patients whom you do not like or whose moral values you do not respect. Some students didn't think they could care for such patients objectively, while other students thought that to be an enlightened physician meant transcending such conflicts and being able to care for all patients equally, irrespective of their core values.

We talked about aligning our goals with that of the patient's. I shared my "litmus test" with them. If I can sleep at night, I have done my best. If I toss and turn, I probably still need to work things out with my patient the next day. I did lose a few nights of sleep when caring for Lorraine. Was I her advocate? Did I do my best in looking beyond the actions to see Lorraine for who she was and what she needed? The engraved words of Dr. L.J. Henderson on the classroom wall continually advise me: "When you talk to a patient you should listen, first, for what he wants to tell you, secondly, for what he does not want to tell, thirdly, for what he cannot tell." Was I giving Lorraine the benefit of the doubt? A chance to rise above? I know that changing the mind of an adult is not only difficult, but, at times, impossible. You must use what you learn from listening to adult patients to correct and challenge misunderstandings. If you see any slight change, continuing to build the relationship is key. If thoughts and behaviors

persist, then it is better to move to the next issue. With Lorraine, I searched for ways to sort out the truth from the dysfunction.

I tried to develop a relationship with her. She was conversant at times. We liked some of the same music, old classics. We spoke of her music frequently as it played in the background on her radio. She responded to my questions with compliance or complaints. Not a day went by that she did not complain of pain. I searched for ways to provide a holistic treatment plan and to move Lorraine forward, including adequate nutrition, daily activity, and prevention of iatrogenic complications. Various consultants were addressing many of these issues, but I carefully reviewed each aspect to make sure nothing was missed. With the help of the nurse practitioner, Grace, I had plastic surgery evaluate and debride her wounds again. In addition, psychiatrists and social workers were involved to help Lorraine deal with her illness.

I listened to see if there was something she could not say. Solutions never came, just more questions. Why did I become a doctor? When faced with a challenging patient like Lorraine, the answer is not always clear. Idealistically, the doctor is a healer, compassionate and empathetic. Was it compassionate to give her pain medication because she complained of pain, or should I "do no harm" and withhold pain medication from an obviously sedated patient? Was it empathetic to listen to her complaints each day and sit back and do nothing, or should I challenge her with my words to expect more from herself and to coerce her to do more than she was willing to do? Through time, one consistent principle became clear to me. Lorraine was set on a path of self-destruction from which there was no return. Although at times she did allow some care, ultimately she was unable, for whatever reason, to hold to a course that might allow her wounds to heal.

Was Lorraine impaired by the large amount of narcotics she was taking? Should treatment be forced on people for their own benefit? Lorraine was actually coherent on an unbelievably large amount of opiate narcotics, and she still complained of pain. In my limited experience, such patients become super-sensitized to pain. With more medication, they seem to have more pain instead of less pain. The idea of narcotic rotation was often presented to Lorraine. Like any other addicted patient, her psychological and physical dependence on these medications would not allow her to agree to a rotation of these medications, medications on which she had come to rely that were now failing to address her problem. I still wonder if we had forced her to rotate her narcotics earlier whether it would have made any difference. I am beginning to believe that forced narcotic rotation may be some patients' only chance at reversing their self-destructive course. This type of treatment plan would entail a forced change of medication against the patient's wishes. When is it reasonable to force such things on a patient? Clearly, if a patient is in imminent danger and lacks insight, the decision is clear. However, when the outcome of treatment is less certain, and the patient has the apparent capacity to make decisions, it is less clear.

As Lorraine continued to refuse care, psychological evaluations were done to see if she had the cognitive capability to make decisions and to determine if she was mentally impaired by depression or suicidal ideation. Each evaluation was the same. She was medically and legally capable of making her own decisions. Thus, we had to respect her refusal of treatments and procedures that we knew could make her better, and allow her to set a course that would lead to her demise. Ultimately, she chose to move toward that fate by changing her code status to "do not resuscitate."

Her refusal of treatment and her decision not to be resuscitated was difficult for her to handle, and appeared almost equally difficult for the nursing staff and healthcare team. This difficulty was not only because of Lorraine's young age, but also because of the unspoken idea that "this could have been prevented."

As a physician, I often feel the weight of the outcome on my shoulders. I am very familiar with the anguish that coincides with perpetually questioning your motivations and

actions regarding a complex patient's well-being and care. The feeling of failure, or the thought, "if only I had done more, or tried harder," can be overwhelming. In situations like caring for Lorraine, these feelings are bound to distress caregivers because of the "love-hate" factor that is inevitably added to relationships with such multifaceted patients. Lorraine had made such poor choices that no healthcare team member could go away from the situation without a smoldering scar.

I repeatedly assured myself that we had done everything possible. I made a decision to dedicate much of my time to supporting the healthcare team who had gathered around this woman. My hope was that I could lighten their burden of responsibility by clearly validating the decisions they made and by consistently reassuring staff that they were doing everything possible.

The time came when I would have to "muster up" the strength to go see Lorraine. Her persistent nagging complaints were almost too much to bear when I knew each of her many needs were being tended to daily by several people. With every visit, I knew it would be a struggle to "enforce the rules." I would set a plan before entering the room to be less swayed by her manipulative behavior.

In the end, I knew I could have another physician who knew Lorraine take over her care, which I did. I felt a sense of despair about Lorraine's decisions and the way that she had isolated the staff who had worked so hard to care for her. I think these feelings stemmed from her unending request for pain medication. I view addiction as separate from the person, and despise where the cycle of addiction brings people. With Lorraine, I felt that her addiction played a large role in the choices she made. As with many addicted patients, there is a point of no return, when addiction irrevocably claims its victim. My recusal from her case was based on the loathsome feeling I have toward the terminal cycle of addiction. I was not upset with Lorraine, but I could no longer deny the strength of her disease.

Lorraine often told me that each day she would make herself get out of bed. This made her feel like she was moving forward. I knew, when she stopped getting out of bed, she had given up hope. The fight was over. By that time, her medical prognosis had become quite grim. Questions continued to arise. How do you assess a patient's quality of life and ensure the best quality possible? When do you make demands? When do you give way? The simple answer is that there is no simple answer. Each question on each day requires evaluation by a team of people who know the details.

When I went to the floor to see Lorraine, I always saw a group of individuals from different backgrounds and of different ages providing compassionate and thoughtful care. Not a day went by that her team did not agonize about each decision, each dressing change, and each pain medication she received.

I remember a particular day that Lorraine was feeling better than usual. She had gone to the hospital salon and had had her hair done and her eyebrows waxed. She was sitting by the nurses' station in her wheelchair "chatting it up." She was talking to the nurses and making jokes. Her enjoyment when visiting with the staff was obvious and tangible. Their relationships had become meaningful. That day, it became very clear to me that Lorraine was receiving the best care that she would allow. She was maintaining the best quality of life possible for her at that moment.

I believe that indifference and neglect result in the worst quality of life and the worst care of patients. In the last months of Lorraine's life and throughout her prolonged hospital stay, she was never treated with indifference or neglect. I don't think that there is any way to figure out why Lorraine made the choices she made. What I do know is that Lorraine received commendable care during her stay, and that her caregivers made sure that her dignity remained intact each day until her last.

Talking with Lorraine's Mother and Sister, Five Months after Her Death

Ellen M. Robinson, Grace Good, and Suzanne Burke

Georgia, Lorraine's mother, and Denise, her sister, shared their experiences as Lorraine's closest family members in an interview five months after her death. We sought to learn about what it was like for them to live through Lorraine's final hospitalization, as well as their reflections on Lorraine's life.

Georgia shared the goodness in Lorraine, describing how her daughter had cared about others. As a teenager, Lorraine had given multiple hours of volunteer service to the patients at Tewksbury Hospital. She loved to hold yard sales to raise money for this and other causes. She was filled with goodness as a child.

Georgia then shared some of the pains that Lorraine experienced during her life. She and Denise expressed regret over a "life lost," a "life that could have been different."

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Lorraine's father moved to California after her parents divorced, and he remarried. Lorraine, at the age of 14, wanted to spend a summer with him, and made plans to do so. But as the time for Lorraine to travel to California to visit with her father grew closer, he rejected these plans, and she learned she was not welcome to come and stay. Georgia believes that this news took an emotional toll. It seemed that, from that point forward, Lorraine was "off and running."

Georgia worried terribly about Lorraine throughout her teens and early twenties, and described how, after Lorraine had been involved with drugs for some time, she came home, so sick, and simply lay in bed. Georgia's husband wanted to call an ambulance, and initially Georgia resisted, saying, "No, at least she'll die in a clean bed." Upon hospitalization, Lorraine was found to have bacterial endocarditis.

Both Georgia and Denise loved Lorraine, and, through all of her difficult behavior and the walls that she had built around herself, they remembered her goodness and childhood smile.

Both spoke of their experiences and stresses in being Lorraine's caregivers at her

home. They spoke of how Lorraine valued being on her own. Denise said, "Home was very important to Lorraine. She used to like to get up in the morning and cook, and she liked to go shopping, and she was very independent." Prior to her final admission to the hospital, Lorraine had started to lose home medical services. This was very stressful for Georgia and Denise, as they worried about her, and knew that her behavior played at least a partial role in the loss of these services. Even then, when Lorraine was admitted to the hospital for what turned out to be her final admission, they did not believe it would be any more than one of her routine hospital admissions. Georgia remembered, "She was losing services, the way everyone was losing services across the board. Going to the hospital was something that we had been used to; it was kind of a cycle, going in for treatment and coming back. Lorraine thought that perhaps she could get some increased services" through being admitted to the hospital.

Denise went on to say, "So we did not see her admission as anything unusual. It was just the visiting nurses having her come in for one week. We didn't realize she was as sick as she was." She continued, "When Lorraine was in the hospital, I was able to change roles and just be her sister. Because she had all the caregivers that she required, I could just be her sister and just care about her. That was very important to me."

There were a few times during her last hospitalization that Lorraine had wanted to go home, and actually demanded to be allowed to do so. Denise recalled, "She had this big thing about home. Like, I ended up coming at 4 a.m. one morning because she was on the phone and saying, 'I want to go home,' and being a real pain the in the neck. And I said, 'I talked to the hospital, and we can have you home in the morning.' She turned a corner when it became real, and she didn't want to go. She said, 'No, I don't want that,' and she fell asleep, and that was a good thing. I told her that she was being a pain the ass — excuse me — but she was. She just needed somebody to say yes or no, be a little bit more real with her. So when it became real, she didn't want to go. She knew she was safe here."

Hospitalization was difficult for Lorraine because it placed limits on her behavior. Georgia said, "She had a lot of self-interest. She didn't really think about how it impacted you. She thought about herself and how it impacted her. She was a social monster. She needed to get downstairs. She needed to smoke a cigarette. I remember the first time when she was restricted to bed. She took it so hard. It's like being grounded. She was not listening to anything. It's one thing to refuse medical advice, but when she wouldn't take any advice and wouldn't listen to anything. . . ."

When discharge from Lorraine's final hospitalization was being considered, Georgia and Denise expressed their fears about how they might not be able to compensate in providing her home care, as services were increasingly being denied, due to Lorraine's past noncompliance and behavioral issues. A plan to discharge Lorraine to home was then abandoned by her professional caregivers, in part because of the toll it would take on her mother and sister.

Denise said, "She stopped saying that she wanted to go home." Georgia added, "She had a difficult time. When she found out that they couldn't fix her hip and that things had gone too far, handling dressings and that type of thing, it was sort of like chemotherapy when you're too far gone. She said, 'There's nothing else they can do for me.' "

Denise believed that Lorraine had confided in her about limiting life-sustaining treatment, and Denise kept this to herself so as not to hurt Georgia, who at that time could not conceive of giving up. As time went on, Georgia came to realize, with the help of the nurse practitioner, and from what she was observing, that Lorraine was not going to get better. Georgia noted, "She lost all the muscle tone in her leg, and it began to atrophy. Things like that. On her last day, if I had known she would have gone that night, I would [have said

to myself], 'Don't go home now, stay a couple more hours.' [To be there when Lorraine died.] She smiled at me, and I told her, 'It's okay, Lorraine, you can leave.' She was like a little girl at that point, with a peaceful, sweet, angelic smile."

Georgia said that she wishes she had not left that night, and wishes that someone had said, " 'Stay with her, she is going to die.' " But Georgia also said she knows that this is not possible even for professionals to predict. She repeated, "I will always remember her smile, like a little girl's smile, she looked so peaceful."

Denise, who had come in after Lorraine had died, expressed it this way: "On the day that Lorraine died, she seemed to be so peaceful, like they had the symphony orchestra there the day that she passed away. She was surrounded by caring people. It was perfect, even the gravestone." Denise shared that she was so impressed by the post mortem care: "Lorraine looked so peaceful, clean, and warm, with her purple monkey."

Their comments in the interview are summarized by their description of how Lorraine was cared for during her final extended hospital stay. They believe that Lorraine felt the compassion of physicians, nurses, and therapists — and her family felt it as well. In a final comment, Georgia said, supported by Denise, "The last six months of her life [while she was hospitalized] were one of the best times for her in her life — she felt safe and cared about."